An exploration of illness perceptions among young people who have a parent with cancer

by

Chloe Fletcher

Thesis
Submitted to Flinders University
for the degree of

Doctor of Philosophy
College of Medicine and Public Health
December 2020

Contents

List of Tables

List of Figures

List of Boxes

List of Abbreviations

Abbreviation	**Full definition**
ACT	Acceptance and Commitment Therapy
APA	American Psychological Association
AUC	Area Under the Curve
AYA	Adolescent and Young Adult
CBT	Cognitive Behaviour Therapy
CRIES	Children’s Revised Impact of Events Scale
CINAHL	Cumulative Index to Nursing and Allied Health Literature
COSMIN	COnsensus-based Standards for the selection of health Measurement INstruments
COPE	Coping Orientation to Problems Experienced scale
CSM	Common-Sense Model
EM	Expectation Maximisation
GRADE	Grading of Recommendations, Assessment, Development and Evaluation
ICC	Intraclass Correlation
ICQ	Illness Cognition Questionnaire
ICS	Illness Cognitions Scale
IES	Impact of Events Scale
IES-R	Impact of Events Scale-Revised
IMIQ	Implicit Models of Illness Questionnaire
IPQ	Illness Perception Questionnaire
IPQ-R	Illness Perception Questionnaire-Revised
KMO	Kaiser-Meyer-Olkin test

KIDCOPE	Child version of Coping Orientation to Problems Experienced scale
LOA	Limits of Agreement
MCAR	Missing Completely at Random
MIC	Minimal Important Change
NPV	Negative Predictive Value
OCNI	Offspring Cancer Needs Instrument
PTGI-SF	Post-Traumatic Growth Inventory-Short form
PPIQ	Perceptions of Parental Illness Questionnaire
PPIQ-C	Perceptions of Parental Illness Questionnaire-Cancer
PPV	Positive Predictive Value
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROSPERO	International Prospective Register of Systematic Reviews
PTGI-C-R	Post-Traumatic Growth Inventory for Children-Revised
PTGI-SF	Post-Traumatic Growth Inventory-Short form
ROC	Receiver Operating Characteristic
RR	Responsiveness Ratio
SDC	Smallest Detectable Change
TMSC	Transactional Model of Stress and Coping

List of Publications

Fletcher, C., Flight, I., Gunn, K., Patterson, P., & Wilson, C. (2020). Comparative systematic review of the psychometric properties of measures of illness perceptions in family members of individuals diagnosed with a chronic physical illness. *Scandinavian Journal of Caring Sciences*, Advance online publication. doi: 10.1111/scs.12822

Fletcher, C., Wilson, C., Flight, I., Gunn, K., & Patterson, P. (2019). Illness cognitions among adolescents and young adults who have a parent with cancer: A qualitative exploration using the Common-Sense Model of Self-Regulation as a framework. *International Journal of Behavioral Medicine*, *26*(5), 531-541. doi:10.1007/s12529-019-09793-4

Fletcher, C., Wilson, C., Flight, I., Gunn, K., Patterson, P. (2019, November). The development and initial validation of the Perceptions of Parental Illness Questionnaire-Cancer. Paper presented at the 5th Victorian Psycho-oncology Conference, Melbourne, Australia.

Fletcher, C., Wilson, C., Flight, I., Gunn, K., Patterson, P. (2019, June). The Perceptions of Parental Illness Questionnaire-Cancer: An instrument to assess illness perceptions among young people who have a parent with cancer. Paper presented at the MASCC/ISOO Annual Meeting on Supportive Care in Cancer, San Francisco, USA.

Fletcher, C., Patterson, P., Wilson, C. Flight, I., Gunn, K. (2018, November). Illness perceptions in young people who have a parent with cancer: A qualitative exploration. Paper presented at the 20th World Congress of Psycho-Oncology, Hong Kong.

Fletcher, C., Wilson, C., Flight, I., Gunn, K., Patterson, P. (2017, August). An exploration of cancer-related beliefs and perceptions in adolescents and young adults who have a parent with cancer. Paper presented at the 19th World Congress of Psycho-Oncology, Berlin, Germany.

Abstract

This thesis describes the validation of an instrument to assess illness perceptions among adolescents and young adults (AYAs) following a parent's cancer diagnosis. Five studies were completed. Study 1 was a comparative systematic review of the psychometric properties of instruments designed to measure illness perceptions in family members of those with chronic physical illness. This study highlighted that better reporting of psychometric information was needed and found that measurement of illness perceptions in children of parents with illness had been largely overlooked.

Only one of the instruments included in the review had been validated for use in adolescents (the Perceptions of Parental Illness Questionnaire; PPIQ). The PPIQ was developed to assess the dimensions of illness perception described in the Common-Sense Model of Self-Regulation (CSM) among adolescents with a parent diagnosed with multiple sclerosis. This finding informed the direction of the research that followed, the aim of which was to adapt and validate a cancer-specific version of the PPIQ.

A qualitative study (Study 2) was conducted to determine whether the CSM adequately explained AYAs' perceptions of their parent's cancer. Deductive thematic analysis of data from eleven semi-structured interviews confirmed that perceptions corresponded with the dimensions of the CSM. Findings guided the adaptation of the PPIQ to produce a cancer-specific version (the Perceptions of Parental Illness Questionnaire-Cancer; PPIQ-C).

The content validity of the PPIQ-C was examined using think-aloud and verbal probing techniques in Study 3. Cognitive interviews were conducted with four young people. Based on findings, the questionnaire was modified to improve the relevance, comprehensiveness, and comprehension of items.

In Study 4, cross-sectional data from 437 AYAs was used to evaluate the psychometric properties of the PPIQ-C. The dimensional structure of the PPIQ-C was determined through exploratory factor analysis. Test-retest reliability, internal consistency, and construct validity were assessed and deemed adequate. The final PPIQ-C consisted of 67 items across 11 core dimensions (*identity*, *emotional representations*, *coherence*, *consequences for personal development*, *consequences for family relationships*, *consequences for daily activities*, *personal control*, *treatment control*, *recovery*, *unpredictability*, and *chronicity*) and three causal dimensions (*behavioural or environmental attributions*, *chance or luck attributions*, *emotional or psychological attributions*).

Using data collected in Study 4, the final study investigated the screening utility of the PPIQ-C (Study 5). Cluster analysis revealed greater psychological morbidity among young people who scored higher on *identity*, *emotional representations*, *consequences for personal development*, *consequences for family relationships*, *consequences for daily activities*, *unpredictability*, *chronicity*, *behavioural or environmental attributions*, and *emotional or psychological attributions* subscales. Scores on these subscales were summed to produce a Total Negative Beliefs score. Optimal cut-off scores for identifying AYAs at risk of psychological distress (137.5) and post-traumatic stress (135.5 for young adults and 140.5 for adolescents) were determined using receiver operating characteristic curves. These findings were discussed in terms of their implications for clinical practice and service provision.

This research provides novel insight into how illness perceptions contribute to psychological morbidity in young people following their parent's cancer diagnosis. The PPIQ-C has important potential for application in both research and clinical settings.

Declaration

I certify that this thesis:

1. does not incorporate without acknowledgment any material previously submitted for a degree or diploma in any university; and

2. to the best of my knowledge and belief, does not contain any material previously published or written by another person except where due reference is made in the text.

Chloe Fletcher

17 December 2020

Acknowledgements

There are a number of people without whom this thesis would not have made it to submission. The four years over which this research was conducted have been incredibly challenging but have also been a period of enormous growth. I have many people to whom I am forever indebted.

Most importantly, thank you to the young people who participated in this research. Thank you for your vulnerability in sharing your experiences with me and for trusting me to handle your stories with care.

To my supervisors – Carlene Wilson, Ingrid Flight, Kate Gunn, and Pandora Patterson – your support and encouragement throughout this process has been invaluable. Thank you for the knowledge that you have contributed, the time and care that you put into providing feedback for me, for believing in me, and for pushing me to reach the potential that you could see in me. Carlene, I would like to especially thank you for taking a chance on me – a biological sciences student – and supporting my transition into behavioural sciences. Your support and guidance have opened many doors for me, and I am truly grateful.

Thank you also to Pandora and the team at Canteen – in particular Fiona McDonald, Bianca Ton, Stephanie Konings, and Adam Wright – for supporting this research, for the work you put into getting young people involved in this research, and for hosting me at the Sydney Division offices while conducting interviews. It has been a pleasure and an absolute privilege to work in this space alongside you.

To my PhD buddies – Donna Hughes-Barton and Jen Fish – thank you for your support, encouragement, and solidarity throughout the PhD journey. I am so excited to finally be joining both of you on the other side!

Special thanks to my little (only in stature) sister, Paige, for contributing as a second coder for the qualitative study. Thank you also for the PhD chats – know that I am always available and ready to listen and empathise as needed throughout the rest of your PhD journey.

Thank you to my parents for cultivating a supportive and loving family environment that always encouraged critical thought and passionate debate. Thank you to my family – Mum, Dad, Paige, Cameron, Shane, and Anne – for providing much laughter and countless thought-provoking conversations over the past few years. Special mention to Shane for being my on-call IT support.

An enormous thank you to my beautiful, warm-hearted, and thoughtful partner, James. Thank you for giving me the quiet space to write and for providing nutritional support in the form of home-cooked meals and emotional support in the form of cuddles and trash TV. Your support means the world to me.

Thank you to my beautiful friends and work colleagues for checking in with me (even if with apprehension) and for cheering me on along the way. Your support has been so wonderful, and I promise that you will never have to hear me talk about my PhD ever again.

Finally, I would like to acknowledge that this research was supported by the Research Training Program Fee Offset provided by the Australian government and a Flinders University Research Scholarship. Education should be a right for all, and not just a privilege given to those who can afford it. I am thankful for the financial support that facilitated my completion of this research and participation in travel opportunities that presented themselves along the way.

Dedications

For the young people who feel forgotten or overlooked by the family and medical professionals after their parent has been diagnosed with cancer.

For my mum, whose experience with cancer inspired this research, and whose strength and determination inspires me daily.

CHAPTER ONE
Introduction

1.0. Preamble

Despite substantial progress in the prevention, detection, and treatment of cancer, which has led to increased life expectancy and greater chance of survival, cancer is still the second leading cause of death globally (Ferlay et al., 2019; World Health Organization, 2020). Current estimates suggest that one in two will receive a cancer diagnosis in their lifetime (Australian Institute of Health and Welfare, 2019; Howlader et al., 2019; Maddams, Utley, & Moller, 2012). In 2018, an estimated 18.1 million new cases of cancer were diagnosed and 9.6 million deaths were attributed to cancer worldwide (Ferlay et al., 2019; World Health Organization, 2020). It is predicted that by 2040 these figures will have doubled (World Health Organization, 2020). Cancer is a leading cause of illness and death in Australia (Australian Institute of Health and Welfare, 2020); in 2016, 33% of deaths from noncommunicable diseases were related to cancer (in contrast with 30% worldwide; Wild, Weiderpass, & Stewart, 2020; World Health Organization, 2018). This year, it is estimated that just under 150,000 Australians will be diagnosed with a new case of cancer and 48,000 will die from cancer (Australian Institute of Health and Welfare, 2020).

Behind each of these figures is a person dealing with the uncertainty and distress that accompanies the cancer diagnosis. For many, a cancer diagnosis is associated with a fear of intense pain, loss of control, stigmatisation, and the threat of death (Singer, 2014). People who are diagnosed with cancer may experience a range of physical, practical, and psychosocial consequences, including changes in their intimate relationships (Hawkins et al., 2009; Stenhammar, Isaksson, Granstrom, Laurell, & Tiblom Ehrsson, 2017), disruptions to their education or employment (Mehnert, 2011; Sodergren et al., 2018; Timmons, Gooberman-Hill, & Sharp, 2013), changes to their physical appearance (Helms, O'Hea, &

Corso, 2008; Nozawa et al., 2013), and spiritual and existential concerns (Henoch & Danielson, 2008; Visser, B., & Vingerhoets, 2009), which may reduce treatment adherence, slow recovery, and contribute to poor physical and mental health outcomes.

It is important to recognise that the psychosocial impacts of cancer extend beyond the person diagnosed, to their family members, friends, and the wider community. For dependent children, it can be especially distressing to learn that their parent has been diagnosed with cancer and to watch them face the many challenges and changes that cancer and its treatment bring (Krattenmacher et al., 2012; Morris, Martini, & Preen, 2016; Walczak, McDonald, Patterson, Dobinson, & Allison, 2018). Older children may find it more difficult to deal with their parent's diagnosis because they are able to better understand the serious implications of cancer for themselves and their family both in the short and longer term (Gazendam-Donofrio et al., 2011; McDonald et al., 2016; Pederson & Revenson, 2005; Weeks, McDonald, Patterson, Konings, & Coad, 2019). Adolescents and young adults (AYAs) themselves experience psychosocial consequences following a parent's cancer diagnosis, with these contributing to high levels of psychological distress and unmet psychosocial needs (McDonald et al., 2016; Patterson, McDonald, White, Walczak, & Butow, 2017; Walczak et al., 2018). In Australia, an estimated 21,000 young people aged 12-25 years have a parent diagnosed with cancer each year and 60% of them report clinically elevated levels of distress (Patterson, McDonald, & Orchard, 2014; Patterson et al., 2017).

Research has examined factors that may impact children's psychological outcomes following their parent's cancer diagnosis. These include, but are not limited to, the child's gender and age, the parent's gender, and variables intrinsic to the parent's illness (e.g., type or stage of cancer) (Krattenmacher et al., 2012; Morris et al., 2016; Osborn, 2007; Visser, Huizinga, van der Graaf, Hoekstra, & Hoekstra-Weebers, 2004). However, much of this research has produced inconsistent findings. Although chronological age is often used as an indicator of cognitive maturity in research examining the impact of parental cancer on children, it is important to note that a child's level of cognitive maturity does not necessarily correspond with their chronological age. Inconsistent findings among research examining the role of age as a predictor of psychological outcomes may be explained by differences in cognitive maturity among children of the same chronological age (Su & Ryan-Wenger, 2007). Furthermore, children's understanding of illness appears to be associated with their level of cognitive maturity (Carson, Gravley, & Council, 1992). Combined, these findings highlight the need to examine the associations between children's psychological adjustment and their conceptualisation of their parent's cancer, rather than their chronological age alone.

A large body of research demonstrates that the way that people think about an illness – their illness perceptions – impacts upon their ability to cope with the illness and, in turn, affects their physical and psychological outcomes (e.g., Hagger, Koch, Chatzisarantis, & Orbell, 2017; Hagger & Orbell, 2003; Leventhal, Brissette, & Leventhal, 2003; Leventhal, Phillips, & Burns, 2016a). Some research suggests that illness perceptions may be an important predictor of children's coping and adjustment following their parent's cancer diagnosis (Compas et al., 1994; Compas, Worsham, Ey, & Howell, 1996). However, knowledge of the specific relationships between illness perceptions, coping, and psychological adjustment in children who have a parent with cancer remains limited, and there are currently no validated instruments to assess illness perceptions in this cohort. Therefore, the purpose of the research outlined in this thesis is to address these gaps in the research through the validation of an instrument that could be used to assess illness perceptions in AYAs who have a parent with cancer and identify those at risk of psychological morbidity.

The first chapter will provide an overview of research examining the psychosocial impact of parental cancer on the parent and dependent children; factors associated with children’s adjustment to parental cancer; theoretical models describing the relationship between illness perceptions and physical and psychological outcomes; and existing instruments used to measure illness perceptions. At the end of the chapter, a summary will be provided, followed by the thesis aims and rationale for the research.

1.1. An Overview of Parental Cancer

Although it is difficult to establish the overall incidence of parental cancer, it is estimated that as many as 20% of individuals diagnosed with cancer have children under the age of 18 years (O'Neill, O'Neill, & Semple, 2020). In Australia, roughly 1 in 5 of those newly diagnosed with cancer are aged 20-54 years (Australian Institute of Health and Welfare, 2020), an age range that encompasses typical childbearing and parenting years (Weaver, Rowland, Alfano, & McNeel, 2010). Improvements in survival rates mean that families impacted by parental cancer are now increasingly living with the longer-term consequences of the illness, its treatment, and the threat of recurrence (Osborn, 2007).

For many adults, being a parent is central to their identity (Moore, Rauch, Baer, Pirl, & Muriel, 2015). Cancer and its treatment pose a significant threat to this identity. Parents who are diagnosed with cancer experience a multitude of fears and worries as they attempt to simultaneously manage their role as a parent and cope with the illness experience (O'Neill et al., 2020). More broadly, family life may be disrupted by changes to routines and household roles, as well as a possible decline in income (Osborn, 2007). Cancer forces the parent into the position of care-receiver, although their greatest concerns usually revolve around the need to function as a provider and caregiver for their family (Hasson-Ohayon & Braun, 2011). Parents are faced with the decision of how to best tell their children about their diagnosis, as well as fears over the possibility of not seeing their children grow up (Inhestern et al., 2016; Konings, McDonald, & Patterson, 2020; Semple & McCance, 2010a).

Once treatment begins, parents may experience changes in their appearance, mobility, memory, mood, and energy levels that can impact their everyday functioning and ability to care for dependent children (Moore et al., 2015). Parents may struggle to balance their own needs with those of their children, often prioritising their children's needs over their own self-care (Rashi, Wittman, Tsimicalis, & Loiselle, 2015). They may feel pressure to be "good" parents; "staying strong" for their children, spending time with them, protecting them from the realities of their illness, and minimising disruptions to family life (Hasson-Ohayon & Braun, 2011; Rashi et al., 2015; Schiena, Hocking, Joubert, Wiseman, & Blaschke, 2019). However, the physical and emotional burden of cancer may limit parents' capacity to respond to the needs of their children (Morris et al., 2016). This can cause parents to question whether they are sufficiently fulfilling their role as a parent and struggle with feelings of guilt and inadequacy (Krauel et al., 2012). These pressures, combined with the stresses associated with coming to terms with a cancer diagnosis as they navigate the healthcare system and undergo treatment, may affect parent's interactions with their children.

Additionally, parents report difficulties in knowing how to communicate appropriately with their children throughout the illness trajectory (Konings et al., 2020; O'Neill et al., 2020; Semple & McCance, 2010b); many feel that they do not have the skills required to talk to their children about their cancer and express a lack of confidence in having these conversations (Konings et al., 2020; Semple & McCaughan, 2013). They may also find their children's questions and concerns confronting or challenging and struggle with uncertainty about whether to share their feelings, especially with younger children (Krauel et al., 2012).

The challenges that parents face following a cancer diagnosis can impact their psychological well-being; significant stress may contribute to them feeling overwhelmed, depressed, and anxious (Fernandes, Muller, & Rodin, 2012; E. M. Park et al., 2016).

Research exploring how parenthood contributes to the psychosocial impact of cancer suggests that parents with children younger than 18 years of age experience higher levels of psychological distress, and that stress is especially elevated when children are older than 6 years (Ernst et al., 2013). In addition, gender-specific differences appear to impact psychological well-being following a cancer diagnosis. Women may be more likely to experience adverse psychological outcomes than men, with one study reporting that the prevalence of depression was 35% among mothers with cancer and 28% among fathers (Schmitt et al., 2008). Another study found that although women reported greater anxiety overall, men who had children were significantly more impacted by anxiety than men without children (Ernst et al., 2013).

Parenting with depression is characterised by decreased psychological availability, which may exacerbate difficulties communicating with children (Semple & McCaughan, 2013). Furthermore, parents may restrict their emotional availability by choosing to give their children information about their illness but avoiding sharing their emotions (Hasson-Ohayon & Braun, 2011; Shands, Lewis, & Zahlis, 2000). As a result, families where a parent has been diagnosed with cancer may be at greater risk of family dysfunction (Semple & McCaughan, 2013).

The changes that occur when a parent is diagnosed with cancer can cause huge disruption among the entire family unit. Many parents report concerns that changes in their mood, cognition, physical functioning, and family routines negatively affect their children (Muriel et al., 2012). It is worth noting that this research did not directly observe parenting behaviour, indicating that parents themselves perceive that their children are affected by changes to their parenting as a result of their cancer. This has important implications; Moore et al. (2015) found an association between parenting self-efficacy beliefs, or expectations about the capacity to parent effectively, and parental concerns about the negative impact of their cancer on their children. Moreover, these findings indicated that higher levels of overall distress, depression, and anxiety, and poorer quality of life (in physical, functional, emotional, and social domains) were associated with reduced parenting self-efficacy beliefs and greater parental concerns about children's emotional distress (Moore et al., 2015). Research by Visser et al. (2006) may provide further context to this relationship; they found that children's emotional functioning was associated with the ill parent's physical and emotional functioning. Although cancer is a source of intense stress for the parent with the disease, these findings highlight the potential wider implications of the parent's psychological well-being for their family.

1.2. Psychosocial Impact of Parental Cancer on Dependent Children

Children are at particular risk of developing psychosocial problems following a parent's cancer diagnosis because they depend on their parent for care and support (Faulkner & Davey, 2002; Krattenmacher et al., 2012). When a parent is diagnosed with cancer, children may express feelings of isolation (Gazendam-Donofrio et al., 2011; Karlsson, Andersson, & Ahlstrom, 2013), loss of control (Phillips & Lewis, 2015), and have difficulty understanding their situation (Karlsson et al., 2013). Children need information about their parent's cancer and support from their family (Forrest, Plumb, Ziebland, & Stein, 2006; Konings et al., 2020; Kristjanson, Chalmers, & Woodgate, 2004; Patterson et al., 2017), but often feel that the impact of parental cancer on them is overlooked by family members and medical professionals (Davey, Tubbs, Kissil, & Nino, 2011).

A number of specific impacts on children have been identified. Children experience significant levels of distress following their parent's cancer diagnosis (Grabiak, Bender, & Puskar, 2007; Harris & Zakowski, 2003; Krattenmacher et al., 2012; Krauel et al., 2012; Patterson et al., 2017), and in some cases may display symptoms of post-traumatic stress disorder (Wozniak & Izycki, 2014). Morris et al. (2016) suggests that children’s distress likely stems from their need for a cohesive and supportive family environment, as well as fears of losing their parent and concerns about the family’s future. In particular, younger children may be more vulnerable to changes within the family because they depend on their parent to meet their daily care needs (Moller et al., 2014). On the other hand, older children are often significant informal carers for their parent and may be more focused on their parent’s well-being than their own (Torp, Thoresen, Gronningsaeter, Grov, & Gustavsen, 2013). Distress may lead to an increased risk of emotional and psychosocial problems (Faulkner & Davey, 2002; Grabiak et al., 2007; Osborn, 2007), such as problematic internalisation (e.g., anxiety, depression) or externalisation (e.g., aggression, oppositional defiance) (Osborn, 2007; Walczak et al., 2018).

Internalising behaviours reflect a child’s emotional or psychological state, and are typically characterised by anxious and depressive symptoms, social withdrawal, and somatic complaints (Gobel, Henning, Moller, & Aschersclehen, 2016; Liu, Chen, & Lewis, 2017). Children experience significant stress-response symptoms, which may present as avoidance and intrusive thoughts (Compas et al., 1994). Younger children may be more likely to experience somatic symptoms such as sleeping difficulties, headaches, and fatigue (Heiney et al., 1997; Hilton & Elfert, 1996; Hymovich, 1993). Adolescents commonly express fears that their parent will die (Grabiak et al., 2007); one study found that adolescents ranked whether their parent was going to survive as their greatest information need (Kristjanson et al., 2004). This is consistent with findings reported by Patterson et al. (2017); they found that two-thirds of AYAs reported that their need for information about the chances of their parent’s recovery was unmet (66.4%; *n* = 256). Some research suggests that fear of losing the parent or of the cancer recurring may be expressed through somatic symptoms (such as migraines; Spira & Kenemore, 2000). Adolescent daughters may express increased perceived vulnerability to cancer because of a possible genetic risk (Grabiak et al., 2007; Kristjanson et al., 2004). Nelson et al. (1994) reported that males experience anxiety about their parent’s cancer but feel unable to discuss their fears with either parent. In general, adolescents refrain from sharing their thoughts, feelings, and fears in an attempt to protect their parent and avoid tension in the relationship (Davey, Askew, & Godette, 2003).

Externalising behaviours are directed outward toward other people or, more generally, to the social context. These may include aggressive, oppositional, and disruptive behaviour in the school or home (Gobel et al., 2016). In children who have a parent with cancer, externalising problems may present as boisterousness, compulsivity, conduct problems, hyperactivity, inattention, or antisocial behaviours (Huizinga, van der Graaf, Visser, Dijkstra, & Hoekstra-Weebers, 2003; Krattenmacher et al., 2013). Children may also experience decreased competencies in school, sports, social relationships, and other activities following their parent’s cancer diagnosis (Huizinga et al., 2003). Despite these findings, it is unclear whether rates of externalising problems in children who have a parent with cancer deviate from age-comparable norms (Osborn, 2007).

The longer-term effects of parental cancer are more difficult to discern due to insufficient longitudinal evidence, although some parents report that children’s problems with anxiety, disordered sleeping, and compulsive behaviour may persist for months, or even years, after the cancer diagnosis (Huizinga et al., 2003). Other studies indicate that children’s acute emotional reactions are unrelated to their longer-term psychological functioning (Gazendam-Donofrio et al., 2011). More recent findings suggest that young adults who experienced parental cancer during their childhood experience higher levels of state and trait anxiety than matched controls (Metcalf, Arch, & Greer, 2017). In addition, Chen et al. (2018) reported that exposure to parental cancer during childhood was associated with greater likelihood of subsequent diagnosis with a psychiatric disorder, particularly stress reaction and adjustment disorders, and psychiatric medication use.

There are positive outcomes that may come from having a parent diagnosed with cancer; these include stronger relationships within the family, a greater appreciation for life and other people, personal growth and development, and positive changes in goals and priorities (Levesque & Maybery, 2012; Walczak et al., 2018). Children may also demonstrate greater self-reliance, increased interest in health and improved health behaviours, and high levels of resilience, particularly among those who are bereaved (Phillips & Lewis, 2015; Walczak et al., 2018).

Children report using a range of strategies to help them cope with their parent's cancer. Most commonly, these include distraction, wishful thinking, spending time with or helping their parent, and accessing peer support (Davey et al., 2003; Krattenmacher et al., 2013; Thastum, Johansen, Gubba, Olesen, & Romer, 2008). Other coping strategies children may use include acceptance, positive reframing, taking time out, using emotional outlets, keeping their thoughts and emotions to themselves, and maintaining normality (Maynard, Patterson, McDonald, & Stevens, 2013; Morris et al., 2016; Thastum et al., 2008). Children may also compromise their own needs or emotions in order to support the parent, through a process known as parentification (i.e., parent-child role reversal) (Davey et al., 2003; Phillips & Lewis, 2015; Thastum et al., 2008). This can potentially be an adaptive coping strategy, if the child's caregiving behaviours are recognised and supported by the family, or may otherwise be destructive, if reciprocity, acknowledgement, and support are absent (Jurkovic, 1997; Thastum et al., 2008).

1.3. Factors Associated with Adjustment to Parental Cancer

There are a range of factors that influence the extent to which a child may be impacted by their parent’s cancer. Su and Ryan-Wenger (2007) propose a theoretical model that describes the relationships between a parent’s cancer diagnosis, moderator and mediator variables, and the child’s psychological adjustment. Contextual influences are identified as moderators of the outcome and include person-defining variables (e.g., the age and gender of the child) or variables that define the illness (e.g., the type and stage of the parent’s cancer). In other words, these variables influence the situation that causes stress (Su & Ryan-Wenger, 2007). Mediator variables, in contrast, exert influence after the parent’s cancer diagnosis; these factors do not impact the intensity of the stressful situation, but rather the way that the family and child respond to the situation (e.g., the quality of the parent-child relationship or the child's coping responses; Su & Ryan-Wenger, 2007).

According to Lazarus and Folkman’s (1984) model of stress and coping, moderator variables are directly related to an individual’s appraisal of a situation (primary appraisal) and indirectly influence the coping strategies that the individual chooses to engage with (secondary appraisal). The primary appraisal of the situation (i.e., perception of parental cancer) and the secondary appraisal of the coping strategy (i.e., evaluation of available coping resources) act as mediators; it is these variables that directly influence the coping strategies that an individual chooses to employ and whether or not they continue to engage with them. This model will be elaborated in further detail in Section 1.4.1.

1.3.1. Moderating variables

Research examining the effect of the child’s age on their psychological adjustment has produced mixed findings. Data collected from 235 families suggest that the proportion of children who experience emotional and behavioural symptoms is higher in preadolescents (6-10 years) than adolescents (11-21 years) (Moller et al., 2014), and in younger adolescents

(11-13 years) than older adolescents (14-21 years) (Moller et al., 2014). Using comparable methodology, but a smaller sample size (167 families), Krattenmacher et al. (2013) found no significant age-effects in adolescents aged 11-18 years. Two smaller studies also found no significant age-effects in adolescents whose parent had early-stage breast cancer (Edwards et al., 2008) or had been diagnosed with cancer within the past four months (Huizinga et al., 2010). On the other hand, Rainville, Dumont, Simard, and Savard (2012) conducted a small study among adolescents with a parent with advanced cancer (defined as metastatic or recurrent cancer, for which there is no curative treatment) and found that older adolescents (15-18 years) experienced more psychological distress than younger adolescents (12-14 years). Another study examined the longer-term impact of parental cancer on children by comparing psychiatric disorder diagnoses in those who had been exposed to parental cancer with a matched cohort who had not been exposed to parental cancer; findings revealed that children who were 13 years or older were at greater risk of developing a psychiatric disorder following their parent's cancer diagnosis (Chen et al., 2018).

Piaget's cognitive-developmental theory may provide additional context for these findings (Piaget & Inhelder, 1969). According to Piaget, preschool children (age 2 to 7) are in the Preoperational stage of cognitive development. At this stage, children's reasoning is characterised by fantasy and magical thinking. Because their moral sense is still developing, they often feel a sense of responsibility for events in their lives and believe that they can cause illness or death with their thoughts, feelings, or wishes. Around the age of 6 or 7 to 11, children achieve the Concrete Operational stage and are able to think more flexibly and can apply logic to concrete information. At this stage, children's abstract thinking ability is still limited; they are better able to engage with cause and effect reasoning when dealing with concrete information, than abstract concepts. This means pre-adolescent children can understand the physical signs and symptoms of their parent's cancer, as these are concrete observables, but may struggle with more abstract concepts such as what it means for their parent to have cancer and the possibility that their parent may die (Su & Ryan-Wenger, 2007). The ability to think about and reason with abstract concepts develops during the Formal Operational stage. This final stage of cognitive development begins around the age of 11 and continues into adulthood. During this stage, adolescents are better able to understand their parent's cancer and the implications for their family (Gazendam-Donofrio et al., 2011; McDonald et al., 2016; Weeks et al., 2019), but they may feel torn between their own need for greater independence and autonomy and their desire to care for their parent (Su & Ryan-Wenger, 2007). The findings of interviews conducted with adolescents supports this; Lewis et al. (1985) found that adolescents experienced conflicting feelings related to an increase in their responsibilities and decrease in their social activities as a result of their parent's cancer. This was further compounded by their desire to care for their parent.

It is important to note that a child's level of cognitive maturity does not necessarily correspond with their chronological age, although age is often used as a proxy for cognitive maturity in research examining children's adjustment to parental cancer. It follows that inconsistencies in the research examining the effect of age on psychological adjustment following a parent's cancer diagnosis may be related to differences in cognitive maturity within age groups and the distribution of this in different studies. Research also indicates that children's understanding of illness corresponds to their level of cognitive maturity (Carson et al., 1992). Su and Wenger (2007) suggest that the child's level of cognitive maturity will impact their appraisal of the cause, severity, and stressfulness of their parent's cancer, and in turn, their psychological adjustment following their parent's diagnosis. Therefore, examining how children conceptualise and make sense of their parent's cancer, and how differences in children's illness perceptions impact their psychological adjustment, may yield more reliable findings than studies that employ chronological age as an indicator of cognitive maturity. This will be discussed further in Section 1.3.2.

Findings regarding the impact of the child's gender are similarly inconsistent. Some studies suggest that daughters experience significantly higher levels of psychological distress (McDonald et al., 2016), post-traumatic stress symptoms (Edwards et al., 2008; Huizinga, Visser, van der Graaf, Hoekstra, & Hoekstra-Weebers, 2005; Huizinga, Visser, van der Graaf, Hoekstra, Klip, et al., 2005), anxiety (E. Nelson & While, 2002; Welch, Wadsworth, & Compas, 1996), and depression (Welch et al., 1996), more emotional and behavioural problems (Visser et al., 2005; Welch et al., 1996), and lower health-related quality of life (Jeppesen, Bjelland, Fosså, Loge, & Dahl, 2016) than sons. Visser, Huizinga, Hoekstra, van der Graaf, Gazendam-Donofrio, et al. (2007) found that adolescent sons reported slightly greater total problems soon after parent's diagnosis, but adolescent daughter reported greater total problems at six- and 12-month follow-ups. The authors highlighted that, in contrast to earlier studies that had found clinically elevated scores primarily among adolescent daughters, their findings revealed clinically elevated scores among adolescent sons as well (Visser, Huizinga, Hoekstra, van der Graaf, Gazendam-Donofrio, et al., 2007). They noted that this may have been a result of their focus on internalising problems, rather than just anxiety and depression (Visser, Huizinga, Hoekstra, van der Graaf, Gazendam-Donofrio, et al., 2007). Another study conducted among adolescents in Turkey found that sons experienced worse outcomes than daughters across multiple psychosocial domains (anxiety, depression, negative identity, somatization, hostility; Küçükoğlu & Çelebioğlu, 2013), however it is possible that this is related to cultural differences in social norms and gender roles. Several studies also point to an interaction between the age and gender of the child, with adolescent daughters reporting more problems than adolescent sons (Moller et al., 2014; Visser et al., 2005; Welch et al., 1996), and preadolescent sons reporting more problems than daughters of the same age (Visser, Huizinga, Hoekstra, van der Graaf, Gazendam-Donofrio, et al., 2007).

Moreover, several studies investigating the impact of the parent’s gender found that children experience poorer outcomes when their father is diagnosed with cancer (Küçükoğlu & Çelebioğlu, 2013; McDonald et al., 2016; Thastum et al., 2009; Visser et al., 2005). There may also be an interaction between the child’s gender and the parent’s gender; Visser et al. (2005) found that adolescent daughters reported more problems when the father was ill than when the mother was ill. Other findings suggested that anxiety and depression were greater in adolescent daughters whose mother had cancer, in comparison with sons or those whose father had cancer (Grant & Compas, 1995; Welch et al., 1996).

Adding to this complexity, some findings suggest that attachment styles within the family contribute to children’s psychological adjustment. Inbar, Ety, Ayala, and Tamer (2013) found that anxious attachment in mothers and avoidant attachment in daughters were both significantly associated with increased levels of psychological distress. Personal attributes, such as temperament (Visser, Huizinga, Hoekstra, van der Graaf, & Hoekstra-Weebers, 2007) and self-esteem (E. Nelson & While, 2002), have also been found to predict children’s emotional and behavioural problems. Family attributes, including socioeconomic status (Edwards et al., 2008), single parenthood (E. Nelson & While, 2002; Visser et al., 2006), and number of children in the family (Visser et al., 2006) may also contribute to psychological adjustment. In particular, children of a single parent with cancer may be at increased risk of poor adjustment (E. Nelson & While, 2002), internalising and externalising problems (Visser et al., 2006), and reduced quality of life (Bultmann et al., 2014) .

Studies have also investigated whether factors intrinsic to the disease predict psychological adjustment, although findings have been largely inconclusive (Krattenmacher et al., 2012). Several studies (Hoke, 2001; Lindqvist, Schmitt, Santalahti, Romer, & Piha, 2007; Moller et al., 2014; Welch et al., 1996) have examined psychological adjustment in relation to cancer type but have found no relationship. This is potentially due to sampling or reporting issues that limit the ability to identify predictor effects (for example, small sample size per cancer type; Walczak et al., 2018). Findings regarding the stage of cancer or prognosis are mixed; several studies found no relationship (Gazendam-Donofrio et al., 2007; Huizinga, Visser, van der Graaf, Hoekstra, & Hoekstra-Weebers, 2005; Lindqvist et al., 2007; Moller et al., 2014), but others indicate that poorer prognosis is associated with poorer psychological adjustment and internalising and externalising problems (Huizinga et al., 2010; Huizinga, Visser, van der Graaf, et al., 2011; Sigal, Perry, Robbins, Gagne, & Nassif, 2003; Visser et al., 2006; Visser, Huizinga, Hoekstra, van der Graaf, & Hoekstra-Weebers, 2007; Visser et al., 2005). Previous research suggests that adolescents experience greater internalising and total problems, as well as more intrusion, avoidance, and total distress when their parent's cancer is recurrent (Huizinga et al., 2010; Huizinga, Visser, van der Graaf, et al., 2011; Visser et al., 2006; Visser, Huizinga, Hoekstra, van der Graaf, & Hoekstra-Weebers, 2007; Visser et al., 2005). Findings are inconclusive for palliative status; Siegel, Karus, and Raveis (1996) found that children whose parents were receiving palliative care experienced higher rates of emotional and behavioural problems, depression, and anxiety, whereas Kühne et al. (2012) found that psychological functioning was better in children whose parents were receiving palliative care than those at other stages of the disease.

Findings regarding longer-term psychological adjustment are also mixed, with studies showing that outcomes improve (Huizinga et al., 2010; McDonald et al., 2016; Visser, Huizinga, Hoekstra, van der Graaf, Gazendam-Donofrio, et al., 2007) or remain stable (Gazendam-Donofrio et al., 2007; Götze, Ernst, Brähler, Romer, & von Klitzing, 2015; Huizinga, Visser, van der Graaf, et al., 2011; Visser, Huizinga, Hoekstra, van der Graaf, & Hoekstra-Weebers, 2007) as time since diagnosis increases. It should be noted that much of this research is cross-sectional; further longitudinal research is needed to more rigorously examine the relationship between psychological adjustment and time since diagnosis. In general, the type of treatment that the parent undergoes does not appear to be significantly related to children's psychological adjustment (Krattenmacher et al., 2012; Walczak et al., 2018), although some findings indicate that more intensive treatment (Visser et al., 2006) and treatment complications (Huizinga, Visser, van der Graaf, et al., 2011; Visser et al., 2006) are predictive of poor adjustment. Again, these findings are subject to sampling and reporting limitations.

1.3.2. Mediating variables

Family functioning has been extensively examined as a predictor of children's psychological adjustment to parental cancer. Findings reveal that positive family functioning, characterised by open communication, greater cohesion, and less conflict, is associated with fewer internalising and externalising problems, reduced distress, fewer unmet needs, and better health-related quality of life (Edwards et al., 2008; Kühne et al., 2012; Lindqvist et al., 2007; McDonald et al., 2016; Moller et al., 2014; Thastum et al., 2009). Consistent with this, several studies found that family dysfunction was the strongest predictor of negative psychosocial outcomes (Edwards et al., 2008; Kühne et al., 2012; Moller et al., 2014; Thastum et al., 2009).

As mentioned in Section 1.1., research suggests that the well-being of individuals within the family is interconnected. Findings show that outcomes in children are predicted by parental depression, anxiety, and general physical and psychosocial functioning (Edwards et al., 2008; Götze et al., 2015; Huizinga, Visser, van der Graaf, et al., 2011; Lindqvist et al., 2007; Thastum et al., 2009). It is likely that there are additive influences from family factors that further compound the impact of parental cancer on children; for example, Schmitt et al. (2008) found that impaired family functioning was predicted by parental depressive symptoms, and that both family functioning and parental depression were independently associated with children’s psychological adjustment.

Although open communication is an important component of positive family functioning, it also appears to be crucial for children’s psychological adjustment when examined separately. Communication has been associated with better coping and lower levels of psychological distress in children (Lindqvist et al., 2007; Phillips & Lewis, 2015; Thastum et al., 2008; Thastum et al., 2009). In children of women with breast cancer, warmth and acceptance-oriented communication between the parent and child were significantly associated with lower levels of internalising problems (Vannatta, Ramsey, Noll, & Gerhardt, 2010). Similarly, Huizinga, Visser, van der Graaf, Hoekstra, and Hoekstra-Weebers (2005) found that more open, less problem-oriented, communication was associated with fewer emotional and behavioural problems. In contrast, avoiding communication about cancer has been associated with poorer psychological outcomes in both parents and children (Krattenmacher et al., 2013), with findings showing associations between poor family communication and increased internalising and externalising problems and stress responses in children (Huizinga, Visser, van der Graaf, et al., 2011; Lindqvist et al., 2007). In a study examining patterns of communication among young people impacted by parental cancer, Rodriguez (2019) found that young people were selective about who they talked to and how much information they shared about their parent’s cancer. They described barriers to communication, with these including perceived unavailability of their parent and fear of the realities of cancer (Rodriguez, 2019).

Parents are an important informant for children, but they too describe challenges in communicating with their children about cancer (Konings et al., 2020; Moore, Shea, Russell, Convery, & Rauch, 2018). This may contribute to discrepancies between the amount of information that children desire and the amount that they receive (Walczak et al., 2018).

AYA children believe that it is important to be well-informed about their parent's cancer (Phillips & Lewis, 2015; Thastum et al., 2008); but, despite this, Patterson et al. (2017) found that many young people described having unmet information needs. This finding was consistent with earlier research that found that the need for information about the parent's side effects and symptoms was the third most commonly reported unmet need among AYA children (Patterson, Pearce, & Slawitschka, 2011). Although the relationship between children's information needs and psychological adjustment has not been explicitly examined, research exploring aspects of family functioning has shown that information sharing is associated with psychological functioning (Osborn, 2007; Rainville et al., 2012). Moreover, J. M. Miller (2008) found that adolescents who received more information about their parents' cancer experienced fewer psychological symptoms.

Open information sharing and communication about cancer may prevent children from forming inaccurate beliefs or having misconceptions about their parent's cancer (Morris et al., 2016; Su & Ryan-Wenger, 2007). Findings reported by Kennedy and Lloyd-Williams (2009) indicate that, without adequate information about their parent's cancer, misconceptions may contribute to undue distress. For example, children may believe that a lack of physical symptoms means that their parent is getting better and that more visible symptoms (e.g., hair loss) mean their parent's condition is worsening (Kennedy & Lloyd-Williams, 2009). This is consistent with research in younger children, which found that children had a number of inaccurate beliefs about their parent's cancer, including that cancer was contagious and could be transmitted by physical contact with their parent (Semple & McCaughan, 2013). Misconceptions of this nature may not always be apparent to parents because of the difficulties that children experience communicating about cancer with their parent (Semple & McCaughan, 2013; Visser et al., 2004).

There is a paucity of research examining how children conceptualise and make sense of their parent's cancer, and even fewer studies have been conducted to examine the specific relationships between children's beliefs about their parent's cancer and their psychological outcomes. Nevertheless, two studies conducted by Compas et al. (1994 and 1996) provide important insights. In their 1994 paper, Compas and colleagues report that perceptions of the seriousness and stressfulness of parental cancer were significantly and positively associated with post-traumatic stress symptoms in AYA children. These findings were extended in their later paper, which reported that greater perceived stressfulness was significantly associated with higher levels of anxiety and depression (Compas et al., 1996). It is possible that inaccurate beliefs and misconceptions, such as a belief that more visible physical symptoms are indicative of the cancer worsening (Kennedy & Lloyd-Williams, 2009), cause children to perceive their parent's cancer as being more serious than it actually is, and in turn this may contribute to poorer psychological adjustment.

Children's appraisal of their parent's cancer may also have implications for the coping strategies they choose to employ (Compas et al., 1996). In turn, the coping strategies that children use may impact psychological outcomes. For example, Krattenmacher et al. (2013) found that support-seeking strategies appeared to be effective approaches to coping and were associated with better psychological outcomes. On the other hand, avoidant coping strategies (e.g., social withdrawal) were associated with poorer outcomes (Krattenmacher et al., 2013; Phillips & Lewis, 2015).

Various other studies suggest that children who are able to accept their parent's cancer diagnosis and talk about cancer with others may have better outcomes than those who employ denial or avoidance-based coping strategies (Krattenmacher et al., 2013; Maynard et al., 2013; Phillips & Lewis, 2015; Thastum et al., 2008). Research examining distraction-based coping strategies (e.g., daydreaming or engaging in other activities as a means of distraction) is inconclusive; some authors suggest that these types of coping strategies are maladaptive (Thastum et al., 2008), however others argue that they are an adaptive attempt to preserve some sense of normality (Kennedy & Lloyd-Williams, 2009; Maynard et al., 2013; Torp et al., 2013). Some findings suggest that children may cycle between approach and avoidant coping strategies (Phillips & Lewis, 2015), indicating an inability to engage with one strategy or one type of strategy as a means of coping (Walczak et al., 2018). Some studies also suggest that parents play an important role in their children's coping and well-being by modelling their own coping behaviours (Maynard et al., 2013).

Children's psychosocial needs also appear to be associated with their psychological adjustment (Kristjanson et al., 2004; McDonald et al., 2016; Patterson et al., 2017). Patterson and colleagues (2011) found that 43% of young people reported 10 or more unmet needs and that there was a significant and positive association between the number of unmet needs and psychological distress. More recent research reports findings consistent with this (McDonald et al., 2016; Patterson et al., 2017). However, many AYAs report that their needs are poorly met (Kristjanson et al., 2004); findings suggest that their top five unmet needs are centred around peer support (e.g., needing understanding from, and open communication with, friends), education and work (e.g., needing help to concentrate at school or work), carer support (e.g., needing information about side effects and symptoms), and dealing with difficult feelings (e.g., problems with self-esteem) (Patterson et al., 2011).

1.4. Theoretical Models Describing the Link between Illness Perceptions and Coping

As noted in the previous section, the cognitive maturity of a child may be an important moderator of the relationship between a parent's diagnosis with cancer and their child's psychological adjustment. In accordance with their theoretical model (described in Section 1.3.), Su and Ryan-Wenger (2007) suggest that the relationship between level of cognitive maturity and psychological adjustment may be mediated by the child’s appraisal (or perceptions) of their parent’s cancer. This means that children’s level of cognitive maturity may influence the extent to which a parental cancer diagnosis affects their psychological functioning by way of differences in how they perceive their parent’s cancer.

In addition to this, examination of children’s perceptions of their parent’s cancer may inform the development of approaches to identify those at risk of poor psychological adjustment, as well as effective interventions to provide children with coping strategies. In contrast, although understanding the predictive ability of moderating variables such as age and gender may be useful for identifying children at risk, it provides limited information for the design and development of intervention strategies. Focusing on these variables also ignores the sociocultural factors that influence how individual children think, feel, and experience the world. An individual’s illness perceptions, on the other hand, are determined by a combination of their personal experience with illness, their personality traits, and the sociocultural context (Hagger & Orbell, 2003; Moss-Morris & Paterson, 1995).

There are a number of theoretical models that have been developed in an effort to understand how illness perceptions influence coping behaviours and, in turn, psychological outcomes. Two of the most prominent models are described below.

1.4.1. The Transactional Model of Stress and Coping

Lazarus and Folkman (Lazarus & Folkman, 1984) integrated research on stress, health, and coping to inform the development of the Transactional Model of Stress and Coping (TMSC). The model describes stress processes as “transactional”, whereby the individual’s coping and emotional responses are determined by their appraisal of the stressful situation (Hulbert-Williams, Morrison, Wilkinson, & Neal, 2013). The central concept of the TMSC is that individuals can appraise identical situations differently, and it is these appraisals that determine their behavioural or health outcomes, rather than the objective characteristics of the situation (Wethington, Glanz, & Schwartz, 2008). Approaching stress as a transactional process between the individual and their environment emphasises the bidirectionality of the relationship. This means that it is an interaction (or transaction) between the individual and their environment that produces the stress response, rather than the response being caused by each factor in isolation (Biggs, Brough, & Drummond, 2017).

According to the TMSC (see Figure 1), primary appraisal, secondary appraisal, and coping responses mediate the relationship between the stressful situation and the individual's stress outcomes (Goh, Sawang, & Oei, 2010). It is proposed that individuals constantly appraise stimuli within their environment. Appraisals may occur consciously or unconsciously and are influenced by situational, cultural, and personal factors (Hulbert-Williams et al., 2013). This process generates emotional responses, and when a particular stimulus is appraised as being threatening, challenging, or harmful to the individual's well-being (i.e., a stressor), a coping response is initiated in order to manage emotions or attempt to directly address the stressor (Biggs et al., 2017). This initial appraisal of the stressor is defined as the primary appraisal; during this stage of the appraisal process, the individual evaluates the stressor in terms of how it may affect their well-being. Depending on how the individual perceives the stressor, they may categorise it as a loss (damage or harm that has already happened), or as a threat or challenge (damage, harm, or growth opportunity that is anticipated) (Berjot & Gillet, 2011). It is important to note the difference between a threat and a challenge; a threat is defined by potential *danger* to the individual's well-being or self-esteem, whereas a challenge refers to a situation that has the potential for *social rewards or personal growth* (Lazarus & Folkman, 1984). Although one perceives potential benefits from the situation, threat and challenge appraisals are not mutually exclusive and can occur simultaneously (Berjot & Gillet, 2011).

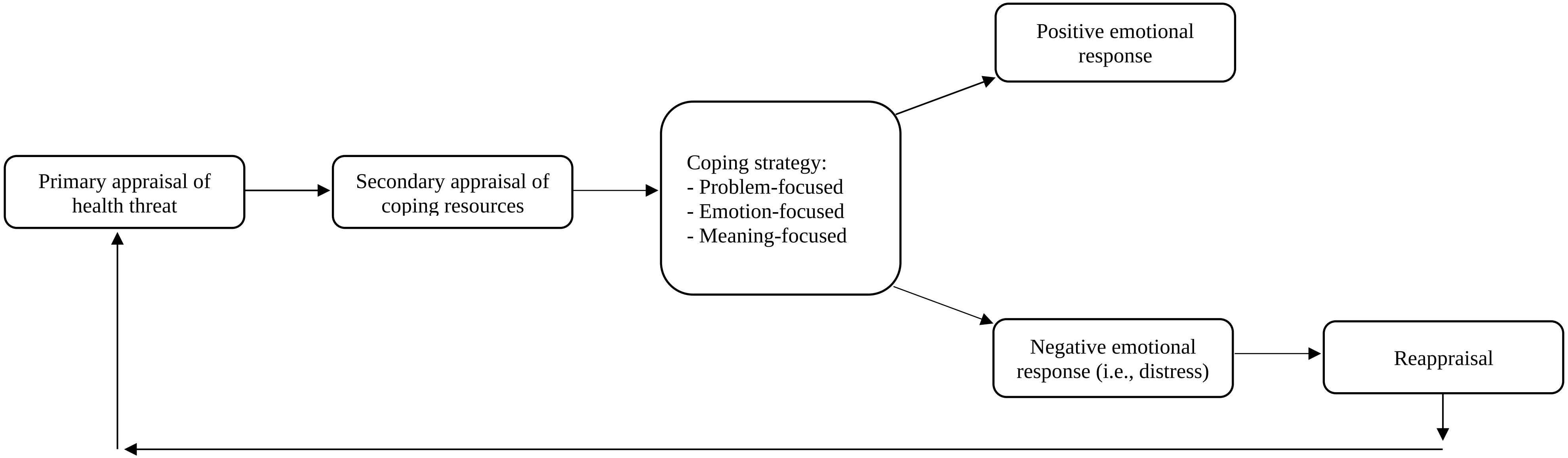


Figure 1. The Transactional Model of Stress and Coping (adapted from Goh et al., 2010)

The secondary appraisal involves the individual evaluating their coping resources and ability to manage the stressful situation (Biggs et al., 2017). Resources may be physical (e.g., health, energy), social (e.g., social support from family, friends, and the broader community), psychological (e.g., self-esteem, perceived control), or material (e.g., financial) (Berjot & Gillet, 2011). Evaluation of coping resources and ability to manage the stressor informs the individual's choice of coping strategy.

The appraisal process continues; the individual's coping response will produce an emotional outcome, and this is then reappraised as favourable, unfavourable, or unresolved (i.e., was the coping strategy effective?). Favourable resolution of the stressor will elicit a positive emotional response, whereas unfavourable or unresolved resolutions will elicit a distress response, prompting the individual to reconsider their coping response and engage with alternative strategies in an effort to resolve the stressor (Biggs et al., 2017). Here, coping can be understood as the cognitive and behavioural effort by the individual to make their situation more manageable. Stress occurs when the transactions between the individual and environment result in the individual perceiving a discrepancy between the demands of the situation and their ability to cope with those demands (Quine & Pahl, 1991).

Within the framework of the TMSC, chronic illness is viewed as a potential stressor. In the context of a child responding to a parent's cancer diagnosis, the TMSC would posit that it is not the cancer diagnosis itself that predicts or causes psychological morbidity in the child, but rather how the child perceives their parent's cancer and their ability to cope with the situation. If the child perceives the demands of the stressor (the parent's cancer) to be greater than their ability to cope with those demands, then they will experience stress.

Later work expanded the concept of appraisal to enable examination of the specific components of primary (goal relevance, goal congruence, and ego-involvement) and secondary (accountability, coping potential, and future expectations) appraisal (Lazarus, 1991, 1993, 1999). The idea is that variation in these components of appraisal determine the core-relational theme (or relational meaning), which is in turn associated with a particular emotional response (i.e., anger, guilt, fear/anxiety, sadness, etc.) (Hulbert-Williams et al., 2013). The emotional response then mediates the individual's choice in coping strategy.

Although the TMSC was conceptualised by Lazarus and Folkman as a linear sequence flowing from primary to secondary appraisals, to coping, and to stress outcomes, it is important to note that this is a reflection of the basic pathways within the overall process, which is much more dynamic (Goh et al., 2010). The primary appraisal does not always precede the secondary appraisal, and neither is judged as being more important than the other (Goh et al., 2010).

1.4.2. The Common-Sense Model of Self-Regulation

Like the TMSC, Leventhal's Common-Sense Model of Self-Regulation (CSM) conceptualises chronic illness as a stressful experience (Hale, Treharne, & Kitas, 2007). The model was developed in effort to understand how people made sense of their experiences with illness and how this guided their coping behaviours (Diefenbach & Leventhal, 1996; A. Richardson & Broadbent, 2017).

The development of the CSM was informed by Leventhal's research exploring the use of fear-based messages to prompt individuals to engage with preventative behaviours such as wearing seat belts or quitting cigarette smoking (Leventhal et al., 2003). Leventhal and Niles (1965) found that the attitudes and actions that individuals had in response to a health threat could be influenced by different types of information. Crucially, though, they found that preventative responses were short-lived. In extension of this work, Leventhal and colleagues investigated the attitudes and coping responses that contributed to maintaining health behaviours in those experiencing chronic illness (Diefenbach & Leventhal, 1996; Leventhal, Meyer, & Nerenz, 1980).

The CSM is comprised of three main constructs: representations of the illness experience (or illness perceptions), coping responses, and appraisal of the coping effort (i.e., was the coping response effective?) (see Figure 2). One element of the CSM that differentiates it from other models of illness perception, is that it is conceptualised as a parallel processing framework, wherein individuals simultaneously regulate their cognitive (i.e., “what is this health threat and what can I do about it?”) and emotional (“how do I feel about it and what can I do to make myself feel better?”) responses to the health threat (Hagger & Orbell, 2003; Hale et al., 2007).

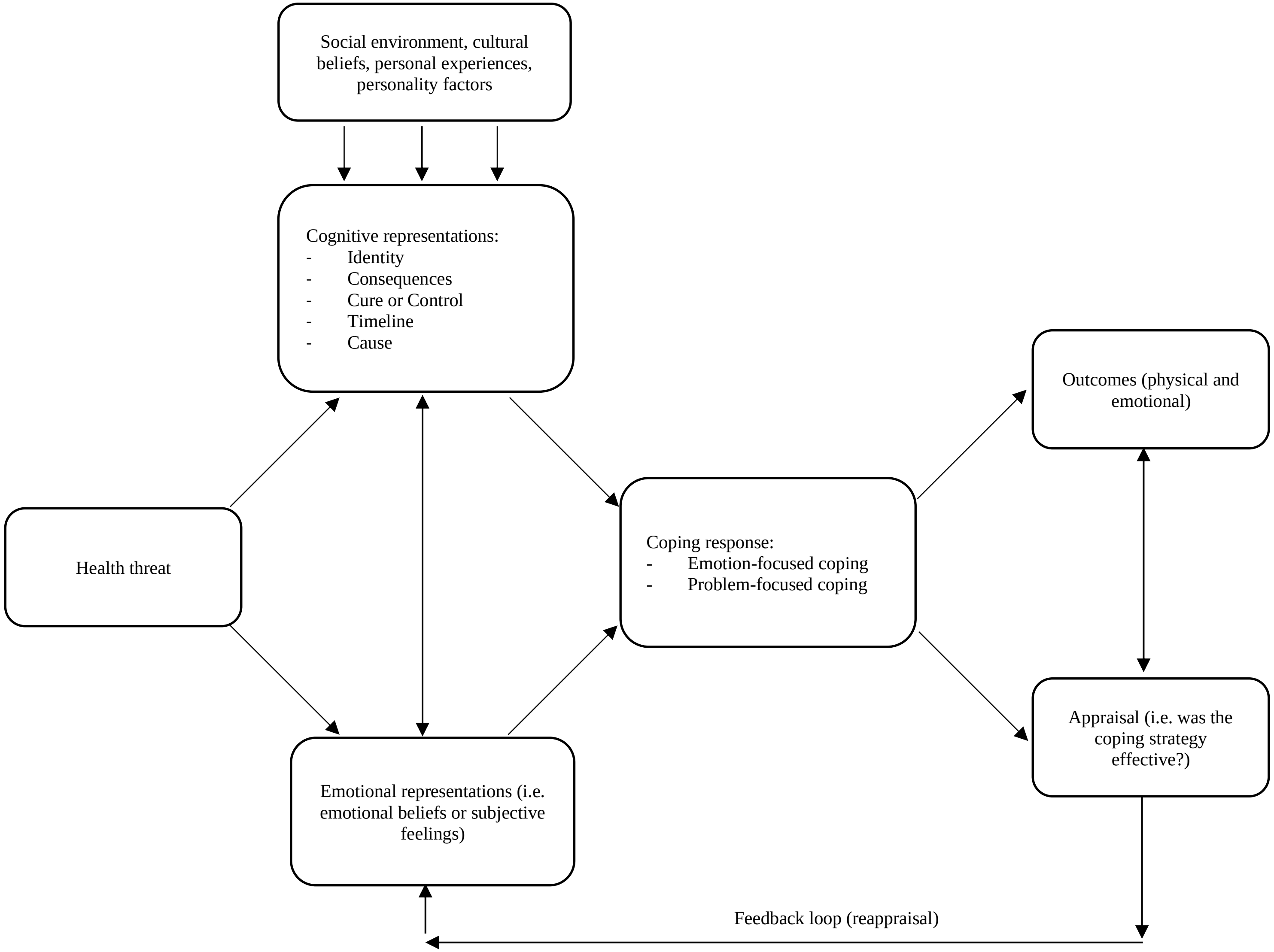


Figure 2. The Common-Sense Model of Self-Regulation (adapted from Hagger & Orbell, 2003)

The key premise of the CSM is that individuals construct representations of their illness based on their own perceptions, interpretation, and understanding of the illness and its treatment (Diefenbach & Leventhal, 1996; Hagger & Orbell, 2003; Petrie & Weinman, 2006). These beliefs assist the individual in making sense of their illness and guide their coping response (Hale et al., 2007), and are informed by three basic sources of information: lay information or cultural knowledge of the illness, the external social environment as represented by important others or authoritative figures (e.g., a family member or doctor), and personal experience with the illness (e.g., current physical symptoms or previous experiences with the illness) (Hagger & Orbell, 2003).

The CSM proposes that cognitive representations of illness are organised around five central dimensions: identity (the label used to describe the illness and the symptoms that are believed to be associated with the illness), consequences (beliefs about the physical, cognitive, or social impact of the illness), cure or control (beliefs about whether the illness can be cured or controlled and the extent to which the individual plays a role in achieving this), timeline (beliefs about how long the illness will last), and cause (beliefs about cause of the illness – medically substantiated or otherwise). As previously mentioned, the CSM has been described as parallel processing model, meaning that individuals simultaneously construct cognitive and emotional representations of their illness. For this reason, it should be noted that emotional representations (emotional beliefs or subjective feelings about the illness) may be important determinants of emotional outcomes (Hagger & Orbell, 2003; Moss-Morris et al., 2002).

Finally, the model is dynamic. Over time, and as the illness progresses, the individual develops new representations of their illness based on new information and evaluation of their coping efforts. This means that illness representations are cumulative and information can be added to the construct, discarded, or adapted as necessary (Hale et al., 2007).

1.4.3. Strengths and limitations of the TMSC and CSM

There are a number of similarities between the TMSC and CSM that are worth noting. Both models place illness perceptions (referred to as “appraisal” in the TMSC and “representation” in the CSM) at the centre of the cognitive processes that influence the individual’s evaluation of the health threat. Also, both models consider the individual components of appraisal/representation that contribute to variation in coping and illness outcomes.

There are also notable differences. The TMSC considers cognitive appraisal of the illness to precede the emotional response, whereas the CSM proposes that this occurs via parallel processing of cognitive and emotional representations. A significant limitation of the TMSC is that many of the hypothesised relationships have not been empirically tested (Hulbert-Williams et al., 2013). As outlined in Section 1.4.1., each emotional response has a unique core-relational theme (i.e., pattern of primary and secondary appraisals), and many of these remain undefined (Hulbert-Williams et al., 2013). In contrast, there is extensive evidence, across a range of different illness groups, demonstrating the relationships between illness perceptions, coping strategies, and illness outcomes as outlined in the CSM (Breland, Wong, & McAndrew, 2020; Hagger et al., 2017; Hagger & Orbell, 2003).

Few studies have tested the predictive utility of the specific components of the TMSC; generally, the concepts outlined by the model have been used as a framework for describing how coping with stress is predictive of stress-related outcomes (Hulbert-Williams et al., 2013). Further empirical testing and development is needed to gain greater understanding of the specific relationships proposed by the TMSC. For example, Hulbert-Williams et al. (2013) examined the validity of the hypothesised relationships between cognitive appraisals and emotional responses and, although their findings supported the general process structure of the TMSC, they were unable to confirm specific hypothesised appraisal-emotion models. One possible explanation for this is the difficulty in isolating a specific stressor (in this case, a diagnosis of cancer) from other potentially stressful experiences. This particular study was conducted over six months, and as Hulbert-Williams et al. (2013) note, it is likely that participants experienced other stressful situations and personal changes within that time, each contributing to the emergence of new stress pathways, with unique cognitive appraisals and emotional responses. This highlights an important shortcoming of applying the TMSC to the context of chronic illness. The TMSC is a general model of stress and coping; therefore, it may not allow for the nuances of the illness experience and the mechanisms by which illness perceptions influence coping and illness outcomes.

In contrast, the components of the CSM have been extensively used to predict variation in illness outcomes (Breland et al., 2020; Hagger et al., 2017; Hagger & Orbell, 2003) and have provided a theoretical foundation for effective interventions aimed at improving treatment adherence (Jones, Smith, & Llewellyn, 2016) and physical and psychological outcomes (Broadbent, Ellis, Thomas, Gamble, & Petrie, 2009b; Cameron & Jago, 2010; McAndrew et al., 2010; Wearden & Peters, 2008). In their meta-analysis of 45 studies, Hagger and Orbell (2003) identified a range of predictive relationships between illness perceptions, coping, and illness outcomes that were consistent with Leventhal’s theoretical framework. In particular, their analysis found that perceptions that the illness was highly symptomatic, chronic, and had serious consequences were significantly associated with avoidance and emotional expression coping strategies. Perceptions in these domains were also negatively associated with psychological well-being, role and social functioning, and vitality. In comparison, stronger perceptions of control were significantly and positively associated with cognitive reappraisal, emotional expression, and problem-focused coping strategies. Perceptions of cure/control were also significantly and positively correlated with adaptive outcomes of psychological well-being, social functioning, and vitality, and negatively correlated with psychological distress and illness outcomes.

The demonstrated predictive utility of the CSM over the TMSC may be related to its use of multi-level concepts. Whereas other models of illness perception assess abstract ideas such as perceived personal vulnerability and illness severity with an overall judgement about how “bad” an illness is, the CSM uses perceptual and behavioural referents of these abstract concepts (Leventhal et al., 2016a). For example, within the framework of the CSM, constructs such as perceived personal vulnerability and illness severity are evaluated using measures of the perceived immediate and projected consequences of the illness, as well as its timeline, curability, and controllability (Leventhal et al., 2016a).

The CSM is also not without limitations. There is some concern about the way that coping has been interpreted within the literature and in context of the CSM. The original CSM conceptualised coping as specific behavioural outcomes (e.g., treatment adherence or self-care), but more recently coping has been interpreted as a broader concept that encompasses cognitive and emotional strategies that the individual might employ to manage their illness and its associated stress (e.g., problem-focused coping, emotion-focused coping, avoidance and denial, seeking social support, etc.). This has resulted in differences in perspectives about how the components of the CSM relate to one another, and specifically whether coping mediates the relationship between illness perceptions and outcomes as hypothesised (A. Richardson & Broadbent, 2017). Hagger et al. (2017) provides additional context; the findings of their meta-analysis of 254 studies indicate that illness perceptions may have both direct effects on outcomes as well as indirect effects mediated by coping. More specifically, their findings suggested that coping only partially accounted for the effects of illness perceptions on outcomes, and perceptions were found to have unique influence on outcomes that were independent of coping. Moreover, there may be significant overlap between constructs that describe illness perception and coping. For example, perceptions relating to personal control may also assess aspects of problem-focused coping. In support of this, some evidence suggests that responses on measures of illness perceptions are confounded by appraisals of coping (Dempster & McCorry, 2012; McCorry, Scullion, McMurray, Houghton, & Dempster, 2013).

It should be noted that although the components of the CSM explain a significant proportion of variance in illness outcomes, a substantial amount remains unexplained (A. Richardson & Broadbent, 2017). Additional psychological variables, such as personality traits or self-efficacy, may also be worth considering (Diefenbach & Leventhal, 1996). As the CSM has been developed over time in response to empirical data and theoretical concepts (Leventhal, Phillips, & Burns, 2016b), there is scope for identifying and including additional aspects of illness perception that are not already defined as part of the model.

1.5. Methods for Assessing Illness Perceptions

Researchers initially assessed illness perceptions via interviews (Hagger & Orbell, 2003; Rosenstock, Hochbaum, & Leventhal, 1960). One of the first studies to examine the predictive role of beliefs in behaviour change used questions based on the Health Belief Model (Rosenstock, 1974; Rosenstock, Strecher, & Becker, 1988). In this study, interviews were conducted to assess participants' beliefs about their personal vulnerability to influenza and its severity, as well as perceptions relating to their personal experiences with the illness (e.g., physical symptoms or observation of influenza in family members) (Rosenstock et al., 1960). Similarly, semi-structured interviews were used to establish and organise the content of illness representations into the dimensions outlined in the CSM (Hagger & Orbell, 2003; Leventhal et al., 2003; Leventhal et al., 1980; Leventhal & Nerenz, 1985; Meyer, Leventhal, & Gutmann, 1985).

Since then, a range of multidimensional instruments have been developed to facilitate more standardised assessment of illness perceptions. These include the Implicit Models of Illness Questionnaire (Schiaffino & Cea, 1995; Turk, Rudy, & Salovey, 1986), the Illness Perception Questionnaire (Weinman, Petrie, Moss-Morris, & Horne, 1996), the Revised Illness Perception Questionnaire (Moss-Morris et al., 2002), the Brief Illness Perception Questionnaire (Broadbent, Petrie, Main, & Weinman, 2006), the Illness Cognition Questionnaire (Evers et al., 2001), and the Illness Cognitions Scale (Berk et al., 2012). Characteristics of these instruments are outlined below.

1.5.1. The Implicit Models of Illness Questionnaire

The Implicit Models of Illness Questionnaire (IMIQ) was developed by Turk et al. (1986) as a measure of illness perceptions and to examine the possible presence of a generic cognitive structure of illness perceptions that applies across illnesses and populations. Turk et al. (1986) described construction of the IMIQ as an attempt to operationalise the constructs of cognitive representation *identity*, *timeline*, *consequences*, and *cause* defined by Leventhal et al. (1980), and *cure* as defined by (Lau & Hartman, 1983). In addition, the scale included items relating to personal responsibility and disruptiveness (Turk et al., 1986).

Factor analysis of the original IMIQ (items = 38) revealed four dimensions of illness representation: *seriousness* (beliefs about the degree to which an illness is serious, contagious, or requires medical attention), *personal responsibility* (beliefs about who or what might be responsible for illness onset), *controllability* (beliefs about the extent to which the illness is controllable by the individual or medical treatment), and *changeability* (beliefs about whether symptoms and other aspects of the illness change over time). The final scale consisted of 24 items (*seriousness*, 9 items; *personal responsibility*, 8 items; *controllability*, 5 items; and *changeability*, 2 items).

Later research conducted by Schiaffino and Cea (1995) examined the factor structure of the IMIQ when applied across illness populations (rheumatoid arthritis, multiple sclerosis, and human immunodeficiency virus). They hypothesised that factor analysis would either reproduce the factor structure described in Turk et al. (1986), reflect the five dimensions of the CSM, or reveal an entirely different factor structure. A unique four-factor solution was identified, although dimensions closely aligned with those described by Turk et al. (1986).

It had been the authors' initial intention to produce a valid measure of illness representations as detailed in the CSM (Schiaffino & Cea, 1995). Although the resultant factor structure was not entirely consistent with theoretical and empirical evidence underlying the model, Schiaffino and Cea (1995) comment that the "spirit of the common-sense model" (p. 544) was captured, to some extent. Given that their analysis was performed using data collected from three different illness groups, they suggest that the search for an underlying factor structure that is common to all illnesses may be misguided, as some items and issues may be more salient for certain illnesses (Schiaffino & Cea, 1995).

1.5.2. The Illness Perception Questionnaire, the Revised Illness Perception Questionnaire, and the Brief Illness Perception Questionnaire

Following this, Weinman et al. (1996) derived items from the CSM to produce a measure that assessed the five dimensions of illness representation (see Section 1.4.2.). The Illness Perception Questionnaire (IPQ) was designed as a general measure of illness perceptions that could be used across illness groups in individuals experiencing chronic illness. It has a core set of items, but additional items can be included for specific illnesses or health threats. Items in the *illness identity* subscale are rated on a four-point Likert scale from 1 = *never* to 4 = *all of the time*. Remaining subscales are rated on a five-point Likert scale from 1 = *strongly disagree* to 5 = *strongly agree.*

Moss-Morris et al. (2002) revised the original IPQ to address minor psychometric problems with two subscales and to include additional subscales assessing *illness coherence* and *emotional representations*. Research conducted with the original IPQ revealed problems with the internal consistency of subscales assessing perceptions relating to the *timeline* and *cure/control* of the illness. In particular, evidence of low internal inconsistency in the *timeline* subscale suggested that the number of items needed to be increased. Furthermore, cyclical timeline beliefs had not been considered in the original IPQ. Therefore, the *timeline* subscale in the revised version of the IPQ (the Revised Illness Perception Questionnaire; IPQ-R) was split into *timeline acute/chronic* and *timeline cyclical*. In addition to this, factor analysis revealed that items in the *cure/control* subscale loaded onto two separate factors. One of these components related to personal control and self-efficacy beliefs, and the other component was concerned with belief in the treatment or recommended medical advice (i.e., response efficacy). Because of this, the *cure/control* subscale was split into two separate subscales in the IPQ-R. These were *personal control* and *treatment control*.

As described in Section 1.4.2., the CSM proposes that individuals develop parallel cognitive and emotional representations in response to an illness or health threat. Combined, these representations influence the individual’s choice of coping strategies, and whether they employ problem-based or emotion-focused approaches. However, the original IPQ was designed to only assess cognitive illness representations, and a subscale assessing emotional representations was not included. Moss-Morris et al. (2002) argued that this was a significant limitation of the IPQ and its capacity to describe an individual’s response to illness. To address this, the *emotional representations* subscale was included in the IPQ-R to assess emotional responses specifically generated by the illness.

A number of other changes were also made. Moss-Morris et al. (2002) included an *illness coherence* subscale to assess the extent to which an individual’s illness representation provided a coherent understanding of the illness or the extent to which the illness “makes sense” to the individual.

They also reported that the *cause* subscale could be further divided into *psychological attributions, risk factors, immune system factors,* and *chance factors*. As with the original IPQ, the *identity* and *cause* subscales could be modified for specific illness groups by including symptoms and possible causes specific to the illness of interest.

A shortened version of the IPQ-R (the Brief IPQ), comprised of only single-item measures for each subscale, was developed to reduce respondent burden and to enable a quick assessment of illness perceptions when time is limited – particularly because the questionnaire is generally completed by individuals with significant illness (Broadbent et al., 2006).

1.5.3. The Illness Cognition Questionnaire

The Illness Cognition Questionnaire (ICQ) was developed to assess three generic illness perceptions thought to reflect different ways of evaluating the aversive nature of a chronic illness (Evers et al., 2001). The three subscales were based on a priori constructs of *helplessness* (e.g., “My illness limits me in everything that is important to me”), *acceptance* (e.g., “I can handle the problems related to my illness”), and *perceived benefits* (e.g., “My illness has taught me to enjoy the moment more”). Factor analysis of the ICQ (items = 18) confirmed that the structure of the instrument was consistent with the three dimensions of illness perceptions proposed and, moreover, was consistent across groups with rheumatoid arthritis and multiple sclerosis (Evers et al., 2001). Items were rated on a four-point Likert scale (1 = *not at all*, 2 = *somewhat*, 3 = *to a large extent*, 4 = *completely*) reflecting the extent to which the respondent agrees with the statement.

1.5.4. The Illness Cognitions Scale

The Illness Cognitions Scale (ICS) consists of 17 items measuring illness perceptions and behaviours relating to investment in the “sick role” and difficulty adjusting out of this role. The scale was developed by Berk et al. (2012), based on a review of the literature and clinical experience, with the intention to capture illness perceptions across illness groups. Factor analysis produced one main factor that captured the majority of variance (*illness investment and sick role inertia*) and three sub-factors assessing maladaptive beliefs about, and attitudes to, the illness (*not accepting the illness and resilience to treatment advice, belief in advantages of the illness,* and *belief by the individual that they need more help with their illness*). The scale was rated on a five-point Likert scale from 1 = *strongly disagree* to 5 = *strongly agree*.

1.6. Illness Perceptions in Family Members

Although theories of illness perception were originally developed to explain variation in outcomes in individuals experiencing the illness or health threat, they could also be used to examine the relationships between perceptions, coping, and outcomes in those close to someone with an illness (e.g., AYAs who have a parent with cancer). As discussed earlier in this chapter, the impacts of chronic illness extend beyond the individual diagnosed with the illness; spouses and family members frequently report increased levels of depression, anxiety, and stress (Li, Mak, & Loke, 2012; Mi et al., 2017; Murray et al., 2010; Patterson et al., 2017; Thomson, Bergman, & Hay, 2018), which are comparable to levels reported by patients themselves (Hodges, Humphris, & Macfarlane, 2005). When an individual is diagnosed with an illness, their spouse, family members, and close friends develop their own understanding and representations of the illness. Evidence suggests that spouses, family members, and other informal carers often form illness perceptions that are significantly more negative than those held by the individual with the illness. This has been demonstrated in a range of chronic illnesses, including cancer (A. Richardson, Morton, & Broadbent, 2015; Szentes, Kökönyei, Békési, Bokrétás, & Török, 2017), diabetes (Klein Woolthuis et al., 2013), cardiovascular conditions (Karademas, Zarogiannos, & Karamvakalis, 2010), neurological conditions (Whitehead, Stone, Norman, Sharpe, & Reuber, 2015), anorexia nervosa (Matthews, Lenz, Peugh, Copps, & Peterson, 2018), and schizophrenia (Kuipers et al., 2007).

The impact of these negative perceptions warrants further investigation. Research suggests that the illness perceptions of family members are likely to impact their own coping and psychological outcomes. For instance, Dempster et al. (2011b) found that illness perceptions (assessed with the IPQ-R) explained 27% of variance in anxiety and 33% of variance in depression among family members caring for survivors of oesophageal cancer. Another study found that relatives of individuals with schizophrenia are more likely to experience higher levels of distress if they perceive the illness as uncontrollable, chronic, highly symptomatic, and as having severe consequences (Fortune, Smith, & Garvey, 2011). Similarly, beliefs about the consequences and the chronicity of the disorder were found to be significantly associated with psychological distress and depressive symptoms in parents of children with autism spectrum disorder (Gatzoyia et al., 2014). Beinke, O'Callaghan, Morrissey, and Duregger (2016) reported that perceptions that the condition was cyclical were related to higher levels of psychological distress in mothers of children with cystic fibrosis. Illness perceptions appear to also be important for children who have a parent with a chronic illness; Bogosian, Moss-Morris, Bishop, and Hadwin (2014) found that adolescents experienced poorer psychological outcomes when they perceived their parent’s multiple sclerosis as being chronic and unpredictable, and as having negative consequences.

Importantly, the illness perceptions of informal carers may impact the way that they respond to the illness and provide support (Quinn, Rees Jones, & Clare, 2017). For example, Al Anbar, Dardennes, Prado-Netto, Kaye, and Contejean (2010) explored the relationships between illness perceptions and treatment choices in parents of children with autism spectrum disorder. They found that perceptions of the seriousness of the disorder were associated with the use of educational tools, whereas perceptions of an unpredictable timeline were associated with pharmaceutical treatments (Al Anbar et al., 2010). Illness perceptions may influence the extent to which carers shield the individual with the illness from their emotional reactions. Consistent with this, Johnson et al. (2013) found that perceptions of control were strongly related to protective buffering (i.e., hiding worries, denying concerns, and avoiding disagreement with partner to reduce upset and burden) in women caring for a spouse with type 2 diabetes.

Caregiver burden is another issue that informal carers face. Matthews et al. (2018) found that the subjective experience of having a child with anorexia nervosa was a stronger predictor of caregiver burden than objective indicators of illness severity. Moreover, negative attributions about the individual’s behaviour, such as perceiving behaviour related to the illness to be manipulative, appears to be linked to resentment and depression in carers (Martin-Cook, Remakel-Davis, Svetlik, Hynan, & Weiner, 2003).

Family members are often the main providers of emotional support and personal care to those experiencing illness (outside of medical professionals) (Wu, Mohamed, Winkel, & Diefenbach, 2013). Given the importance of this role and the potential impact on the carer, it is worth considering the accuracy of the carer’s illness perceptions. Particularly because illness perceptions may influence the carer’s own chosen behaviours as well as the person with the illness. For example, Quinn et al. (2017) suggest that individuals may be discouraged from attempting to manage their illness if an important other believes that nothing can be done to treat it. Consistent with this, Sim and Matthews (2012) note that for treatment of anorexia nervosa in adolescent girls to be effective, parents must have a coherent understanding of the illness and also believe it to be treatable. Moreover, the way that carers respond to the illness may influence the individual’s experience of their illness – especially for children diagnosed with a chronic illness. Ringlever, Otten, van Schayck, and Engels (2012) found that mothers’ perceptions of illness, specifically relating to aspects of identity, consequences, concern, and emotional influence, were associated with children’s asthma symptoms and symptom-related quality of life.

There is other evidence that family members’ illness perceptions have implications for the well-being of the individual with the illness. Wu et al. (2013) found that interactions between spousal beliefs about treatment control and the timeline of the illness were associated with quality of life in individuals with prostate cancer. Another study reported that variance in quality of life in individuals with head and neck cancer was explained by interactions between their own illness perceptions and those of their carer (A. Richardson et al., 2015). Findings reported by Dempster et al. (2011b) indicate that carers’ illness perceptions significantly contribute to psychological distress among individuals with oesophageal cancer. Moreover, carer perceptions were found to moderate the relationship between the patients’ perceptions and psychological distress (Dempster et al., 2011b).

Discrepancies between spousal and patient illness perceptions may be responsible for adverse psychological outcomes. Giannousi, Karademas, and Dimitraki (2016) found that differences in perceived consequences were associated with increased psychological symptoms in patients diagnosed with cancer. This is consistent with findings reported by A. Richardson, Morton, and Broadbent (2016a), where discrepancies at diagnosis in perceptions about the timeline, personal control, and illness identity between patients and their carers predicted lower patient health-related quality of life after six months. Combined, these findings suggest that adaptation to chronic illness may be a dyadic process whereby the patient and partner’s well-being is impacted by each other’s understanding of the illness (Giannousi et al., 2016).

Much of this research has been conducted using the CSM as a theoretical framework, but it is important to note that the applicability of the model is dependent on the availability of measurement instruments that are appropriate for use in the population of interest (Heyduck-Weides, Bengel, Farin, & Glattacker, 2019). Moreover, the validity of findings is dependent on the quality of the measurement instruments used (Kimberlin & Winterstein, 2008). Although the IPQ/IPQ-R demonstrates good reliability and validity among individuals diagnosed with the illness (Abubakari et al., 2012; Ashley et al., 2013; Moss-Morris et al., 2002), its appropriateness for family members and informal carers is questionable and the validity of its use within this group remains unclear. Furthermore, it is clear from the literature that illness perceptions are highly individual and that patterns of illness perceptions vary across different illness groups. Hence, there is a need for further research to examine the utility and applicability of the CSM for describing illness perceptions among family members and informal carers of an individual with a chronic illness. In addition to this, research examining the relationships between illness perceptions and psychological outcomes among children with a parent with a chronic illness is limited. Research is required to validate a measure of illness perceptions for use among children of people with chronic illness – and specifically, in the context of the present thesis, cancer.

1.7. Chapter Summary and Rationale for the Research Presented in the Thesis

This chapter has provided a broad overview of research examining the impact of parental cancer on dependent children, the relationships between illness perceptions and physical and psychosocial outcomes, and the potential role that illness perceptions play in predicting coping and psychosocial outcomes among family members of an individual diagnosed with a chronic illness.

The literature reviewed in this chapter highlights several key points. Firstly, children who have a parent with cancer remain an understudied group and knowledge of the factors that impact children's adjustment to parental cancer is limited. Secondly, research examining the role of age as a predictor of psychosocial outcomes has produced inconsistent findings, potentially as a result of differences in the way that children of the same chronological age conceptualise their parent's cancer. Thirdly, although the way that children perceive their parent's cancer also appears to impact their adjustment, there has been very little research conducted to examine the specific relationships between children's illness perceptions and their psychosocial outcomes. Fourthly, a large body of research confirms significant relationships between illness perceptions, coping, and outcomes in individuals diagnosed with an illness and their family members. Finally, and crucially, although a range of instruments have been developed to measure illness perceptions in those diagnosed with the illness, the validity of their use among family members, including children, remains unclear.

In light of these findings, the overall aim of this thesis was to validate an instrument that may be used to assess illness perceptions in AYAs who have a parent with cancer. The specific aims of the research were to:

1. Identify the published instruments developed to measure illness perceptions in family members and carers of people with chronic physical health conditions;
2. Assess and compare the psychometric properties and characteristics of the identified instruments using quality criteria for measurement properties;
3. Provide an in-depth qualitative exploration of illness perceptions among AYAs who have a parent with cancer;

4. Adapt an existing measure of illness perceptions that is appropriate for use in AYAs who have a parent with cancer, based on the findings of the systematic review and qualitative study (conducted to address Aims 1 and 2);
5. Determine the content validity, factor structure, internal consistency, construct validity, and test-retest reliability of the new measure; and,
6. Assess the discriminative validity and screening utility of the measure for identifying AYAs who are at risk of psychological distress and post-traumatic stress symptoms following a parent's cancer diagnosis.

CHAPTER TWO

Comparative systematic review of the psychometric properties of measures of illness perceptions in family members of individuals diagnosed with a chronic physical illness

Statement of Authorship

The following chapter presents a systematic review published online by the Scandinavian Journal of Caring Sciences on 31 January 2020. The authors included PhD candidate, Chloe Fletcher, and her supervisory team: Prof Carlene Wilson, Dr Ingrid Flight, Dr Kate Gunn, and Dr Pandora Patterson. Full citation: Fletcher, C., Flight, I., Gunn, K., Patterson, P., & Wilson, C. (2020). Comparative systematic review of the psychometric properties of measures of illness perceptions in family members of individuals diagnosed with a chronic physical illness. *Scandinavian Journal of Caring Sciences*, Advance online publication. doi: 10.1111/scs.12822

Candidate Statement

I confirm that I was the primary author of the systematic review manuscript, and that guidance and supervision were provided by the co-authors throughout the manuscript preparation, submission, and revision processes. I was responsible for developing the research protocol; conducting the literature search; selecting papers for inclusion based on the eligibility criteria; extracting and analysing data; evaluating methodological quality of the included studies; evaluating quality of psychometric properties of included instruments; and writing the manuscript. I was listed as corresponding author on the submitted manuscript and was also responsible for revising the paper based on reviewer feedback and corresponding with the refereed journal.

Co-authors provided on-going supervision throughout the candidature resulting in this publication. Specific to this study, their role included providing guidance throughout the review process, feedback on drafts of the manuscript, and advice on responding to the journal reviewers. In addition, Prof Carlene Wilson contributed as a second reviewer to evaluation of quality of psychometric properties of the included instruments; Dr Ingrid Flight contributed as a second reviewer to selection of papers for inclusion based on the eligibility criteria and to evaluation of methodological quality of included studies; and Dr Kate Gunn contributed as a second reviewer to selection of papers for inclusion based on the eligibility criteria. The percentage contribution of each author was as follows:

- Research design: Chloe Fletcher (60%), Dr Ingrid Flight (10%), Dr Kate Gunn (10%), Dr Pandora Patterson (10%), and Prof Carlene Wilson (10%)
- Data collection and analysis: Chloe Fletcher (100%)
- Writing and editing: Chloe Fletcher (80%), Dr Ingrid Flight (5%), Dr Kate Gunn (5%), Dr Pandora Patterson (5%), and Prof Carlene Wilson (5%)

2.0. Preamble

As highlighted in Chapter 1, there is substantial evidence that illness perceptions are associated with coping and psychological outcomes in family members caring for an individual with a chronic illness. However, much of this research was conducted using instruments that were originally developed and validated for use in individuals experiencing the illness themselves (such as the Illness Perception Questionnaire and Revised Illness Perception Questionnaire; IPQ/IPQ-R). It is important to acknowledge this distinction because it may have consequences for the validity of findings regarding the relationships between illness perceptions, coping, and psychological outcomes among family members and informal carers. Accurate examination of illness perceptions requires the availability of instruments that are valid for use in the population of interest (Heyduck-Weides et al., 2019; Zamanzadeh et al., 2015).

The overall aim of this thesis was to develop an instrument that may be used to assess illness perceptions in adolescents and young adults (AYAs) who have a parent with cancer. To address this aim, a preliminary systematic review was conducted to identify instruments designed to measure illness perceptions among family members and informal carers of people with chronic physical illnesses. Because the validity of findings is dependent on the quality of the instrument used, the psychometric properties and characteristics of the identified instruments were evaluated using published quality assessment criteria (Terwee et al., 2007). The systematic review was conducted in order to identify gaps in research describing measurement of illness perceptions among family members and informal carers and to identify instruments that may be appropriate to adapt for use among AYAs impacted by parental cancer.

The study presented in Chapter 2 is in the same manuscript form as it was when accepted for publication. Minor modifications have been made to manuscript formatting in line with APA 6th style requirements. The co-author approval form and published manuscript for this study are presented in Appendices A and B, respectively.

Abstract

Although illness perceptions have significant implications for psychological morbidity in those diagnosed with a physical illness, the strength of this relationship in their family members remains understudied. The validity of findings is dependent on the quality of the instruments used; therefore, it is essential that psychometrically robust measures of illness perceptions are available. The purpose of this systematic review was to identify, assess, and compare the psychometric properties of instruments designed to measure illness perceptions in family members of individuals with chronic physical illnesses. A systematic search was conducted using MEDLINE, PubMed, CINAHL, Scopus, and PsycINFO databases, and supplemented with forward and backward searches. Studies were included in the review if they described the development, adaptation, or psychometric evaluation of an instrument designed to measure illness perceptions in family members of an individual with a chronic physical illness. The methodological quality of included studies was assessed using the COSMIN Risk of Bias checklist. The psychometric quality of instruments was evaluated using published quality assessment criteria. Eleven articles describing nine different instruments were included in the review. Almost all instruments were designed for parents of a child with a chronic illness. There was wide variation in the quality of methods used to develop, adapt, or evaluate the instruments and missing information restricted the evaluation of psychometric properties. Further validation is needed for all instruments before meaningful conclusions can be drawn. Findings indicate that measurement of illness perceptions in children or siblings of an individual with a chronic physical illness has been largely ignored. Future research addressing this gap would be an important addition to the current body of work examining illness perceptions in family members.

Introduction

The impacts of chronic illness are far-reaching, extending beyond the person diagnosed with the illness to their parents, siblings, and children. An emerging body of evidence highlights the high levels of distress and psychological morbidity experienced by family members and carers of someone diagnosed with a chronic illness (e.g., Li et al., 2012; Mi et al., 2017; Murray et al., 2010; Patterson et al., 2017; Thomson et al., 2018), with some findings indicating that levels of anxiety and depression experienced by informal carers are comparable to those with the illness and significantly greater than healthy controls (Mitchell, Ferguson, Gill, Paul, & Symonds, 2013; Patterson et al., 2017).

When an individual experiences a health threat, either through identifying a physical symptom or receiving a clinical diagnosis, they form beliefs about the illness that are based on their perception, interpretation, and understanding of the illness and its treatment (Diefenbach & Leventhal, 1996; Hagger & Orbell, 2003; Petrie & Weinman, 2006). These beliefs play an important mediating role between their physical experience of the illness and their subsequent coping and well-being. Differences in physical and psychological outcomes between individuals diagnosed with the same illness can be, at least partly, explained by differences in the way that they perceive and think about their illness (Evers et al., 2001; Hagger & Orbell, 2003; Lazarus, 1999; Leventhal, Diefenbach, & Leventhal, 1992; Petrie & Weinman, 2006).

It is likely that the mechanisms by which illness perceptions influence coping and psychological adjustment are also implicated in outcomes for family members and carers of those with the illness. For example, Beinke et al. (2016) found that perceptions of the cyclical nature of their child’s illness were associated with distress in mothers of young adults with cystic fibrosis. Similar findings have been found in parents who have a child with cancer; parental distress has been linked with perceptions of the stressfulness and severity of the illness, the impact of the illness on the family, the intensity of treatment, and the potential for the illness to be life-threatening (Kazak et al., 2004; Sloper, 2000; Sultan, Leclair, Rondeau, Burns, & Abate, 2016). Some findings indicate that this association is independent of the child's treatment and illness status (Sloper, 2000). Likewise, children's perceptions of the stressfulness of their parent's cancer have been shown to be associated with anxiety and depression (Compas et al., 1996). In adolescents with a parent with multiple sclerosis, beliefs about the impact of the illness on the family environment and the unpredictable course of the illness were found to be strongly correlated with emotional and behavioural difficulties (Bogosian et al., 2014).

A range of theoretical models have been utilised to describe the relationships between illness perceptions and health outcomes (e.g. Lazarus and Folkman's Transactional Model of Stress and Coping (Lazarus & Folkman, 1984), the Health Belief Model (Rosenstock, 1974), and the Theory of Planned Behaviour (Ajzen, 1985)). Diversity in approaches used to conceptualise illness perceptions and describe their relationships with health outcomes has led to the development of a number of multidimensional instruments designed to measure this construct. These include the Illness Cognitions Scale (Berk et al., 2012), the Illness Cognition Questionnaire (Evers et al., 2001), the Illness Perception Questionnaire (Weinman et al., 1996), and the Revised Illness Perception Questionnaire (Moss-Morris et al., 2002), all of which were originally developed to measure illness perceptions among people experiencing ill-health. Since then, the Illness Perception Questionnaire and the Illness Cognition Questionnaire have been adapted to measure illness perceptions in adolescents who have a parent with multiple sclerosis (Bogosian et al., 2014) and parents who have a child with cancer (Sint Nicolaas et al., 2016), respectively. However, neither has been validated for other illnesses and no one instrument has been consistently used across illness populations to assess illness perceptions in family members of an individual with a chronic illness.

To date, a systematic review of instruments developed and validated to measure illness perceptions in family members of an individual with a chronic illness has not been undertaken. The ability to identify illness perceptions in family members of people with a chronic illness is essential, given the significant impact on carers and supporters and the growing body of research indicating that these beliefs may be important predictors or mediators of psychological outcomes in this cohort. Moreover, the validity of findings in this area is dependent on the quality of the instruments used; it is essential that psychometrically robust measures of illness perceptions are available for use with family members, particularly where these instruments are used to screen for potential psychological morbidity. Therefore, the purpose of this review was to:

(1) Identify instruments designed to measure illness perceptions in family members and carers of individuals with chronic physical illnesses;
(2) Assess and compare the psychometric properties of identified instruments using published quality assessment criteria (Terwee et al., 2007); and
(3) Assess the methodological quality of studies describing the development and/or evaluation of identified instruments using the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) Risk of Bias checklist (Mokkink et al., 2018; Prinsen et al., 2018; Terwee et al., 2018).

Given the heterogeneous nature of research exploring illness perceptions, and that the relationship between illness perceptions and psychological outcomes in family members of a person with a chronic illness has attracted little hypothesis testing, this review was not limited to instruments developed with a specific theoretical underpinning. “Illness perceptions” were broadly defined to encompass a diverse range of conceptualisations. Consequently, we identified relevant papers as describing measures that capture an individual’s cognitive and affective representation of a family member’s illness.

Method

The review protocol was registered at the PROSPERO international prospective register of systematic reviews (registration number: CRD42017072845).

Search strategy. Databases MEDLINE (from 1946-), PubMed (excluding MEDLINE; from 1946-), Cumulative Index to Nursing and Allied Health Literature (CINAHL), Scopus (Social Sciences and Humanities subject areas; from 1823-), and PsycINFO (from 1806-) were searched using a combination of terms relating to illness cognitions, perceptions or beliefs, measurement, family members or carers, and chronic illness or disease. Searches were limited to studies conducted with human participants and published in English. The full search string run in MEDLINE is outlined in Table 1. The search was performed on 27 July 2017.

Following systematic screening procedures, forward and backward searching techniques were employed to identify relevant studies that cited, or were cited by, studies that were included. Google Scholar was used to identify studies that had cited included studies and the reference lists of included studies were hand searched. When an included study described the adaptation or evaluation of an existing instrument, the reference list was searched to identify the original development study.

Inclusion criteria. Studies were included if they were published in peer-reviewed journals; they described the development, adaptation, or psychometric evaluation of an instrument; the instrument was designed to measure illness perceptions; the instrument was designed for use in family members or carers of an individual; the individual had been diagnosed with a chronic physical illness; and there was a hypothesised predictive relationship between the construct being measured and psychological outcomes in the respondent.

Table 1

Advanced Search String Run in MEDLINE

1	(illness cognition* or illness perception* or illness belief* or cognition* or perception* or belief*).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
2	(scale or instrument or measure or survey or questionnaire or assessment or checklist).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
3	(family member or parent or mother or father or m*m or dad or child or children or adolescent or young adult).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
4	(chronic illness or chronic disease or palliative or terminal or cancer or diabetes or multiple sclerosis or heart disease or stroke or asthma or arthritis or osteoporosis or chronic obstructive pulmonary disease or chronic kidney disease).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
5	1 and 2 and 3 and 4
6	Limit 5 to (English language and humans)
7	exp "Surveys and Questionnaires"/st [Standards]
8	health care surveys/ or questionnaires/
9	7 or 8
10	6 and 9

Exclusion criteria. Studies were excluded if the instrument was designed for use in the individual diagnosed with the illness or measured illness perceptions relating to mental illness. Studies not published in English were also excluded if no translation was readily available.

Study selection. In total, 6,360 articles were retrieved from initial searches on MEDLINE (*n* = 2,092), PubMed (excluding MEDLINE; *n* = 2,246), CINAHL (excluding MEDLINE; *n* = 1,650), Scopus (*n* = 347), and PsycINFO (*n* = 25; see Figure 3 for PRISMA flow diagram of study selection process). Citations were imported into reference management software EndNote and duplicates (*n* = 623) were removed. Titles and abstracts were screened by the first author (CF) for relevance according to the inclusion and exclusion criteria resulting in a further 5,683 citations being excluded. A subset (10%) of abstracts were screened independently by a second reviewer (IF), with excellent agreement (95%) observed between reviewers (Kappa = 0.877, $p < 0.001$). Following title and abstract screening, 54 full text articles were retrieved and considered for inclusion. A second reviewer (KG) independently screened a subset (10%) of full text articles, with 100% agreement observed between reviewers. Fifty articles were excluded for the reasons outlined in Figure 3 and the remaining 4 were included in the review. Forward and backward searches were conducted for each of the included studies and an additional 7 articles were identified and included in the review.

Data extraction. Descriptive information was extracted for each of the included studies. Information related to characteristics of the study (type of study, sample type, sample size, mean age, gender distribution, and the country where the study was conducted) and characteristics of the instrument (instrument name, construct measured, the population the instrument was developed or adapted for, number of subscales, number of items, scale type, response options, sample items, information about instrument development, and the available languages of the instrument). Psychometric data addressing the relevant quality criteria (described below and in Table 2) were also extracted.

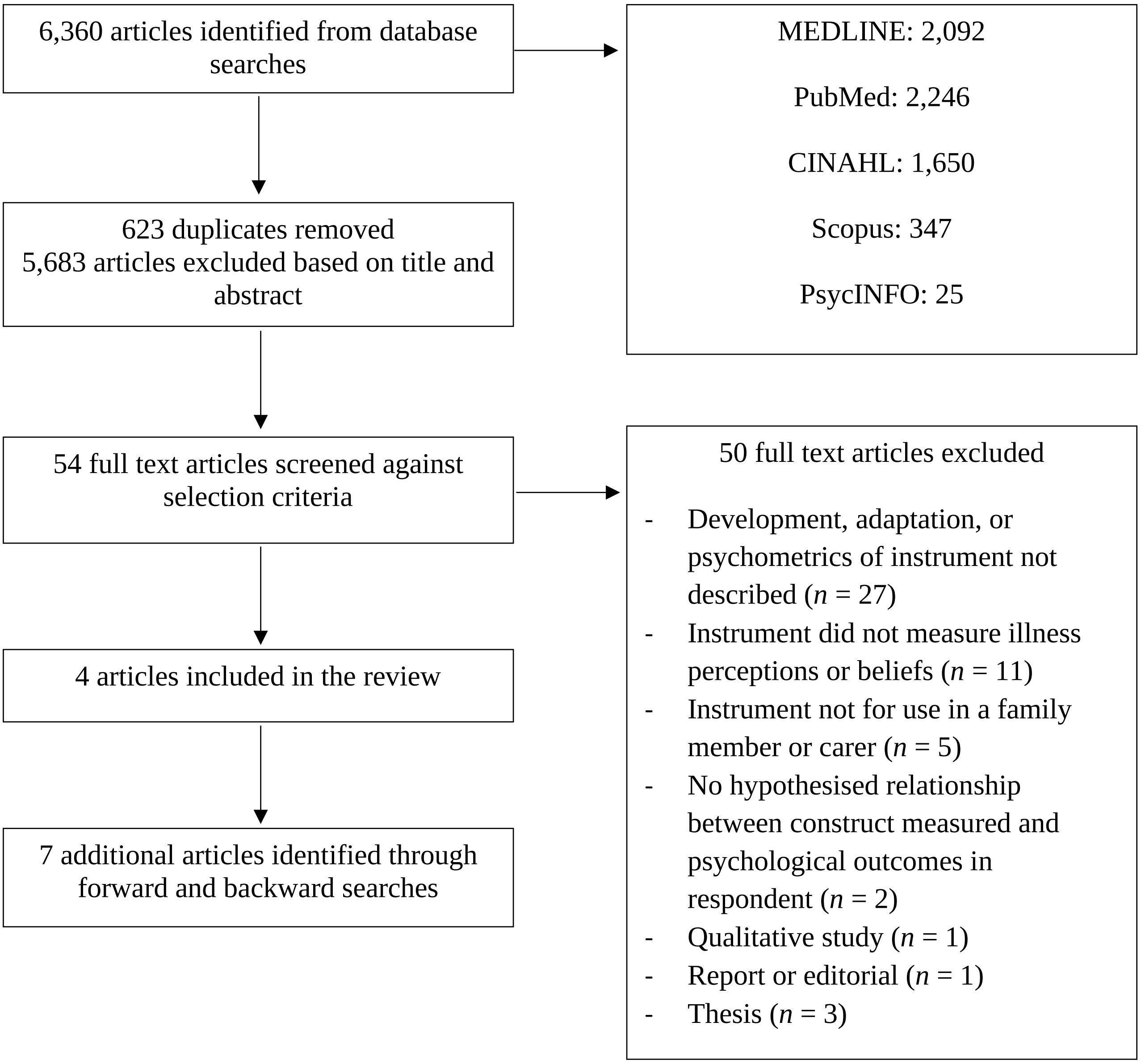


Figure 3. Flow Diagram of Study Selection Process. CINAHL = Cumulative Index to Nursing and Allied Health Literature; *n* = number of articles.

Quality assessment. The psychometric properties of each instrument were evaluated using published quality assessment criteria originally developed for the purpose of evaluating the psychometric properties of health status measures (Terwee et al., 2007). The criteria address content validity, structural validity, internal consistency, criterion validity, construct validity, measurement error, test-retest reliability, responsiveness, floor and ceiling effects, and interpretability. Each study was assessed individually, with findings rated as positive (+), negative (-), or indeterminate (?) for each property (see Table 2 for detailed quality criteria). Only properties that were reported in the original study were assessed.

The methodological quality of included studies was evaluated using the COSMIN Risk of Bias checklist (Mokkink et al., 2018; Prinsen et al., 2018; Terwee et al., 2018). The COSMIN checklist is a standardised tool for evaluating the methodological quality of studies describing measurement properties. It was developed in an international Delphi study involving 57 researchers with backgrounds in epidemiology, psychometrics, qualitative research, health care, and development and evaluation of outcome measurement instruments (Mokkink, Prinsen, Bouter, de Vet, & Terwee, 2016; Mokkink et al., 2010).

The checklist contains ten boxes used to assess whether a study meets standards for good methodological quality for testing psychometric properties. Where reported in the original study, methods used to evaluate structural validity, internal consistency, cross-cultural validity, reliability, measurement error, criterion validity, construct validity, and responsiveness were assessed using the designated COSMIN boxes. Methods were rated as very good, adequate, doubtful, or inadequate according to the criteria outlined in each box. As recommended in the COSMIN guidelines (Mokkink et al., 2018; Prinsen et al., 2018; Terwee et al., 2018), the ‘worst score counts’ method was used to determine the overall rating for the quality of methods used to evaluate each psychometric property. This meant that the lowest rating of any of the items in the designated box for each psychometric property was taken as the overall score for that particular property. Boxes for properties that were not evaluated in the study did not receive a score at all. Most studies reported very little information on the methods used to develop the instrument and assess content validity, meaning that the boxes for instrument development and content validity could not be completed. Instead the development of the instrument and any cognitive interview studies or other pilot studies were qualitatively described and outlined in the results (e.g., instrument development and content validity testing of the Perceptions of Parental Illness Questionnaire is described in Table 4 as follows: *"Questionnaire items were developed based on interviews conducted with adolescents with a parent with multiple sclerosis and Leventhal's Common-Sense Model of Self-Regulation. Questionnaire items were refined based on cognitive interviews conducted with six adolescents. Two subscales were removed and item wording was modified based on feedback."*)

Assessments were conducted by the first author (CF), with a subset (10%) of studies independently assessed by a second reviewer (CW assessed the quality of psychometric properties of instruments and IF assessed the methodological quality of included studies). At each stage of assessment, disagreement in ratings were discussed among reviewers until consensus was reached.

It should be noted that although the quality of methodology used to evaluate content validity could not be properly assessed, following the criteria developed by Terwee et al. (2007), instruments were given a positive rating for content validity if the development involved input from the target population.

Table 2

Quality Criteria for Assessment of Psychometric Properties

	Property	Definition	Quality criteria[a, b]
1	Content validity	The extent to which the domain of interest is comprehensively sampled by the items in the questionnaire	+ A clear description is provided of the measurement aim, the target population, the concepts that are being measure, and the item selection AND target population and (investigators OR experts) were involved in item selection; ? A clear description of above-mentioned aspects is lacking OR only target population involved OR doubtful design or method; - No target population involvement; 0 No information found on target population involvement
2	Structural validity	The extent to which scores on a particular questionnaire are an adequate reflection of the dimensionality of the construct to be measured	+ Factors analyses performed on adequate sample size (7 * # items and ≥100); ? No factor analysis OR doubtful design or method; 0 No information found on structural validity
3	Internal consistency	The extent to which items in a (sub)scale are intercorrelated, thus measuring the same construct	+ Cronbach’s alpha(s) calculated per dimension AND Cronbach’s alpha(s) between 0.70 and 0.95; ? Cronbach’s alpha(s) NOT calculated for each dimension OR doubtful design or method; - Cronbach’s alpha(s) <0.70 or >0.95, despite adequate design and method; 0 No information found on internal consistency
4	Criterion validity	The extent to which scores on a particular questionnaire relate to a gold standard	+ Convincing arguments that gold standard is “gold” AND correlation with gold standard ≥0.70; ? No convincing arguments that gold standard is “gold” OR doubtful design or method; - Correlation with gold standard <0.70, despite adequate design and method; 0 No information found on criterion validity
5	Construct validity	The extent to which scores on a particular questionnaire relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are being measured	+ Specific hypotheses were formulated AND at least 75% of the results are in accordance with these hypotheses; ? Doubt design or method (e.g. no hypotheses); - Less than 75% of hypotheses were confirmed, despite adequate design and methods; 0 No information found on construct validity
6	Measurement error	The extent to which scores on repeated measures are close to each other (absolute measurement error)	+ SDC < MIC OR MIC outside the LOA OR convincing arguments that agreement is acceptable; ? Doubtful design or method OR (MIC not defined AND no convincing arguments that agreement is acceptable); - SDC ≥ MIC OR MIC equals or inside LOA, despite adequate design and method; 0 No information found on agreement
7	Reliability	The extent to which respondents can be distinguished from each	+ ICC or weighted Kappa ≥ 0.70; ? Doubtful design or method (e.g. time interval not mentioned);

	Property	Definition	Quality criteria[a, b]
		other, despite measurement errors (relative measurement error)	- ICC or weighted Kappa < 0.70, despite adequate design and method; 0 No information found on reliability
7	Responsiveness	The ability of a questionnaire to detect clinically important changes over time	+ SDC or SDC < MIC OR MIC outside the LOA OR RR > 1.96 OR AUC ≥ 0.70; ? Doubtful design or method; - SDC or SDC ≥ MIC OR MIC equals or inside LOA OR RR ≤ 1.96 OR AUC < 0.70, despite adequate design and methods; 0 No information found on responsiveness
8	Floor and ceiling effects	The number of respondents who achieved the lowest or highest possible score	+ ≤15% of the respondents achieved the highest or lowest possible scores; ? Doubtful design or method; - >15% of the respondents achieved the highest or lower possible scores, despite adequate design and methods; 0 No information found on floor and ceiling effects
9	Interpretability	The degree to which one can assign qualitative meaning to quantitative scores	+ Mean and SD scores presented of at least four relevant subgroups of respondents and MIC defined; ? Doubtful design or method OR less than four subgroups OR no MIC defined; 0 No information found on interpretation

Note. MIC = minimal important change; SDC = smallest detectable change; LOA = limits of agreement; ICC = intraclass correlation; RR = responsiveness ratio; AUC = area under the curve; SD = standard deviation.

[a] + = positive rating; ? = indeterminate rating; - = negative rating; 0 = no information available.

[b] Doubtful design or method = lacking of a clear description of the design or methods of the study, sample size smaller than 50 participants (should be at least 50 in every (subgroup) analysis), or any important methodological weakness in the design of the study.

Table adapted from Terwee et al. (2007).

Results

Instruments identified. The search string was developed to identify studies that used specific terms relating to illness cognitions, perceptions, or beliefs. During the screening process, we identified a number of instruments that had been developed to measure perceptual constructs that were conceptually related, although not specifically described as illness cognitions, perceptions, or beliefs by the study authors. These constructs were worry, uncertainty, chronic sorrow, perceived emotional resources, and perceived impact of illness.

Worry was defined by the study authors as “thoughts and images that relate to possible negative or threatening outcomes” and they predicted a clear relationship between worry, coping, and psychological outcomes in the respondent (de Vet & Ireys, 1998). Mishel (1983) proposed uncertainty as “a major perceptual variable influencing parents’ experiences during their child’s illness” that affects appraisal of the illness and the ability to cope. Bonner et al. (2006) sought to develop a measure of the important aspects of psychological adjustment that encompassed constructs of uncertainty, chronic sorrow (defined as “prolonged feelings of grief, disappointment, and fear in response to the continual losses experienced over the trajectory of an illness”), and perceived emotional resources (defined as “cognitive appraisals of internal resources and perceived self-efficacy”). Bemister (2014) developed a measure of perceived psychosocial impact of illness that captured parents’ sense of guilt and blame in relation to their child’s diagnosis. Because each of these constructs captures a cognitive or affective representation of a family member’s illness, and a clear relationship between each construct and coping in the face of a health threat was predicted, these instruments were included in the review. For this reason, a majority of the included articles were identified through forward and backward searching techniques, rather than through the initial database search.

Table 3 describes the eleven articles included in the review. Five articles described the development and initial psychometric properties of an original instrument. An additional four articles described the validation of adapted (three articles) or translated (one article) instruments. The remaining two articles provided further validation of an existing instrument. Instruments were primarily developed to be applicable for any chronic illness, although three instruments were developed for specific use in family members of an individual with cancer (Family Illness Beliefs Inventory; Kazak et al., 2004), multiple sclerosis (Perceptions of Parental Illness Questionnaire; Bogosian et al., 2014), and perinatal stroke (Parental Outcome Measure; Bemister et al., 2014) (see Table 4). All of the instruments were designed for parents with a child with a chronic illness, except for the Perceptions of Parental Illness Questionnaire (Bogosian et al., 2014).

Table 3

Characteristics of Included Studies

Instrument/Study	Type of study	Sample type	Sample size	Age, mean (SD)	Female (%)	Country
Parental Outcome Measure						
(Bemister et al., 2014)	Development and validation	Parents of children with perinatal stroke	Time 1: 110, time 2: 58	39.5 (7.4) years	Time 1: 74.5	Canada
Perceptions of Parental Illness Questionnaire						
(Bogosian et al., 2014)	Development and validation	Adolescents with a parent with multiple sclerosis	Development: 15, cognitive interviews: 6, validation: 104	Validation: 15.4 (1.97) years	59.6	UK
Parental Experience of Child Illness						
(Bonner et al., 2006)	Development and initial validation	Parents of children with brain tumours	Pilot testing: 6, validation: 148	39.5 (5.9) years	82.6	USA
(Bonner, Hardy, Willard, Hutchinson, & Guill, 2008)	Validation	Parents of children diagnosed with cancer	Time 1: 125, time 2: 75	39.6 years	Time 1: 80.8	USA
Maternal Worry Scale						
(de Vet & Ireys, 1998)	Development and psychometric evaluation	Mothers of children with juvenile rheumatoid arthritis, sickle cell anaemia, or diabetes	140	-	100	USA
Family Illness Beliefs Inventory						
(Kazak et al., 2004)	Development and validation	Parents of children with cancer	119 mothers, 56 fathers	-	68	USA
Parental Perception of Uncertainty Scale						
(Mishel, 1983)	Adaptation and validation	Parents of children hospitalised with an illness	272	-	80.1	USA
(Molzon et al., 2014)	Validation	Parents of children with cancer	183	-	86.9	USA
Parental Perception of Uncertainty Scale-Diagnosis						
(Santacroce, 2001)	Adaptation and validation	Mothers of infants who are HIV seropositive	Content validity testing: 11, preliminary psychometric evaluation: 25	29 years	100	USA
Parental Perception of Uncertainty Scale-Chinese						
(Ye et al., 2017)	Adaptation and validation of translated version	Parents of children diagnosed with cancer	377	-	-	China
Illness Cognition Questionnaire-Parent version						
(Sint Nicolaas et al., 2016)	Adaptation and validation	Parents of children with cancer	242	39.98 (6.30) years	76	The Netherlands

Table 4

Characteristics of the Included Instruments

Instrument	Construct	Target Population	Dimensions (number of items)	Total number of items	Response options (range)	Instrument development and content validity testing	Language
Parental Outcome Measure (Bemister et al., 2014)	Perceived Impact of Illness, Guilt, and Blame	Parents of children with perinatal stroke	Three dimensions – Psychosocial Impact (17) Guilt (4) Blame (5)	31 items, revised to 26 items based on exploratory factor analysis	5-point scale (0 = strongly disagree to 4 = strongly agree)	Questionnaire items were developed by a multidisciplinary team based on their experience with perinatal stroke and the literature on caring for children with disabilities. A draft 34-item questionnaire was piloted among 10 health care professionals and six parents of children with perinatal stroke to collect feedback on the content and face validity and wording of the questionnaire. The questionnaire was modified based on the feedback. The resulting questionnaire consisted of 31 items. This was later revised to 26 items based on exploratory factor analysis.	English
Perceptions of Parental Illness Questionnaire (Bogosian et al., 2014)	Illness Beliefs	Adolescents with a parent with multiple sclerosis	11 dimensions – Internal items: Emotional representation (5) Adolescents' control (4) Negative consequences for family (3) Positive consequences for adolescents (4) Negative consequences for adolescents (3) Chronic timeline (4) Unpredictable timeline (2) Causal items: Psychological attributions (2) Central nervous system (2) External/environmental attributions (2) Hereditary/chance (2)	33 items	Not described	Questionnaire items were developed based on interviews conducted with adolescents with a parent with multiple sclerosis and Leventhal's Common-Sense Model of Self-Regulation. Questionnaire items were refined based on cognitive interviews conducted with six adolescents. Two subscales were removed and item wording was modified based on feedback.	English
Parental Experience of Child Illness (Bonner et al., 2006; Bonner et al., 2008)	Chronic Sorrow, Uncertainty, and Emotional Resources	Parents of children with a chronic illness	Four dimensions – Emotional Resources (3) Guilt and Worry (11) Unresolved Sorrow and Anger (7) Long-term Uncertainty (4)	25 items	5-point scale (0 = never to 4 = always)	A preliminary list of items reflecting issues relevant to parenting a child with chronic illness was developed by a multidisciplinary team. The initial 60-item draft covered theoretical domains derived from the literature. Discussion among research team	English

Instrument	Construct	Target Population	Dimensions (number of items)	Total number of items	Response options (range)	Instrument development and content validity testing	Language
						resulted in removal of 24 items that were redundant, ambiguous, or had low relevance to theoretical constructs. The remaining 36 items were reviewed by six parents of children diagnosed with cancer. A further 11 items were removed following feedback provided by the parents.	
Maternal Worry Scale (de Vet & Ireys, 1998)	Worry	Mothers of children with a chronic illness	One dimension – Maternal worry (11)	11 items	4-point scale (1 = not at all to 4 = most of the time)	Questionnaire items were developed through discussions with five mothers of young adults who had arthritis since childhood. Mothers were participants in a parent support intervention. The theme of "worrying about your kid" emerged repeatedly throughout group discussions. The content of worries were recorded over a six-month period and were used to develop an initial set of 30 items. Additional items were included based on previous research and clinical experience. Items were worded to apply to children with any ongoing physical illness. The initial set of items was piloted among five mothers of children with arthritis. Items that were redundant or related to infrequently mentioned worries were removed or combined with other items to produce the final 11-item questionnaire.	English
Family Illness Beliefs Inventory (Kazak et al., 2004)	Illness Beliefs	Parents of children with cancer	Five dimensions – Treatment-related Suffering (11) Death and Devastation (8) Caregiver Competence (9) Connection (8) Finding Meaning (5)	41 items	Card-sort method, with cards sorted into one of four piles: 1 = not at all true for me, 2 = just a little bit true for me, 3 = pretty much true for me, and 4 = very true for me	A set of 75 items was developed by a multidisciplinary team based on clinical experience and the literature. Items were structured in six important areas: affect, finding meaning, optimism, self-efficacy, connection, and illness-specific beliefs. Items were worded to reflect growth promoting and growth inhibiting beliefs. Items were reviewed by five parents of pediatric oncology patients. One item relating to fear of relapse was added based on their feedback.	English
Parental Perception of Uncertainty Scale (Mishel, 1983; Molzon et al., 2014)	Uncertainty	Parents of children hospitalised with a serious illness	One dimension – Global uncertainty (25)	25 items in final structure proposed by Molzon et al. (Molzon et al., 2014)	5-point scale (1 = strongly disagree to 5 = strongly agree)	The original Mishel Uncertainty in Illness Scale (Mishel, 1981) was modified to produce a measure of perceived uncertainty in parents. Items were modified from the original format and reviewed by a group of pediatric nurses for content validity. Items were evaluated to assess relevance to the	English

Instrument	Construct	Target Population	Dimensions (number of items)	Total number of items	Response options (range)	Instrument development and content validity testing	Language
						pediatric hospitalisation experience and parental concerns.	
Parental Perception of Uncertainty Scale-Diagnosis (Santacroce, 2001)	Uncertainty	Parents of children undergoing diagnosis of a serious illness	One dimension – Global parental uncertainty (24)	32 items in original questionnaire, revised to 24 items after psychometric analysis	As above	The original Parental Perception of Uncertainty Scale was modified to produce a measure of parental uncertainty during diagnosis. Content validity was assessed by five mothers of seropositive infants undergoing diagnosis for perinatally acquired HIV infection and six advanced practice nurses. Mothers and nurses were asked to provide feedback on the face validity of the questionnaire, comment on the clarity of items, indicate items that seemed redundant or unrelated, and suggest additional items or alternative wording.	English
Parental Perception of Uncertainty Scale-Chinese (Ye et al., 2017)	Uncertainty	Parents of children with cancer	Two dimensions – Ambiguity (9) Comprehension (5)	19 items in original questionnaire, revised to 14 items after psychometric analysis	As above	The Parental Perception of Uncertainty Scale was translated into Chinese by two researchers. The questionnaire was revised by removing redundant or irrelevant items through discussion amongst six psychology professors and nine clinical nursing professors. Content validity was assessed using the content validity index.	Chinese
Illness Cognition Questionnaire-Parent version (Sint Nicolaas et al., 2016)	Illness Beliefs	Parents of children with a chronic illness	Three dimensions – Helplessness (6) Acceptance (6) Perceived Benefits (6)	18 items	4-point scale (1 = not at all to 4 = completely)	The original Illness Cognition Questionnaire was modified to measure illness cognitions in parents with a child with cancer. No information available on content validity.	Full questionnaire published in English, but validated in Dutch

Psychometric properties of instruments. Table 5 summarises the quality of psychometric properties of instruments evaluated in included studies.

Content validity. Most instruments were comprised of items generated by the researchers, informed by literature reviews and/or input from clinical experts, and involved the target population in item selection through cognitive interview studies or other pilot testing. Both strategies resulted in a positive score for content validation. The Parental Perception of Uncertainty Scale and the Illness Cognition Questionnaire-Parent Version were both adapted from existing measures that were originally developed to measure uncertainty and illness cognitions in the individual with the illness. Neither Mishel (1983) nor Sint Nicolaas et al. (2016) described including the new target population (i.e. parents) during the adaptation process to ensure content validity. Similarly, the new target population was not involved during the translation of the Parental Perception of Uncertainty Scale to Chinese (Ye et al., 2017). The remaining studies both involved further validation of an existing measure and did not include additional assessment of content validity.

Structural validity. In a large proportion of the included studies, factor analyses were conducted using an inadequate sample size (i.e. if the sample size was less than 7x the number of items OR less than 100) (Bemister et al., 2014; Bogosian et al., 2014; Bonner et al., 2006; Kazak et al., 2004; Molzon et al., 2014). These studies were given an indeterminate rating. Bonner et al. (2008) and Santacroce (2001) did not conduct factor analysis and were also given indeterminate ratings.

Internal consistency. Cronbach’s alphas were reported to be ≥0.70 for all subscales in a number of studies (Bemister et al., 2014; Bonner et al., 2006; Bonner et al., 2008; de Vet & Ireys, 1998; Kazak et al., 2004; Mishel, 1983; Molzon et al., 2014; Sint Nicolaas et al., 2016; Ye et al., 2017). Bogosian et al. (2014) reported a Cronbach’s alpha of 0.64 for the Chronic Timeline subscale of the Perceptions of Parental Illness Questionnaire.

Santacroce (2001) did not report separate Cronbach's alphas for each unidimensional subscale of the Parental Perception of Uncertainty Scale-Diagnosis.

Criterion validity. There is no apparent "gold standard" for validation of illness perceptions in family members of an individual with a chronic illness. Although several studies reported that they had assessed criterion validity, many used a comparator measure that may have been more appropriate to assess construct validity (Bemister et al., 2014; Bogosian et al., 2014; Ye et al., 2017). These studies were assessed using criteria for construct validity.

Construct validity. Construct validity was assessed for all of the instruments, however only seven studies received a positive rating (Bemister et al., 2014; Bogosian et al., 2014; de Vet & Ireys, 1998; Kazak et al., 2004; Santacroce, 2001; Sint Nicolaas et al., 2016; Ye et al., 2017). The remaining studies were given an indeterminate rating because they did not outline specific theoretically-derived hypotheses (Bonner et al., 2006; Bonner et al., 2008; Mishel, 1983; Molzon et al., 2014). Construct validity was assessed primarily by examining convergent validity; specifically, the extent to which scores on the instrument correlated with measures of psychological outcomes, including the Hospital Anxiety and Depression Scale (Bemister et al., 2014; Sint Nicolaas et al., 2016), the Profile of Mood States (Sint Nicolaas et al., 2016), the Strengths and Difficulties Questionnaire (Bogosian et al., 2014), the Brief Symptom Inventory (Bonner et al., 2006; Kazak et al., 2004; Molzon et al., 2014), the Impact of Event Scale (Bonner et al., 2006; Kazak et al., 2004; Molzon et al., 2014), the Beck Depression Inventory (de Vet & Ireys, 1998), and the Psychiatric Symptom Index (de Vet & Ireys, 1998).

Measurement error. The standard error of measurement was not calculated for any of the instruments.

Reliability. Test-retest reliability was assessed for four of the instruments (Bemister et al., 2014; Bonner et al., 2008; de Vet & Ireys, 1998; Ye et al., 2017), all of which assessed test-retest reliability using a Pearson or Spearman correlation coefficient. According to the criteria developed by Terwee et al. (2007), this is not considered to be an adequate reliability parameter because systematic differences are not taken into account. Terwee et al. (2007) recommend that test-retest reliability is assessed using the intraclass correlation coefficient (with a cut-off of >0.70) and a sample size of >50. Despite the study authors reporting large Pearson correlation coefficients, following Terwee's criteria, indeterminate ratings were given for the Parental Outcome Measure (r = 0.87; Bemister et al., 2014), the Parental Experience of Child Illness (r = 0.83 - 0.86 across the four subscales; Bonner et al., 2008), the Maternal Worry Scale (r = 0.84; de Vet & Ireys, 1998), and the Parental Perception of Uncertainty Scale-Chinese version (r = 0.88; Ye et al., 2017).

Responsiveness. Bogosian et al. (2014) reported that they assessed the relationship between illness perceptions at baseline and adjustment at 6 months follow-up, however insufficient information was provided to give a positive or negative rating.

Floor/ceiling effects. Potential floor and ceiling effects were examined for the Parental Outcome Measure (Bemister et al., 2014), the Maternal Worry Scale (de Vet & Ireys, 1998), and the Parental Perception of Uncertainty Scale-Diagnosis (Santacroce, 2001). None of these instruments showed evidence of a floor or ceiling effect.

Interpretability. Three studies were given an indeterminate rating for interpretability because they did not report mean scores and standard deviations for at least four subgroups (Kazak et al., 2004; Mishel, 1983; Sint Nicolaas et al., 2016). Ye et al. (2017) did not provide descriptive statistics for subgroups, but did recommend a cut-off score for the Parental Perception of Uncertainty Scale-Chinese version, suggesting that parents who scored higher than 42.5 may require further attention.

Table 5

Quality of Psychometric Properties of Instruments Assessed in Included Studies

Instrument/Study	Content validity	Structural validity	Internal consistency	Construct validity	Criterion validity	Measurement Error	Reliability	Responsiveness	Floor or ceiling effect	Interpretability
Parental Outcome Measure										
(Bemister et al., 2014)	+	?	+	+	0	0	?	0	+	0
Perceptions of Parental Illness Questionnaire										
(Bogosian et al., 2014)	+	?	-	+	0	0	0	?	0	0
Parental Experience of Child Illness										
(Bonner et al., 2006)	+	?	+	?	0	0	0	0	0	0
(Bonner et al., 2008)	0	?	+	?	0	0	?	0	0	0
Maternal Worry Scale										
(de Vet & Ireys, 1998)	+	+	+	+	0	0	?	0	+	0
Family Illness Beliefs Inventory										
(Kazak et al., 2004)	+	?	+	+	0	0	0	0	0	?
Parental Perception of Uncertainty Scale										
(Mishel, 1983)	-	+	+	?	0	0	0	0	0	?
(Molzon et al., 2014)	0	?	+	?	0	0	0	0	0	0
Parental Perception of Uncertainty Scale-Chinese										
(Ye et al., 2017)	-	+	+	+	0	0	?	0	0	?
Parental Perception of Uncertainty Scale-Diagnosis										
(Santacroce, 2001)	+	?	?	+	0	0	0	0	+	0
Illness Cognition Questionnaire-Parent version										
(Sint Nicolaas et al., 2016)	-	+	+	+	0	0	0	0	0	?

Note. + = positive rating; ? = indeterminate rating; - = negative rating; 0 = no information available.

Table 6

Methodological Quality of Included Studies

Instrument/Study	Structural validity	Internal consistency	Cross-cultural validity	Reliability	Measurement error	Criterion validity	Hypotheses testing	Responsiveness
Parental Outcome Measure								
(Bemister et al., 2014)	Inadequate[a]	Very Good		Doubtful[g]			Adequate[h]	
Perceptions of Parental Illness Questionnaire								
(Bogosian et al., 2014)	Inadequate[a]	Very Good					Adequate[h]	Very Good
Parental Experience of Child Illness								
(Bonner et al., 2006)	Adequate[b]	Very Good					Adequate[h]	
(Bonner et al., 2008)		Very Good		Doubtful[g]			Adequate[h]	
Maternal Worry Scale								
(de Vet & Ireys, 1998)	Doubtful[c]	Very Good		Doubtful[g]			Adequate[h]	
Family Illness Beliefs Inventory								
(Kazak et al., 2004)	Inadequate[a]	Very Good					Adequate[h]	
Parental Perception of Uncertainty Scale								
(Mishel, 1983)	Adequate[e]	Very Good					Inadequate[i]	
(Molzon et al., 2014)	Adequate[b]	Very Good					Doubtful[j]	
Parental Perception of Uncertainty Scale-Chinese								
(Ye et al., 2017)	Very Good	Very Good		Doubtful[g]			Doubtful[j]	
Parental Perception of Uncertainty Scale-Diagnosis								
(Santacroce, 2001)		Inadequate[f]					Doubtful[j]	
Illness Cognition Questionnaire-Parent version								
(Sint Nicolaas et al., 2016)	Adequate[e]	Very Good					Adequate[h]	

Note. [a]Sample size <5 times the number of items. [b]Sample size at least 5 times the number of items and >100. [c]Factor analysis and rotation method not described. [e]Confirmatory factor analysis was not performed. [f]Internal consistency was not assessed for each unidimensional subscale. [g]Pearson or Spearman correlation coefficient calculated without evidence of no systematic change. [h]Assumable that statistical methods adequate. [i]No information on measurement properties of comparator instruments. [j]Insufficient information on measurement properties of comparator instruments.

Methodological quality of studies. Table 6 summarises the methodological quality of studies for each psychometric property. None of the studies assessed or reported on the full range of properties recommended in the COSMIN guidelines. There was some variation in the quality of methods used to assess each of the properties across the studies. Almost all of the studies involved assessment of structural validity, however the methodology was rated as inadequate or doubtful in several studies because the sample size was not large enough for the number of items included in the factor analysis or because there were other minor methodological flaws (e.g. the type of factor analysis performed was not described). Overall, most studies were rated as very good for methodology used to assess internal consistency. Santacroce (2001) was rated as inadequate because Cronbach's alphas were not reported for all unidimensional subscales. Only one study reported cross-cultural adaptation of an existing measure (Ye et al., 2017), however the cross-cultural validity of the translated measure was not assessed. Reliability was assessed in four of the studies; the methodology was rated as doubtful in all of the studies because they assessed reliability using a Pearson or Spearman correlation coefficient. None of the studies assessed measurement error or criterion validity. Ratings for hypotheses testing for construct validity were varied, mostly because of a lack of information on comparator instruments. Only one study assessed responsiveness (Bogosian et al., 2014); the methods used were rated as very good.

Discussion

Measurement of illness perceptions in family members of an individual with a chronic illness has the potential to help identify those at risk of poor psychological outcomes and inform the development of interventions to target unhelpful beliefs and perceptions. However, this requires the availability of instruments that have been psychometrically validated as appropriate for predicting these outcomes in the population in which they are to be used. The purpose of this review was to identify, assess, and compare the psychometric properties of instruments designed to measure illness perceptions in family members and carers of individuals with chronic physical illness with the goal of identifying instruments that could predict individual differences in risk for poor psychological outcomes. The review identified nine instruments that assessed aspects of illness perception and were developed or validated for use in family members and carers. Most instruments were developed to assess illness perceptions in parents of a child with a chronic illness; only the Perceptions of Parental Illness Questionnaire was validated in children of a parent with a chronic illness. Eight instruments were developed in English; the Illness Cognition Questionnaire-Parent version was developed and validated in Dutch. Of the eight instruments developed in English, only one was translated into a different language (i.e. The Parental Perception of Uncertainty Scale-Chinese).

The findings of this review suggest that 1) further validation is needed for all of the instruments assessed, and 2) reporting of instrument development and validation needs to be improved. Information on measurement error, test-retest reliability (assessed using the intraclass correlation coefficient), responsiveness, floor and ceiling effects, and interpretability was missing for most of the instruments. Many studies also failed to use adequate methodology or provide adequate information on content, structural, and construct validity. At a minimum, adequate information should be provided on content validity, structural validity, internal consistency, and construct validity to demonstrate that the instrument measures a construct that is theoretically sound, that items within subscales are adequately correlated (to indicate that they are measuring the same construct), and that analyses will be able to discriminate between and/or predict the relevant outcome variables (Windle, Bennett, & Noyes, 2011). Additional information about measurement error and reliability is required depending on whether the instrument is intended for evaluative or discriminative purposes (Terwee et al., 2007). Moreover, information about responsiveness and interpretability – specifically, information about what cut off or change in score would be clinically meaningful – is imperative for instruments intended for clinical use (Terwee et al., 2007). Inclusion of this information in development and validation studies will also enable researchers to make informed decisions about which instrument is most appropriate to address the aims of their study.

The most widely used measures of illness perceptions in individuals diagnosed with a chronic physical illness are the Illness Perception Questionnaire (Weinman et al., 1996) and the Revised Illness Perception Questionnaire (Moss-Morris et al., 2002). Only one of the instruments included in the present review, the Perceptions of Parental Illness Questionnaire (Bogosian et al., 2014), was adapted from the Illness Perception Questionnaire. Although the Illness Perception Questionnaire is commonly used to assess illness perceptions in spouses of an individual diagnosed with an illness (Karademas et al., 2010; Searle, Norman, Thompson, & Vedhara, 2007; Wu et al., 2013), limited data on the validity of its application in this population exists. Psychometric evidence is relative to the target population and measures of illness perceptions need to be appropriately adapted and validated for the groups in which they are intended to be used. Based on the findings of the present review, it is recommended that research be conducted to examine the psychometric properties of the Illness Perception Questionnaire when adapted for use in family members in a range of illness groups, age groups, cultural groups, and for different genders.

It should also be noted that only one of the instruments examined in the present review was designed for use in children, under the age of 18 years, who have a parent with a chronic illness. Furthermore, none of the reviewed instruments were validated for use in children under the age of 12 years. This highlights a huge gap in research examining illness perceptions and is concerning given that children’s perceptions of their parent’s illness may have significant implications for their affective development. The only study that validated a measure of illness perceptions in young people with a parent with a chronic illness was conducted by Bogosian et al. (2014). They found that beliefs about the impact of the illness on the family environment and the unpredictable course of the illness were strongly correlated with emotional and behavioural difficulties in adolescents with a parent with multiple sclerosis (Bogosian et al., 2014). Future research may potentially be conducted to validate this measure for use with other illness groups. The availability of an evidence-based and psychometrically robust measure of illness perceptions that has been validated for use in children who have a parent with a diverse range of chronic illnesses would facilitate a more thorough examination of how children think about their parent's illnesses and how illness perceptions impact psychological adjustment following their parent's diagnosis.

The findings of the present review should be considered in light of potential limitations. First, the heterogeneous nature of research exploring illness perceptions and differences in conceptualisations of "illness perceptions" posed significant limitations. Although the search string was developed to identify studies that used specific terms relating to illness cognitions, perceptions, or beliefs, a number of instruments that measured perceptual constructs that were associated with coping and psychological outcomes were included in the review. For the purposes of the review, illness perceptions were broadly defined as an individual's cognitive and affective characterisation of a family member's illness. This definition was used to guide decisions about whether an instrument should be included in the review and led to the inclusion of some instruments not specifically described as measures of illness perceptions by the study authors. To address limitations posed by the search string not including terms relating to perceived impact of illness, worry, and uncertainty, and hence ensure completeness of study selection, the initial search was supplemented using forward and backward searching techniques that involved searching for articles that cited or were cited by the articles included in the review.

Second, the psychometric properties of many instruments were only described in one of the included studies, although two had been evaluated in more than one study. Previous systematic reviews that have assessed and compared the psychometric properties of instruments measuring the same (or similar) construct(s) have determined the overall quality of properties by reference to results reported across multiple studies. For example, Park et al. (2013) determined an overall rating for each of the instruments in their study by using a level of evidence approach that considered the number of studies that had evaluated the instrument, their methodological quality, and the consistency of their findings. Furthermore, the COSMIN guidelines recommend using the GRADE approach to summarise evidence of the quality of each psychometric property of an instrument based on the quality of methodology used to assess the property (Mokkink et al., 2018; Prinsen et al., 2018; Terwee et al., 2018). In the present study, only two of the instruments (i.e. the Parental Experience of Child Illness questionnaire and the Parental Perception of Uncertainty Scale) had been assessed in more than one study and neither had been assessed in sufficient studies for a level of evidence approach to support meaningful conclusions. The methodological quality of studies included in the review was assessed primarily for descriptive purposes; however the assessments may be used by researchers and clinicians to guide instrument selection based on the quality of psychometric properties of an instrument and the quality of methodology used to assess psychometric properties in studies that have evaluated the instrument.

Third, a lack of published psychometric information limited the evaluation of psychometric properties for each of the instruments. Missing information or indeterminate ratings are not necessarily indicative of poor instrument quality, but instead may mean that the properties have not yet been evaluated or that the instrument is newly developed (Terwee et al., 2007). It is worth noting that the initial search excluded articles that were not published in English, even though studies that described translated instruments were included in the review. It is possible that additional studies describing the development or psychometric evaluation of translated measures were published in languages other than English, in which case they would have been excluded from this review.

Finally, there are no standardised criteria to evaluate the quality of self-report instruments. However, the criteria published by Terwee et al. (2007) were developed as a first attempt to establish a standardised assessment tool for the psychometric properties of measurement instruments (Rosenkoetter & Tate, 2018). These criteria, or variations thereof, have been used in systematic reviews to identify the strengths and weaknesses of a range of self-report instruments, including those that measure health-related patient-reported outcomes (Bot et al., 2004; de Boer et al., 2004), psychological constructs (T. Park et al., 2013; Windle et al., 2011), and social constructs (Brohan, Slade, Clement, & Thornicroft, 2010).

Despite these limitations, the present review provides important insights into the quality of psychometric properties of instruments designed to measure illness perceptions in family members and carers of an individual with a chronic physical illness. This information may be utilised by researchers and clinicians concerned with identifying individuals at risk for psychological morbidity following a family member’s diagnosis. In general, findings highlight that further psychometric evaluation of instruments is needed and reporting of development and validation studies should be improved. Future research should aim to report as much information on psychometric properties and the methodology used to evaluate psychometric properties as possible; as a minimum, information on content, structural, and construct validity and internal consistency is needed to enable researchers to make informed decisions when choosing an instrument to use in their studies. Furthermore, almost all of the instruments were designed for parents with a child with a chronic illness. Research that aims to develop instruments to measure illness perceptions in children who have a parent with a chronic illness, or to validate the existing Perceptions of Parental Illness Questionnaire for use in different illness populations, is a much-needed addition to fill this gap in the current body of work examining the relationship between illness perceptions and psychological morbidity in family members of a person diagnosed with a chronic illness.

2.1. Chapter Summary and Future Directions

Chapter 2 presented a systematic review of instruments designed to measure illness perceptions among family members and carers of an individual with a chronic physical illness. Nine different instruments were identified, the majority of which were designed for use among parents of a child with a chronic illness.

Overall, findings indicated that further psychometric evaluation is needed for all the identified instruments. A lack of published information on many of the psychometric properties restricted the extent to which properties could be evaluated and compared between instruments. There is a clear need for better reporting of studies describing the development and validation of instruments. Findings also highlighted the dearth of research examining illness perceptions among children or siblings of an individual with a chronic illness; only one instrument designed for use among children was identified, and none for siblings.

In relation to the thesis aims, the Perceptions of Parental Illness Questionnaire (PPIQ) was the only identified instrument that was designed for use among children impacted by parental chronic illness (specifically, multiple sclerosis). Unlike other identified instruments, it had a strong theoretical underpinning; it was the only identified instrument that was adapted from the original IPQ/IPQ-R, and therefore assessed the aspects of illness perceptions outlined in the Common-Sense Model of Self-Regulation (CSM). As a result, the PPIQ demonstrated good content and construct validity. For these reasons, the PPIQ was the most appropriate identified instrument to adapt for use in AYAs impacted by parental cancer.

The findings of the study presented in Chapter 2 were used to inform the design of the following studies. The quality criteria published by Terwee et al. (2007) and COSMIN guidelines (Mokkink et al., 2010; Prinsen et al., 2018; Terwee et al., 2018) were referred to throughout the conduct of this research to ensure that the adaptation of the PPIQ adhered to best-practice guidelines for the development of measurement instruments.

CHAPTER THREE

Illness cognitions among adolescents and young adults who have a parent with cancer: A qualitative exploration using the Common-Sense Model of Self-Regulation as a framework

Statement of Authorship

An earlier version of the following chapter was published online by the International Journal of Behavioral Medicine on 3 June 2019. The original paper has since been published in vol. 29, pp. 531-41. The authors included PhD candidate, Chloe Fletcher, and her supervisory team: Prof Carlene Wilson, Dr Ingrid Flight, Dr Kate Gunn, and Dr Pandora Patterson. Full citation: Fletcher, C., Wilson, C., Flight, I., Gunn, K., & Patterson, P. (2019). Illness cognitions among adolescents and young adults who have a parent with cancer: A qualitative exploration using the Common-Sense Model of Self-Regulation as a framework. *International Journal of Behavioral Medicine, 26*(5), 531-541. doi:10.1007/s12529-019-09793-4

Candidate Statement

I confirm that I was the primary author of the qualitative study manuscript, and that guidance and supervision were provided by the co-authors throughout the manuscript preparation, submission, and revision processes. I was responsible for developing the research protocol and interview schedule; conducting individual interviews; conducting the thematic analysis; and writing the manuscript. I was listed as corresponding author on the submitted manuscript and was also responsible for revising the paper based on reviewer feedback and corresponding with the refereed journal.

Co-authors provided on-going supervision throughout the candidature resulting in this publication. Their role involved providing guidance regarding the research design, data collection, and data analysis, as well as feedback on drafts of the manuscript, and advice on responding to the journal reviewers. The percentage contribution of each author was as follows:

- Research design: Chloe Fletcher (60%), Prof Carlene Wilson (10%), Dr Ingrid Flight (10%), Dr Kate Gunn (10%), and Dr Pandora Patterson (10%)
- Data collection and analysis: Chloe Fletcher (100%)
- Writing and editing: Chloe Fletcher (80%), Prof Carlene Wilson (5%), Dr Ingrid Flight (5%), Dr Kate Gunn (5%), and Dr Pandora Patterson (5%)

3.0. Preamble

Although the Common-Sense Model of Self-Regulation (CSM) has been used previously to describe the relationships between illness perceptions, coping, and psychological outcomes among family members and informal carers (Fortune et al., 2011; Gatzoyia et al., 2014; Karademas et al., 2010; Klein Woolthuis et al., 2013; Matthews et al., 2018; A. Richardson, Morton, & Broadbent, 2016b; Scerri, Saliba, Saliba, Scerri, & Camilleri, 2019a), it was originally developed to describe these relationships in individuals with an illness (Hagger & Orbell, 2003; Leventhal et al., 2016a). Prior to adapting the Perceptions of Parental Illness Questionnaire (PPIQ), a qualitative study was conducted to ensure that the aspects of illness perceptions outlined in the CSM corresponded with the illness perceptions reported by young people who have a parent with cancer.

The study presented in Chapter 3 is in the same manuscript form as it was when accepted for publication. Minor modifications have been made to manuscript formatting in line with APA 6th style requirements. Additional changes to the original paper are denoted by footnotes. The co-author approval form and published manuscript for this study are presented in Appendices A and C, respectively.

Abstract

Background: Individuals construct beliefs about an illness based on their own perceptions, interpretation, and understanding of the illness and its treatment. These beliefs (collectively referred to as "illness cognitions" or "representations") can have implications for psychological outcomes in family members and carers of an individual with an illness. The aim of this study was to explore young people's perceptions of their parent's cancer using the Common-Sense Model of Self-Regulation as a theoretical framework.

Methods: Semi-structured, one-on-one interviews were conducted with young people who had a parent diagnosed with cancer. Interview transcripts were analysed using deductive thematic analysis techniques.

Results: Eleven young people aged 15-24 years participated in the study. Major themes aligned with the dimensions of the Common-Sense Model of Self-Regulation. Young people described their experiences with parental cancer with reference to cognitive representations (beliefs about the illness identity, their understanding or coherence of the illness, and consequences, curability or controllability, timeline, and cause of the illness) and emotional representations (emotional beliefs and subjective feelings about the illness).

Conclusions: Findings indicate that young people's perceptions of their parent's cancer can be usefully described within the framework of the Common-Sense Model of Self-Regulation. Future research should investigate the relationships between young people's illness cognitions, coping strategies, and psychological adjustment following their parent's cancer diagnosis. This will provide valuable insights for the development of interventions that target specific types of illness cognitions associated with maladaptive coping strategies and poor adjustment.

Introduction

A parent's diagnosis of cancer can have a significant impact on the family and can be especially distressing for dependent children. Adolescents and young adults, who are at an age where they have the capacity to understand the potential implications of cancer for themselves and their family (Gazendam-Donofrio et al., 2011; Pederson & Revenson, 2005), have been identified as experiencing higher levels of distress than younger children (Compas et al., 1996; Huizinga, Visser, Zelders-Steyn, et al., 2011; Visser et al., 2004). In Australia, it is estimated that 21,000 young people aged 12-25 years have a parent diagnosed with cancer each year (Patterson et al., 2014), and more than half experience clinically-elevated levels of distress (Patterson et al., 2017).

A range of factors are known to moderate young people's psychological adjustment following a parent's cancer diagnosis, including their age and gender, their parent's gender, time since parent's diagnosis, having unmet needs, and poor family functioning (Ellis, Wakefield, Antill, Burns, & Patterson, 2016; Krattenmacher et al., 2012; McDonald et al., 2016; Su & Ryan-Wenger, 2007; Visser et al., 2004). Some findings also suggest that the way that children think about their parent's cancer is associated with their ability to cope with, and adjust to, the illness. For example, Compas et al. (1996) found that children's perception of the stressfulness of their parent's cancer was associated with the development of anxiety and depression, and that this had a more significant impact than the stage of their parent's disease and their prognosis. Furthermore, they found that children generally perceived low levels of personal control over their parent's cancer, which may potentially prevent them from using problem-focused approaches to coping (Compas et al., 1996). Despite these important findings, to the best of our knowledge, there has been no recent research exploring the beliefs that young people form about their parent's cancer and the potential impact of these beliefs on their psychological adjustment to the illness.

Leventhal’s Common-Sense Model of Self-Regulation proposes an explicit causal link between an individual’s beliefs about an illness, the coping strategies that they employ in response to the illness, and their physical health and psychological outcomes (see Figure 4) (Gray & Rutter, 2007; Hagger & Orbell, 2003; Hoving, van der Meer, Volkova, & Frings-Dresen, 2010). An individual constructs beliefs about an illness based on their own perceptions, interpretation, and understanding of the illness and its treatment (Diefenbach & Leventhal, 1996; Hagger & Orbell, 2003; Petrie & Weinman, 2006). These beliefs are collectively defined as “illness cognitions” or “representations”, and are not necessarily scientifically or medically substantiated (Browning, Wewers, Ferketich, Otterson, & Reynolds, 2010). They are formulated based on information provided by three broad domains of information: previous social interactions, personal experiences, and cultural knowledge of the illness; external social influences from perceived important others or authoritative figures; and current experiences with the illness (Hagger & Orbell, 2003). Illness cognitions are complex and dynamic (Leventhal et al., 2016a), evolving over time in response to internal and external stimuli, for example; emerging side effects of treatment or public opinion about the causes of the illness (Browning et al., 2010).

The Common-Sense Model of Self-Regulation is conceptualised as a parallel processing framework, whereby an individual simultaneously constructs cognitive and emotional representations of an illness (Hagger & Orbell, 2003). Cognitive representations are organised around five central themes or dimensions: *identity* (the label used to describe the illness and perceptions of symptoms associated with the illness), *consequences* (beliefs about the anticipated or experienced impact of the illness on quality of life or physical, cognitive, or social capacity), *cure or control* (beliefs about the efficacy of treatment or the extent to which the individual has personal control over the illness), *timeline* (beliefs about the duration or progression of the illness), and *cause* (beliefs about the factors responsible for causing the illness). These dimensions are considered to be the “basic building blocks” of investigations into how individuals construct beliefs about illness. The Common-Sense Model of Self-Regulation reflects an ongoing interplay between empirical data and theoretical concepts (Leventhal et al., 2016b). As a result, the original model has been extended through the inclusion of an additional cognitive representation dimension of *coherence* (personal understanding or comprehension of the illness) (Moss-Morris et al., 2002). Other changes have included incorporating a *cyclical timeline* dimension, and splitting the *cure or control* dimension into separate dimensions of *treatment control* and *personal control* (Moss-Morris et al., 2002).

In response to an illness or health threat, an individual is thought to employ coping strategies that are consistent with their own representation of the illness or threat. For example, research indicates that perceptions of serious consequences are associated with avoidance and denial coping strategies, such as behavioural disengagement (Moss-Morris, Petrie, & Weinman, 1996). Furthermore, perceptions that the illness is uncontrollable, chronic, and highly symptomatic have also been associated with avoidance and denial coping strategies (Hagger & Orbell, 2003). In contrast, perceptions that the illness is controllable have been associated with active coping strategies, including problem-focused coping (Kemp, Morley, & Anderson, 1999). This research highlights the importance of understanding these associations; by identifying and modifying beliefs that are associated with maladaptive coping strategies, an individual’s distress may be reduced, potentially resulting in better psychological adjustment (Hagger & Orbell, 2003).

Although the model was originally developed to describe “lay” understanding of illness experiences in individuals diagnosed with an illness (Corbett, Groarke, Walsh, & McGuire, 2016), it has also been used to explore illness cognitions in family members and significant others caring for someone experiencing ill-health (Bogosian et al., 2014; Del

Castillo, Godoy-Izquierdo, Vazquez, & Godoy, 2011; Dempster et al., 2011a; Fortune et al., 2011; Gatzoyia et al., 2014; Graham, Dempster, McCorry, Donnelly, & Johnston, 2016; Scerri, Saliba, Saliba, Scerri, & Camilleri, 2019b; Yu, Lingler, Sereika, & Erlen, 2017). Bogosian et al. (2014) used the model to describe illness cognitions among adolescents who have a parent with multiple sclerosis. Using a quantitative measure they developed for specific use in this cohort, they found that stronger subjective beliefs about the negative consequences of their parent’s illness and a perception that it was chronic and unpredictable were associated with poorer psychological adjustment, independent of parental report of illness severity (Bogosian et al., 2014).

Based on these findings, the Common-Sense Model of Self-Regulation is proposed as an appropriate theoretical framework for exploring illness cognitions in young people who have a parent with cancer. The aim of this study was to explore young people’s perceptions of their parent’s cancer and examine how these perceptions correspond with the dimensions of illness cognitions as understood within the Common-Sense Model of Self-Regulation.

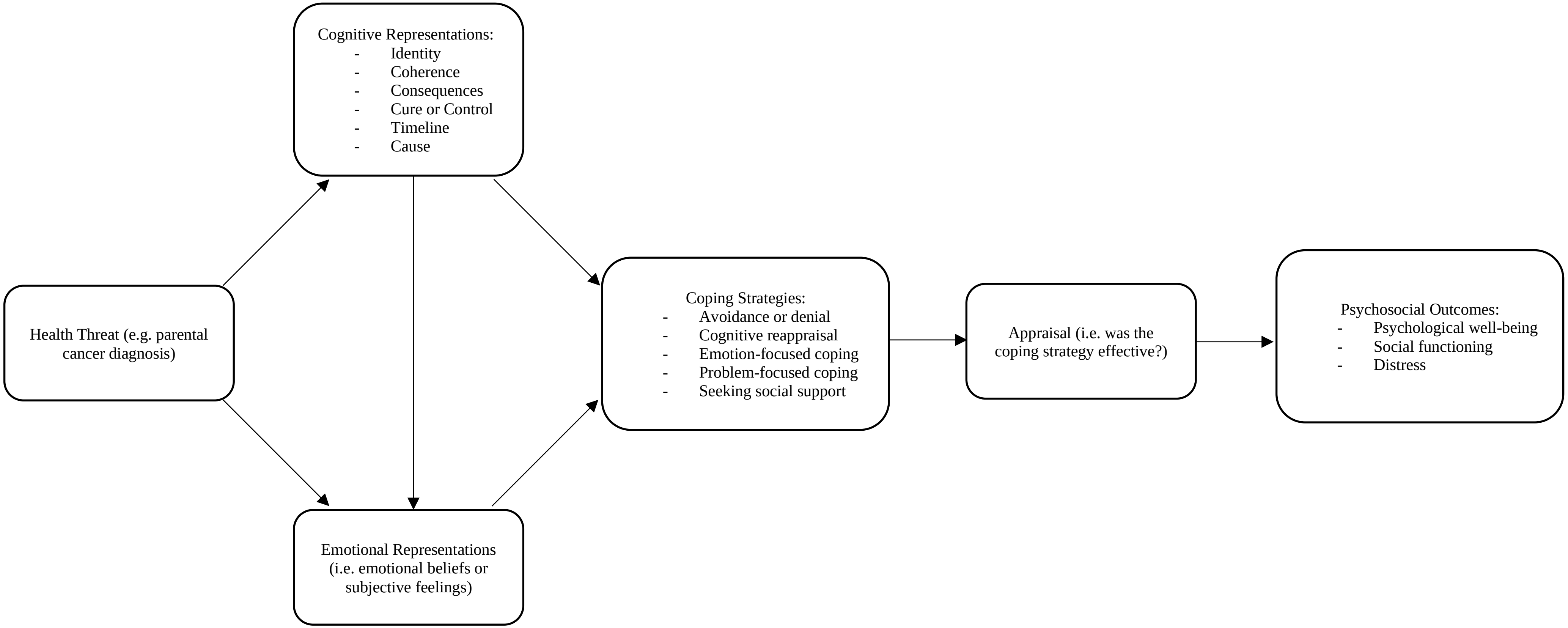


Figure 4. Leventhal’s Common-Sense Model of Self-Regulation Applied to Parental Cancer (adapted from Hagger et al., 2017; Joice, 2012)

Methods

Methodology and epistemology.[1] A qualitative methodology was considered most appropriate to address the aim of the study. It was important for this study to employ an exploratory approach, given the dearth of research exploring young people's perceptions of their parent's cancer. A qualitative methodology seeks to understand the way that people interpret and make sense of their experiences and the world around them. Where a quantitative approach would simply report measures of behaviour or internal cognitions, a qualitative approach aims to uncover patterns of meaning within people's behaviour, perceptions, feelings, and experiences (Braun & Clarke, 2013). In doing so, this approach can facilitate a deeper and richer understanding of the phenomenon of interest (Braun & Clarke, 2013). By employing a qualitative methodology in the present study, it was possible to gain insight into how young people perceive their parent's cancer and examine how these perceptions corresponded with the dimensions of illness cognitions as outlined within the Common-Sense Model of Self-Regulation.

Qualitative research methods are underpinned by epistemological assumptions. For the present study, an interpretivist epistemological position was taken. Interpretivism aims to understand phenomena through the lens of lived experience; the interpretivist position argues that people are complex, and that different people experience and understand the same "objective reality" in different ways. This was central to the aim of the study, which sought to examine how young people thought about (perceived) and made sense of (interpreted) their parent's cancer. Although an existing model was used as a framework for examining young people's illness perceptions, it was important that the analysis captured the broad and varied ways in which young people constructed representations of their parent's cancer.

---

[1] This section was added after the original paper was published.

This approach was chosen based on previous research examining illness perceptions among people with physical and mental illnesses. Qualitative methods have been used to explore illness perceptions in a range of different groups (e.g., Corbett et al., 2016; Heffernan, Coulson, Henshaw, Barry, & Ferguson, 2016; Huston & Houk, 2011; Simons et al., 2017), and in some instances, have provided a theoretical foundation for the adaptation or development of quantitative instruments to enable illness perceptions to be measured (e.g., Bogosian et al., 2014).

Design. Semi-structured, one-on-one interviews were conducted with young people who had a parent diagnosed with cancer. Interviews explored young people's experiences following their parent's diagnosis and their cognitive and emotional representations of their parent's cancer. Open-ended questions included in the interview schedule (Table 7) were based on the Common-Sense Model of Self-Regulation.

Participants. A convenience sample was recruited through advertisements distributed online through cancer support and advocacy groups, including Canteen[2] and Australian-based cancer support groups on social media. All advertisements contained information about the study and interested young people (or their parents) were asked to contact the researcher via telephone or email if they (or their child) was interested in participating in the study.

Young people aged 12-24 years were eligible to participate if they had a parent who had been diagnosed with any type or stage of cancer, were aware of their parent's cancer diagnosis, and were living at home during their parent's diagnosis and treatment. This included young people who had a parent who had been diagnosed with curable, metastatic, or advanced cancer, and young people who had a parent who had died from cancer. Although

---

[2] Canteen is an Australian support organisation for young people affected by cancer; including young people diagnosed with cancer, and young people who have a brother, sister, or parent diagnosed with cancer.

there are various definitions of “youth” (e.g. 15-24 years as defined by the United Nations (2017) and 10-24 years as defined within the scientific literature (Sawyer, Azzopardi, Wickremarathne, & Patton, 2018)), the age range of 12-24 years was chosen because it aligns with definitions provided by the Australian Institute of Health and Welfare (2011).

Eleven young people from ten families participated in the interviews (age range = 15-24 years; *M* = 19.1 years; female = 7). Of those, two participants had both parents who had been diagnosed with cancer, two participants were bereaved, and two had a parent with advanced or metastatic cancer, one had a parent who was still undergoing treatment for a curable form of cancer, and four had a parent who had finished active treatment. One participant’s father had been diagnosed with several unrelated types of cancer. Time since initial diagnosis ranged from six months to eight years. Parental cancer diagnoses included non-Hodgkin’s lymphoma, lung, bowel, pancreatic, kidney (including collecting duct carcinoma), testicular, ovarian, cervical, and breast cancer. Participants described various household structures; two participants lived alone with a single parent, three participants lived with a single parent and other relatives (two with siblings and one with grandparents), and six participants lived with both parents (four with siblings and one with an aunt). Of those that lived with both parents, two participants lived with parents who were separated but had moved in together in order to fulfil a caregiver role.

Table 7

Interview Schedule Outlining Relevant Dimension of the Common-Sense Model of Self-Regulation

1. Could you tell me about your experience with parental cancer? (illness identity, treatment control)
    a. *Which of your parents was diagnosed with cancer?*
    b. *What type of cancer were they diagnosed with?*
    c. *What type of treatment have they had?*
2. What was it like when your parent was first diagnosed with cancer? (emotional representations)
    a. *How did you react when you first found out?*
    b. *How did your family deal with your parent's diagnosis*?
3. In general, how do you feel when you think about your parent's cancer? (emotional representations)
4. How has your parent's cancer impacted your life? (consequences)
    a. *In what way has your parent's cancer negatively impacted your life?*
    b. *Has your parent's cancer had a positive impact on your life in any way?*
5. How has cancer affected your parent? (illness identity)
6. What symptoms and side effects has your parent experienced as a result of their cancer? (illness identity)
7. How do you think your parent's cancer might affect them in the long-term?[a] (timeline, treatment control)
8. Can you tell me about any changes you've noticed in the way you act around your parent since their diagnosis? (personal control)
    a. *What sort of things do you do that you feel make a difference to their illness or how they're feeling?*
9. Can you tell me about the sort of things that you might talk about with your parent or with your family about your parent's cancer? (coherence)
10. Can you tell me about what you thought about the information that was shared with you about your parent's cancer? (coherence)
    a. *Do you feel like you know enough about your parent's cancer?*
    b. *Was there anything that you might have liked to know more about?*
11. Do you think there is anything your parent could've done to avoid cancer? (cause)
    a. *Do you have any thoughts on what might've caused your parent's cancer?*
12. Is there anything else you would like to add to what's already been said?

Note. Questions asked in past tense for bereaved participants and participants whose parent had finished active treatment. [a]Bereaved participants were not asked this question.

Interviews. Interviews were conducted by the first author (CF). CF is a PhD researcher with a background in social health sciences and previous research experience in cancer support. The researcher had no contact with participants prior to the study commencement. Interviews were conducted either face-to-face, via telephone, or via a web-conferencing program. Face-to-face interviews were conducted at the Canteen Division office in Sydney, Australia. Interviews ranged from nine to 37 minutes in length (M = 23 minutes). Interviews were audio-recorded and transcribed verbatim using a professional transcription service. The interview schedule was used flexibly to explore participants' cognitive and emotional representations of their parent's cancer in relation to the dimensions of the Common-Sense Model of Self-Regulation. All participants were asked each of the questions outlined in the interview schedule, with the exception of bereaved participants who were not asked about their thoughts on how their parent's cancer might affect them long-term. Probing questions were used to obtain more specific, or in-depth information, on emerging themes. Similar themes were apparent within all interviews and recruitment was finalised when no new themes emerged in three consecutive interviews (i.e., data saturation was reached).

Ethical considerations. The study was reviewed and approved by the Social and Behavioural Research Ethics Committee at Flinders University (project number 7436). All participants received a research pack containing written information about the study, formal written consent forms, and a questionnaire to collect participant demographic and parent cancer-related medical information. A copy of the interview schedule was also included to enable potential participants to make a fully informed decision about participating in the study. Parental consent was obtained for participants younger than 18 years. Participants returned signed consent forms and completed demographics questionnaires to the researcher prior to the interview. Participants were given assurances regarding the confidentiality and anonymity of the information they shared during the interview. Participants were informed that they were able to withdraw from the study at any time.

Analysis.[3] Following previous qualitative research exploring illness perceptions using the Common-Sense Model of Self-Regulation as a framework (Bogosian et al., 2014; Corbett et al., 2016; Heffernan et al., 2016), thematic analysis methods were used to analyse the data. Thematic analysis involves identifying themes or patterns of meaning across a qualitative data set (Braun & Clarke, 2013). It is a flexible method that does not prescribe methods for data collection, theoretical positions, or epistemological or ontological frameworks (Braun & Clarke, 2013). In the present study, a deductive (theory-driven) approach was employed to identify and organise themes using the Common-Sense Model of Self-Regulation as a framework.

Braun and Clarke’s (Braun & Clarke, 2006, 2013) steps for coding and analysing qualitative data were followed to conduct the thematic analysis. Interview transcripts were verified for accuracy prior to analysis and were read-through several times for familiarisation. Interview transcripts were coded by the first author (CF) according to the dimensions of the model. Themes were reviewed to ensure that data within themes were coherent, and that there were clear and identifiable distinctions between themes. Following this, a codebook was developed by the first author to outline definitions of codes. A sub-set of transcripts (30%) were coded independently by a second researcher with a background in psychology and qualitative research (see Acknowledgements), who was not part of the research team, through reference to the codebook. Discrepancies in coding were resolved through discussion until consensus on definition of codes and interpretation of data was reached.

---

[3] This section has been modified since the original paper was published.

Results

In general, participants expressed appreciation for the opportunity to share their experience and they hoped that this would, in some way, help other young people in a similar situation. Most young people spoke openly during the interview, with very minimal prompting. Two participants required more direct prompting with probing questions; these interviews were the two shortest in length (9 and 14 minutes). One of these participants had a parent with terminal cancer, possibly contributing to her difficulty in answering some of the questions. The other participant's parent had a recent diagnosis, and hence was at the beginning of the cancer experience, also potentially contributing to his short responses, as some of the questions asked may not have been relevant to his individual experience. Analyses confirmed that the Common-Sense Model of Self-Regulation could be utilised to describe young people's experience of parental cancer and that these experiences were varied. Definitions and illustrative quotes for themes and sub-themes are presented in Table 8.

Identity. All participants were able to describe the type of cancer that their parent had been diagnosed with; many were also able to describe the stage of cancer. Participants spoke about the impact of common side effects including fatigue, hair loss, nausea, weight gain, weight loss, and problems with memory or concentration. Some participants spoke about the high cost of symptoms like fatigue or problems with memory or concentration; in particular for participants who had a parent with terminal cancer. This was noted by one participant as she recounted the last few months spent with her dad: *"…we wanted to spend time with him but he just, I don't know, couldn't – didn't have the thinking capacity to do it."* This experience was similar to another participant who described the impact of her mother's fatigue:

> *"…in the last few months, probably just her being really exhausted all the time – having to not just rest sort of once a day or occasionally but being – becoming, I guess, less … around and available in some ways and probably … energy wise, just not being able to sort of talk with people for a longer period of time."*

Young people also spoke about the emotional side effects of cancer; cancer didn't just impact their parent physically, it was also a *"mental game"*. One participant described the changes in her mother since her diagnosis with ovarian cancer: *"…emotionally she's a lot more anxious about things and she cries a lot and gets down a fair bit"*. Some participants felt that although their parent was likely experiencing a number of physical symptoms, this was not always expressed; for example, two participants described their parent's pain as being something that was not really *"complained about"* or *"overemphasised"*, despite being ever-present.

Emotional representations. Young people described a mixture of initial feelings after learning of their parent's cancer diagnosis: shock (e.g. *"it felt like all the breath had been knocked out of me"*), confusion (e.g. *"I didn't really understand what, the magnitude of what it was, or like, how to feel or what to do"*), numbness (e.g. *"the immediate reaction, it was for me, it was a bit muted"*), a loss of control (e.g. *"It felt like I had no control back over my life"*), and, in contrast, a level of acceptance (e.g. *"I guess I was prepared for it to be cancer"*). Participants described their experiences since their parent's diagnosis as *"surreal"*, *"a bumpy ride"* and *"very full on, very confronting"*. In general, young people felt fear over the possibility of losing their parent and uncertainty about the future, despite whether their parent was in remission, undergoing curative treatment, or receiving palliative care.

Table 8

Themes and Sub-themes Identified from the Deductive Analysis

Main theme (dimension of Common-Sense Model)	Definition	Sub-theme	Illustrative quotes from young peoples' interviews
1. Identity	*Young peoples' perceptions of the side effects and symptoms that their parent experiences due to their cancer*		"She didn't really eat, she was very nauseous and she spent a lot of time sleeping."
2. Emotional Representations	*Young peoples' emotional beliefs and subjective feelings about their parent's cancer*		"I think, shock and confusion, just because I didn't really understand what, the magnitude of what it was, or like, how to feel or what to do."
3. Coherence	*Young peoples' understanding or comprehension of their parent's cancer*		"I wanted to know enough information to get me by, but not enough information to know every detail."
4. Consequences	*Young peoples' perceptions of the impact their parent's cancer has had on their daily life*	1. Young Person	"I've just become a lot more anxious kind of person, yeah. It's really changed my perspective on things and there's been like actually a bit of role reversal, so because my mum's a single parent, yeah, so it's actually like I am kind of mothering her in a sense."
	Young peoples' perceptions of the impact their parent's cancer has had on their family life	2. Family	"…it's caused a lot of tension in our house as well because this is so much stress all of the time."
	Young peoples' perceptions of the impact their parent's cancer has had on their parent's life	3. Parent	"…she's also figured out who her true friends are, and emotionally that has really helped her, she feels a lot lighter, she even says 'That whole experience, I know who my true friends are, because they were the ones that offered to come to a round of chemo, or offered to take me to radiation' …"
5. Cure or Control	*Young peoples' perceptions of the personal control they have over their parent's cancer symptoms, side effects, and distress*	1. Personal Control – Relieving Symptoms and Distress	"…you had to sort of have a calming presence, you couldn't be loud and yelling or anything like that, because if it stressed her out … she would feel sicker and you didn't want to her feel unhappy and unwell."
	Young peoples' perceptions of the personal control they have over maintaining a normal life separate from their parent's cancer	2. Personal Control – Maintaining Normality	"…when they're sick and that sort of idea of someone's helpless, I guess, is kind of pretty prevalent in society in general. So, I think just making sure that you don't replicate that as much as possible at home and treating family members as human…"
	Young peoples' perceptions of the control treatment can have over their parent's cancer	3. Treatment Control	"…she's got like medically it's stage 4 breast cancer, secondary breast cancer so it's caused by the production of the

Main theme (dimension of Common-Sense Model)	Definition	Sub-theme	Illustrative quotes from young peoples' interviews
			hormones but it's coming from her bones, so it's in her bones so it's like it's inoperable. So essentially it's pretty much just keeping it under control but it's not going to go away."
6. Timeline	*Young peoples' perceptions of the expected duration and progression of their parent's cancer*		"…that's all pretty uncertain, they've sort of said to us, this is incurable and we know that the end is coming soon but they won't give us a time frame … it's kind of like switched between denial and … she'll be fine and then thinking that she's going to die like tomorrow."
7. Cause	*Young peoples' perceptions of the causes of their parent's cancer*	1. Chance	"…to me it just feels like it picks a random person in the world there's no actual reason for it, because there's no history of it going through our family either so."
		2. Environmental or External Attributions	"I've heard that people have said that, other bad people in their lives can cause cancer, …"
		3. Genetic / Hereditary Attributions	"…cancer has also been a fairly genetic sort of disease going through – I mean her father passed away when she was fifteen or something of cancer and he had a melanoma in the eye or something and she had a few other relatives that've passed away early because of cancer. So whether or not there is some sort of gene in your DNA that has something to do with the likelihood of her developing cancer, …"
		4. Lifestyle Attributions	"…she eats everything I eat, she drinks whereas I don't, but drinking usually is a liver issue not just any random cancer issue. She smokes but it wasn't lung cancer it wasn't anything that could have linked to that. So I worry more now that she's still smoking because well I feel like that might cause it to come back sooner or things like that."
		5. Psychological Attributions	"…I think she told me that it was stress."

Coherence. Young people described a desire to be informed about their parent's cancer, and they achieved this by accompanying their parent to doctor's appointments, speaking with their parent's treating doctors, researching the disease in their own time, and having open communication with their parent about their cancer. One participant described checking in with his mother to *"see how it's all going, how the treatment's going ... [and to] see if she's alright."* Another participant commented that he found knowing as much as he could about his mother's cancer beneficial:

> *"...I just tried to get as much information as possible about it, and I think that helped me to accept that, helped me to really, like reduce the anxiety just knowing this, like percentages of chance of it coming back, and all that, I think, because that was one of the ways I coped by like, information and reading about it, and knowing as much as I could about it."*

In contrast, some participants felt that knowing too much would have been overwhelming:

> *"I wanted to know enough information to get me by, but not enough information to know every detail. ...because it almost made it too real, and I kind of wanted to keep this fantasy land that we were all okay."*

A similar sentiment was shared by another participant who commented that *"it's good to be a little bit oblivious"*.

Consequences. Young people described a range of consequences that their parent's cancer had for them, their family, and their parent. For many, the most prominent consequence for themselves was having to take on extra household responsibilities:

> *"...I would often come home from school and I'd cook dinner and I'd clean the house and I found myself taking on a lot more responsibilities just because my mum wasn't able to..."*

Many young people also accompanied their parent to treatment and other medical appointments and provided emotional support. Two participants likened this to fulfilling a parental role, with one stating that: *"... it's actually like I am kind of mothering her in a sense."* Taking on extra responsibilities had a flow-on effect on schooling:

> *"...my results went down the drain, because I didn't have time to study for exams on top of looking after mum, and looking after my little brother after he got home from school."*

One participant spoke about how she had considered dropping out of school to work full-time and care for her mother. Some described this not necessarily as a negative consequence of their parent's cancer, but as a shift in their priorities. In particular, many participants spoke about how the experience brought their family closer together and taught them to be more appreciative of the time they had together. One participant described it in this way:

> *"...we care more about going to things and it's important to go watch someone in the football grand final, because you don't know what could happen the next day. So I think everything has become more important to us."*

Young people also spoke about the consequences for their parent. In particular, two participants described how breast cancer had negatively impacted their mothers' self-esteem. One participant reflected on her mother losing her hair after chemotherapy: *"I went from seeing my mum being such a strong, beautiful woman to someone who felt so insecure."* Another participant stated that her mother no longer wore swimsuits because of the visible scars on her breasts.

Young people also highlighted the positive consequences for their parent. One participant stated that his mother was *"pretty proud of herself that she's made it"*. Several participants spoke about how cancer had improved their relationships by helping them to *"figure out who [their] true friends are"* and had given their parent *"a new fresh start ... a start again, and at a new place, with different people"*.

Cure or Control. Young people were very well-informed about what could be done medically to treat their parent's cancer. One participant, whose mother was diagnosed with metastatic breast cancer, described the difference between treating to cure and treating to control: *"...essentially it's pretty much just keeping it under control, but it's not going to go away."* Another participant spoke about how chemotherapy reduced her mother's risk of cancer recurrence, although she acknowledged that the risk was not completely eliminated. Frustration was expressed anger by another participant who stated that he had been *"stonewalled"* by an oncologist who deemed his mother too unwell to try an experimental treatment for her rare form of cancer.

Young people gained a sense of personal control over the cancer experience by doing anything that they could to *"make [their parent's] life easier or less stressful"*. One participant said that she felt that this *"made it just so much easier for [her mother] because she didn't have to worry about those things, she could concentrate on the things that she felt were most important to her."* Another participant described how changing the way he behaved around his mother could help to make her feel better:

> *"...you had to sort of have a calming presence, you couldn't be loud and yelling or anything like that, because if it stressed her out ... she would feel sicker and you didn't want to her feel unhappy and unwell."*

For some participants this meant shielding their parent from their emotional reactions to their cancer. One participant stated that she "*hid away [her] feelings because it – she [mother] was really upset and we just needed to get things done..."* Another participant stated that she *"didn't want her [mum] to know that it was affecting me in such a great way* *until I knew she would be able to cope with the fact that, it was obviously affecting me as well as her."*

Personal control over the cancer experience was as much about what could be done to support the parent or relieve their symptoms and distress as it was about preventing cancer from becoming the parent's identity; one participant stated that it was important to make sure that she did not make her mother feel like she was helpless just because she was sick; as much as possible she wanted to avoid her mother *"becoming the cancer patient … in every facet of life"*. There was also a desire to maintain a normal life outside of the cancer experience; that although cancer was a big aspect of their family's lives, *"it [didn't] have to be the focus"*.

Timeline. Young people spoke about cancer as being a "battle". One participant expressed this through her belief that *"my mum's not going to die, my mum's going to win this battle."* For others, their parent's future was much less certain. One participant, reflected on his father's the unpredictable timeline of his father's cancer: *"it's a battle for him, and it's touch and go, whether or not he lives or dies"*. Similarly, other young people who had a parent with terminal cancer expressed a lot of uncertainty over their parent's future and a desire to know exactly how much time they had left with their parent:

> *"…that's all pretty uncertain, they've sort of said to us, this is incurable and we know that the end is coming soon but they won't give us a time frame … it's kind of like switched between denial and … she'll be fine and then thinking that she's going to die like tomorrow."*

Although this uncertainty was a continuous stress for those with a parent with terminal cancer, young people showed resilience through their ability to adapt to the situation. One participant commented that she and her family had learned to take it *"day by day … and well we're kind of figuring out like a pattern now."*

Those who had a parent in remission spoke about their fears that the cancer might come back in the future, although one participant commented that these fears *"have slowly dissipated and gone away"* over time. Another participant was able to rationalise her fears that her mother's cancer would return: *"...I figured with the amount of chemicals and treatment they gave her, at the moment, for the next couple of years, I know that we're okay and it's not going to come back."*

Cause. Although they were able to offer a range of potential causes for their parent's cancer, including biological, environmental, lifestyle, or psychological causes, many participants felt that cancer was something that *"just happens"*. One participant felt that in her mother's case *"[cancer] picks a random person in the world, there's no actual reason for it, because there's no history of it going through our family"*.

Young people also talked about their fears about their own risk of developing cancer in the future (e.g. *"I get worried of is it going to be me in five years or ten years or however long..."*) and how their attitude towards cancer prevention behaviours has changed in response to their parent's cancer (e.g. *"Even things like putting sun block on, I'm so crazy, obsessive about putting it on now..."*).

Discussion[4]

These findings extend previous research examining young people's experiences with parental cancer by providing an insight into young people's perceptions of, and emotional reactions to, their parent's cancer. Results confirmed the relevance of the dimensions of the Common-Sense Model of Self-Regulation for describing illness cognitions in young people following their parent's diagnosis with cancer.

The eleven young people who participated in interviews were a diverse sample; but despite this, there were common themes in the experiences they reported. Participants

---

[4] Discussion has been modified since the original paper was published.

described negative emotional reactions, such as feelings of fear, uncertainty, and a loss of control, in response to their parent's cancer. These feelings were not just limited to participants who had a parent with terminal cancer or who were reflecting on the time before their parent had died; they were shared by participants who had a parent in remission. Many still feared that the cancer could return.

Participants described having an active involvement in helping their parent during their cancer experience; through attending their parent's appointments, researching their parent's cancer in their own time, or talking with their parent about treatment options. They had a coherent understanding of their parent's cancer, their treatment, and what could be expected in terms of the progression of the disease, and indicated that knowing as much information as possible about their parent's cancer was beneficial. This is consistent with findings reported by Maynard et al. (2013), where adolescent participants described that having information about their parent's cancer and its treatment was important in helping them cope. Using a problem-focused coping strategy, whereby young people play an active role in supporting their parent through their illness, may improve their sense of control over the situation. This sentiment was not shared by all participants however; one participant felt that knowing too much about her parent's cancer would erase the illusion that everything was okay, suggesting a personal preference for a more emotion-focused coping strategy, such as denial or avoidance.

Participants described taking on extra responsibilities that would have otherwise been fulfilled by their parent, following their parent's diagnosis. These included preparing meals, cleaning the house, looking after pets, and babysitting younger siblings. Although almost all participants spoke about taking on these extra responsibilities, this was particularly notable for those with a single parent. Many participants described how having these responsibilities impacted their schooling. For some, this meant considering whether or not to drop out of school to find full-time work and care for their parent. Others felt that this shift in their priorities was a positive experience, because it made them focus on what was important in their lives – their family. Benefit finding, defined as "the process of deriving positive growth from adversity" (Cassidy, McLaughlin, & Giles, 2014), has been shown to lead to positive outcomes among family caregivers (Cassidy, 2012; Cassidy, Giles, & McLaughlin, 2014), and may be an important aspect of coping for young people following a parent's cancer diagnosis.

Several participants described their parent's cancer as a "battle". The use of violence-related metaphors for the cancer experience has been described previously both in peer-reviewed literature (R. S. Miller, 2010; Semino et al., 2015) and in the media (Cooper, 2014; Granger, 2014; James, 2018; Semino, 2015) as potentially detrimental for people diagnosed with cancer. These kinds of metaphors position cancer as an opponent – an enemy to defeat – and may contribute to feelings of helplessness and anxiety. In particular for people with terminal cancer, this can cause feelings of failure or guilt for losing the "fight" against cancer (Semino, 2015). For young people who have a parent with terminal cancer, this may generate feelings of resentment against their parent for not "fighting hard enough" and lead to difficulties accepting their parent's diagnosis and eventual death. Instead it may be more helpful to view cancer as a natural process – as something that "just happens" – as several participants described when reflecting on possible causes of their parent's cancer. This belief is substantiated by research showing that two-thirds of all cancers are caused by unavoidable random errors that occur during DNA replication (Tomasetti, Li, & Vogelstein, 2017). Changing the rhetoric around the cancer experience by avoiding the use of violence-related metaphors that place blame on the person with the diagnosis and emphasising the random nature of the disease may be helpful for young people to reach an acceptance of their parent's cancer and prognosis.

Facing the potential death of a parent is not an experience that is normal in adolescence or young adulthood (Phillips & Lewis, 2015). Despite their difficult and unusual circumstances, many participants did what they could to maintain a sense of normality in their life. For some this involved ensuring that family life did not revolve around their parent's cancer, by drawing a distinction between their parent as a mother or father versus a "cancer patient". This attempt to separate the disease from the family has been previously described as adaptive. In particular, a family identity that revolves around the parent with cancer for an extended period of time may negatively affect the family's ability to meet the developmental needs of all family members, particularly dependent children (Pederson & Revenson, 2005). Although taking ownership of the disease appears to be an important aspect of perceived control for the person with cancer (Karnilowicz, 2011), maintaining a sense of normality – an identity separate from the cancer – may be an important coping strategy for young people when their parent has cancer.

The findings of this study should be considered in light of some potential limitations. Participants were recruited primarily through an organisation aimed at supporting young people through a cancer experience. Furthermore, participants were self-selected. As a result, the findings may not be representative of the experiences of young people outside of the organisation. The sample size was quite small, however this is a small and difficult to recruit population. Talking about parental cancer can be uncomfortable and potentially distressing, and it is possible that many young people may have not wanted to participate in the study for this reason. The young people who participated in the study had varied experiences with parental cancer and varied in the length of time since their parent's initial diagnosis. This included young people whose parent had been diagnosed with metastatic cancer or had died from cancer, which may have coloured how they perceived their parent's cancer and remembered their experiences. Despite this, there were common themes among the experiences that young people reported and those whose experiences was some time ago were easily able to recall and recount them in great detail.

The findings of this research indicate that young people's perceptions of their parent's cancer can be usefully described within the framework of the Common-Sense Model of Self-Regulation. It should be noted that it was not the aim of this research to make conclusions about the relationships between specific types of illness cognitions, coping strategies, and psychological morbidity, but instead to examine how young people's perceptions of their parent's cancer correspond with the dimensions of the Common-Sense Model of Self-Regulation. The present study extends previous research by exploring young people's perceptions of their parent's cancer in the context of a theoretical framework. This enables the opportunity for measures based on the model to be developed and used in further research, and potentially clinical practice, to identify those at risk of poor psychological adjustment following a parent's cancer diagnosis. Hence, this work may provide a basis for the development of quantitative measures of illness cognitions in young people who have a parent with cancer.

Future quantitative research should expand on these findings by determining the relationships between young people's illness cognitions, coping strategies, and psychological adjustment following their parent's cancer diagnosis. Quantitative research should be conducted, using a larger sample size, to examine differences in illness cognitions among young people of different ages, and to ensure that the findings may be generalised and replicated within the wider cancer population. Future research may also explore how young people's perceptions of their parent's cancer impact their own coping with personal cancer symptoms as an adult, using a longitudinal design. This research may provide valuable insight for the development of interventions that target harmful beliefs and promote positive coping strategies to support young people through the cancer experience.

Reflexive statement[5]

Reflexivity is considered to be an integral part of rigorous qualitative research. When employing a qualitative research, a double hermeneutic is created. An individual's experiences are context-bound; that is, they cannot be free of the setting and situation in which they occurred, or the mind of the individual. The experiences of the individual are interpreted and analysed by the researcher who is inseparable from the qualitative research process. The researcher brings with them their own experiences, assumptions, and beliefs, which will influence the research process. It is important for qualitative researchers to acknowledge the socially constructed nature of the world and recognise that complete objectivity is impossible to achieve because they are part of the social world under study (Green & Thorogood, 2018; Jootun, McGhee, & Marland, 2009). The process by which researchers consider their own experiences, assumptions, and beliefs, and how these may influence the interpretation of qualitative data, is referred to as reflexivity (Jootun et al., 2009). This involves critical reflection about the cultural, political, social, ideological, and experiential origins of one's own perspective. The following section contains the personal reflections of the researcher and will be written from first-person perspective.

It is important to disclose that my interest in researching young people's experiences of parental cancer stems from my own experience with parental cancer as a teenager. My mother was diagnosed with early-stage breast cancer when I was 14 years old. Her treatment involved an initial lumpectomy, followed by six cycles of chemotherapy, and six weeks of daily radiotherapy. She is currently 13 years post-diagnosis and living cancer-free. This experience was instrumental in shaping the person that I am today. After I finished high school, I studied at university with the intention of someday contributing to cancer research. Initially, I completed a Bachelor of Science specialising in molecular biosciences and

---

[5] Reflexive statement was not included in the published paper.

microbiology, but then transitioned into psycho-oncology and began working with the Cancer and Behavioural Science team at the Flinders Centre for Innovation in Cancer. It was here that I felt I could make a meaningful contribution to the lives of people diagnosed with cancer and their families.

My experience with parental cancer was influenced by the sociodemographic context in which it occurred. My family are working class white Australian (of European ancestry) and were living in rural South Australia at the time of my mother's cancer diagnosis. My mother was the primary income earner and had to resign from her job in order to undergo treatment – which, because of our rural location, she had to travel to receive. Fortunately, the upfront costs of my mother's cancer treatment were minimal, owing to Australia's publicly funded universal health care system, Medicare. My parents were married – and still are to this day – and my father's income was enough to cover our living costs. Because I lived in a two-parent household, additional household and caring responsibilities were able to be split between my father, my sister, and me. Although my mother was very open about her cancer and how it affected her, it is likely that my living situation sheltered me from the brunt of her diagnosis. I imagine that this would have been different had my mother been a single parent. My experience with parental cancer was likely influenced by mine and my mother's gender and the quality of our relationship. Importantly, my mother's cancer was caught early, and her treatment has thus far been effective in keeping her in remission for 12 years and counting.

A reflexive practice involves considering how one's own experiences, assumptions, and beliefs might influence the research process. Throughout the undertaking of this research, I was aware of the potential for my own experience of parental cancer to influence each stage of the research process – from the study design, data collection, and data analysis, through to the presentation of findings. I was aware that my experience of parental cancer could be perceived as a weakness in the research – and, indeed, I felt hesitant to disclose my experience to participants and other researchers alike. My interpretation of the data was through the lens of my own experience. Within a positivist epistemological framework, subjectivity is seen as a threat to validity (Newton, Rothlingova, Gutteridge, LeMarchand, & Raphael, 2011), and it could be argued that this chapter would have presented an entirely different set of findings had the study been designed and conducted by a researcher with different life experiences. But Gough (2003) argues that reflexivity should be approached with the intention of making use of subjectivity; acknowledging and embracing subjectivity within qualitative research can strengthen the credibility of findings (Gough, 2003).

It was important that I disclosed my own experience of parental cancer to participants at the beginning of interviews. This was done with the intention of creating a safe space for young people to talk about their experiences, thoughts, and feelings, and to facilitate a level of detachment between myself and the content of the interview. I felt that this disclosure was helpful for building rapport with the participant – by giving them some assurance that I understood what they were going through and that the experiences they shared would be safe with me. Moreover, by naming my own experience of parental cancer at the beginning of the interview, I was then able to separate myself from the experiences shared by the participant. This process of detachment is called bracketing or phenomenological reduction; it is the cognitive process by which the researcher puts aside their beliefs (or, in this case, experiences), and refrains from making judgements about what they are observing or hearing (Jootun et al., 2009).

A consideration for how my own experience might influence my interpretation of the experiences of the young people I interviewed was built into the study design. The CSM was used as a theoretical framework to explore young people’s perceptions of their parent’s cancer and a deductive approach was employed to analyse data. This meant that the patterns of meaning that were identified within young people’s experiences were grounded within a theoretical framework rather than being solely interpreted through the lens of my own personal experience. The interview schedule was developed based on the dimensions of the CSM and served as a helpful reminder that my role as the interviewer was to prompt, probe, and encourage participants to share their personal experiences. Although I had disclosed my own experience with parental cancer to participants, I strived to maintain broad, open questioning and neutral responses to the experiences, thoughts, and feelings that they shared. Through this research, I felt a great sense of personal responsibility to handle the experiences that were shared with me with honesty and integrity; I felt that I owed it as much to the participants as I did to my 14-year-old self.

3.1. Chapter Summary and Future Directions

Chapter 3 presented a qualitative exploration of young people's perceptions of their parent's cancer using the CSM as a theoretical framework. Semi-structured interviews were conducted with eleven young people aged 15-24 years who had a parent diagnosed with cancer. Overall, findings demonstrated that the CSM was relevant to the experiences they described and their perceptions of their parent's cancer. Importantly, these findings provide a sound theoretical basis from which to adapt the PPIQ for use in this cohort.

The overall aim of this thesis was to develop and validate an instrument that may be used to assess illness perceptions in adolescents and young adults (AYAs) who have a parent with cancer. Chapter 2 presented a systematic review that compared instruments measuring illness perceptions that were developed and validated for use among family members caring for an individual with a chronic physical illness. The PPIQ was identified as the most appropriate measure to adapt for use among AYAs with a parent with cancer. As explained in Section 2.1, the PPIQ assesses the aspects of illness perceptions outlined in the CSM. Prior to adapting the PPIQ, it was necessary to examine whether the CSM was an appropriate framework for describing young people's illness perceptions related to their parent's cancer.

The qualitative study presented in this chapter confirms that young people's perceptions of their parent's cancer correspond to the dimensions of the CSM, and therefore supports the adaptation of the PPIQ for use among AYAs impacted by parental cancer.

CHAPTER FOUR

Content validity of a cancer-specific version of the Perceptions of Parental Illness Questionnaire: A cognitive interview study using the think-aloud method and verbal probes

4.0. Preamble

Content validity, defined as the extent to which the constructs of interest are comprehensively represented by the items in the questionnaire (Terwee et al., 2007), is regarded as one of the most important measurement properties because it is a prerequisite for other types of validity (Zamanzadeh et al., 2015). Cognitive interviewing can be used to evaluate the content validity of a questionnaire when used for a particular purpose and in a specific population. The overall aim of cognitive interviewing is to understand how respondents interpret questionnaire items and to identify potential problems that respondents may experience when completing the questionnaire (Drennan, 2003). More specifically, cognitive interviewing can be used to identify problems in questionnaires related to the relevance, comprehensiveness, comprehension, and interpretation of questionnaire items (Peterson, Peterson, & Powell, 2017).

Chapter 4 describes the adaptation of the original Perceptions of Parental Illness Questionnaire (PPIQ) to produce a cancer-specific version of the questionnaire and the subsequent cognitive interview study conducted to evaluate the content validity of the adapted questionnaire items among adolescents and young adults (AYAs) impacted by parental cancer.

Abstract

Objective: To evaluate the content validity of a cancer-specific version of the PPIQ by identifying the problems that AYAs experience when completing the questionnaire.

Methods: Cognitive interviews were conducted with young people impacted by parental cancer. A think-aloud approach was employed in combination with verbal probes to assess relevance, comprehensiveness, comprehension, and accurate interpretation of items, and to identify problems with recall and the response format. Interview transcripts were analysed using a directed qualitative content analysis approach.

Results: Four young people (age range = 15-23 years; *M* = 19.5 years; female = 4) participated in cognitive interviews. In total, 197 problems were identified, the majority of which were related to difficulties using the think-aloud method. Items in the *cause, identity, emotional representations*, and *consequences* subscales yielded the most difficulties. There were no problems identified related to the response format.

Conclusions: Feedback provided by participants supported the content validity of the PPIQ-C, with items appearing to be highly relevant to their perceptions of their parent's cancer. Problems identified were addressed through several changes to the questionnaire; six items were reworded to address problems with comprehension, additional items were included to assess aspects of illness perceptions that were not covered by the questionnaire, and revisions were made to instructions for the *identity* and *cause* subscales.

Introduction

Leventhal’s Common-Sense Model of Self-Regulation (CSM) is the most widely applied model for describing how people cope in the face of an illness or health threat. According to the model, people form beliefs about their illness (collectively defined as “illness perceptions” or “illness representations”) that directly influence their coping behaviours, and in turn, their physical health and psychological outcomes (Diefenbach & Leventhal, 1996; Leventhal et al., 2016a; Petrie & Weinman, 2006). Much of the research exploring the relationships between illness perceptions and health outcomes has utilised the Illness Perception Questionnaire (IPQ) and its derivatives, the Revised Illness Perception Questionnaire (IPQ-R; Moss-Morris et al., 2002) and the Brief Illness Perception Questionnaire (Brief IPQ; Broadbent et al., 2006).

Weinman and colleagues (1996) originally developed the IPQ to assess cognitive representations of illness in individuals experiencing chronic illness. It is comprised of five subscales, derived from the dimensions of illness representation outlined in the CSM; these are, *identity* (ideas about the label and symptoms associated with the illness), *cause* (ideas about aetiology), *timeline* (perceived duration of the illness and whether the illness is acute, chronic, or cyclical/episodic), *consequences* (expected effects and outcomes), and *cure/control* (perceptions of how one controls or recovers from the illness). The IPQ was specifically designed to be a general measure of illness perceptions that could be used across illness groups. Although it has a core set of items, additional items can be included for specific illnesses or health threats; in particular, the *identity* and *cause* subscales can be modified by including symptoms and possible causes specific to the illness of interest.

Since its original development, the IPQ has been revised to address minor psychometric problems with the *timeline* and *cure/control* subscales (Moss-Morris et al., 2002). The IPQ-R incorporated a *cyclical timeline* subscale and included separate subscales to assess perceptions of *treatment control* and *personal control*. An additional two subscales were included to assess perceptions of *coherence* (personal understanding of the illness or the extent to which the illness “makes sense”) and *emotional representations* (emotional beliefs and subjective feelings about the illness).

Although the IPQ/IPQ-R were originally developed for use in individuals who were experiencing the illness or health threat themselves, they have since been adapted by researchers to enable assessment of illness perceptions in their family members or significant others (e.g., Bassi et al., 2016; Felnhofer et al., 2019; S. Nelson, Slusar, Albert, Liu, & Riedy, 2016; A. Richardson et al., 2016b). Despite their widespread use in family members and significant others, there is limited (published) information on the validity of their use in these groups (in particular for chronic physical illness[6]).

It is important to acknowledge that validity is not an inherent property of an instrument, but rather is a property of the scores obtained when using an instrument for a specific purpose and within a specific group of respondents (Zamanzadeh et al., 2015). Therefore, the validity of the IPQ/IPQ-R should be examined each time it is adapted for use in family members or significant others across illness groups, age groups, cultural groups, and genders. Without evidence of the validity of these instruments when applied to a particular respondent group, and for a specific purpose, findings related to the strength of the relationship between illness perceptions and psychological outcomes in family members caring for someone with a chronic illness are questionable.

Validity is defined as the ability of an instrument to measure the properties of the construct of interest. There are three separate types of validity that are examined to determine the overall validity of an instrument; these are, content, construct, and criterion-related

---

[6] See systematic review of the psychometric properties of measures of illness perceptions developed for use among family members presented in Chapter 2.

validity. Content validity, which refers to the extent to which items capture the aspects of the constructs that they are intended to measure (Terwee et al., 2007; van Oort, Schroder, & French, 2011), is a prerequisite for other forms of validity (Zamanzadeh et al., 2015), and therefore should be regarded as the highest priority during initial instrument development or adaptation. One way to ensure content validity during the adaptation of an existing measure is by involving individuals from the target population during the adaptation process (French & Weinman, 2008). This allows researchers to identify what items might need to be modified or whether additional items should be included to reflect an aspect of the construct of interest that is unique to their context.

Furthermore, involving the target population provides insight into whether the people who complete the questionnaire are interpreting items in the way that they were originally intended (McCorry, Scullion, et al., 2013). There are four steps, or cognitive operations, that a respondent should go through when formulating a response to a questionnaire item: 1) understanding and interpreting what the question is asking, 2) retrieving relevant information from memory, 3) integrating this information into a summarised thought, and 4) translating this thought into the format of the response scale (Tourangeau, 1984; Tourangeau & Rasinki, 1988). Each of these cognitive operations represents a potential source for error. It is therefore essential to identify and address problems in cognitive operations because the respondent's interpretation of questionnaire items forms the basis of any inferences that are made from data collected using the questionnaire (Peterson et al., 2017). Moreover, having an understanding of how individuals interpret the items that are used to measure the constructs that define illness representation is essential for researchers involved in the design and development of interventions that aim to modify individuals' perceptions of their family member's illness (McCorry, Scullion, et al., 2013).

There are a number of cognitive interview techniques that may be used to examine potential problems with questionnaire items, provide evidence of content validity, and gain insight into the response processes and cognitive operations that respondents engage when completing a questionnaire. Two commonly used techniques are the “think-aloud” method and verbal probes. The former involves respondents describing their thinking as they respond to each item whereas the latter involves retrospective examination of respondent reasoning (Peterson et al., 2017). Using these techniques, researchers can identify differences between the respondent’s interpretation and the intended meaning of a particular item, and explore how problematic items may be improved based on the participant’s response (Peterson et al., 2017).

The systematic review reported in an earlier paper[7] found that young people with a parent with a chronic illness represent an understudied group. Of nine instruments included in the review, only one had been validated for use in adolescents; the Perceptions of Parental Illness Questionnaire (PPIQ; Bogosian et al., 2014). Additionally, the PPIQ was the only instrument included in the review that measured the aspects of illness representation outlined in the CSM. The PPIQ was developed as an age-appropriate measure of adolescents’ perceptions of their parent’s multiple sclerosis, but the authors suggest that it may be used in adolescents with a parent with other chronic illnesses with appropriate changes to the wording. Furthermore, the authors recommend additional validation research if the PPIQ is to be applied in the context of other chronic illnesses.

On this basis, the PPIQ was adapted to produce a cancer-specific version to assess illness perceptions in AYAs with a parent with a cancer. Therefore, the aim of this study was to evaluate, using the think-aloud method and verbal probing techniques, the content validity of the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C).

---

[7] Presented in Chapter 2.

Methods

Initial development of the Perceptions of Parental Illness Questionnaire-Cancer. The original PPIQ was developed, based on the IPQ and its derivatives, as a measure of adolescents' perceptions of their parent's multiple sclerosis. Prior to adapting the IPQ/IPQ-R, Bogosian and colleagues (2014) collected interview data to examine whether the CSM was an appropriate framework with which to describe adolescents' perceptions of their parent's multiple sclerosis. They generated questionnaire items directly from interview quotes, and then conducted cognitive interviews to assess the items and inform refinements.

The structure of the PPIQ deviates somewhat from the original subscales of the IPQ/IPQ-R and dimensions of the CSM. The PPIQ is comprised of seven subscales assessing *emotional representations* (5 items)*, adolescents' control* (4 items), *negative consequences for family* (3 items)*, positive consequences for adolescents* (4 items), *negative consequences for adolescents* (3 items), *timeline chronic* (4 items), and *timeline unpredictable* (2 items). A further four subscales assess beliefs about the *cause* of the illness; *psychological attributions* (2 items), *central nervous system attributions* (2 items)*, external/environmental attributions* (2 items), and *hereditary/chance attributions* (2 items).

Unlike the IPQ/IPQ-R, the PPIQ does not include subscales assessing cognitive representations relating to dimensions *identity*, *coherence*, and *treatment control*. This is because these subscales failed to load coherently onto factors during principal components analysis (Bogosian et al., 2014). Cognitive interviews also revealed problems with the *identity* and *treatment control* subscales; adolescents were not familiar with available treatments for multiple sclerosis and were confused about which symptoms could be attributed to multiple sclerosis and which to medical treatments (Bogosian et al., 2014). Despite this, the final version of the PPIQ demonstrated good content and construct validity and was the most appropriate instrument identified through the systematic review[8] to adapt and validate for use in AYAs with a parent with cancer.

Following the methodology used by Bogosian et al. (2014) to develop the PPIQ, qualitative interviews were conducted with eleven AYAs to examine whether the CSM was relevant to their perceptions of their parent's cancer.[9] Findings confirmed that young people's perceptions did correspond with the dimensions of the CSM, therefore interview data were used to adapt the PPIQ to produce the PPIQ-C.

A summary of the initial modifications made to the original PPIQ is shown in Table 9. The first modification made was to replace the words 'multiple sclerosis' with 'cancer'. Second, subscales for *identity*, *coherence,* and *treatment control* were added based on quotes from the qualitative interviews[10] and items from the IPQ/IPQ-R. Minor modifications were made to items that referred to cancer symptoms to also include side effects because the two are generally indistinguishable for people undergoing treatment for cancer (Ashley et al., 2013; Cleeland, 2007). Items in the *cause* subscale were amended to include the phrase 'my parent's cancer was caused by' leading into the statement being rated and additional items were included based on quotes from the qualitative interviews and items from the IPQ/IPQ-R.

---

[8] Presented in Chapter 2.
[9] See paper presented in Chapter 3.
[10] Findings reported in Chapter 3.

Table 9

Initial Adaptations to PPIQ Items to Produce the PPIQ-C

Subscale/Item	Change(s)	Rationale
Identity		
1. Fatigue (tiredness)	Added	Item included based on qualitative interview study
2. Pain	Added	Item included based on qualitative interview study
3. Weakness	Added	Item included based on qualitative interview study
4. Nausea (feeling sick in the stomach)	Added	Item included based on qualitative interview study
5. Vomiting	Added	Item included based on qualitative interview study
6. Hair loss	Added	Item included based on qualitative interview study
7. Weight gain	Added	Item included based on qualitative interview study
8. Weight loss	Added	Item included based on qualitative interview study
9. Skin irritation	Added	Item included based on qualitative interview study
10. Loss of appetite	Added	Item included based on qualitative interview study
11. Depression	Added	Item included based on qualitative interview study
12. Problems with concentration, thinking, or memory (chemo brain)	Added	Item included based on qualitative interview study
Emotional Representations		
13. My parent’s cancer symptoms are confusing to me	Moved to *coherence* subscale (note: original PPIQ did not include *coherence* subscale)	Item included in coherence subscale of IPQ-R
14. When I think about my parent’s cancer I get upset	-	
15. My parent’s cancer makes me feel angry	-	
16. My parent’s cancer worries me	-	
17. My parent having cancer makes me feel stressed	-	
18. I try not to think about my parent’s cancer	Added	Item included based on qualitative interview study
19. I feel overwhelmed when I think about my parent’s cancer	Added	Item included based on qualitative interview study
20. I try to hide my feelings about my parent’s cancer	Added	Item included based on qualitative interview study

Subscale/Item	Change(s)	Rationale
21. I feel numb when I think about my parent's cancer	Added	Item included based on qualitative interview study
Coherence		
22. I have a good understanding of my parent's cancer	Added	Item adapted from IPQ-R
23. I have enough information about my parent's cancer	Added	Item included based on qualitative interview study
24. I would like to know more about my parent's cancer	Added	Item included based on qualitative interview study
25. I would rather not know details about my parent's cancer	Added	Item included based on qualitative interview study
Personal Control		
26. I can help my parent manage their symptoms by looking after them	Wording changed to "I can help my parent manage their symptoms/side effects by looking after them"	Item adapted to refer to "symptoms/side effects" as the two are generally indistinguishable in the context of cancer
27. My parent's cancer symptoms get better when I do not stress them out (e.g. staying out late, arguing with brother or sister)	Wording changed to "My parent's cancer symptoms/side effects get better when I do not stress them out (e.g. staying out late, arguing with brother or sister)"	Item adapted to refer to "symptoms/side effects" as the two are generally indistinguishable in the context of cancer
28. If I'm not playing up, I can make my parent's symptoms get better	Wording changed to "If I'm not playing up, I can make my parent's symptoms/side effects get better"	Item adapted to refer to "symptoms/side effects" as the two are generally indistinguishable in the context of cancer
29. My parent not being stressed or worried can make their symptoms get better	Wording changed to "My parent not being stressed or worried can make their symptoms/side effects get better"	Item adapted to refer to "symptoms/side effects" as the two are generally indistinguishable in the context of cancer
30. There isn't anything I can do to make my parent's symptoms/side effects get better	Added	Item adapted from IPQ-R
31. I do what I can to help make my parent's life easier	Added	Item included based on qualitative interview study
Treatment Control		
32. There isn't a lot that can be done to treat my parent's cancer	Added	Item adapted from IPQ
33. My parent's treatment will be effective in curing their cancer	Added	Item adapted from IPQ
34. My parent's treatment can control their cancer	Added	Item adapted from IPQ-R
35. There is nothing that can be done to treat my parent's cancer	Added	Item adapted from IPQ-R
Consequences		
36. My parent's cancer causes arguments in the family	-	
37. My parent's cancer puts strain on the family	-	
38. My parent's cancer makes it more difficult to do family activities	-	
39. My parent's cancer has made me more responsible	-	
40. My parent's cancer has made me more independent	-	

Subscale/Item	Change(s)	Rationale
41. My parent's cancer has made me grow up quicker	Added	Item included based on qualitative interview study
42. My parent's cancer has made more understanding of other people	-	
43. My parent's cancer brought me closer to my family	-	
44. Because of my parent's cancer, I spend less time doing social activities (e.g. hobbies, sports)	-	
45. Because of my parent's cancer, I spend more time doing housework	-	
46. Because of my parent's cancer, I spend less time with my friends	-	
47. Because of my parent's cancer, I spend less time doing schoolwork	Added	Item included based on qualitative interview study
Timeline (acute/chronic)		
48. My parent's cancer will get worse	-	
49. My parent's cancer suddenly got worse and never got better	-	
50. I expect my parent to have cancer for the rest of their life	-	
51. My parent's cancer will stay the same	-	
52. My parent's cancer will improve in time	Added	Item adapted from IPQ-R
53. My parent will recover from their cancer	Added	Item included based on qualitative interview study
Timeline (cyclical/unpredictable)		
54. The severity of my parent's cancer symptoms change a great deal from day to day	Wording changed to "The severity of my parent's cancer symptoms/side effects change a great deal from day to day"	Item adapted to refer to "symptoms/side effects" as the two are generally indistinguishable in the context of cancer
55. The number of my parent's symptoms change a great deal from day to day	Wording changed to "The number of my parent's symptoms/side effects change a great deal from day to day"	Item adapted to refer to "symptoms/side effects" as the two are generally indistinguishable in the context of cancer
56. My parent's symptoms/side effects come and go in cycles	Added	Item adapted from IPQ-R
57. I never know how my parent will be feeling one day to the next	Added	Item included based on qualitative interview study
58. My parent goes through cycles where their cancer symptoms/side effects get better or worse	Added	Item adapted from IPQ-R
59. My parent's cancer is very unpredictable	Added	Item adapted from IPQ-R
Cause		
60. Stress or worry	Wording changed to "My parent's cancer was caused by stress or worry"	"My parent's cancer was caused by" added to improve comprehension by young people
61. Family problems or worries	Wording changed to "My parent's cancer was caused by family problems or worries"	"My parent's cancer was caused by" added to improve comprehension by young people
62. Scars on the spine	Removed	Not relevant to cancer

Subscale/Item	Change(s)	Rationale
63. Nerve damage	Removed	Not relevant to cancer
64. A germ or virus	Wording changed to “My parent’s cancer was caused by a germ or virus”	“My parent’s cancer was caused by” added to improve comprehension by young people
65. Hereditary – it runs in the family	Wording changed to “My parent’s cancer is hereditary – it runs in the family”	“My parent’s cancer was caused by” added to improve comprehension by young people
66. Chance or bad luck	Wording changed to “My parent’s cancer was caused by change or bad luck”	“My parent’s cancer was caused by” added to improve comprehension by young people
67. My parent’s cancer was caused by poor diet	Added	Item adapted from IPQ-R
68. My parent’s cancer was caused by sun exposure	Added	Item included based on qualitative interview study
69. My parent’s cancer was caused by lack of exercise	Added	Item included based on qualitative interview study
70. My parent’s cancer was caused by smoking	Added	Item adapted from IPQ-R
71. My parent’s cancer was caused by drinking alcohol	Added	Item adapted from IPQ-R
72. My parent’s cancer was caused by chemicals or radiation in the environment	Added	Item adapted from IPQ
73. My parent’s cancer was caused by bad or toxic people in their life	Added	Item included based on qualitative interview study

The initial version of the PPIQ-C consisted of nine subscales assessing *identity*, *emotional representations, coherence*, *personal control*, *treatment control, consequences, timeline (acute/chronic), timeline (cyclical/unpredictable)*, and *cause* (initial draft of PPIQ-C presented in Appendix D). Although the items assessing the *cause* dimension of the CSM have a factor structure that is independent to the rest of the scale (Bogosian et al., 2014; Moss-Morris et al., 2002; Weinman et al., 1996), at this stage in the adaptation process, and for the purposes of the cognitive interviews, this was treated as a single subscale. Items in the *identity* subscale were rated on a four-point Likert scale, where 1 = *no impact*, 2 = *some impact,* 3 = *quite a bit of an impact*, 4 = *a very big impact*. Items in all other subscales were rated on a five-point Likert scale with responses ranging from *strongly disagree* to *strongly agree*. This initial version of the PPIQ-C was evaluated in the cognitive interview study described below.

Cognitive interviews.

Design. Structured, one-on-one cognitive interviews were conducted with young people who had a parent diagnosed with cancer. A think-aloud approach was used in combination with verbal probes to assess relevance, comprehensiveness, comprehension, and accurate interpretation of items, and to identify any other problems with recall and the response format. Cognitive interviewing techniques were chosen in accordance with de Leeuw and colleagues (2004) who recommend that they are suitable for use in adolescents and young adults.

Participants. Young people who participated in the qualitative interviews described in an earlier paper[11] were invited to attend another interview to provide feedback on the PPIQ-C. Snowball sampling techniques were also employed, whereby young people who

---

[11] See paper presented in Chapter 3.

participated in a cognitive interview were asked to forward information about the study to others who might be interested in participating.

Young people aged 12-24 years were eligible to participate if they had a parent who had been diagnosed with, or received treatment for, any type or stage of cancer within the last five years, and were living at home at the time of their parent's diagnosis and treatment. Four young people participated in cognitive interviews (age range = 15-23 years; *M* = 19.5 years; female = 4). Two participants had a mother with breast cancer (one had finished active treatment, and the other was receiving palliative care for metastatic breast cancer), one participant had a father who had completed treatment for non-Hodgkin's lymphoma, and the remaining participant had a mother who had died from ovarian cancer. The time since initial diagnosis ranged from 18 months to eight years.

Interviews. Participants were invited to participate in the interview using the format that they were most comfortable with (i.e., face-to-face, telephone, or web-conferencing program). All participants chose to participate in the interview via telephone. Interviews ranged from 38 to 85 minutes in length (*M* = 58 minutes). Interviews were audio-recorded and transcribed verbatim.

At the beginning of the interview, the interviewer (CF) provided verbal instructions on how to use the think-aloud technique while answering the questionnaire items (Box 1). Participants were asked to familiarise themselves with the think-aloud technique using a warm-up task, during which they were asked to describe their thought processes as they imagined their house and counted the number of windows it had (Peterson et al., 2017; Willis, 2005). Following this, participants were prompted to use the think-aloud technique as they answered each individual item of the questionnaire. Participants whose parent had completed treatment or had died were asked to think back to when their parent was undergoing treatment for cancer as they responded. Participants were asked to stop at the end of each subscale, where a combination of scripted and spontaneous probes were used to further explore problems experienced while thinking aloud and to assess comprehension and interpretation of items (e.g. *"what does the word 'numb' mean to you as it is used in Question 9?"* and *"can you repeat Question 19 in your own words?"*), retrieval of information needed to answer items (e.g. *"how easy or difficult is it to remember the impact of each of the side effects on your parent?"* and *"before you answered the questions about the cause of your parent's cancer, how much had you thought about it?"*), applicability and adequacy of content (e.g. *"how comfortable did you feel answering these questions?"*, *"how well does each of these statements apply to you?"*, and *"was there anything not included in these questions that should be added about your perceptions of the consequences of your parent's cancer on your personal and family life?"*), and suitability of the response format (e.g. *"were you able to find an accurate answer for each statement from the response options shown?"*).

Box 1

Verbal Instructions Provided to Participants

I wanted to first of all thank you for participating in this interview today and for agreeing to give us your feedback on the questionnaire that we have developed to help health professionals understand more about young peoples' thoughts and beliefs about their parent's cancer. We really appreciate your participation. Your feedback is really valuable and will help us to improve the questionnaire that we have designed.

Today's interview will probably go for about an hour. During the interview we will go through each section of the questionnaire separately. The first thing I will ask you to do is to read each questionnaire item aloud and then think aloud as you choose your each of your answers. I will then ask you some specific questions about the questionnaire items that will help us to see if they could be improved in any way. Do you have a copy of the questionnaire in front of you?

I will be recording the interview so that it can be typed up and then I can analyse the feedback that you've given. I'll be using a voice recorder to do this. Anything you say will remain anonymous and your name will not be attached to any quotes that I might use when presenting or publishing the research – is that okay with you?

The questionnaire that you will be giving feedback on asks questions about your parent's cancer. You might find that some of the questionnaire items are difficult or upsetting to think about or answer. Or you might find that it doesn't really bother you. People react differently to their parent's cancer, and that's perfectly okay.

Please be assured that this is a safe environment. Please don't feel bad or embarrassed if you do find that going through the questionnaire is upsetting or uncomfortable. Remember that your participation in the interview is voluntary and you can choose not to answer any of the questions and if you would like to leave the interview at any time you are welcome to do that as well.

As I mentioned we're interested in what you thinking about while you are answering each of the questions, so I'm going to ask you to talk through your thinking as you work through each of the questions. You can say whatever you're thinking as if I'm not listening to you and it doesn't matter if you say anything that's negative about what you're reading, I won't be offended; if you say that something sounds strange or if it's confusing or anything like that it would actually be really helpful for us to know that and that's really why we're getting this feedback from you. You might also have thoughts that don't relate to the questions and that's okay as well, you don't have to worry about if what you're saying makes sense to me. When we get to the end of each section we'll go through your responses.

The technique that I'm going to ask you to use – the talking through your responses or talking through your thinking as you answer the questions – is called thinking aloud. It can be a really strange thing to do because we're not really used to talking through all of the thoughts that we might have, so before we move on to the questionnaire items, I have practice activity to help get you used to talking through your thinking. What I want you to do is to visualise your house and I want you to think about how many windows there are in your house. I want you to talk through what you're thinking and seeing in your mind as you count the windows in your house, does that make sense?

Ethical considerations. The study was reviewed and approved by the Social and Behavioural Research Ethics Committee at Flinders University (project number 8022). All participants received written information about the study, formal written consent forms, and a copy of the PPIQ-C to refer to during the telephone interview. Participants aged younger than 18 years required parental consent to participate. Signed consent forms were returned to the researcher prior to the interview. Demographic and parent cancer-related medical information was collected through the completion of a questionnaire. Participants were assured that the information they shared during the interview would remain confidential and that their personal information would not be published. Participants were informed that they were able to decline to answer any questions asked during the interview and they could withdraw from the study at any time.

Analysis. Prior to analysis, interview transcripts were verified for accuracy and were read-through several times for familiarisation. Data were analysed using a directed qualitative content analysis approach (Braun & Clarke, 2006, 2013). Traditionally, content analysis is a method whereby data are coded and analysed numerically. A *qualitative* approach was used because this allowed data to be summarised numerically as well as examined for patterns, themes, and meaning (V. D. Goodman, 2011; Zhang & Wildemuth, 2009). Predetermined categories were used to code the data, which meant that the analysis was *directed* in approach (Assarroudi, Nabavi, Armat, Ebadi, & Vaismoradi, 2018; Zhang & Wildemuth, 2009). Because two different cognitive interviewing techniques were used in conjunction (think-aloud method and verbal probing), transcripts were divided into sections of text referring to each technique, and the analysis was conducted in two separate stages.

During the first stage of analysis, participants’ thinking processes were coded according to the type of problem they experienced as they responded to each item. Codes were defined according to specific categories that have been used in previous think-aloud studies (Aujla, Vedhara, Walker, & Sprigg, 2018; McCorry, Scullion, et al., 2013; van Oort et al., 2011). These were; (1) no problems identified, (2) insufficient thinking aloud, (3) reread question, (4) difficulty generating an answer, (5) difficulty with response format, (6) questioned content, (7) confusion or misinterpretation, (8) incongruent response, and (9) missing answer. Definitions for each code are described in Table 10.

Table 10

Definitions of Codes used to Categorise Problems Identified using the Think-Aloud Method

Code (type of problem experienced)	Definition
No problems identified	Participant demonstrated no problems with the item
Insufficient thinking aloud	Participant did not engage in adequate thinking aloud while responding to the question
Reread question	Participant reread question or seriously stumbled in answering the question
Difficulty generating an answer	Participant expressed that they were not sure of the response that they would provide, either because of problems with understanding the question or because the item was not relevant to their experience
Difficulty with response format	Participant expressed problems with indicating their answer using the response format or understanding the response options
Questioned content	Participant suggested problems with how an item was worded or the relevance of the item
Confusion or misinterpretation	Participant expressed that they did not fully understand the question, answered the question in a way that highlighted that they had misinterpreted, or misread what the question was asking
Incongruent response	Participant gave a written and verbal response that did not match
Missing answer	Participant did not respond to the question (e.g. unintentionally skipped question)

Note. Table adapted from Aujla et al. (2018), McCorry, Scullion, et al. (2013), and van Oort et al. (2011).

Following this, participants' responses to verbal probes were coded according to the source of confusion of any problems identified: (1) comprehension or understanding of item, (2) recall or retrieval of information, (3) judgement or acceptability of item, (4) instructions or response options, and (5) comprehensiveness or adequacy of content (see Table 11). Data collected using the think-aloud technique and verbal probes were then combined and problems identified were thematically organised.

Table 11

Definitions of Codes used to Categorise Problems Identified using Verbal Probing Techniques

Code (source of confusion)	Definition
Understanding or comprehension of item	Participant demonstrated their level of understanding of the item wording, terminology, and structure
Recall or retrieval of information	Participant commented on whether they had the necessary information to answer an item (including if they experienced difficulty retrieving information needed to answer an item)
Judgement or acceptability of item	Participant expressed an opinion relating to the content of an item or subscale (e.g. they were uncomfortable with an item)
Instructions or response options	Participant provided feedback on the instructions or response options
Comprehensiveness or adequacy of content	Participant commented on the whether a subscale covered all aspects of the construct (including suggestions of additional items for inclusion)

Note. Table adapted from Peterson et al. (2017).

Results

Summary of problems experienced while completing the PPIQ-C. The frequency of problems experienced by participants while thinking aloud is outlined in Table 12. The total number of problems experienced was 197 out of a possible 288 (68.4%).[12] Participants most frequently experienced problems with items in the cause (17.6%), identity (16.2%), emotional representations (15.2%), and consequences (15.2%) subscales. The majority of problems were related to missing answers or insufficient thinking aloud (72.6%), suggesting that problems generally stemmed from difficulty with the think-aloud method rather than the questionnaire content. Difficulty generating an answer was the second most common problem experienced by participants (20.3%), followed by confusion or misinterpretation of the item (4.1%). None of the participants experienced difficulty with the response format.

---

[12] Total number of problems experienced calculated by summing the number of “no problems” and “total problems” experienced by participants when responding to each question.

Table 12

Frequency of Problems Experienced Across PPIQ-C Subscales

	No problems	Type of problem experienced						Total problems, *N* (%)
		Missing answer or insufficient thinking aloud	Re-read question	Difficulty generating an answer	Questioned content	Confusion or misinterpretation	Incongruent response	
Identity	16	17	0	14	0	1	0	32 (16.2)
Emotional representations	7	18	0	10	0	0	2	30 (15.2)
Coherence	3	9	0	4	0	0	0	13 (6.6)
Personal control	11	12	0	0	0	1	0	13 (6.6)
Treatment control	5	6	2	5	0	1	0	14 (7.1)
Consequences	18	25	0	3	0	2	0	30 (15.2)
Timeline (acute/chronic)	10	14	0	0	0	0	0	14 (7.1)
Timeline (cyclical/unpredictable)	7	12	0	4	0	1	0	17 (8.6)
Cause	14	30	0	0	2	2	0	34 (17.6)
Total problems, *N* (%)	91	143 (72.6)	2 (1)	40 (20.3)	2 (1)	8 (4.1)	2 (1)	197

After participants had answered each item in a subscale, they were prompted to elaborate on problems that they experienced while using the think-aloud technique. A summary of the problems identified using both the think-aloud technique and verbal probes is provided below.

Challenges with thinking aloud. Cognitive interviews were conducted remotely via telephone and participants were provided with a copy of the questionnaire prior to the interview taking place. As a result, participants were familiar with the questionnaire content, and many appeared to have determined their answers to the items prior to the interview. In general, insufficient thinking aloud appeared to arise when the participant was responding to the item without any problems. Insufficient thinking aloud was most prominent in two participants, although both appeared to be comfortable identifying problems (e.g., when they did not understand the meaning of a particular word) and responding to verbal probes.

Difficulty generating an answer or choosing a single answer. Several participants experienced difficulty generating an answer for items in the identity subscale. This appeared to be related to problems with recall or retrieval of the information necessary to provide an answer to the item (e.g., *"I don't actually know the answer to that one"*). One participant stated:

> *"I can't remember her [mother] losing her appetite, but she might, I don't know, I can't recall if she lost her appetite significantly, so I don't want to like give an answer to that cos I don't actually remember and I don't want to give a false answer."* (in response to item 10)

Participants also experienced difficulty generating an answer when they could answer the item in multiple ways. For example, in reference to item 19 ("I feel overwhelmed when I think about my parent's cancer"), one participant said: *"I think it's something that can* *actually be overwhelming; I think it would just depend on [my] mood"*. This was also expressed by another participant, whose mother had finished active treatment for breast cancer, as she reflected on her experience to answer item 33 ("My parent's treatment will be effective in curing their cancer"):

> *"Oh, that's a hard one. Because I was very indecisive because I thought that she was going to pass away, but at the same time I thought that she was going to come out strong."*

One participant, whose mother had died from ovarian cancer, expressed a lot of difficulty summarising her experience, which spanned a number of years, within the confines of the questionnaire. This caused her to have difficulty choosing a single answer for many items, often opting to choose multiple answers or "neither agree nor disagree" (e.g., *"...for some of them I just had a line that went between a couple or connected two responses"*). In reference to item 38 ("My parent's cancer makes it more difficult to do family activities"), the participant stated that:

> *"I think probably for the first at least 2, 2-3 years it didn't really make that much of a difference, it would probably affect it a little bit but not massively. ... I think it's probably the last 6 months there would be a massive turning point, we'd have to cancel a holiday or have a trip the week that she died that, this thing that we had planned and she was too ill to go, there were significant things like that but all that was in the last 6 months ... so it wasn't across the entire period. So that's why that was a bit mixed."*

Participants whose parent had completed curative treatment did not appear to share this difficulty. When asked how easy or difficult it was for her to recall her experience in order to respond to the questionnaire items, one participant stated that:

> *"…it was an easy thing to remember because of how much I saw of my mum at that point in time and I feel like if anyone asked me questions about that time of my life, where mum was going through cancer, I'd be able to easily say it because of how much I saw and how much it affected me."*

Problems with wording or interpretation of items. Several problems with wording and interpretation of items were identified. While thinking aloud, one participant expressed confusion over the meaning of an item in the *identity* subscale (item 3 – weakness):

> *"As for weakness, is that like, I'm just not 100% sure what that means… She [mother] didn't show many kinds of weakness I guess you could say, but I mean like anybody going through chemo[therapy] or radio[therapy] or cancer, there are moments of weakness, but I wouldn't say that she's weak at all. If anything, she's one of the strongest people I know really."*

When prompted to share her thoughts on the *identity* subscale, the participant identified her own misinterpretation:

> *"…the only one that I was a bit confused about was the weakness one, which I guess now I'm thinking about it, is more like loss of strength…"*

Although none of the participants expressed any problems when answering the item using the think-aloud technique, two participants admitted that they weren't *"really sure"* when they were prompted to explain their interpretation of item 9 (skin irritation). Despite this, both participants demonstrated an accurate understanding of the item and were able to suggest an example of skin irritation: *"I think it would just be having some bad reaction to your skin"* and *"I'd think of a rash or really dry skin, just not normal skin I guess."*

The youngest participant also identified problems with wording of items in the *personal control* (item 28 – "If I'm not playing up I can make my parent's symptoms get better"), treatment control (item 33 – "My parent's treatment will be effective in curing their cancer"), timeline (item 54 – "The severity of my parent's cancer symptoms or side effects change a great deal from day to day"), and cause subscales (item 72 – "My parent's cancer was caused by chemicals or radiation in the environment"). Specifically, the participant was unsure of the meaning of the phrases "playing up" and "chemicals or radiation in the environment" and the words "curing" and "severity". This seemed to stem primarily from the participant doubting her own interpretation or reading the item too quickly. When prompted to elaborate on her comprehension of the item in the *treatment control* subscale, the participant stated:

> *"I had one of those moments where you just completely say the sentence too fast and you just don't take in what any of the words mean … and I said the complete wrong word, but everyone will know that says 'curing' … it was just because I was mashing it up in my head."* (in reference to item 33)

Problems with identity subscale. The *identity* subscale is designed to assess respondents' perceptions of the impact of a range of symptoms and side effects that their parent may have experienced as a result of their cancer diagnosis and treatment. Several participants suggested that "anxiety" should be included in the subscale. One participant also highlighted that respondents may not necessarily perceive their parent's symptoms and side effects as having a *negative* impact. The instructions for the *identity* subscale ask respondents to *"select the answer that best reflects overall how much of an impact you think each symptom and side effect has had on your parent"*. The participant expressed that problems with concentration, thinking, or memory (item 12) were something that had a significant impact on her mother, but that it wasn't *"a negative thing"* and was something that her family could make a joke of. When prompted to elaborate on this, she stated that if she were to rate the *negative* impact of problems with concentration, she would have rated this as “only some impact”.

Problems with cause subscale. The *cause* subscale is designed to assess respondents’ personal beliefs about the cause of their parent’s cancer, and items are comprised of a range of perceived causes of cancer, regardless of whether they are scientifically or medically substantiated. Two participants questioned whether item 66 (“My parent’s cancer was caused by chance or bad luck”) should be split into two separate items. Both participants said that they agreed that their parent’s cancer was caused by chance or was just “something that happened to occur” but disagreed that it was bad luck. One participant also suggested that “personal salvation” or “sin” could be included as a potential item.

Some of the items included in the cause subscale elicited a strong response in participants. One participant felt that including items such as “My parent’s cancer was caused by bad or toxic people in their life” was *“taking the piss”*. She elaborated:

> *“I don’t, honestly I don’t really like that section. Because for me I don’t know much about like other types of cancers, but like can cancer be caused by stress or worry? Or like, can it be caused by family problems? I guess I don’t actually know if that is an accurate representation, to be like that’s why my mum got it because they were stressed. Like I think the hereditary one is good because that’s a legit thing. Poor diet, I’m not 100% sure if that’s actually a thing or not, I don’t know. But like the ones that say that alcohol, smoking, and the chemicals, I think they're all legit ones, but like [item] 12, like bad or toxic people in their life, that, like that to me doesn’t feel like a reason for why someone would have cancer, that’s not a medically diagnosed thing as to how someone would have cancer, and that’s kind of like what this section to me would be, like what's the actual reason as to why they got cancer.”*

This was supported by another participant who said that *"a lot of them [items] are things that personally I wouldn't attribute it [her mother's cancer] to"*. In contrast, another participant felt strongly that her mother's cancer was caused by stress:

> *"So when my mum was going through cancer she told me that she was diagnosed because of how much you know, stress, because of her mum dying at the age of sixteen."*

This participant also answered "strongly agree" to items 61 ("My parent's cancer was caused by family problems or worries") and 73 ("My parent's cancer was caused by bad or toxic people in their life"), validating the inclusion of items that refer to psychological or superstitious attributions.

Sensitivity to content. The potential for questionnaire items to cause discomfort or distress was raised by two participants. When prompted to elaborate on her level of comfort responding to questions in the *personal control* subscale, one participant said:

> *"I can't speak for every single kid that's gone through it [parental cancer] because even if I am comfortable I still do feel sad and upset about some things when some people bring stuff up, so I think that with any question that anyone asks someone going through cancer is still going to trigger something, doesn't matter if you ask something big or something small it's still going to set back a memory and could possibly trigger someone; so I don't think it's necessarily something that you have control over, I think it just goes for the person's experience and how bad it was."*

Another participant pointed out that respondents may find answering questions in the *timeline* subscale particularly challenging:

> *"I think you could break that section up ... with a gap, and then put a bit of a trigger warning. Because like I said I'm fine with talking about it and just the way it is, but*

> *not everyone might be. Because like the bit before is all just about different symptoms and stuff that is happening, but this is getting, this bits like the, kind of like the heaviness of it ... These are heavier questions."*

Overall feedback on the PPIQ-C. In general, participants found the questionnaire simple to complete and the instructions easy to understand. Participants described the questionnaire content as highly relevant and acknowledged that questions implied an understanding by the developers of the experiences and feelings that young people might have following their parent's cancer diagnosis. This was summarised by one participant, who stated that *"the words were taken out of my mouth"* and *"it's nice to know that someone kind of put [the questionnaire item] there and understands"*. Participants also suggested that the questionnaire could help young people make sense of their emotional responses to their parent's cancer diagnosis:

> *"…it'd help people, especially teenagers to identify those of kinds of feelings inside themselves, there are certain triggers and that, that will help them be like, oh that's what that is."*

Furthermore, participants felt that the questionnaire could be used as a "discussion starter" to open up a conversation about their parent's cancer – both as a way for young people to be able to organise and express their thoughts and feelings, but also for health professionals to use as a tool to check in with them. One participant explained:

> *"I feel like it's a good way to have like a little seminar about it really. Like a good seminar to have with people, just to have a chat about it. ... I'm lucky where we're very open about discussing how I'm feeling and whatever, but a lot of people wouldn't. And like if a professional had something like this, and for teenagers or whatever, they could make it like ... about how you feel the day went…"*

Final modifications made to the PPIQ-C. A summary of the final modifications made to the PPIQ-C is shown in Table 13. Based on cognitive interviews, changes were made to the wording of six items and an additional four items were included in the questionnaire. One item was added to the *cause* subscale ("My parent's cancer was caused by a higher power (e.g., it was an act of God") and one item was separated into two parts ("My parent's cancer was caused by chance" and "My parent's cancer was caused by bad luck"). Instructions for the *identity* subscale were modified to clarify that respondents should select the answer that reflects their perception of how much of a *negative* impact each symptom and side effect has had on their parent (i.e., *"please select the answer that best reflects overall how much you believe each symptom and side effect has negatively impacted your parent"*). Instructions for the *cause* subscale were also modified to specify that items *"describe things that young people commonly believe might have caused their parent's cancer"*. Furthermore, a brief content warning was added to the beginning of the questionnaire: *"You may find that some of the questions are upsetting or difficult to think about and answer. Please answer as many questions as you feel able to."* Although no problems were identified with the response format, a decision was made to amend the response options for the *identity* subscale to 1 = *no impact*, 2 = *some impact*, 3 = *quite a big impact*, 4 = *a very big impact* for the purpose of clarity and conciseness. The final version of the PPIQ-C, including instructions for each section of the questionnaire, can be found in Appendix E.

Table 13

Final Modifications Made to Refine PPIQ-C Items

Subscale/Item	Change(s)	Rationale
Identity		
1. Fatigue (tiredness)	-	-
2. Pain	-	-
3. Weakness	Wording changed to “Physical weakness (loss of strength)”	Feedback from one participant in the cognitive interviews suggested that the original wording could be interpreted in multiple ways (i.e., emotional weakness or physical weakness).
4. Nausea (feeling sick in the stomach)	-	-
5. Vomiting	-	-
6. Hair loss	-	-
7. Weight gain	-	-
8. Weight loss	-	-
9. Skin irritation	Wording changed to “Skin irritation (dry skin, rashes, or itchiness)”	Feedback from participants in the cognitive interviews suggested that it was unclear what original wording referred to.
10. Loss of appetite	-	-
11. Depression	-	-
12. Anxiety	Added	Multiple participants in the cognitive interviews suggested that this item should be included.
13. Problems with concentration, thinking, or memory (chemo brain)	-	-
Emotional Representations		
14. When I think about my parent’s cancer I get upset	-	-
15. My parent’s cancer makes me feel angry	-	-
16. My parent’s cancer worries me	-	-
17. My parent having cancer makes me feel stressed	-	-
18. My parent having cancer makes me feel worried about the future	Added	Feedback from one participant in the cognitive interview suggested that this item should be included.
19. I try not to think about my parent’s cancer	-	-
20. I feel overwhelmed when I think about my parent’s cancer	-	-
21. I try to hide my feelings about my parent’s cancer	-	-
22. I feel numb when I think about my parent’s cancer	-	-

Subscale/Item	Change(s)	Rationale
Coherence		
23. My parent’s cancer symptoms are confusing to me	Wording changed to “My parent’s cancer is confusing to me”	Wording revised to be consistent with other items in subscale.
24. I have a good understanding of my parent’s cancer	-	-
25. I have enough information about my parent’s cancer	-	-
26. I would like to know more about my parent’s cancer	-	-
27. I would rather not know details about my parent’s cancer	-	-
Personal Control		
28. I can help my parent manage their symptoms/side effects by looking after them	-	-
29. My parent’s cancer symptoms/side effects get better when I do not stress them out (e.g. staying out late, arguing with brother or sister)	-	-
30. If I’m not playing up, I can make my parent’s symptoms/side effects get better	Wording changed to “If I’m not misbehaving, I can make my parent’s symptoms/side effects get better”	Wording revised to improve comprehension by younger age group.
31. My parent not being stressed or worried can make their symptoms/side effects get better	-	-
32. There isn’t anything I can do to make my parent’s symptoms/side effects get better	-	-
33. I do what I can to help make my parent’s life easier	-	-
Treatment Control		
34. There isn’t a lot that can be done to treat my parent’s cancer	-	-
35. My parent’s treatment will be effective in curing their cancer	-	-
36. My parent’s treatment can control their cancer	-	-
37. There is nothing that can be done to treat my parent’s cancer	-	-
Consequences		
38. My parent’s cancer causes arguments in the family	-	-
39. My parent’s cancer puts strain on the family	-	-
40. My parent’s cancer makes it more difficult to do family activities	-	-
41. My parent’s cancer has made me more responsible	-	-

Subscale/Item	Change(s)	Rationale
42. My parent's cancer has made me more independent	-	-
43. My parent's cancer has made me grow up quicker	-	-
44. My parent's cancer has made more understanding of other people	-	-
45. My parent's cancer brought me closer to my family	-	-
46. My parent's cancer brought me closer to my parent	Added	Feedback from one participant in the cognitive interview suggested that this item should be included separately from item 45.
47. Because of my parent's cancer, I spend less time doing social activities (e.g. hobbies, sports)	-	-
48. Because of my parent's cancer, I spend more time doing housework	-	-
49. Because of my parent's cancer, I spend less time with my friends	-	-
50. Because of my parent's cancer, I spend less time doing schoolwork	-	-
Timeline (acute/chronic)		
51. My parent's cancer will get worse	-	-
52. My parent's cancer suddenly got worse and never got better	-	-
53. I expect my parent to have cancer for the rest of their life	-	-
54. My parent's cancer will stay the same	-	-
55. My parent's cancer will improve in time	-	-
56. My parent will recover from their cancer	-	-
Timeline (cyclical/unpredictable)		
57. The severity of my parent's cancer symptoms/side effects change a great deal from day to day	Wording changed to "The intensity of my parent's cancer symptoms/side effects change a great deal from day to day"	Wording revised to improve comprehension by younger age group.
58. The number of my parent's symptoms/side effects change a great deal from day to day	-	-
59. My parent's symptoms/side effects come and go in cycles	-	-
60. I never know how my parent will be feeling one day to the next	-	-
61. My parent goes through cycles where their cancer symptoms/side effects get better or worse	-	-
62. My parent's cancer is very unpredictable	Wording changed to "My parent's symptoms/side effects are very unpredictable"	Wording revised to be consistent with other items in subscale.

Subscale/Item	Change(s)	Rationale
Cause		
63. My parent’s cancer was caused by stress or worry	-	-
64. My parent’s cancer was caused by family problems or worries	-	-
65. My parent’s cancer was caused by a germ or virus	-	-
66. My parent’s cancer is hereditary (it runs in the family)	-	-
67. My parent’s cancer was caused by chance or bad luck	Separated into two items: “My parent’s cancer was caused by chance” and “My parent’s cancer was caused by bad luck”	Feedback from multiple participants in the cognitive interviews suggested that this item should be split into two separate items.
68. My parent’s cancer was caused by poor diet	-	-
69. My parent’s cancer was caused by sun exposure	-	-
70. My parent’s cancer was caused by lack of exercise	-	-
71. My parent’s cancer was caused by smoking	-	-
72. My parent’s cancer was caused by drinking alcohol	-	-
73. My parent’s cancer was caused by chemicals or radiation in the environment	Wording changed to “My parent’s cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)	Feedback from participants in the cognitive interviews suggested that it was unclear what original wording referred to.
74. My parent’s cancer was caused by bad or toxic people in their life	-	-
75. My parent’s cancer was caused by a higher power (e.g., it was an act of God)	Added	Feedback from one participant in the cognitive interviews suggested that this item should be included.

Discussion

Young people with a parent with a chronic illness remain a largely forgotten group in research examining the relationships between illness perceptions and psychological adjustment in family members and carers. The development of the PPIQ-C represents an important step towards addressing this gap in the literature. The purpose of this study was to evaluate the content validity of the PPIQ-C by examining the nature and source of problems young people experienced while responding to questionnaire items.

Cognitive interviews identified a number of problems, most of which were attributed to difficulty using the think-aloud method rather than the questionnaire content. In particular, some participants struggled to consistently “think aloud” throughout the interview. The think-aloud method relies on participants being able to verbalise their cognitions while responding to questionnaire items (McCorry, Scullion, et al., 2013). De Leeuw et al. (2004) argue that young people have a natural inclination to articulate their thought processes, but may require more extensive probing than adult populations. In the present study, participants were provided with the questionnaire in advance, which meant that they were familiar with the questionnaire content prior to participating in the interview. This was done to ensure that participants were fully informed about what they would be asked to do as part of the study, however this may have restricted the effectiveness of the think-aloud method to identify problems that young people experienced while responding to the questionnaire items.

It is also possible that participants felt uncomfortable using the think-aloud method to talk through their responses, or alternatively, that they found it difficult to verbalise their thoughts about an experience that may have been traumatic and likely still caused them distress. On the other hand, insufficient thinking aloud generally appeared to occur when the participant was responding to the item without any problems. Moreover, direct verbal probes were able to identify problematic items that would have otherwise been overlooked due to insufficient thinking aloud. Aujla et al. (2018) support the use of a verbal probes in combination with the think-aloud method; they argue that this approach could help to facilitate a conversation between the researcher and respondent to identify the sources of confusion in problematic questions and determine how they could be addressed. Indeed, this appeared to be true for the present study.

Problems with the questionnaire content were predominantly related to difficulty generating an answer or choosing a single response to an item. In general, this was expressed by participants who were reflecting on a past experience (i.e., their parent had completed treatment or had died) as a tendency to select the middle response option (*"neither agree nor disagree"*). In terms of the cognitive operations described by Tourangeau (1984), this may be interpreted as the respondent experiencing difficulty retrieving the information needed to select an accurate answer (step 2: retrieval) or having made a judgement that the item did not apply to their specific situation (step 3: judgement). This finding suggests that the PPIQ-C may have reduced validity when used to retrospectively assess the illness perceptions of young people with a parent with cancer, in particular when respondents are referring to their parent's experience as a whole rather than at a specific point in time. In the present study, the participant whose parent had died appeared to have greater difficulty selecting an accurate response than participants whose parents had completed curative treatment. Respondents were prompted to think back to when their parent was undergoing treatment, although this may not have been specific enough; for example, "treatment" may be interpreted as meaning curative treatment or palliative care, and perceptions of *illness identity* (i.e., symptoms and side effects) may vary significantly between active treatment and end of life phases of the cancer trajectory. In addition to this, their parent may have received more than one type of treatment, and each treatment may be associated with different side effects of varying intensity, potentially having implications for illness perceptions relating to *consequences* and *personal control*.

Cognitive interviews informed several changes to the questionnaire. First, changes were made to the wording of six items to address problems with comprehension. The original PPIQ was designed using language appropriate for adolescents aged 12-19 years and evaluated among adolescents in the UK. In some cases, problematic wording of items seemed to arise from differences in the English language used within the British and Australian contexts (e.g., the phrase "playing up" as used in item 28). For other items, misinterpretation stemmed from the item lacking clarity or being open to interpretation (e.g., item 3 – weakness). Second, participants identified aspects of illness perception that were not covered by the questionnaire content. Specifically, participants provided suggestions for additional items in the *identity, consequences,* and *cause* subscales, all of which were included in the final PPIQ-C (i.e., item 12 – anxiety, item 18 – *"My parent having cancer makes me feel worried about the future"*, and item 75 – *"My parent's cancer was caused by a higher power (e.g., it was an act of God)"*). Third, the instructions for the *identity* subscale were amended to specify that respondents should answer according to perceived *negative* impacts of their parent's cancer symptoms and side effects. Finally, participants suggested that the *cause* subscale had the potential to be polarising and alienating. Amendments were made to the instructions for this subscale to emphasise that items reflected things that young people *believe* may have caused their parent's cancer, although these beliefs may not necessarily be substantiated by medical and scientific research.

Importantly, young people highlighted that extra care needed to be taken to inform respondents of the potentially triggering nature of topics covered by questionnaire items. In particular, the *timeline* subscale asks young people to consider their beliefs about whether their parent will recover from their cancer. This was addressed by including a brief content warning at the beginning of the questionnaire. It should be noted that both participants who raised this concern also highlighted that the questionnaire provided an important opportunity for young people to identify and explore their feelings about their parent's cancer. Moreover, they expressed that this experience led them to feel less isolated in their experience and, in a sense, to normalise the beliefs that they held about their parent's cancer. In other words, exposure to the content of the questionnaire indicated to the participants that someone understood what they were going through and that they were not alone in their experience. For clinicians using the questionnaire, this may act not only as a tool to identify young people who might be at risk of poor psychosocial outcomes, but also as a pathway to conversation with young people about how they are coping with their parent's cancer diagnosis in order to identify where extra support may be needed.

Although the findings of the present study provide important preliminary insight into the content validity of the PPIQ-C, they should be considered in light of potential limitations. Two important limitations have already been discussed: 1) that participants were provided with the questionnaire prior to participating in the cognitive interview, and 2) that almost all participants were responding to the questionnaire retrospectively. Additional limitations should also be noted. First, the study sample consisted of four young people, all of whom were female. Other studies utilising the think-aloud approach tend to involve a much larger sample (Belzer et al., 2013; French, Cooke, Mclean, Williams, & Sutton, 2007; McCorry, Scullion, et al., 2013; van Oort et al., 2011), although, Aujla and colleagues (2018) found that data saturation was reached after six interviews. Furthermore, the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) Risk of Bias checklist, which is used to assess the methodological quality of studies that evaluate the psychometric properties of quantitative instruments, confirms that 4-6 cognitive interviews are adequate for assessing content validity during instrument development (Mokkink et al., 2018; Prinsen et al., 2018; Terwee et al., 2018). Second, although the PPIQ-C is intended for use in young people aged 12-24 years, the youngest participant was 15 years old. Further research should be conducted to examine the content validity of the PPIQ-C using a larger sample that is representative of the intended target population in terms of age, gender, cultural background, type of parent’s cancer, and time since parent’s cancer diagnosis. This would allow for a more comprehensive analysis of the validity of the PPIQ-C within specific subgroups to be undertaken.

Despite these limitations, feedback provided by the young people who participated in cognitive interviews has contributed to improving the relevance, comprehensiveness, and comprehension of PPIQ-C items. The next stage in the development and initial testing of the PPIQ-C will determine the factor structure of the scale. In addition, further psychometric evaluation will be undertaken to assess the internal consistency, construct validity, and test-retest reliability of the PPIQ-C. The availability of a psychometrically robust measure of illness perceptions in young people who have a parent with cancer will enable more comprehensive examination of how young people think about their parent’s cancer and how this impacts their ability to cope in the face of their parent’s diagnosis.

4.1. Chapter Summary and Future Directions

Chapter 4 presented a cognitive interview study conducted to evaluate the content validity of the PPIQ-C. Four young people aged 15-23 years who had a parent diagnosed with cancer participated in cognitive interviews. A think-aloud approach was employed in combination with verbal probes to evaluate the relevance, comprehensiveness, comprehension, and accurate interpretation of items, and to identity problems with recall and the response format.

Overall, findings provided strong evidence supporting the content validity of the PPIQ-C; participants described the questionnaire content as highly relevant to their experiences with parental cancer. Cognitive interviews facilitated identification of problems with the questionnaire content, including items that were difficult for respondents to interpret accurately and aspects of illness perception that were not covered by the questionnaire items. Other problems related to difficulty generating an answer or choosing a single response to an item; this was observed in participants who were reflecting on a past experience because their parent had completed treatment or had died. This finding may indicate reduced validity of the questionnaire when used to retrospectively assess illness perceptions among AYAs impacted by parental cancer.

The feedback provided by participants informed several changes to the PPIQ-C; six items were reworded to address problems with comprehension, additional items were included to assess aspects of illness perceptions that had not been covered by the questionnaire, and revisions were made to instructions for the *identity* and *cause* subscales. Ultimately, these changes contributed to improving the relevance, comprehensiveness, and comprehension of the PPIQ-C.

In summary, the overall aim of this thesis was to develop and validate an instrument to assess illness perceptions in AYAs who have a parent with cancer. When developing a measurement instrument, it is important to ensure that it is valid for use in a specific population and for a particular purpose. Content validity is considered one of the most important measurement properties because it is a prerequisite for other types of validity (Zamanzadeh et al., 2015). The findings of the study presented in Chapter 4 support the content validity of the PPIQ-C. The next stage of this research will determine the factor structure, and evaluate the internal consistency, construct validity, and test-retest reliability, of the PPIQ-C.

CHAPTER FIVE

Psychometric evaluation of the Perceptions of Parental Illness Questionnaire-Cancer among adolescents and young adults who have a parent with cancer

5.0. Preamble

The availability of a psychometrically robust measure of illness perceptions among young people who have a parent with cancer may facilitate investigation of the mechanisms by which illness perceptions impact young people’s coping and psychological outcomes following their parent’s cancer diagnosis. However, in order for an instrument to be used with psychometric confidence, its factor structure, reliability, and validity must be determined.

Chapter 4 described the development of the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C) and the cognitive interview study conducted to establish its content validity among young people with a parent with cancer. Following this, a cross-sectional survey study was conducted to determine the factor structure of the PPIQ-C and to evaluate its internal consistency, construct validity, and test-retest reliability. This study is presented in Chapter 5.

Abstract

Objective: To evaluate the psychometric properties of a cancer-specific version of the Perceptions of Parental Illness Questionnaire (PPIQ-C).

Methods: Adolescents and young adults (aged 12-24 years) who had a parent diagnosed with cancer were invited to participate in the study. Young people (*n* = 437) completed the PPIQ-C and measures of psychological distress, post-traumatic stress symptoms, post-traumatic growth, and coping strategies. A subsample (*n* = 23) retested the PPIQ-C after two weeks. Exploratory factor analysis was conducted to determine the dimensional structure of the PPIQ-C. Internal consistency and construct validity were also evaluated.

Results: The final PPIQ-C is comprised of 67 items across 14 dimensions. There were minor differences between the structure of the original PPIQ and the PPIQ-C, including the addition of subscales measuring perceptions of illness *identity*, *coherence*, and *treatment control*. Test-retest correlations provided initial support for the stability of the instrument. Correlations between PPIQ-C subscales and validated measures of psychological distress, post-traumatic stress, and post-traumatic growth provided support for construct validity.

Conclusions: This study provides preliminary evidence of the reliability and validity of the PPIQ-C. The availability of the PPIQ-C will enable further examination of the relationships between illness perceptions, coping, and psychosocial outcomes among young people who have a parent with cancer. The PPIQ-C has many potential applications; its use may assist in identifying young people at risk of poor psychosocial outcomes and provide insights for potential targets for supportive interventions.

Introduction

Although many young people cope well in the face of a parent's cancer diagnosis, a large proportion experience high levels of psychological distress and unmet psychosocial needs (Patterson et al., 2017), potentially contributing to the development of significant, on-going psychological problems. Previous research has tended to focus on describing the prevalence and severity of poor psychosocial outcomes (Faccio, Ferrari, & Pravettoni, 2018; Grabiak et al., 2007; Morris, Turnbull, Preen, Zajac, & Martini, 2018; Osborn, 2007; Visser et al., 2004), and while this has provided important insights into how coping and psychological development may be disrupted as a result of a parent's cancer diagnosis, less is known about the factors that influence the extent to which functioning may be impacted. Furthermore, where factors that influence psychosocial outcomes have been established in a research context, evidence for their predictive ability varies in consistency, and the clinical utility of significant findings remains largely unexplored (Walczak et al., 2018).

Our understanding of the mechanisms by which parental cancer affects adolescent and young adult (AYA) children may be extended through the identification of factors that put young people at risk for, or protect them from, poor psychosocial outcomes. In practice, this knowledge is essential to the development and implementation of effective prevention and early intervention strategies designed to support young people through the cancer experience. Moreover, this knowledge can be applied in the development and validation of specific screening tools designed to identify young people who are at risk of developing more serious psychological problems and would benefit from supportive intervention.

A significant proportion of the literature describing the psychosocial impact of parental cancer on young people has focused on factors related to the *illness or medical treatment* (i.e., type of cancer, time since diagnosis, stage of illness, treatment type), the *family* (i.e. family functioning, parent-child communication, parenting style), or the *parent* (i.e., parent gender, negative mood or depressive affect, level of distress) (Faccio et al., 2018; Krattenmacher et al., 2012; Morris et al., 2016; Osborn, 2007; Visser et al., 2004). Where studies have examined *child*-related factors, they have primarily focused on the impact of the child's age and gender on psychosocial adjustment, with isolated studies examining personality traits (E. Nelson & While, 2002; Visser, Huizinga, Hoekstra, van der Graaf, & Hoekstra-Weebers, 2007). The development and validation of the Offspring Cancer Needs Instrument (OCNI) was an urgently needed step towards addressing this gap (Patterson et al., 2013; Patterson et al., 2011). Using the OCNI, McDonald and colleagues (2016) found that higher levels of unmet psychosocial needs were associated with higher levels of distress among young people with a parent with cancer, and that this relationship was independent of other known risk factors, such as poor family functioning and child gender.

Despite these important findings, further research is needed to achieve a comprehensive understanding of the factors that influence young people's adjustment to their parent's cancer. In particular, the potential impact of young people's perception of their parent's illness has been largely overlooked. The limited available research in this area suggests that illness perceptions may be an important predictor of psychosocial outcomes among young people. Research conducted by Compas et al. (1994) found that children's distress was related to their perceptions of the seriousness and stressfulness of their parent's cancer. This finding was supported by further research conducted by Huizinga et al. (2005), although the relationship was only found to be significant in daughters and not sons. Further investigation is warranted; especially given that illness perceptions are widely recognised as having significant implications for coping and psychological adjustment in those diagnosed with the illness themselves (Hagger et al., 2017).

The Common-Sense Model of Self-Regulation (CSM) provides a framework for understanding how individuals make sense of their experience when faced with a health threat (Leventhal et al., 2003; Leventhal et al., 2016a). The model is based on the premise that individuals are active problem solvers and construct cognitive and emotional representations of their illness by integrating information gathered from current experiences with the illness (i.e., symptoms and side effects related to the illness), interactions with external social influences (i.e., information from medical professionals, family members, friends, or the media), and pre-existing knowledge of the illness (i.e., past experience with the illness themselves or with someone close to them being diagnosed) (Hagger & Orbell, 2003). Therefore, an individual’s illness perceptions are subjective and are not necessarily scientifically or medically substantiated. This makes them an appropriate target for personalised interventions that aim to challenge inaccurate or unrealistic perceptions and improve understanding of the illness (A. Richardson & Broadbent, 2017).

A large body of research has tested the validity of the CSM for a range of health conditions. The Illness Perception Questionnaire (IPQ; Weinman et al., 1996), and its derivatives the Revised Illness Perception Questionnaire (IPQ-R; Moss-Morris et al., 2002) and the Brief Illness Perception Questionnaire (Brief IPQ; Broadbent et al., 2006), were developed to enable quantitative assessment of illness perceptions according to the dimensions of the CSM (i.e., *identity*, *timeline*, *cause*, *consequences*, and *controllability*). Although originally developed for use in individuals diagnosed with an illness, the IPQ/IPQ-R have been used to assess illness perceptions in spouses, family members, and informal carers (for example; Graham et al., 2016; Karademas, Dimitraki, Thomadakis, & Giannousi, 2019; A. Richardson et al., 2016a, 2016b; Wu et al., 2013). Bogosian et al. (2014) produced the first adaptation of the IPQ/IPQ-R for specific use in young people with a parent with a chronic illness (the Perceptions of Parental Illness Questionnaire; PPIQ). The PPIQ was developed and validated for adolescents with a parent with multiple sclerosis, however Bogosian and colleagues (2014) propose that with appropriate changes to wording and additional validation research, it may be used in other illness groups.

Earlier papers describe the qualitative exploration of young people's perceptions of their parent's cancer using the CSM as a framework[13] and the development of a cancer-specific version of the PPIQ to assess illness perceptions in young people with a parent with cancer.[14] Following on from this work, the aim of this study was to evaluate the psychometric properties of the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C).

Methods

Design. A cross-sectional design was used to assess the psychometric properties of the PPIQ-C, including its factor structure, internal consistency, construct validity, and test-retest reliability. Data were collected via an online survey that contained the PPIQ-C and measures of psychological distress (Kessler Psychological Distress Scale), post-traumatic stress symptoms (Impact of Events Scale-Revised and Children's Revised Impact of Events Scale), post-traumatic growth (Post-Traumatic Growth Inventory-Short form and Post-Traumatic Growth Inventory for Children-Revised), and coping strategies (Brief COPE and KIDCOPE). Psychological outcomes and coping responses were assessed to facilitate construct validity testing of the PPIQ-C. Socio-demographic information about the participant and their parent, and medical information about the parent's cancer was also collected. Test-retest reliability was assessed among a subsample of participants who completed a two-week retest of the PPIQ-C.

---

[13] See paper presented in Chapter 3.
[14] See paper presented in Chapter 4.

Participants. Young people aged 12-24 years were eligible to participate if they had a parent who had been diagnosed with, or received treatment for, any type or stage of cancer within the last five years, and were living at home at the time of their parent's diagnosis and treatment. This included young people who had a parent who had been diagnosed with curable, metastatic, or advanced cancer, and young people who had a parent who had died from cancer.

Recruitment. Several recruitment strategies were employed to reach an extensive and broad sample of young people affected by parental cancer. First, the study was advertised through cancer support and advocacy groups, including Canteen, Breast Cancer Network Australia, Prostate Cancer Foundation Australia, Australian Melanoma Research Foundation, and Cancer Voices. Second, a study-specific Facebook page[15] and Instagram account[16] were used to target advertisements to young people within the eligible age range. Third, offspring members of Canteen were contacted by a research assistant via telephone and email and invited to complete the survey. Finally, potential participants based in Australia, as well as the UK and Ireland, the United States of America, Canada, and New Zealand, were recruited through Prolific,[17] an online platform for participant recruitment for research studies.

Procedure. Potential participants younger than 16 years required parental consent to complete the survey. Online advertisements contained a link to written information about the study and a screening questionnaire that assessed the young person's eligibility to participate in the study and determined whether parental consent needed to be obtained. The screening questionnaire asked potential participants to select their age group and confirm (through self-report) that they had a parent who had been diagnosed with cancer and that they had lived

---

[15] https://www.facebook.com/ParentalCancerStudy
[16] https://www.instagram.com/parentalcancerstudy/
[17] https://www.prolific.co/

with their parent at the time of their diagnosis. Potential participants who met the eligibility criteria and were aged 16 years or over were redirected to the online version of the survey. Those who met the eligibility criteria but were aged under 16 years were redirected to a short form that informed them that they would need their parent's permission to participate in the study and invited them to submit their contact details. These participants were contacted by a research assistant and provided with written information about the study as well as a written consent form for their parent to sign and return. Once the parental consent form had been returned, they were provided with a link to the online survey. Following completion of the survey, participants were given the option of providing an email address in order to receive a $10 iTunes voucher as reimbursement for their time and effort. Participants recruited through Prolific received a £5 reimbursement.

Ethical considerations. The study was reviewed and approved by the Social and Behavioural Research Ethics Committee at Flinders University (project number 8023). It also received ethical clearances from organisations involved in the recruitment process. Participants indicated their informed consent through the completion of the survey. Parental consent was obtained for participants younger than 16 years.

Survey.

Socio-demographic and medical questions. The survey contained items that asked for demographic information about the participant (i.e., age, gender, country of residence, level of education), demographic information about the parent diagnosed with cancer (i.e., gender), and cancer-related medical information (i.e., type of cancer, time since diagnosis, metastasis status, stage of cancer care continuum at time of survey completion).

Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C). The PPIQ-C is a 76-item measure of illness perceptions in young people who have a parent with cancer.

Item development and content validity testing is described in an earlier paper.[18] The version of the questionnaire subjected to psychometric testing is presented in Appendix E. The PPIQ-C is comprised of three sections that measure the different dimensions of illness perceptions outlined in the CSM. Each section of the questionnaire has a separate dimensional structure.

Items in the first section measure the *identity* dimension of the CSM and represent symptoms and side effects that can be attributed to cancer and its treatment. Participants were asked to think about their parent's cancer symptoms and side effects and select an answer that best reflected how much they believed each symptom and side effect had negatively impacted their parent. Responses were indicated on a four-point Likert scale, where 1 = *no impact*, 2 = *some impact*, 3 = *quite a big impact*, and 4 = *a very big impact*.

Items in the second section measure the *emotional representations, coherence, personal control*, *treatment control*, *consequences, timeline (acute/chronic),* and *timeline (cyclical/unpredictable)* dimensions. Items comprised statements that described thoughts and beliefs that young people may have about their parent's cancer. Participants were asked to select an answer that indicated their level of agreement with each statement. Items were rated on a five-point Likert scale (1 = *strongly disagree*, 2 = *disagree*, 3 = *neither agree nor disagree*, 4 = *agree*, 5 = *strongly agree*).

Finally, items in the third section measure the *cause* dimension and represent common beliefs about the cause of cancer (e.g., "My parent's cancer was caused by bad luck" and "My parent's cancer was caused by smoking"). Participants were asked to select an answer that indicated their level of agreement with each item. Items were rated on a five-point Likert scale, where 1 = *strongly disagree*, 2 = *disagree*, 3 = *neither agree nor disagree*, 4 = *agree*, 5 = *strongly agree*.

---

[18] See paper presented in Chapter 4.

Kessler Psychological Distress Scale (K10). The K10 is a widely used 10-item measure of general psychological distress that is also sensitive to symptoms of both anxiety and depression (Kessler et al., 2002; Kessler et al., 2003). Participants were asked to reflect on how they had been feeling during the previous 30 days and respond to items using a five-point Likert scale (1 = *none of the time*, 2 = *a little of the time*, 3 = *some of the time*, 4 = *most of the time*, 5 = *all of the time*). Total scores range from 10 to 50, with higher scores indicating higher levels of distress. The following categories are used by the Clinical Research Unit for Anxiety and Depression, a joint initiative of St Vincent's Hospital and the University of New South Wales, and in primary healthcare settings to monitor distress: 10-19 = *likely to be well*, 20-24 = *likely to have a mild mental disorder*, 25-29 = *likely to have a moderate mental disorder*, 30-50 = *likely to have a severe mental disorder* (Andrews & Slade, 2001; Australian Bureau of Statistics, 2012).

Although the measure was originally developed for use in adults, it has since been validated in Hong Kong for use in children as young as 12 years (Chan & Fung, 2014) and has been used in Australia to measure distress among young people aged 12-24 years (McDonald et al., 2016; Patterson et al., 2013). The scale demonstrated excellent internal consistency in the present sample (Cronbach's α = .93).

Impact of Events Scale-Revised (IES-R). The IES-R is a 22-item measure of subjective distress caused by traumatic events (Horowitz, Wilner, & Alvarez, 1979; Weiss & Marmar, 1997). In the present study, the IES-R was used to assess post-traumatic stress symptoms in the subsample of young adult participants aged 18 to 24 years. Items comprised difficulties that individuals may experience following a stressful life event (e.g., "Any reminder brought back feelings about it" and "I avoided letting myself get upset when I thought about it or was reminded of it"). Participants were asked to think about their parent's cancer diagnosis and treatment and indicate how much they were distressed or bothered by each item during the previous seven days. Responses were indicated on a five-point Likert scale (0 = *not at all*, 1 = *a little bit*, 2 = *moderately*, 3 = *quite a bit*, 4 = *extremely*). Total scores range from 0-88, with scores ≥24 suggestive of PTSD being a clinical concern (Weiss & Marmar, 1997). Internal consistency in the present sample was excellent (Cronbach's α = .94).

Children's Revised Impact of Events Scale (CRIES). In the subsample of adolescent participants aged 12 to 17 years, the CRIES was used to assess post-traumatic stress responses to parental cancer. The scale consists of eight items that were adapted from the original IES for use in children aged eight years and above (Horowitz et al., 1979; Perrin, Meiser-Stedman, & Smith, 2005; P. Smith, Perrin, Dyregrov, & Yule, 2003). Participants were asked to think about their parent's cancer diagnosis and treatment and indicate how often they experienced each item during the previous seven days. Items were rated on a non-linear scale, where 0 = *not at all*, 1 = *rarely*, 3 = *sometimes*, and 5 = *often*. Total scores range from 0-40, with scores ≥17 indicating high likelihood of PTSD (Perrin et al., 2005). Internal consistency in the present sample was good (Cronbach's α = .89).

Post-Traumatic Growth Inventory-Short form (PTGI-SF). The PTGI-SF is a 10-item measure of post-traumatic growth (Cann et al., 2010; Tedeschi & Calhoun, 1996). In the present study, the PTGI-SF was used to assess the extent to which parental cancer led to the experience of significant positive change (i.e., post-traumatic growth) in the young adult subgroup. Items consist of positive outcomes that individuals may experience in the aftermath of a traumatic event (e.g., "I have a great appreciation for the value of my own life" and "I know better that I can handle difficulties"). Participants were asked to rate each item based on the extent to which they had experienced the change in the time since their parent's cancer diagnosis. Items were rated on a six-point Likert scale (0 = *I did not experience this change*, 1 = *I experienced this change to a very small degree*, 2 = *I experienced this change* *to a small degree*, 3 = *I experienced this change to a moderate agree*, 4 = *I experienced this change to a great degree*, 5 = *I experienced this change to a very great degree*). Total scores range from 0-50, with higher scores indicating greater perceived positive change. The scale demonstrated excellent internal consistency in the present sample (Cronbach's α = .90).

The Post-Traumatic Growth Inventory for Children-Revised (PTGI-C-R). The PTGI-C-R is an adaptation of the PTGI (Kilmer et al., 2010; Tedeschi & Calhoun, 1996) and was used to assess post-traumatic growth in the subgroup of adolescent participants. The scale consists of 10 items, each of which are rated on a four-point Likert scale (0 = *no change*, 1 = *a little change*, 2 = *some change*, 3 = *a lot of change*). Total scores range from 0 to 30. Internal consistency in the present sample was good (Cronbach's α = .86).

Brief COPE. The Brief COPE is a 28-item measure of strategies that may be used to cope with a stressful life event (Carver, 1997; Carver, Scheier, & Weintraub, 1989). In the present study, the Brief COPE was used to assess engagement with approach (e.g., active coping, positive reframing, seeking emotional support, etc.) or avoidance (e.g., denial, behavioural disengagement, self-distraction, self-blame, etc.) coping strategies in the subsample of young adults. Participants were asked to select the response that best reflected how frequently they had been using each strategy to help them cope with their parent's cancer diagnosis (e.g., "I've been refusing to believe that it has happened" and "I've been getting comfort and understanding from someone"). Items were rated on a four-point Likert scale, where 1 = *I haven't been doing this at all*, 2 = *I've been doing this a little bit*, 3 = *I've been doing this a medium amount*, 4 = *I've been doing this a lot*. Scores on items relating to active coping, positive reframing, planning, acceptance, seeking emotional support, and seeking information support were summed to produce total scores for the *approach strategies* subscale. Scores on items relating to denial, substance use, venting, behavioural disengagement, self-distraction, and self-blame were summed to produce total scores for the *avoidance strategies* subscale. Both subscales demonstrated good internal consistency in the present sample (*approach strategies*, Cronbach's α = .86; *avoidance strategies*, Cronbach's α = .81).

KIDCOPE. Adolescent participants completed the frequency scale of the KIDCOPE to assess engagement with coping strategies in response to their parent's cancer diagnosis. The KIDCOPE consists of 10 items rated on a four-point Likert scale whereby participants indicate how often they use the strategy described in each item (0 = *not at all*, 1 = *sometimes,* 2 = *a lot of the time,* 3 = *almost all the time*) (Spirito, Stark, & Williams, 1988). Examples of items include: "I thought about something else; tried to forget it; and/or went and did something like watch TV or play a game to get it off my mind" and "I just accepted the problem because I knew I couldn't do anything about it". The factor structure of the KIDCOPE has been found to vary in different populations (Spirito, 1996). Spirito (1996) suggests that this is because the coping strategy "…cannot be separated from the situation", meaning that categorisation of a particular coping strategy (as positive/negative, adaptive/maladaptive, approach/avoidance, etc.) does not remain the same across different circumstances. Therefore, an initial factor analysis was conducted to determine the factor structure of the KIDCOPE in the present study. Two components were identified. The first component consisted of items relating to social support, cognitive restructuring, resignation (or, alternatively, acceptance), and emotional regulation (item 7b). Scores on these items were summed to produce total scores for the *approach strategies* subscale. The second component consisted of items related social withdrawal, self-criticism, blaming others, wishful thinking, and emotional regulation (item 7a). Similarly, scores on these items were summed to produce total scores for the *avoidance strategies* subscale. In the present study, the *avoidance strategies* subscale demonstrated acceptable internal consistency (Cronbach's α = .72), however internal consistency in the *approach strategies* subscale was low

(Cronbach's α = .47). Similar findings were observed by T. F. Smith and colleagues (2013) in their study of coping among adolescents with spinal cord injury; they suggested that the KIDCOPE may not adequately measure the range of approach coping strategies used by adolescents within this particular cohort, especially as the KIDCOPE was designed as a general measure of coping in youth. This may also be the case for young people impacted by parental cancer who face many unique challenges and stressors.

Statistical analysis. Analysis was conducted using IBM SPSS Statistics version 25. Statistical analyses used to evaluate psychometric properties of the PPIQ-C are summarised in Table 14.

Factor analysis of the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C).

Identity subscale. Following methods used by Moss-Morris et al. (2002) to determine the factor structure of the IPQ-R, items assessing the *identity* dimension did not undergo factor analysis as they are rated on a different scale than items in the core subscales.

Core subscales. Core subscales were comprised of items representing the *emotional representations*, *coherence*, *personal control*, *treatment control*, *consequences*, *timeline (acute/chronic)*, and *timeline (cyclical/unpredictable)* dimensions of the CSM.

Prior to factor analysis, Little's test was conducted to test the assumption that data were missing completely at random. Little's test was non-significant, indicating that data were missing completely at random (MCAR) and could therefore be appropriately managed through pairwise deletion or simple imputation ($\chi^2 = 823.016$, $df = 847$, $p = .716$). To reduce the potential impact of bias resulting from data loss through pairwise deletion, missing data were imputed through expectation maximisation (EM). Data distribution was assessed using Kolmogorvo-Smirnov and Shapiro-Wilk tests of normality; all p's = .000 indicating violation of the assumption of normality. The suitability of data for factor analysis were assessed using the Kaiser-Meyer-Olkin (KMO) test for sampling adequacy and Bartlett's test for sphericity (KMO > 0.7 and $p < 0.01$ indicates suitability; test statistics presented below each pattern matrix).

Factor analysis was conducted using principal axis factoring due to the non-normal distribution of data. An oblique rotation method (direct oblimin) was employed to allow for correlation between factors. To determine whether the structure of the PPIQ-C was consistent with the original PPIQ, an initial confirmatory factor analysis was conducted using the 25 core items adapted directly from the original PPIQ. For this factor analysis, factor extraction was fixed to seven factors in accordance with the original structure identified by Bogosian et al (2014).

Following this, exploratory factor analysis was conducted on the full scale including the new items developed based on qualitative interviews. The correlation matrix was inspected to inform removal of items that risked extreme multicollinearity ($r \geq .8$). Inspection of the scree plot and use of Kaiser's criterion rule guided selection of the number of factors for extraction. The pattern matrix was examined to identify items with low factor loadings or high cross-loadings. Items with low factor loadings were considered for removal; items with high cross-loadings were allocated to the factor with which they were most closely conceptually related. Items that loaded onto factors that were not conceptually related were removed. Factor analysis was re-run following removal of items, until the cleanest and most interpretable factor structure was determined. The internal consistency of each factor was evaluated using Cronbach's alpha. Finally, the subscales derived from the final factor solution were interpreted and named according to the conceptual relationship between the items they contained.

Cause subscales. Unlike the other dimensions, items representing the *cause* dimension can be grouped into a number of factors. Therefore, a separate factor analysis was conducted to determine the factor structure of the *cause* subscales. Prior to factor analysis, Little's test was conducted and confirmed that data were MCAR ($\chi^2 = 114.964$, $df = 104$, $p = .217$). Missing data were imputed using EM. Moreover, Kolmogorvo-Smirnov and Shapiro-Wilk tests of normality confirmed that causal data were also non-normally distributed (all *p*'s = .000). Exploratory factor analysis was conducted using principal axis factoring and direct oblimin rotation. Inspection of the scree plot and use of Kaiser's criterion rule guided selection of the number of factors for extraction; however, because the break in the scree plot was ambiguous, factor analyses were conducted to explore factor structure when three, four, and five factors were extracted. The remainder of the analysis followed the methods outlined above for the analysis of core subscales.

Validity and reliability testing. Content validity of the *identity* scale was assessed by determining the proportion of respondents endorsing each symptom. Similarly, the proportion of respondents endorsing each cause was calculated to assess validity of items included in the *cause* subscales. Inclusion was considered valid if the item was endorsed by any percentage of respondents. Reliability analysis using Cronbach's alpha was conducted to evaluate the internal consistency of the subscales identified through the factor analyses. To facilitate evaluation of construct validity and test-retest reliability, scores on items representing each subscale were summed to produce total subscale scores. Validity and reliability analyses were conducted using non-parametric tests. Construct validity was evaluated using Spearman rank-order correlation analysis to examine correlations between the PPIQ-C subscales and measures of psychological distress (K10), post-traumatic stress symptoms (IES-R and CRIES), post-traumatic growth (PTGI-SF and PTGI-C-R), and coping strategies (Brief COPE and KIDCOPE). Test-retest reliability was evaluated using intraclass correlations and Spearman rank-order correlations of PPIQ-C subscale scores at baseline and a two-week retest.

Table 14

Statistical Analyses used to Assess Psychometric Properties of PPIQ-C

Assessment	Purpose	Statistical analysis
Extraction of Factors	To determine the optimal number of factors and guide item reduction.	Scree plots and exploratory factor analysis on full scale (i.e., including both items adapted from original PPIQ and new items developed based on qualitative interviews).
Tests of Validity		
Structural validity	To determine the extent to which scores on the PPIQ-C adequately reflect the dimensions of the original PPIQ (i.e., to validate whether previous hypothetical structure fits the items when applied to parental cancer).	Confirmatory factor analysis on items adapted from original PPIQ.
Construct validity	To determine the extent to which scores on PPIQ-C subscales relate to scores on measures of psychological distress, post-traumatic stress symptoms, post-traumatic growth, and coping strategies in a way that is consistent with hypotheses derived from the Common-Sense Model.	Spearman rank-order correlation analysis used to estimate associations between PPIQ-C subscale scores and theoretically related constructs; stronger correlation coefficients suggest support for convergent validity.
Tests of Reliability		
Internal consistency	To determine the extent to which items in each PPIQ-C subscale are intercorrelated and thus measuring the same construct.	Reliability analysis using Cronbach’s alpha.
Test-retest reliability	To determine the extent to which scores on repeated measures are close to one another (i.e., are the scores consistent over time?).	Intraclass correlation analysis and Spearman rank-order correlation analysis used to estimate strength of the relationship between PPIQ-C subscale scores at baseline and two-week retest.

Results

Participants. In total, 437 young people completed the survey. Of those, 85 were aged 12-17 years (adolescent subgroup) and 352 were aged 18-24 years (young adult subgroup). Mean participant age was 19.99 years (*SD* = 2.84 years). Time since their parent's initial diagnosis ranged from two months to 17 years (*M* = 34.70 months, *SD* = 31.33 months). Further participant demographic characteristics are outlined in Table 15.

Psychological morbidity. Young people reported high levels of psychological distress (*M* = 29.26, *SD* = 9.73, possible range = 10-50), with 82.4% reporting scores equal to or above the clinical cut-off (i.e., 20; Australian Bureau of Statistics, 2012). Overall, 16.2% of young people reported scores indicative of a mild psychological disorder (i.e., 20-24), 16.8% reported scores indicative of a moderate psychological disorder (i.e., 25-29), and 49.5% reported scores indicative of a severe psychological disorder (i.e. 30-50). Similarly, scores on measures of post-traumatic stress symptoms were consistent with PTSD being a clinical concern for both young adult (*M* = 32.43, *SD* = 19.42, possible range = 0-88) and adolescent (*M* = 23.62, *SD* = 11.69, possible range = 0-40) participants. The proportion of young adults who reported scores equal to or above the recommended clinical cut-off (i.e., 24; Weiss & Marmar, 1997) was 65.2%. In adolescents, 75.3% reported scores equal to or above the clinical cut-off (i.e., 17; Perrin et al., 2005).

Identity scale. Percentages of respondents endorsing items on the *identity* scale are presented in Table 16. The most widely endorsed symptom was fatigue (96.6%), followed by pain (94.3%), and physical weakness (91.5%). All symptoms were endorsed by over half of respondents, except for weight gain which was the least endorsed (28.7%). Cronbach's α for the scale was .815, indicating a high level of internal consistency.

Table 15

Demographic Characteristics of Young People who Completed the Survey

Characteristic	Number	Percentage
Gender		
Female	267	61.1
Male	164	37.5
Non-binary	5	1.1
Did not specify	1	0.2
Age		
12-17 years	85	19.5
18-24 years	352	80.5
Country		
Australia	176	40.3
United Kingdom	144	33.0
United States of America	91	20.8
Canada	13	3.0
New Zealand	2	0.5
Ireland	2	0.5
Other[a]	9	2.1
Highest level of education completed		
Years 7-10	57	13.0
Years 11-12	157	35.9
Certificate or diploma	100	22.9
Undergraduate degree	108	24.7
Postgraduate degree	15	3.4
Young people with more than one parent diagnosed with cancer	28	6.4
Gender of parent with cancer		
Female	275	62.9
Male	158	36.2
Did not specify	3	0.7
Type of cancer		
Breast	161	36.8
Lung	40	9.2
Prostate	39	8.9
Bowel	36	8.2
Melanoma	36	8.2
Ovarian	25	5.7
Non-Hodgkin's lymphoma	19	4.3
Leukaemia	17	3.9
Other[b]	81	18.5
Time since diagnosis		
≤2 years	225	51.5
>2 years to ≤5 years	147	33.6
>5 years	62	14.2
Metastatic diagnosis	125	28.6
Stage of cancer continuum		
Recently diagnosed (hasn't started treatment)	4	0.9
Having treatment	147	33.6
Finished treatment	203	46.5
Receiving palliative care	15	3.4
Parent died	56	12.8
Unsure	12	2.7

Note. [a]Belgium, France, Netherlands, Poland, Portugal, and Spain. [b]Other cancer types included bladder, brain, cervical, colon, endometrial, oesophageal, head and neck, kidney, liver, pancreatic, multiple myeloma, squamous cell carcinoma, testicular, throat, thyroid, and tongue.

Table 16

Percentage of Young People Endorsing Items on Identity Scale

Identity scale item	N endorsing symptom[a]	Percentage of respondents	Sample N
PPIQ-C1. Fatigue (tiredness)	422	96.6%	437
PPIQ-C2. Pain	410	94.3%	435
PPIQ-C3. Physical weakness (loss of strength)	399	91.5%	436
PPIQ-C4. Nausea (feeling sick in the stomach)	376	86%	437
PPIQ-C10. Loss of appetite	368	84.6%	435
PPIQ-C11. Depression	349	79.9%	437
PPIQ-C13. Problems with concentration, thinking, or memory (chemo brain)	329	75.3%	437
PPIQ-C12. Anxiety	325	75.1%	433
PPIQ-C9. Skin irritation (dry skin, rashes, or itchiness)	302	69.6%	434
PPIQ-C8. Weight loss	286	65.9%	434
PPIQ-C5. Vomiting	287	65.7%	437
PPIQ-C6. Hair loss	286	65.7%	435
PPIQ-C7. Weight gain	123	28.7%	428

Note. [a]Young people were considered to have endorsed symptom if they had perceived symptom as having “some impact”, “quite a big impact”, or “a very big impact” on their parent.

Core subscales. The pattern matrix of the initial confirmatory factor analysis of items adapted from the original PPIQ differed from the factor structure published by Bogosian et al. (2014) (see Table 17). Two items did not load onto any of the extracted factors (PPIQ-C28 and PPIQ-C23). In the original PPIQ, these items sat within the *adolescents' control* and *emotional representations* subscales, respectively. Moreover, the item "My parent's cancer brought me closer to my family", which loaded onto the *positive consequences for adolescents* subscale of the original PPIQ, loaded most strongly onto the factor representing *consequences for family relationships* (Component 5) in the present study.

The pattern matrix of the initial exploratory factor analysis of the 49 core PPIQ-C items is presented in Table 18. Eleven components were extracted, explaining 63.1% of variance in illness perceptions in total. Several items were removed based on this factor analysis. Inspection of the correlation matrix revealed one item pair with a correlation coefficient >.8 (items PPIQ-C57 and PPIQ-C58; r = .815). PPIQ-C58 was removed. One item was removed because it had no loading above .3 (item PPIQ-C21). PPIQ-C22 loaded onto a factor that was not conceptually related, and consequently was removed. Items PPIQ-C27, PPIQ-C26, and PPIQ-C19 formed a separate factor that was perhaps more closely related to coping strategies (i.e., thought suppression or avoidance) than illness perceptions and these were also removed.

Table 17

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Seven Factor Solution of Items Adapted from Original PPIQ

PPIQ core scale item	1	2	3	4	5	6	7
PPIQ-C41. My parent's cancer has made me more responsible	**.762**	-.027	-.009	-.007	.051	-.036	.126
PPIQ-C42. My parent's cancer has made me more independent	**.730**	-.001	.021	.063	.143	.003	-.039
PPIQ-C44. My parent's cancer has made me more understanding of other people	**.592**	.044	.034	.023	-.035	-.035	.057
PPIQ-C30. If I'm not misbehaving, I can make my parent's symptoms/side effects get better	.011	**.777**	.024	.066	.019	.054	-.030
PPIQ-C29. My parent's cancer symptoms/side effects get better when I do not stress them out (e.g., staying out later, arguing with brother or sister)	-.031	**.744**	.055	-.077	.015	.016	-.025
PPIQ-C31. My parent not being stressed or worried can make their symptoms/side effects get better	-.018	**.639**	-.034	-.084	.053	-.024	.055
PPIQ-C28. I can help my parent to manage their symptoms/side effects by looking after them	.133	.286	-.007	.073	-.208	-.147	.024
PPIQ-C57. The intensity of my parent's symptoms/side effects change a great deal from day to day	-.063	.032	**.999**	.045	-.056	.022	.003
PPIQ-C58. The number of my parent's symptoms/side effects change a great deal from day to day	.048	.015	**.818**	-.010	.080	.036	.020
PPIQ-C53. I expect my parent to have cancer for the rest of their life	.030	-.019	.020	**.863**	-.040	.011	-.032
PPIQ-C54. My parent's cancer will stay the same	.055	.034	-.034	**.662**	-.021	.034	-.082
PPIQ-C51. My parent's cancer will get worse	.013	-.089	.043	**.645**	.011	-.040	.112
PPIQ-C52. My parent's cancer suddenly got worse and never got better	-.047	-.093	.118	**.451**	.093	-.091	.014
PPIQ-C38. My parent's cancer causes arguments in the family	.084	.059	.038	.038	**.670**	-.067	-.035
PPIQ-C39. My parent's cancer puts strain on the family	.140	-.046	.083	.064	**.641**	-.067	.105
PPIQ-C40. My parent's cancer makes it more difficult to do family activities	.048	-.014	.132	.039	**.460**	-.220	.081
PPIQ-C45. My parent's cancer brought me closer to my family	.295	.077	.031	.039	**-.341**	-.124	.167
PPIQ-C23. My parent's cancer is confusing to me	-.040	.072	-.043	.160	.184	-.013	.099
PPIQ-C47. Because of my parent's cancer, I spend less time doing social activities (e.g., hobbies, sports)	-.068	-.021	-.016	.062	.044	**-.855**	.046
PPIQ-C49. Because of my parent's cancer, I spend less time with my friends	.021	-.002	-.020	.036	.124	**-.700**	.030
PPIQ-C48. Because of my parent's cancer, I spend more time doing housework	.218	.044	.081	-.104	-.078	**-.439**	-.059
PPIQ-C16. My parent's cancer worries me	.057	.014	-.047	-.030	-.035	.096	**.823**

PPIQ core scale item	1	2	3	4	5	6	7
PPIQ-C17. My parent having cancer makes me feel stressed	.106	-.033	.005	.016	.031	.008	**.725**
PPIQ-C14. When I think about my parent’s cancer, I get upset	-.060	.003	.056	-.042	-.058	-.075	**.706**
PPIQ-C15. My parent’s cancer makes me feel angry	.004	.024	.046	.033	.099	-.061	**.459**

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 12 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .827. Bartlett’s Test of Sphericity, χ^2 (300) = 3877.234, *p* = .000. Coefficients >.30 shown in bold.

Table 18

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Initial Eleven Factor Solution of 49 Core PPIQ-C Items

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10	11
PPIQ-C16. My parent’s cancer worries me	**.816**	-.055	-.064	.006	.056	-.012	-.014	-.085	-.036	-.056	.053
PPIQ-C17. My parent having cancer makes me feel stressed	**.738**	-.052	.021	.002	-.001	-.047	-.004	.001	-.080	.019	-.051
PPIQ-C18. My parent having cancer makes me feel worried about the future	**.716**	.001	-.018	.029	.022	-.016	-.042	-.002	.004	.046	-.013
PPIQ-C14. When I think about my parent’s cancer, I get upset	**.675**	-.085	.073	.017	.020	.024	-.031	-.059	-.041	-.114	-.055
PPIQ-C20. I feel overwhelmed when I think about my parent’s cancer	**.671**	.030	.041	.012	-.040	.055	-.078	.151	.064	.136	-.111
PPIQ-C15. My parent’s cancer makes me feel angry	**.461**	.008	.063	.017	.015	-.093	.078	.101	.117	-.060	-.061
PPIQ-C33. I do what I can to help make my parent’s life easier	**.314**	.071	.090	-.184	-.034	-.066	-.269	-.067	-.018	-.062	-.004
PPIQ-C21. I try to hide my feelings about my parent’s cancer	.204	.063	.025	.113	-.024	-.193	.174	.084	.079	.111	.059
PPIQ-C35. My parent’s treatment will be effective in curing their cancer	-.032	**.767**	-.062	-.027	.016	.034	-.015	.025	-.125	-.022	-.002
PPIQ-C56. My parent will recover from their cancer	-.010	**.755**	-.041	-.002	.056	.085	-.033	-.041	.003	-.276	-.059
PPIQ-C55. My parent’s cancer will improve in time	-.062	**.647**	.012	-.004	.100	.059	-.060	.031	-.078	-.227	-.089
PPIQ-C36. My parent’s treatment can control their cancer	-.080	**.577**	.086	-.106	-.025	.050	-.064	.036	-.184	.080	.070
PPIQ-C57. The intensity of my parent’s symptoms/side effects change a great deal from day to day	-.039	.007	**.823**	-.061	-.069	-.008	-.008	-.004	.049	-.026	.035
PPIQ-C58. The number of my parent’s symptoms/side effects change a great deal from day to day	-.022	.027	**.785**	-.010	-.076	-.082	.044	.019	.061	-.052	.037
PPIQ-C61. My parent goes through cycles where their symptoms/side effects get better or worse	.042	-.029	**.764**	-.015	.095	.085	-.019	-.031	-.029	-.001	.005
PPIQ-C62. My parent’s symptoms/side effects are very unpredictable	-.027	.008	**.704**	.043	.007	-.059	.060	.008	.040	.012	.011
PPIQ-C60. I never know how my parent will be feeling one day to the next	.065	.028	**.703**	.041	.037	-.027	.022	-.012	-.053	.005	.000
PPIQ-C59. My parent’s symptoms/side effects come and go in cycles	-.024	-.010	**.676**	.056	.071	.036	-.089	.017	-.033	.024	-.041
PPIQ-C24. I have a good understanding of my parent’s cancer	.004	.063	-.015	**-.814**	.023	-.056	.074	-.075	.040	.034	.021
PPIQ-C25. I have enough information about my parent’s cancer	-.015	.082	-.044	**-.806**	.085	.015	.021	.045	.118	.057	-.058
PPIQ-C23. My parent’s cancer is confusing to me	.053	.133	.000	**.515**	.025	-.004	.002	.042	.203	.140	-.085
PPIQ-C30. If I’m not misbehaving, I can make my parent’s symptoms/side effects get better	-.025	-.009	.020	-.001	**.760**	-.067	.025	.085	.135	.021	.045

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10	11
PPIQ-C29. My parent’s cancer symptoms/side effects get better when I do not stress them out (e.g., staying out late, arguing with brother or sister)	-.007	-.009	.051	-.015	**.706**	-.011	.004	.036	.078	-.089	.026
PPIQ-C31. My parent not being stressed or worried can make their symptoms/side effects get better	.058	.038	.013	.003	**.667**	.000	.072	.038	-.131	.034	-.049
PPIQ-C32. There isn’t anything I can do to make my parent’s symptoms/side effects get better	-.008	.020	.008	.033	**-.574**	-.071	.065	.106	.183	.037	-.024
PPIQ-C28. I can help my parent to manage their symptoms/side effects by looking after them	.032	.032	-.003	-.189	**.310**	-.108	-.200	-.054	.029	.087	-.066
PPIQ-C43. My parent’s cancer has made me grow up quicker	-.013	-.061	.000	-.026	-.020	**-.800**	-.044	.076	.068	-.006	-.109
PPIQ-C42. My parent’s cancer has made me more independent	-.049	-.063	.030	.028	.021	**-.756**	.058	-.017	-.029	.017	-.021
PPIQ-C41. My parent’s cancer has made me more responsible	.111	-.058	.016	-.035	.020	**-.696**	-.049	-.035	-.096	-.017	-.075
PPIQ-C44. My parent’s cancer has made me more understanding of other people	.015	.012	.079	-.033	.060	**-.577**	-.139	-.062	.031	.022	.016
PPIQ-C46. My parent’s cancer brought my closer to my parent	.182	.070	.008	.046	.089	-.116	**-.665**	-.020	-.035	.142	-.164
PPIQ-C45. My parent’s cancer brought me closer to my family	.106	.075	.066	.076	.047	-.215	**-.633**	-.086	-.018	.114	-.053
PPIQ-C38. My parent’s cancer causes arguments in the family	.028	-.006	.106	.053	.047	-.040	**.445**	-.004	.000	.130	-.281
PPIQ-C39. My parent’s cancer puts strain on the family	.157	-.086	.138	-.085	-.021	-.044	**.420**	-.089	-.066	.174	-.290
PPIQ-C27. I would rather not know details about my parent’s cancer	-.088	-.011	-.001	.131	.095	.097	-.042	**.640**	.034	.016	-.057
PPIQ-C26. I would like to know more about my parent’s cancer	.018	.053	-.034	**.339**	.150	-.070	.052	**-.527**	.171	.123	-.043
PPIQ-C19. I try not to think about my parent’s cancer	.102	.062	-.035	.028	.028	-.088	.080	**.390**	.045	.068	.069
PPIQ-C37. There is nothing that can be done to treat my parent’s cancer	-.033	-.280	-.030	-.010	-.003	.014	-.010	.025	**.728**	-.006	-.088
PPIQ-C34. There isn’t a lot that can be done to treat my parent’s cancer	-.072	-.215	.144	.036	-.091	-.062	-.014	.098	**.595**	.031	-.075
PPIQ-C52. My parent’s cancer suddenly got worse and never got better	.004	-.243	.122	-.043	-.067	.072	-.029	-.015	**.546**	.095	-.138
PPIQ-C22. I feel numb when I think about my parent’s cancer	.227	.212	.063	-.001	-.005	-.071	.075	-.101	**.313**	.192	.020
PPIQ-C54. My parent’s cancer will stay the same	-.085	-.030	-.013	.043	-.020	-.020	-.056	.031	.026	**.683**	-.009
PPIQ-C53. I expect my parent to have cancer for the rest of their life	-.033	**-.334**	.047	-.039	-.042	-.051	.006	.017	.022	**.632**	-.029
PPIQ-C51. My parent’s cancer will get worse	.128	**-.359**	.093	-.105	-.083	.023	-.019	.002	.015	**.449**	-.101
PPIQ-C47. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports)	.060	-.034	-.005	.031	-.008	-.103	-.073	-.004	.028	-.030	**-.736**

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10	11
PPIQ-C49. Because of my parent's cancer, I spend less time with my friends	.029	.039	-.003	-.026	.000	-.113	-.009	.030	.028	.006	**-.719**
PPIQ-C50. Because of my parent's cancer, I spend less time doing schoolwork	.070	.046	.019	.013	-.014	.009	.071	-.058	.160	.052	**-.457**
PPIQ-C40. My parent's cancer makes it more difficult to do family activities	.094	-.017	.179	.055	-.010	-.024	.279	-.076	-.031	.105	**-.346**
PPIQ-C48. Because of my parent's cancer, I spend more time doing housework	-.056	.012	.071	-.054	.043	**-.311**	-.107	-.046	-.085	-.089	**-.319**

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 16 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .872. Bartlett's Test of Sphericity, χ^2 (1176) = 9858.771, *p* = .000. Coefficients >.30 shown in bold.

Factor analysis was re-run using the remaining 43 items (pattern matrix presented in Table 19). Ten components were extracted, explaining 64.8% of variance in illness perceptions. A further three items were removed based on this factor analysis: PPIQ-C33, PPIQ-C28, and PPIQ-C52. PPIQ-C28 had no factor loadings above .3 and PPIQ-C33 and PPIQ-C52 loaded onto factors that were not conceptually related.

The final factor structure of the remaining 40 items is presented in Table 20. Items PPIQ-C23, PPIQ-C32, PPIQ-C45, PPIQ-C46, PPIQ-C34, and PPIQ-C37 were reverse-scored to ensure that items in each subscale measured the construct in the same direction. Internal consistency was assessed for each subscale, with Cronbach's α exceeding .7 for all subscales, except for the subscale representing *consequences for family relationships* (α = .589; Table 20). Cronbach's α for Component 2 increased from .869 to .881 with the item "My parent's treatment can control their cancer" excluded. Likewise, exclusion of the item "My parent's cancer is confusing to me" from Component 5 increased Cronbach's α from .737 to .790. Despite this, the decision was made to retain these items due to the small number of items within each subscale and as each subscale demonstrated adequate internal consistency (i.e., Cronbach's α >.7) with these items included.

The ten subscales accounted for 66.7% of variance in illness perceptions in total. Component 1 contained items PPIQ-C16, PPIQ-C17, PPIQ-C18, PPIQ-C14, PPIQ-C20, and PPIQ-C15, relating to young people's emotional representations of their parent's cancer and accounted for 20.9% of variance. Component 2 combined items that originally sat within *treatment control* and *timeline acute/chronic* subscales of the IPQ-R: PPIQ-C35, PPIQ-C56, PPIQ-C55, and PPIQ-C36. These items related to young people's beliefs about their parent's recovery from cancer and accounted for 12.0% of variance. Items relating to young people's beliefs about the unpredictability of their parent's cancer comprised Component 3 and explained 6.3% of variance. Component 4 consisted of items relating to perceived consequences for personal development and accounted for 5.7% of variance. Component 5 combined item PPIQ-C23, which originally sat within the *emotional representations* subscale of the original PPIQ, with new items PPIQ-C24 and PPIQ-C25. These items related to young people's coherence or understanding of their parent's cancer and accounted for 4.8% of variance. Items relating to young people's beliefs about their personal ability to control their parent's symptoms comprised Component 6 and explained 4.3% of variance. Component 7 consisted of items relating to perceived consequences for family relationships and accounted for 4.2% of variance. Component 8 contained items relating to young people's beliefs that treatment would be able to control their parent's cancer and explained 3.0% of variance. Items relating to young people's beliefs about the chronicity of their parent's cancer comprised Component 9 and accounted for 2.9% of variance. Component 10 consisted of items relating to perceived consequences for daily activities and explained 2.7% of variance. Interestingly, the item "My parent's cancer makes it more difficult to do family activities" loaded more strongly onto Component 10, than Component 7, which contained items relating to consequences for family relationships.

Table 19

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Initial Ten Factor Solution of 43 Core PPIQ-C Items

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10
PPIQ-C43. My parent’s cancer has made me grow up quicker	**.801**	-.020	-.020	-.018	.016	.026	-.016	.105	.013	-.061
PPIQ-C42. My parent’s cancer has made me more independent	**.796**	-.024	.001	.017	-.043	-.015	.090	-.010	.036	.023
PPIQ-C41. My parent’s cancer has made me more responsible	**.718**	-.007	.004	-.134	.033	-.005	-.023	-.057	.005	-.025
PPIQ-C44. My parent’s cancer has made me more understanding of other people	**.566**	.002	.085	-.023	.018	-.052	-.143	.015	.005	.007
PPIQ-C35. My parent’s treatment will be effective in curing their cancer	.007	**.811**	-.066	-.013	.014	-.008	.018	-.091	.014	.018
PPIQ-C56. My parent will recover from their cancer	-.069	**.757**	-.031	-.018	.001	-.048	-.024	.010	-.258	-.076
PPIQ-C55. My parent’s cancer will improve in time	-.046	**.652**	.013	.045	-.006	-.094	-.044	-.058	-.206	-.092
PPIQ-C36. My parent’s treatment can control their cancer	-.019	**.615**	.089	.051	.098	.037	-.036	-.143	.102	.095
PPIQ-C61. My parent goes through cycles where their symptoms/side effects get better or worse	-.072	-.009	**.828**	-.027	.022	-.068	-.014	.002	-.018	.038
PPIQ-C60. I never know how my parent will be feeling one day to the next	.051	.057	**.729**	-.065	-.035	-.012	.038	-.019	.001	.029
PPIQ-C59. My parent’s symptoms/side effects come and go in cycles	-.031	.006	**.722**	.042	-.049	-.048	-.088	.005	.008	-.015
PPIQ-C57. The intensity of my parent’s symptoms/side effects change a great deal from day to day	.007	-.032	**.702**	.032	.061	.062	-.005	.021	-.024	-.028
PPIQ-C62. My parent’s symptoms/side effects are very unpredictable	.085	.018	**.699**	.019	-.045	.010	.077	.052	.007	.021
PPIQ-C16. My parent’s cancer worries me	.034	-.031	-.074	**-.835**	.000	-.047	.003	-.046	-.045	.081
PPIQ-C17. My parent having cancer makes me feel stressed	.059	-.022	.008	**-.747**	-.006	.006	.012	-.065	.033	-.015
PPIQ-C18. My parent having cancer makes me feel worried about the future	.020	.034	-.029	**-.732**	-.027	-.020	-.034	.019	.061	.012
PPIQ-C14. When I think about my parent’s cancer, I get upset	-.025	-.081	.076	**-.662**	-.009	-.012	-.032	-.046	-.112	-.044
PPIQ-C20. I feel overwhelmed when I think about my parent’s cancer	-.056	.018	.034	**-.655**	-.026	.036	-.067	.063	.120	-.091
PPIQ-C15. My parent’s cancer makes me feel angry	.100	.018	.047	**-.466**	-.034	-.019	.092	.123	-.049	-.036
PPIQ-C33. I do what I can to help make my parent’s life easier	.068	.064	.084	**-.308**	.192	.050	-.257	-.036	-.070	-.002
PPIQ-C24. I have a good understanding of my parent’s cancer	.035	.042	-.007	-.009	**.848**	.010	.069	.010	.010	-.004
PPIQ-C25. I have enough information about my parent’s cancer	-.019	.099	-.043	.009	**.769**	-.070	.028	.123	.058	-.041
PPIQ-C23. My parent’s cancer is confusing to me	.024	.136	-.011	-.088	**-.517**	-.047	.008	.197	.144	-.086
PPIQ-C30. If I’m not misbehaving, I can make my parent’s symptoms/side effects get better	.080	.024	.000	.000	-.014	**-.767**	.043	.153	.046	.067

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10
PPIQ-C29. My parent’s cancer symptoms/side effects get better when I do not stress them out (e.g., staying out later, arguing with brother or sister)	.009	-.010	.041	.004	.007	**-.703**	.005	.071	-.081	.020
PPIQ-C31. My parent not being stressed or worried can make their symptoms/side effects get better	-.006	.046	.018	-.055	-.013	**-.661**	.063	-.115	.040	-.046
PPIQ-C32. There isn’t anything I can do to make my parent’s symptoms/side effects get better	.084	.051	-.007	-.019	-.026	**.566**	.089	.214	.051	.010
PPIQ-C28. I can help my parent to manage their symptoms/side effects by looking after them	.099	.018	.008	-.029	.190	-.296	-.206	.004	.071	-.077
PPIQ-C46. My parent’s cancer brought my closer to my parent	.078	.046	.013	-.157	-.036	-.086	**-.678**	-.046	.122	-.179
PPIQ-C45. My parent’s cancer brought me closer to my family	.177	.040	.076	-.085	-.062	-.042	**-.653**	-.045	.084	-.086
PPIQ-C38. My parent’s cancer causes arguments in the family	.054	.012	.091	-.055	-.066	-.052	**.440**	.011	.144	-.286
PPIQ-C39. My parent’s cancer puts strain on the family	.061	-.100	.135	-.171	.077	.032	**.412**	-.098	.164	**-.309**
PPIQ-C37. There is nothing that can be done to treat my parent’s cancer	-.001	-.228	-.042	-.004	.002	-.016	.004	**.716**	.022	-.082
PPIQ-C34. There isn’t a lot that can be done to treat my parent’s cancer	.106	-.105	.142	.027	-.040	.081	.027	**.682**	.066	-.003
PPIQ-C52. My parent’s cancer suddenly got worse and never got better	-.060	-.211	.126	-.028	.047	.062	-.023	**.530**	.100	-.138
PPIQ-C54. My parent’s cancer will stay the same	.037	.012	-.023	.046	-.057	.005	-.053	.043	**.689**	.006
PPIQ-C53. I expect my parent to have cancer for the rest of their life	.059	-.288	.041	.011	.032	.031	.004	.042	**.639**	-.006
PPIQ-C51. My parent’s cancer will get worse	-.005	**-.325**	.101	-.131	.100	.082	-.011	.027	**.442**	-.063
PPIQ-C47. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports)	.056	-.065	-.004	-.027	-.026	.001	-.107	.027	-.034	**-.764**
PPIQ-C49. Because of my parent’s cancer, I spend less time with my friends	.087	.022	.006	-.010	.030	-.001	-.028	.040	.006	**-.714**
PPIQ-C50. Because of my parent’s cancer, I spend less time doing schoolwork	-.017	.041	.022	-.075	-.011	.009	.051	.150	.054	**-.477**
PPIQ-C40. My parent’s cancer makes it more difficult to do family activities	.032	-.037	.164	-.103	-.059	.012	.266	-.059	.101	**-.375**
PPIQ-C48. Because of my parent’s cancer, I spend more time doing housework	.283	-.041	.062	.084	.052	-.037	-.126	-.117	-.110	**-.355**

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 14 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .880. Bartlett’s Test of Sphericity, χ^2 (903) = 8721.712, *p* = .000. Coefficients >.30 shown in bold.

Table 20

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Final Ten Factor Solution of 40 Core PPIQ-C Items

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10
PPIQ-C16. My parent’s cancer worries me	**.838**	-.019	-.073	-.040	.012	-.039	-.009	-.058	-.053	.093
PPIQ-C17. My parent having cancer makes me feel stressed	**.755**	-.008	.010	-.060	.008	.014	-.003	-.073	.024	-.003
PPIQ-C18. My parent having cancer makes me feel worried about the future	**.733**	.037	-.025	-.018	-.010	-.017	-.045	.022	.057	.012
PPIQ-C14. When I think about my parent’s cancer, I get upset	**.664**	-.069	.080	.026	.008	-.007	-.041	-.034	-.122	-.039
PPIQ-C20. I feel overwhelmed when I think about my parent’s cancer	**.660**	.015	.041	.054	-.013	.046	-.068	.051	.114	-.091
PPIQ-C15. My parent’s cancer makes me feel angry	**.470**	.009	.053	-.095	-.027	-.010	.087	.126	-.055	-.039
PPIQ-C35. My parent’s treatment will be effective in curing their cancer	.018	**.839**	-.071	-.014	.013	-.010	.011	-.045	.015	.020
PPIQ-C56. My parent will recover from their cancer	.022	**.761**	-.032	.062	-.001	-.046	-.024	.038	-.256	-.082
PPIQ-C55. My parent’s cancer will improve in time	-.039	**.663**	.010	.039	-.003	-.093	-.051	-.038	-.202	-.093
PPIQ-C36. My parent’s treatment can control their cancer	-.051	**.647**	.082	.003	.083	.034	-.024	-.096	.095	.101
PPIQ-C61. My parent goes through cycles where their symptoms/side effects get better or worse	.042	-.011	**.838**	.082	.039	-.057	-.035	.000	-.023	.036
PPIQ-C60. I never know how my parent will be feeling one day to the next	.066	.066	**.727**	-.046	-.030	-.013	.036	.004	-.002	.027
PPIQ-C59. My parent’s symptoms/side effects come and go in cycles	-.034	.004	**.725**	.035	-.038	-.042	-.092	.002	.006	-.018
PPIQ-C57. The intensity of my parent’s symptoms/side effects change a great deal from day to day	-.028	-.033	**.698**	-.015	.057	.062	.002	-.002	-.020	-.027
PPIQ-C62. My parent’s symptoms/side effects are very unpredictable	-.019	.016	**.696**	-.087	-.049	.012	.084	.045	.008	.023
PPIQ-C43. My parent’s cancer has made me grow up quicker	.021	-.023	-.020	**-.798**	.019	.017	-.023	.107	.019	-.064
PPIQ-C42. My parent’s cancer has made me more independent	-.013	-.012	-.003	**-.794**	-.046	-.016	.082	.003	.030	.034
PPIQ-C41. My parent’s cancer has made me more responsible	.139	.010	.000	**-.728**	.031	-.006	-.027	-.060	.000	-.010
PPIQ-C44. My parent’s cancer has made me more understanding of other people	.037	.002	.089	**-.566**	.036	-.051	-.166	.009	.008	.007
PPIQ-C24. I have a good understanding of my parent’s cancer	.038	.042	.007	-.042	**.871**	.012	.036	.001	.019	-.015
PPIQ-C25. I have enough information about my parent’s cancer	.008	.084	-.026	.022	**.768**	-.073	.001	.135	.067	-.070
PPIQ-C23. My parent’s cancer is confusing to me (r)	.076	.113	-.012	-.003	**-.501**	-.049	.013	.212	.150	-.099

PPIQ-C core scale item	1	2	3	4	5	6	7	8	9	10
PPIQ-C30. If I'm not misbehaving, I can make my parent's symptoms/side effects get better	.000	.000	.001	-.070	.009	**-.769**	.014	.171	.060	.048
PPIQ-C29. My parent's cancer symptoms/side effects get better when I do not stress them out (e.g., staying out later, arguing with brother or sister)	-.010	-.025	.039	-.008	.019	**-.701**	-.008	.078	-.069	.007
PPIQ-C31. My parent not being stressed or worried can make their symptoms/side effects get better	.044	.058	.005	-.002	-.006	**-.671**	.054	-.103	.050	-.040
PPIQ-C32. There isn't anything I can do to make my parent's symptoms/side effects get better (r)	.022	.034	.005	-.056	-.020	**.558**	.082	.247	.051	-.011
PPIQ-C45. My parent's cancer brought me closer to my family (r)	.099	.040	.088	-.171	-.024	-.040	**-.680**	-.058	.098	-.107
PPIQ-C46. My parent's cancer brought my closer to my parent (r)	.171	.047	.022	-.084	-.003	-.081	**-.672**	-.065	.130	-.193
PPIQ-C38. My parent's cancer causes arguments in the family	.042	.008	.081	-.054	-.081	-.060	**.432**	-.014	.150	-.270
PPIQ-C39. My parent's cancer puts strain on the family	.156	-.089	.119	-.073	.051	.020	**.420**	-.147	.173	-.286
PPIQ-C34. There isn't a lot that can be done to treat my parent's cancer (r)	-.027	-.197	.163	-.086	-.036	.084	.034	**.635**	.090	-.051
PPIQ-C37. There is nothing that can be done to treat my parent's cancer (r)	.003	**-.334**	-.018	.005	-.006	-.002	.021	**.572**	.064	-.124
PPIQ-C54. My parent's cancer will stay the same	-.047	.010	-.023	-.034	-.051	.001	-.052	.046	**.689**	.002
PPIQ-C53. I expect my parent to have cancer for the rest of their life	-.012	-.292	.041	-.063	.027	.033	.014	.018	**.635**	-.002
PPIQ-C51. My parent's cancer will get worse	.124	**-.328**	.099	-.004	.093	.074	.004	-.009	**.447**	-.060
PPIQ-C47. Because of my parent's cancer, I spend less time doing social activities (e.g., hobbies, sports)	.036	-.074	.004	-.055	-.016	.008	-.098	.011	-.039	**-.764**
PPIQ-C49. Because of my parent's cancer, I spend less time with my friends	.020	.013	.013	-.077	.046	.000	-.036	.034	.005	**-.724**
PPIQ-C50. Because of my parent's cancer, I spend less time doing schoolwork	.077	.019	.027	.019	-.008	.009	.056	.124	.060	**-.482**
PPIQ-C40. My parent's cancer makes it more difficult to do family activities	.092	-.032	.154	-.036	-.071	.005	.270	-.093	.108	**-.359**
PPIQ-C48. Because of my parent's cancer, I spend more time doing housework	-.086	-.022	.057	-.297	.044	-.044	-.103	-.113	-.110	**-.342**
Number of items	6	4	5	4	3	4	4	2	3	5
Cronbach's alpha	.838	.869[a]	.854	.846	.737[b]	.765	.589	.809	.769	.749

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 15 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .876. Bartlett's Test of Sphericity, χ^2 (780) = 8059.088, p = .000. Coefficients >.30 shown in bold. [a]Cronbach's alpha = .881 if item "My parent's treatment can control their cancer" removed. [b]Cronbach's alpha = .790 if item "My parent's cancer is confusing to me" removed.

Cause subscales. All items were endorsed by a moderate percentage of participants, confirming the validity of the range of causal attributions included in the scale. Sixty-seven percent of respondents agreed that their parent's cancer was caused by chance (Table 21). This was followed by 58.6% of respondents who endorsed bad luck as the cause of their parent's cancer, and 32.3% who felt that their parent's cancer was hereditary. The remaining items in the *cause* subscales were endorsed by fewer than 20% of respondents.

A series of factor analyses were conducted to determine the factor structure of the *cause* subscales. The pattern matrix of the initial exploratory factor analysis is presented in Table 22. Inspection of the scree plot and eigenvalues suggested extraction of four factors. The four-factor solution included four items that did not have loadings >.3 for any of the factors: PPIQ-C70, PPIQ-C65, PPIQ-C76, and PPIQ-C66. Components 1 and 2 demonstrated low internal consistency with Cronbach's α of .535 and .660, respectively.

Following this, factor analyses were conducted with the number of factors fixed to three and five. Pattern matrices for the three-factor and five-factor solutions are presented in Tables 23 and 24. The three-factor solution had four items with factor loadings <.3: PPIQ-C70, PPIQ-C65, PPIQ-C76, and PPIQ-C66. Cronbach's α was adequate for Components 1 and 3 (.714 and .753, respectively), but was below the recommended threshold for Component 2 (.660). The five-factor solution had three items with factor loadings <.3: PPIQ-C65, PPIQ-C76, and PPIQ-C66. Moreover, three of the five components demonstrated low internal consistency, with Cronbach's α ranging from .391 to .660.

The three-factor solution was determined to be the best fit in terms of internal consistency and conceptual coherence of subscales: Component 1 consisted of items relating to possible behavioural or environmental causes of cancer, Component 2 contained items relating to chance or bad luck, and Component 3 was comprised of items relating to possible emotional or psychological causes of cancer. The final three-factor solution explained 46.8% of variance in total, with 24.6% attributed to Component 1, 11.7% to Component 2, and 10.5% to Component 3.

Although items PPIQ-C70, PPIQ-C65, PPIQ-C76, and PPIQ-C66 had factor loadings <.3, they were retained because they either represented medically substantiated causes of cancer or were endorsed by a percentage of respondents as a possible cause of their parent's cancer. It should be noted that these items were not included in subsequent *cause* subscale scores. As with the original IPQ and IPQ-R (Moss-Morris et al., 2002; Weinman et al., 1996), researchers are encouraged to modify the *cause* scale for use in particular cultural settings or populations and determine the dimensionality of the scale with each modification.

Table 21

Percentage of Young People Endorsing Items on Cause Scale

Cause scale item	N endorsing cause[a]	Percentage of respondents	Sample N
PPIQ-C67. My parent's cancer was caused by chance	292	67%	436
PPIQ-C68. My parent's cancer was caused by bad luck	256	58.6%	437
PPIQ-C66. My parent's cancer is hereditary (it runs in the family)	141	32.3%	436
PPIQ-C74. My parent's cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)	85	19.5%	437
PPIQ-C69. My parent's cancer was caused by poor diet	80	18.4%	436
PPIQ-C72. My parent's cancer was caused by smoking	70	16.1%	436
PPIQ-C70. My parent's cancer was caused by sun exposure	70	16%	437
PPIQ-C63. My parent's cancer was caused by stress or worry	70	16%	437
PPIQ-C64. My parent's cancer was caused by family problems or worries	63	14.4%	437
PPIQ-C71. My parent's cancer was caused by lack of exercise	49	11.2%	436
PPIQ-C76. My parent's cancer was caused by a higher power (e.g., it was an act of God)	48	11%	437
PPIQ-C73. My parent's cancer was caused by drinking alcohol	38	8.7%	434
PPIQ-C65. My parent's cancer was caused by a germ or virus	37	8.5%	436
PPIQ-C75. My parent's cancer was caused by bad or toxic people in their life	26	5.9%	436

Note. [a]Young people were considered to have endorsed cause if they answered "agree" or "strongly agree".

Table 22

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Four-Factor Solution of Causal Items of PPIQ-C

PPIQ-C cause scale item	1	2	3	4
PPIQ-C72. My parent’s cancer was caused by smoking	**.596**	-.063	.095	.004
PPIQ-C73. My parent’s cancer was caused by drinking alcohol	**.564**	.046	.030	-.124
PPIQ-C74. My parent’s cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)	**.377**	-.019	-.081	-.093
PPIQ-C70. My parent’s cancer was caused by sun exposure	.293	-.017	-.045	-.013
PPIQ-C65. My parent’s cancer was caused by a germ or virus	.267	.120	-.254	-.038
PPIQ-C67. My parent’s cancer was caused by chance	-.176	**.782**	.145	-.029
PPIQ-C68. My parent’s cancer was caused by bad luck	.067	**.644**	-.031	.083
PPIQ-C64. My parent’s cancer was caused by family problems or worries	-.070	-.030	**-.729**	-.078
PPIQ-C63. My parent’s cancer was caused by stress or worry	-.149	-.071	**-.721**	.000
PPIQ-C75. My parent’s cancer was caused by bad or toxic people in their life	.134	-.030	**-.686**	.018
PPIQ-C76. My parent’s cancer was caused by a higher power (e.g., it was an act of God)	.006	.140	-.284	-.129
PPIQ-C66. My parent’s cancer is hereditary (it runs in the family)	.039	-.007	-.102	.007
PPIQ-C69. My parent’s cancer was caused by poor diet	.150	-.037	.072	**-.810**
PPIQ-C71. My parent’s cancer was caused by lack of exercise	-.020	-.053	-.069	**-.805**
Number of items	3	2	3	2
Cronbach’s alpha	.535	.660	.753	.823

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 9 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .732. Bartlett’s Test of Sphericity, χ^2 (91) = 1324.832, p = .000. Coefficients >.30 shown in bold.

Table 23

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Three-Factor Solution of Causal Items of PPIQ-C

PPIQ-C cause scale item	1	2	3
PPIQ-C69. My parent’s cancer was caused by poor diet	**.776**	.027	-.026
PPIQ-C71. My parent’s cancer was caused by lack of exercise	**.617**	.016	-.164
PPIQ-C73. My parent’s cancer was caused by drinking alcohol	**.592**	.032	.045
PPIQ-C72. My parent’s cancer was caused by smoking	**.499**	-.082	.113
PPIQ-C74. My parent’s cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)	**.422**	-.023	-.059
PPIQ-C70. My parent’s cancer was caused by sun exposure	.278	-.028	-.020
PPIQ-C65. My parent’s cancer was caused by a germ or virus	.271	.109	-.233
PPIQ-C67. My parent’s cancer was caused by chance	-.121	**.819**	.145
PPIQ-C68. My parent’s cancer was caused by bad luck	-.011	**.602**	-.009
PPIQ-C64. My parent’s cancer was caused by family problems or worries	-.021	-.028	**-.758**
PPIQ-C63. My parent’s cancer was caused by stress or worry	-.161	-.073	**-.750**
PPIQ-C75. My parent’s cancer was caused by bad or toxic people in their life	.094	-.043	**-.666**
PPIQ-C76. My parent’s cancer was caused by a higher power (e.g., it was an act of God)	.116	.150	-.292
PPIQ-C66. My parent’s cancer is hereditary (it runs in the family)	.030	-.009	-.096
Number of items	5	2	3
Cronbach’s alpha	.714[a]	.660	.753

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 7 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .732. Bartlett’s Test of Sphericity, χ^2 (91) = 1324.832, p = .000. Coefficients >.30 shown in bold.

[a] Cronbach’s alpha = .719 if item “My parent’s cancer was caused by chemicals or radiation in the environment” removed.

Table 24

Pattern Matrix for Principal Axis Factor Analysis with Oblimin Rotation of Five-Factor Solution of Causal Items of PPIQ-C

PPIQ-C cause scale item	1	2	3	4	5
PPIQ-C69. My parent's cancer was caused by poor diet	**.870**	-.054	-.032	.088	-.026
PPIQ-C71. My parent's cancer was caused by lack of exercise	**.812**	.059	-.047	-.053	-.003
PPIQ-C63. My parent's cancer was caused by stress or worry	-.014	**.758**	-.053	-.019	-.108
PPIQ-C64. My parent's cancer was caused by family problems or worries	.073	**.738**	-.008	.030	-.042
PPIQ-C75. My parent's cancer was caused by bad or toxic people in their life	-.006	**.612**	-.024	.086	.210
PPIQ-C67. My parent's cancer was caused by chance	.015	-.147	**.754**	-.121	-.099
PPIQ-C68. My parent's cancer was caused by bad luck	-.073	.031	**.671**	.082	-.002
PPIQ-C72. My parent's cancer was caused by smoking	-.017	.033	-.045	**.743**	-.016
PPIQ-C73. My parent's cancer was caused by drinking alcohol	.191	.045	.050	**.487**	.068
PPIQ-C74. My parent's cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)	.098	-.053	-.037	.145	**.498**
PPIQ-C70. My parent's cancer was caused by sun exposure	.021	-.064	-.033	.086	**.398**
PPIQ-C65. My parent's cancer was caused by a germ or virus	.061	.183	.117	.131	.274
PPIQ-C76. My parent's cancer was caused by a higher power (e.g., it was an act of God)	.130	.179	.140	-.111	.245
PPIQ-C66. My parent's cancer is hereditary (it runs in the family)	-.016	.026	-.016	-.060	.205
Number of items	2	3	2	2	2
Cronbach's alpha	.823	.753	.660	.587	.391

Note. Extraction method: Principal Axis Factoring; Rotation method: Oblimin with Kaiser Normalisation. Rotation converged in 8 iterations. Kaiser-Meyer-Olkin Measure of Sampling Adequacy = .732. Bartlett's Test of Sphericity, χ^2 (91) = 1324.832, p = .000. Coefficients >.30 shown in bold.

Construct validity. Based on the relationships proposed by the CSM and previous literature testing the model (Hagger et al., 2017; Hagger & Orbell, 2003), predictions were made about the expected relationships between PPIQ-C subscales and measures of psychological distress, post-traumatic stress symptoms, post-traumatic growth, and coping strategies.

Higher scores on the *identity*, *emotional representations, consequences for family relationships, consequences for daily activities*, *unpredictability*, and *chronicity* subscales represent strongly held negative emotions and negative beliefs about each factor. It was anticipated that small to moderate positive correlations would be observed between these subscales and measures of psychological distress, post-traumatic stress symptoms, and avoidance coping strategies. Small to moderate negative correlations were anticipated between scores on *identity*, *emotional representations*, *consequences for family relationships*, *consequences for daily activities, unpredictability,* and *chronicity* subscales and measures of post-traumatic growth and approach coping strategies.

Conversely, higher scores on the *coherence*, *consequences for personal development*, *personal control*, *treatment control*, and *recovery* subscales represented more positive reactions to parental cancer in each of the factor domains. It was anticipated that small to moderate negative correlations would be observed between scores on these subscales and measures of psychological distress, post-traumatic stress symptoms, and avoidance coping strategies. Small to moderate positive correlations were anticipated between scores on *coherence*, *consequences for personal development*, *personal control*, *treatment control*, and *recovery* subscales and measures of post-traumatic growth and approach coping strategies.

Table 25 presents correlations between PPIQ-C subscales and measures of psychological outcomes and coping strategies.

Table 25

Spearman’s Rho Correlations Between PPIQ-C Subscales, Coping Strategies, and Psychological Outcomes

PPIQ-C subscale	Psychological distress (K10 total)	Post-traumatic stress (IES-R total)	Post-traumatic stress (CRIES total)	Post-traumatic growth (PTGI-SF total)	Post-traumatic growth (PTGI-C-R total)	Avoidance strategies (Brief COPE)	Avoidance strategies (KIDCOPE)	Approach strategies (Brief COPE)	Approach strategies (KIDCOPE)
1. Identity	.442**	.481**	.299**	.336**	.278*	.416**	.432**	.307**	.041
2. Emotional Representations	.328**	.417**	.556**	.390**	.408**	.329**	.572**	.253**	.009
3. Coherence	-.188**	-.167**	-.108	-.029	.125	-.201**	-.268*	.132*	.140
4. Consequences for Personal Development	.188**	.325**	.329**	.537**	.402**	.261**	.427**	.385**	.297**
5. Consequences for Family Relationships	.201**	.129*	.179	-.227**	-.268*	.171**	.167	-.205**	-.288**
6. Consequences for Daily Activities	.365**	.387**	.424**	.311**	.117	.478**	.455**	.262**	.087
7. Personal Control	-.006	-.006	.146	.227**	.365**	-.026	.323**	.272**	.289**
8. Treatment Control	-.202**	-.339**	-.275*	-.162**	-.010	-.304**	-.201	-.079	.037
9. Recovery	-.147**	-.229**	.030	.003	.247*	-.224**	.088	.064	.008
10. Unpredictability	.297**	.406**	.022	.242**	.163	.362**	.195	.190**	.137
11. Chronicity	.264**	.360**	.133	.119*	.003	.378**	.249*	.110*	-.049
12. Behavioural or Environmental Attributions	.150**	.185**	.091	.044	-.045	.206**	.032	.057	.043
13. Chance or Luck Attributions	.131**	.054	.105	-.005	.095	.028	.209	-.018	.137
14. Emotional or Psychological Attributions	.159**	.178**	.100	.079	.105	.178**	.203	.114*	.120

Note. **p < .01 (2-tailed). *p < .05 (2-tailed).

Psychological distress. As predicted, higher levels of psychological distress (assessed using the K10) were significantly associated with higher scores on the *identity*, *emotional representations*, *consequences for family relationships*, *consequences for daily activities, unpredictability,* and *chronicity* subscales. Also as predicted, there were significant negative correlations between psychological distress and scores on the *coherence*, *treatment control*, and *recovery* subscales. Although scores on the *personal control* subscale appeared to be negatively associated with psychological distress, this relationship was not significant. Interestingly, scores on the *consequences for personal development* subscale were significantly positively correlated with psychological distress scores, suggesting that young people who perceive greater positive consequences for their personal development experience higher levels of psychological distress in the face of their parent's cancer.

Post-traumatic stress. Post-traumatic stress in young adults (measured by the IES-R) was significantly positively correlated with the *identity*, *emotional representations*, all *consequences*, *unpredictability*, and *chronicity* subscales and significantly negatively correlated with *coherence*, *treatment control*, and *recovery* subscales. In adolescents, post-traumatic stress (assessed using the CRIES) was significantly positively correlated with *identity*, *emotional representations*, *consequences for personal development* and *consequences for daily activities* subscales and significantly negatively correlated with *treatment control* subscales. This was consistent with predictions, except for the finding that higher scores on the *consequences for personal development* subscale was positively associated with post-traumatic stress in both young adults (r = .325) and adolescents (r = .329).

Post-traumatic growth. As predicted, post-traumatic growth in young adults (measured by the PTGI-SF) was significantly positively correlated with the *consequences for personal development* and *personal control* subscales and significantly negatively correlated with the *consequences for family relationships*. However, contrary to prediction, scores on the PTGI-SF were significantly positively correlated with scores on the *identity*, *emotional representations*, *consequences for daily activities*, and *unpredictability* subscales. Furthermore, there was a significant negative correlation between perceptions relating to *treatment control* and post-traumatic growth (r = -.162), suggesting that greater belief in ability of treatment to control their parent's cancer was associated with lower post-traumatic growth in young adults.

Similar relationships were observed in adolescents; predictions that higher scores on the *consequences for personal development* and *personal control* subscales and lower scores on the *consequences for family relationships* subscale would be associated with greater post-traumatic growth were supported. Furthermore, as predicted, greater belief in the likelihood of their parent's recovery from cancer was significantly associated with higher levels of post-traumatic growth in adolescents (r = .247). As observed in young adults, higher scores on *identity* and *emotional representations* subscales were also associated with greater post-traumatic growth in adolescents (r = .278 and r = .408, respectively).

Avoidance coping strategies. Predictions that higher scores on *identity*, *emotional representations*, *consequences for family relationships*, *consequences for daily activities*, *unpredictability*, and *chronicity* subscales would be significantly associated with greater use of avoidance coping strategies were supported in young adults. Furthermore, higher scores on *coherence*, *treatment control*, and *recovery* subscales were significantly associated with reduced engagement with avoidance coping strategies in young adults. Contrary to prediction, greater perceived consequences for personal development were also associated with greater use of avoidance coping strategies (r = .261).

In adolescents, there were significant positive associations between scores on the *avoidance strategies* subscale of the KIDCOPE and scores on *identity, emotional representations, consequences for daily activities,* and *chronicity* subscales. As observed in young adults, higher scores on the *consequences for personal development* subscale were significantly associated with greater use of avoidance coping strategies in adolescents (r = .427). Moreover, there was a significant positive correlation between scores on the *personal control* and *avoidance strategies* subscales (r = .323). Consistent with prediction, greater coherence and understanding of parental cancer was associated with reduced engagement with avoidance coping strategies (r = -.268).

Approach coping strategies. Predictions that higher scores on *coherence, consequences for personal development*, and *personal control* subscales, and lower scores on the *consequences for family relationships* subscale, would be associated with greater approach coping strategies were supported in young adults. Scores on *identity, emotional representations, consequences for daily activities*, *unpredictability*, and *chronicity* subscales were also significantly positively correlated with use of approach coping strategies in young adults.

In adolescents, engagement with approach coping strategies was significantly positively correlated with scores on *consequences for personal development* and *personal control* subscales (r = .297 and r = .289, respectively), and significantly negatively correlated with scores on the *consequences for family relationships* subscale (r = -.288), providing further support for predictions.

Test-retest reliability. Twenty-three young people completed the PPIQ-C at baseline and two-week retest. According to Terwee et al. (2007), ICC = .70 is recommended as a minimum standard for test-retest reliability. Six subscales met this criterion; *identity*

(ICC = .787), *coherence* (ICC = .715), *consequences for daily activities* (ICC = .716), *personal control* (ICC = .759), chronicity (ICC = .791), and *emotional or psychological attributions* (ICC = .741) (Table 26). The remaining subscales demonstrated moderate test-retest reliability (Koo & Li, 2016), ranging from ICC = .603 (*recovery*) to ICC = .673 (*unpredictability*), except for the *treatment control* subscale, which had poor test-retest reliability (ICC = .423).

Table 26

Two-Week Test-Retest Reliabilities for PPIQ-C Subscales

PPIQ-C subscale	Intraclass correlation	Spearman's rho	N
1. Identity	.787**	.818**	20
2. Emotional Representations	.605**	.598**	23
3. Coherence	.715**	.712**	23
4. Consequences for Personal Development	.631**	.612**	22
5. Consequences for Family Relationships	.666**	.645**	23
6. Consequences for Daily Activities	.716**	.711**	21
7. Personal Control	.759**	.772**	23
8. Treatment Control	.423*	.398	23
9. Recovery	.603**	.540**	22
10. Unpredictability	.673**	.625**	22
11. Chronicity	.791**	.844**	21
12. Behavioural or Environmental Attributions	.653**	.620**	22
13. Chance or Luck Attributions	.627**	.619**	23
14. Emotional or Psychological Attributions	.741**	.653**	23

**p < .01. *p < .05.

Discussion

The purpose of this study was to evaluate the psychometric properties of a measure of illness perceptions for young people who have a parent with cancer. The adaptation and validation of the PPIQ-C extends the literature on the impact of parental cancer by providing a tool to facilitate examination of how young people's perceptions of their parent's cancer affects their coping and psychological adjustment. In particular, because the PPIQ-C was developed to assess the dimensions of illness perceptions as described by the CSM, it will enable further exploration of the utility of this model for understanding the psychosocial implications of parental cancer for AYA children.

As in the original IPQ/IPQ-R and PPIQ, the PPIQ-C is comprised of three sections, each with a separate dimensional structure. The final version of the PPIQ-C was determined following exploratory factor analysis. The unidimensional *identity* subscale consists of 13 items; the core subscales are comprised of 40 items in ten dimensions; and the *cause* subscales contain 14 items across three dimensions. Unlike the original PPIQ, the PPIQ-C includes subscales representing young people's perceptions of illness *identity*, *coherence*, and *treatment control*.

Factor analysis revealed minor differences in the factor structure of the original PPIQ and the PPIQ-C. The most significant difference was the factor structure of items measuring the *consequences* dimension. In the original PPIQ, these items were factored into three subscales (*negative consequences for family, positive consequences for adolescents*, and *negative consequences for adolescents*). In the present study, items assessing perceived consequences were factored according to the aspect of the young person's life that was impacted, rather than whether the consequence was perceived as positive or negative (i.e., *consequences for personal development*, *consequences for family relationships*, and *consequences for daily activities*). Because of this, value judgements categorising consequences as “positive” or “negative” were excluded from subscale titles. Furthermore, the PPIQ-C included an additional subscale comprised of items assessing young people’s beliefs about their parent’s *recovery* from cancer; these items originally sat within *treatment control* and *timeline acute/chronic* subscales of the IPQ-R.

Preliminary evidence for the reliability and validity of the PPIQ-C was obtained from 437 young people who had a parent diagnosed with cancer. Structural validity was assessed using principal axis factoring and each analysis was conducted using an adequate sample size (defined by Terwee et al. (2007) as 7x the number of items and ≥100 participants). Cronbach’s alphas were acceptable for all subscales, except *consequences for family relationships* ($\alpha = .589$) and *chance or luck attributions* ($\alpha = .660$). Most subscales demonstrated evidence for good test-retest reliability over two weeks (defined by Terwee et al. (2007) as ICC ≥.7), with intraclass correlation coefficients ranging from .791 to .715. Poor test-retest reliability was observed in subscales assessing beliefs about *recovery, unpredictability,* and *treatment control* (ICC = .603, .673, and .423, respectively). Participants included young people who were reflecting on a past experience with parental cancer (i.e., in cases where the parent had completed treatment or had died), which meant that some participants made judgements about how they remembered perceiving their parent’s cancer, whereas others were commenting on recent experience. It is possible that poor test-retest reliability in *recovery*, *unpredictability*, and *treatment control* subscales stemmed from difficulties in recalling beliefs at the time of the parent’s diagnosis and treatment. For example, selecting a response to the statement “My parent will recover from their cancer” that reflected their level of agreement *at the time of their parent’s diagnosis and treatment* may have been challenging for young people whose parent had completed treatment and was no longer living with cancer.

Post hoc analyses were undertaken to examine differences in test-retest reliability between respondents who completed the questionnaire concurrently (i.e., at the time of their parent's diagnosis and treatment) and retrospectively (i.e., after parent had completed treatment or had died) (see table in Appendix F). The majority of subscales demonstrated good test-retest reliability over two weeks in participants who completed the questionnaire concurrently, including *recovery* (ICC = .891) and *unpredictability* (ICC = .787) subscales. Interestingly, the *treatment control* subscale did not demonstrate adequate test-retest reliability in either subgroup (concurrent response ICC = .082 vs. retrospective response ICC = .482). This may be reflective of day-to-day changes in AYAs' beliefs about the effectiveness of treatment to control their parent's cancer. There were also notable differences in test-retest reliabilities in *coherence* (concurrent response ICC = .426 vs. retrospective response ICC = .787) and *personal control* (concurrent response ICC = .621 vs. retrospective response ICC = .825) subscales, which demonstrated better test-retest reliability among those who responded retrospectively. In general, findings provide initial support for the stability of the instrument, but further research is needed to examine test-retest reliability in a larger sample of young people whose parent has been recently diagnosed and/or is undergoing treatment for cancer at the time of data collection. Further work may be needed to address problems in the *treatment control* subscale.

Correlations between PPIQ-C subscales and measures of psychological distress, post-traumatic stress, and post-traumatic growth provided support for construct validity. All PPIQ-C subscales were significantly correlated with psychological distress, except for beliefs about *personal control*. Beliefs in this domain were significantly and positively correlated with post-traumatic growth and approach coping strategies in both age groups. This finding may be reflective of young people's use of cognitive reappraisal strategies to reframe their initial perception of their parent's cancer or problem-focused coping strategies to enhance their sense of control over the illness. In their meta-analysis, E. M. Richardson et al. (2017) found that higher levels of perceived personal control were associated with both higher levels of cognitive reappraisal and problem-focused coping strategies in people diagnosed with cancer. Other research in caregivers has found that cultivating a sense of control can promote resilience and buffer distress (Applebaum & Breitbart, 2013; Teixeira, Applebaum, Bhatia, & Brandao, 2018; Waldron, Janke, Bechtel, Ramirez, & Cohen, 2013), further supporting the finding that higher levels of perceived *personal control* were significantly correlated with higher levels of post-traumatic growth and were not associated with psychological distress.

Furthermore, findings revealed positive correlations between perceived *consequences for personal development* and psychological distress, post-traumatic stress, and post-traumatic growth. This suggests that although perceptions of greater independence and responsibility may facilitate positive psychological change over time, they also cause young people a significant level of stress. This pattern was also observed in correlations between *identity* and *emotional representations* subscales and psychological distress, post-traumatic stress, and post-traumatic growth. It translates that an experience of trauma is needed for the development of post-traumatic growth (Turner, Hutchinson, & Wilson, 2018). This is reflected in a large body of research that indicates that distress and growth co-occur (Zieba, Wiechec, Bieganska-Banas, & Mieleszczenko-Kowszewicz, 2019). In the context of parental cancer, a young person may experience significant distress and intrusive thoughts in the face of their parent’s diagnosis, but they may also gain a greater sense of personal strength and a greater appreciation for life. Research examining post-traumatic growth in this cohort supports this assertion (Levesque & Maybery, 2012; Morris, Turnbull, Martini, Preen, & Zajac, 2020; Wong, Cavanaugh, Macleamy, Sojourner-Nelson, & Koopman, 2009).

Although not specifically examined in the present study, the trauma literature suggests that negative psychological change, or post-traumatic depreciation, is another important indicator of coping and psychological adjustment following a traumatic life event. Post-traumatic depreciation is the opposite of growth; it is defined as a reduced or impaired sense of psychological adjustment, cognitive development, and emotional awareness in the same domains as post-traumatic growth (i.e., changes to sense of self, changes to relationships with others, changes to spirituality, changes to perceived opportunities, and changes to appreciation for life) (Barrington & Shakespeare-Finch, 2013). Research suggests that post-traumatic depreciation and post-traumatic growth are independent constructs (Zieba et al., 2019), and although it may seem paradoxical to measure both, they have different predictors and outcomes (Barrington & Shakespeare-Finch, 2013). Further research should be conducted to tease out the relationships between illness perceptions, psychological distress, post-traumatic stress, post-traumatic growth, and post-traumatic depreciation in this cohort.

The findings of the present study also revealed significant, negative associations between perceived *consequences for family relationships* and post-traumatic growth, highlighting the importance of a strong family network in supporting young people through the cancer experience. Social support is proposed to buffer against the effects of stressful life events (Cohen & Wills, 1985). Research has consistently shown that social support is associated with lower levels of post-traumatic stress symptoms and depression among cancer populations (Beatty & Kissane, 2017; Manne et al., 2018; Romeo et al., 2019; Shand, Brooker, Burney, Fletcher, & Ricciardelli, 2018). Calhoun and Tedeschi (2006) suggest that the relationship between social support and post-traumatic growth may be more specific. They propose that the degree to which post-traumatic growth is experienced is impacted by the response given to behaviours displayed by the person in crisis (Calhoun & Tedeschi, 2006). Social support may help to alter the perception and appraisal of a stressful life event, by facilitating deliberate rumination and allowing the individual to engage in sense and meaning making, and in turn, facilitating post-traumatic growth (Shakespeare-Finch, Obst, &

Rogers, 2019). Further research may seek to clarify these relationships by directly examining social support in relation to illness perceptions and post-traumatic growth in young people who are impacted by parental cancer.

The findings of the present study should be interpreted with considerations for its limitations. The generalisability of the findings is limited by the study's sample. Participants were comprised of young people who had a parent who had been diagnosed with, or received treatment for, any type or stage of cancer, within the last five years. This included participants whose parent had completed treatment or had died; these participants were asked to respond to items on the PPIQ-C based on how they would have felt at the time of their parent's diagnosis and treatment (i.e., retrospectively). This may have led to inaccuracies or inconsistencies in the data. In addition, young people aged 12-17 years were underrepresented given that they only comprised 19.5% of the sample. There were considerable difficulties recruiting minors, potentially due to the need for young people aged 15 years and under to obtain parental consent in order to participate in the study. This research covered a topic that is sensitive in nature, and it is not unreasonable to assume that the need to obtain parental consent discouraged young people from participating in the study. Moreover, test-retest reliability of the PPIQ-C was only assessed in young adult participants (aged 18-24 years). Further research is needed to explore the validity of the PPIQ-C when used in adolescents aged 12-17 years.

In conclusion, this study provides preliminary evidence that the PPIQ-C is a reliable, valid, and useful tool for assessing illness perceptions among young people who have a parent with cancer. The PPIQ-C has many potential applications to clinicians, researchers, and service providers who support young people affected by parental cancer. Although the PPIQ-C is newly developed and validated, the findings of this study support its use to identify young people who may be at greater risk of poorer psychological outcomes (based on subscale scores). Further longitudinal work should be conducted to determine patterns of illness perceptions that are associated with maladaptive coping strategies and psychological morbidity. In addition, use of the PPIQ-C may assist in highlighting strongly held beliefs that are disempowering, inaccurate, or medically unsubstantiated (based on items endorsed) and provide insight for potential targets for interventions that aim to support young people adjusting to a parent’s cancer diagnosis.

5.1. Chapter Summary and Future Directions

Chapter 5 presented a cross-sectional survey study conducted to evaluate the psychometric properties of the PPIQ-C. Survey responses were collected from adolescents (*n* = 85) and young adults (*n* = 352) impacted by parental cancer, a subsample (*n* = 23) of which completed a two-week retest of the PPIQ-C. Respondents completed the PPIQ-C as well as measures of psychological distress (K10; Kessler et al., 2002; Kessler et al., 2003), post-traumatic stress (IES-R and CRIES; Horowitz et al., 1979; Perrin et al., 2005; P. Smith et al., 2003; Weiss & Marmar, 1997), post-traumatic growth (PTGI-SF and PTGI-C-R; Cann et al., 2010; Kilmer et al., 2010; Tedeschi & Calhoun, 1996) and coping strategies (Brief COPE and KIDCOPE; Carver, 1997; Carver et al., 1989; Spirito et al., 1988).

The PPIQ-C is comprised of three sections (*identity* subscale, core subscales, and *cause* subscales), each of which has a separate factor structure. Analyses were carried out to determine the factor structure of the core and *cause* subscales, however the *identity* subscale did not undergo factor analysis as it is a unidimensional scale. The final PPIQ-C is comprised of 67 items across 14 dimensions (the complete questionnaire, including instructions for each section, is presented in Appendix G). The *identity* subscale consists of 13 items, measuring young people’s perceptions of the negative impact of their parent’s cancer symptoms and side effects. Core items were factored into ten subscales: *emotional representations* (6 items), *coherence* (3 items), *consequences for personal development* (4 items), *consequences for family relationships* (4 items), *consequences for daily activities* (5 items), *personal control* (4 items), *treatment control* (2 items), *recovery* (4 items), *unpredictability* (5 items), and *chronicity* (3 items). Items measuring perceptions about the *cause* of cancer were factored into three subscales: *behavioural or environmental attributions* (5 items), *chance or luck attributions* (2 items), and *emotional or psychological attributions* (3 items). There were minor structural differences between the original PPIQ and the PPIQ-C, including the addition of subscales measuring perceptions of illness *identity*, *coherence*, and *treatment control*.

All PPIQ-C subscales demonstrated acceptable internal consistency (α > .714), except *consequences for family relationships* (α = .589) and *chance or luck attributions* (α = .660). Test-retest correlations provided initial support for the stability of the PPIQ-C; most subscales demonstrated good test-retest reliability over two weeks (ICC > .715), however poor test-retest reliability was observed in subscales assessing beliefs about *recovery*, *unpredictability*, and *treatment control* (ICC = .603, .673, and .423, respectively). One possible explanation for this finding is that participants were responding to the questionnaire from various post-diagnosis perspectives (i.e., parent was recently diagnosed, undergoing curative treatment, completed treatment, receiving palliative care, or had died). Respondents who were completing the questionnaire retrospectively were asked to select answers that best reflected their beliefs and perceptions at the time of their parent's diagnosis and treatment. This may have been particularly challenging for *recovery*, *unpredictability*, and *treatment control* subscales which assessed young people's beliefs about their parent's likelihood of recovery, the unpredictable nature of their parent's cancer, and the effectiveness of treatment in controlling their parent's cancer. Post hoc analyses revealed that, as expected, *recovery* and *unpredictability* subscales demonstrated good test-retest reliability in participants who completed the questionnaire concurrently (ICC = .891 and .787, respectively). However, the *treatment control* subscale did not demonstrate adequate test-retest reliability in either subsample (concurrent response ICC = .082 vs. retrospective response ICC = .482). Interestingly, *coherence* (concurrent response ICC = .426 vs. retrospective response ICC = .787) and *personal control* (concurrent response ICC = .621 vs. retrospective response ICC = .825) subscales demonstrated better test-retest reliability in those who completed the questionnaire retrospectively.

Correlations between PPIQ-C subscales and validated measures of psychological distress, post-traumatic stress, and post-traumatic growth provided support for construct validity. Psychological distress was significantly and positively associated with perceptions relating to *identity*, *emotional representations*, *consequences for family relationships, consequences for personal development*, *consequences for daily activities*, *unpredictability*, and *chronicity*. In contrast, psychological distress was significantly and negatively associated with perceptions relating to *coherence*, *treatment control*, and *recovery*.

A similar correlation pattern was observed for post-traumatic stress; in young adults (*n* = 352), post-traumatic stress was significantly and positively associated with perceptions relating to *identity*, *emotional representations*, *consequences for family relationships, consequences for personal development, consequences for daily activities, unpredictability,* and *chronicity*. Conversely, post-traumatic stress was negatively associated with perceptions relating to *coherence*, *treatment control*, and *recovery*. In adolescents (*n* = 85), post-traumatic stress was significantly and positively associated with perceptions relating to *identity*, *emotional representations*, *consequences for personal development* and *consequences for daily activities*. Moreover, post-traumatic stress was significantly and negatively associated with perceptions relating to *treatment control*.

Findings revealed that post-traumatic growth in young adults was positively associated with perceptions relating to *identity*, *emotional representations*, *consequences for personal development*, *consequences for daily activities*, *personal control*, and *unpredictability*, and negatively associated with perceptions relating to *consequences for family relationships* and *treatment control*. Similarly, post-traumatic growth in adolescents was positively associated with perceptions relating to *identity*, *emotional representations*, *consequences for personal development*, *personal control*, and *recovery*, and negatively associated with perceptions relating to *consequences for family relationships*.

It is worth highlighting that many of the subscales that were found to be associated with poorer psychosocial outcomes were also associated with greater post-traumatic growth. This finding indicates that an experience of trauma was necessary for the development of post-traumatic growth and is consistent with findings in survivors of childhood and adolescent cancer (Turner et al., 2018).

Overall, this study provided preliminary evidence that the PPIQ-C is a reliable and valid tool for assessing young people's perceptions of their parent's cancer. The following study aims to extend these findings by exploring whether the PPIQ-C may be suitable for use as a screening tool for identifying young people at risk of psychological distress and post-traumatic stress.

CHAPTER SIX

Do scores on the Perceptions of Parental Illness Questionnaire-Cancer predict psychological morbidity in young people following a parent's cancer diagnosis?

6.0. Preamble

The findings presented in Chapter 5 indicated that strongly held negative emotions and negative beliefs about the impact of symptoms and side effects, the consequences of the illness, and the chronic or unpredictable nature of the illness were related to significantly higher levels of psychological distress and post-traumatic stress. Given that certain domains of illness perceptions are more strongly associated with poor psychological outcomes, the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C) has potential application as a screening tool for identifying young people who may be at risk of psychological morbidity. This has important implications for the early diagnosis and treatment of distress among young people following their parent's cancer diagnosis, potentially preventing the development of significant, on-going psychological morbidity.

Chapter 6 presents the final study conducted as part of this thesis. The aim of this study was to determine the discriminative validity and screening utility of the PPIQ-C and establish cut-off scores for identifying young people at risk of psychological distress and post-traumatic stress following their parent's cancer diagnosis.

Abstract

Objective: To determine the discriminative validity and screening utility of the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C) and to establish optimal cut-off scores for identifying young people at risk of poor psychological outcomes following their parent's cancer diagnosis.

Methods: Data from 85 adolescents (aged 12-17 years) and 352 young adults (aged 18-24 years) were analysed to determine the utility of the PPIQ-C for identifying individuals at risk of psychological morbidity in these age groups. Cluster analysis, independent sample t-tests, and chi-square analyses were conducted to identify PPIQ-C subscales most closely associated with poor psychological outcomes. Based on these analyses, a Total Negative Beliefs subscale score was produced. Receiver operating characteristic curves were constructed to examine the ability of the Total Negative Beliefs subscale score to predict psychological distress and post-traumatic stress.

Results: The Total Negative Beliefs subscale consists of 48 items, with a possible total minimum score of 48 and maximum score of 227. Optimal cut-off scores for identifying psychological morbidity were determined; a cut-off score of 137.5 was optimal for identifying psychological distress (sensitivity = 0.746; specificity = 0.836; AUC = 0.829), and scores of 135.5 for young adults (sensitivity = 0.815; specificity = 0.688; AUC = 0.804) and 140.5 for adolescents (sensitivity = 0.830; specificity = 0.60; AUC = 0.731) were optimal for identifying those experiencing post-traumatic stress.

Conclusions: This study provides the first insights into the pattern of illness perceptions that contribute to psychological morbidity in young people following their parent's cancer diagnosis. The Total Negative Beliefs subscale of the PPIQ-C demonstrates good

discriminative validity and may be used as a screening tool for identifying young people at risk of psychological distress or post-traumatic stress.

Introduction

Adolescence and young adulthood is a period of intense physical, emotional, and psychological development (Meeus, 2016). During this time, young people begin to establish their identity independent of their family, pursue education and employment, learn the mechanisms of intimate personal relationships, and gain financial independence (McDonald et al., 2016; Meeus, 2016; Walczak et al., 2018).

Having a parent diagnosed with cancer can cause significant disruption to this process; additional responsibilities at home may pressure young people to remain close to the family at a time when they are seeking greater autonomy (Fletcher, Wilson, Flight, Gunn, & Patterson, 2019; Grabiak et al., 2007; Walczak et al., 2018). In addition, the responsibility of care may shift from the parent being the primary carer to the young person caring for their parent (Harris & Zakowski, 2003). In order to fulfil the role as carer, young people may put their own lives on hold, by withdrawing from education, or delaying moving out of home or seeking full time employment (Fletcher et al., 2019; McDonald et al., 2016).

Adolescents and young adults (AYAs) are particularly vulnerable to negative psychosocial outcomes in the face of a parental cancer diagnosis. They may be more aware than younger children of the seriousness of their parent’s cancer and the potential implications for their family (Gazendam-Donofrio et al., 2011), but may not have developed the ability to cope with the gravity of the situation (Karlsson et al., 2013). Parents may also lean on their older children for emotional support (Gazendam-Donofrio et al., 2011), putting extra pressure on AYAs at this turbulent stage in their development.

Many young people cope well following a parent’s cancer diagnosis, but there remains a large percentage who experience clinically significant levels of distress (Patterson et al., 2017). Early diagnosis and treatment of distress has the potential to reduce emotional suffering and prevent the development of significant, on-going psychological problems, as well as reduce the impact of poor psychosocial outcomes on other important aspects of life (e.g., education, recreational activities, and relationships with friends and family). Self-report questionnaires may be used to screen young people to identify those at risk of psychological morbidity and to determine those who would benefit from supportive intervention. Hence, there is a need to develop screening tools specific to this underserved population.

In an earlier paper,[19] the psychometric properties of the Perceptions of Parental Illness Questionnaire-Cancer (PPIQ-C) were described. This scale was developed to enable assessment of illness perceptions in young people who have a parent with cancer. The findings of this study suggested that strongly held negative emotions and negative beliefs about the impact of symptoms and side effects, the consequences of the illness, and the chronic or unpredictable nature of the illness were related to significantly higher levels of psychological distress and post-traumatic stress.

There is substantial evidence that an individual's illness perceptions are not only related to, but are also predictive of, behavioural and psychological outcomes (Hagger et al., 2017; Hagger & Orbell, 2003). Given that earlier findings suggest that young people's illness perceptions contribute to poor psychosocial outcomes following their parent's cancer diagnosis,[20] it is worth examining whether the PPIQ-C may be suitable for use as a screening tool for identifying young people at risk of psychosocial morbidity. Although there are general measures of distress that have been validated for use in young people aged 12 years and older (e.g., the Kessler Psychological Distress Scale (Kessler et al., 2002; Kessler et al., 2003; Smout, 2019) and the Depression, Anxiety and Stress Scale (Henry & Crawford, 2005; Szabó, 2010)), the aetiology, characteristics, and assessment and treatment needs of distress

---

[19] See paper presented in Chapter 5.

[20] See findings reported in Chapter 5.

in young people impacted by parental cancer may be quite different from the broader population. This has been observed across illness populations, where findings indicate that items from general measures of distress function differently than in norm groups (Waller, Compas, Hollon, & Beckjord, 2005) and thresholds for detecting distress vary according to the illness type (Wang & Gorenstein, 2013). Hence, there is a need for measures of psychological morbidity that are specific to young people affected by parental cancer. Moreover, by using a screening tool that assess young people's illness perceptions, those at risk of psychological distress or post-traumatic stress may be identified *prior to* the onset of symptoms. This is crucial, given that young people may initially present with mild or subclinical psychological problems that, without appropriate intervention, will likely increase in severity over time (Malla et al., 2016; McGorry, Hickie, Yung, Pantelis, & Jackson, 2006).

On this basis, the aims of this study were to determine the discriminative validity and screening utility of the PPIQ-C and to establish optimal cut-off scores for identifying young people at risk of psychological distress and post-traumatic stress following their parent's cancer diagnosis.

Methods

Data collected as part of a previous study were analysed to determine the clinical utility of the PPIQ-C. Methods for participant recruitment and data collection are described in the previous paper.[21]

Statistical analysis. Analysis was conducted using IBM SPSS Statistics version 25.

---

[21] Presented in Chapter 5.

Cluster analysis. Previous research, including research conducted among this cohort,[22] suggests that certain types or patterns of illness perception are associated with better or worse psychosocial outcomes. Therefore, the first step in the analysis was to identify the broader illness schemata that exist within the sample. Hierarchical cluster analysis of PPIQ-C subscale scores was conducted in order to group young people who shared a similar pattern of illness perceptions. Subscale scores were converted to Z scores and Ward's linkage algorithm using the squared Euclidean distance proximity measure was used to identify the number of clusters present (Ward Jr., 1963). Based on inspection of the agglomeration schedule and dendrogram, two clusters were determined to be the optimal solution.

Descriptive statistics were calculated to examine differences in sample characteristics between clusters. Differences in psychological morbidity among clusters were examined using independent samples t-tests and chi-square analysis. Prior to chi-square analysis, scores on measures of psychological distress and post-traumatic stress were dichotomised according to whether they were above or below recommended clinical cut-offs (i.e., at risk vs. not at risk for psychological distress or post-traumatic stress symptoms). In the present study, clinical cut-off scores were as follows: Kessler Psychological Distress Scale (K10) = 20 (Australian Bureau of Statistics, 2012), Impact of Events Scale-Revised (IES-R) = 24 (Weiss & Marmar, 1997), and Children's Revised Impact of Events Scale (CRIES) = 17 (Perrin et al., 2005).

Illness perception clusters informed selection of PPIQ-C subscales that appeared most salient for psychological morbidity; scores on identified subscales were summed to produce a Total Negative Beliefs score.

---

[22] See findings reported in Chapter 5.

Receiver operating characteristic (ROC) analysis. ROC curves were constructed using the Total Negative Beliefs score as the test (independent) variable and the dichotomised level of risk as the outcome (dependent) variable for each measure of psychological morbidity (psychological distress, K10; post-traumatic stress in young adults, IES-R; post-traumatic stress in adolescents, CRIES). The area under the curve (AUC) was interpreted as follows: <0.70 indicates low diagnostic accuracy, 0.70-0.90 indicates moderate diagnostic accuracy, and ≥0.90 indicates high diagnostic accuracy (Bara, Farias, Felden, & Cordeiro, 2017). Youden's index, defined as the sum of sensitivity and specificity minus one, was calculated for each possible Total Negative Beliefs score (see Appendix H). The maximum value of Youden's index was used to determine the optimal cut-off score for identifying young people at risk for each outcome variable.

The discriminative validity of the Total Negative Beliefs subscale was further examined by comparing the predictive ability of the Total Negative Beliefs score to the predictive ability of the *emotional representations* subscale score. Although the CSM proposes that cognitive and emotional responses to illness are regulated simultaneously (Hagger & Orbell, 2003; Hale et al., 2007), the *emotional representations* subscale assesses strongly held negative emotions, and may be reflective of distress. Consistent with this, previous research among this cohort suggests that emotional representations are strongly correlated with psychological distress and post-traumatic stress symptoms.[23] To examine whether the *emotional representations* subscale score was a stronger predictor of psychological morbidity than the Total Negative Beliefs score, an additional set of ROC curves were constructed using the *emotional representations* subscale score as the test

---

[23] See findings reported in Chapter 5.

(independent) variable and the dichotomised level of risk as the outcome (dependent) variable for each measure of psychological morbidity.

Results

Participants characteristics. Participant characteristics are described in the previous paper.[24]

Total subscale scores. Mean total subscale and item level subscale scores for each PPIQ-C subscale are provided in Table 27.

Patterns of illness perceptions. Table 28 provides means and standard deviations for scores on PPIQ-C subscales in each of the two clusters identified by the cluster analysis.

Young people in Cluster 1 had a more coherent understanding of their parent’s cancer, more positive beliefs about their personal ability to control their parent’s symptoms, more positive beliefs about the effectiveness of treatment to control their parent’s cancer, and more positive beliefs about their parent’s likelihood of recovering from their cancer. They also had greater endorsement of chance or bad luck as the cause of their parent’s cancer.

Young people in Cluster 2 scored higher on average on the *identity, emotional representations,* all *consequences, unpredictability,* and *chronicity* subscales. These findings suggest that young people in Cluster 2 have a less positive perception of their parent’s cancer than those in Cluster 1. Specifically, young people in Cluster 2 have greater perceptions of the negative impact of their parent’s cancer symptoms, more strongly held negative emotions about their parent’s cancer, greater perceived consequences, and greater perceptions of the

---

[24] See Table 15 presented in Chapter 5 detailing demographic characteristics of young people who completed the survey.

unpredictable and chronic nature of their parent’s cancer. They also had greater endorsement of behavioural or emotional causes of their parent’s cancer.

Independent samples t-tests were conducted to examine differences in mean subscales scores between the clusters. There was a significant difference between clusters in mean scores for all subscales, except for *chance or bad luck attributions* (p = .180).

Table 27

Descriptive Statistics and Interpretation of PPIQ-C Total Subscale Scores

PPIQ-C subscale	Possible score range	Mean total subscale score	Mean item level subscale score	*n*	Meaning of a high subscale score
Identity	13-52	31.81	2.45	415	Greater perceived negative impact of symptoms on parent
Core subscales					
1. Emotional Representations	6-30	25.11	4.19	433	More strongly held negative emotions associated with parent's cancer
2. Coherence	3-15	10.05	3.35	436	More coherent understanding of parent's cancer
3. Consequences for Personal Development	4-20	15.87	3.97	434	Greater perceived positive consequences for personal growth
4. Consequences for Family Relationships	4-20	10.89	2.72	436	Greater perceived negative consequences for family relationships
5. Consequences for Daily Activities	5-25	16.95	3.39	435	Greater perceived negative consequences for daily activities
6. Personal Control	4-20	13.19	3.29	434	Positive beliefs about the effectiveness of their ability to control parent's symptoms
7. Treatment Control	2-10	7.12	3.56	436	Positive beliefs about the effectiveness of treatment to control parent's cancer
8. Recovery	4-20	14.61	3.65	433	Positive beliefs about parent's likelihood of recovery from cancer
9. Unpredictability	5-25	17.65	3.53	435	Perceptions that parent's cancer is unpredictable in nature
10. Chronicity	3-15	8.25	2.75	433	Perceptions that parent's cancer is chronic in nature
Cause subscales					
1. Behavioural or Environmental Attributions	5-25	10.15	2.03	431	Belief that parent's cancer was caused by behavioural or environmental factors
2. Chance or Bad Luck	2-10	6.95	3.48	436	Beliefs that parent's cancer was caused by chance or bad luck
3. Emotional or Psychological Attributions	3-15	5.96	1.99	436	Belief that parent's cancer was caused by emotional or psychological factors

Table 28

Descriptive Statistics of PPIQ-C Total Subscale Scores for Illness Perception Clusters

PPIQ-C subscale	Scale midpoint	Cluster 1 (*n* = 217) M (SD)	Cluster 2 (*n* = 176) M (SD)	*t*	*p*
Identity	32.5	28.94 (7.10)	35.24 (6.02)	-9.52	.000
Core subscales					
1. Emotional Representations	18	23.89 (4.43)	26.45 (3.42)	-6.45	.000
2. Coherence	9	10.82 (2.43)	9.23 (2.70)	6.06	.000
3. Consequences for Personal Development	12	14.79 (3.47)	17.19 (2.46)	-8.02	.000
4. Consequences for Family Relationships	12	10.24 (3.20)	11.78 (3.09)	-4.80	.000
5. Consequences for Daily Activities	15	15.18 (4.05)	19.11 (3.34)	-10.57	.000
6. Personal Control	12	13.83 (3.16)	12.42 (3.15)	4.41	.000
7. Treatment Control	6	8.39 (1.57)	5.70 (2.26)	13.41	.000
8. Recovery	12	16.29 (2.59)	12.69 (3.93)	10.44	.000
9. Unpredictability	15	15.92 (4.52)	19.64 (3.03)	-9.73	.000
10. Chronicity	9	6.75 (2.59)	10.05 (2.43)	-12.89	.000
Cause subscales					
1. Behavioural or Environmental Attributions	15	9.30 (3.47)	11.18 (4.08)	-4.83	.000
2. Chance or Bad Luck Attributions	6	7.09 (1.99)	6.82 (2.10)	1.34	.180
3. Emotional or Psychological Attributions	9	5.62 (2.48)	6.17 (2.89)	-2.02	.044

Sample characteristics among illness perception clusters. Table 29 outlines characteristics of young people in each of the illness perception clusters. Cluster 1 contained greater percentages of respondents who were male and older, in comparison with Cluster 2 (45.6% vs. 25.6% male and 86.2% vs. 73.9% aged 18-24 years, respectively). Moreover, a greater percentage of those in Cluster 1 reported that their parent with cancer was female and that their parent had finished treatment (66.4% vs. 56.8% female parent and 59.4% vs. 33.5% finished treatment). In contrast, Cluster 2 contained greater percentages of respondents who reported that their parent had been diagnosed with metastatic cancer (45.5%, compared with 15.2% in Cluster 1) or had died (26.7%, compared with 2.3% in Cluster 1).

Differences in psychological morbidity among illness perception clusters. Independent samples t-tests were conducted to examine differences in psychological morbidity among the illness perception clusters (Table 30). Although mean scores for psychological distress (K10) and post-traumatic stress (IES-R and CRIES) were above clinical cut-offs in both clusters, they were significantly greater for young people with a pattern of illness perceptions represented by Cluster 2 (all *p*’s <.05).

Chi-square tests were performed to examine differences in level of risk of psychological morbidity among the illness perception clusters (Table 31). The percentage of respondents who were identified as being “at risk” of psychological morbidity was greater in Cluster 2 for both psychological distress and post-traumatic stress (all *p*’s <.05).

Table 29

Characteristics of Participants in Illness Perception Clusters

Characteristics	Cluster 1 *n* (%)	Cluster 2 *n* (%)
Gender		
Female	114 (52.5)	130 (73.9)
Male	99 (45.6)	45 (25.6)
Non-binary	4 (1.8)	1 (0.6)
Age		
12-17 years	30 (13.8)	46 (26.2)
18-24 years	187 (86.2)	130 (73.9)
Country		
Australia	64 (29.5)	96 (54.5)
United Kingdom	80 (36.9)	47 (26.7)
United States of America	51 (23.5)	31 (17.6)
Canada	11 (5.1)	2 (1.1)
New Zealand	1 (0.5)	-
Ireland	2 (0.9)	-
Other	8 (3.7)	-
Highest level of education completed		
Years 7-10	20 (9.2)	32 (18.2)
Years 11-12	70 (32.3)	71 (40.3)
Certificate or diploma	63 (29.0)	30 (17.0)
Undergraduate degree	52 (24.0)	40 (22.7)
Postgraduate degree	12 (5.5)	3 (1.7)
Gender of parent with cancer		
Female	144 (66.4)	100 (56.8)
Male	71 (32.7)	76 (43.2)
Did not specify	2 (0.9)	-
Metastatic diagnosis	33 (15.2)	80 (45.5)
Stage of cancer continuum		
Recently diagnosed (hasn’t started treatment)	1 (0.5)	3 (1.7)
Having treatment	70 (32.3)	55 (31.3)
Finished treatment	129 (59.4)	59 (33.5)
Receiving palliative care	6 (2.8)	6 (3.4)
Parent passed away	5 (2.3)	47 (26.7)
Unsure	6 (2.8)	6 (3.4)

Table 30

Differences in Scores on Measures of Psychological Morbidity between Illness Perception Clusters

	Scale midpoint	Recommended cut-off score[a]	Cluster 1 M (SD)	Cluster 2 M (SD)	*t*	*p*
Psychological distress (K10)	30	≥20[b]	25.93 (9.32) *n* = 210	32.02 (9.16) *n* = 173	-6.42	.000
Post-traumatic stress (IES-R)	44	≥24[c]	24.26 (16.67) *n* = 180	41.58 (17.65) *n* = 126	-8.73	.000
Post-traumatic stress (CRIES)	20	≥17[d]	18.46 (12.96) *n* = 28	26.00 (9.84) *n* = 45	-2.64	.011

Note. [a]Scores equal to or above recommended cut-off indicate that psychological distress or PTSD is a clinical concern. [b]Australian Bureau of Statistics (2012). [c]Weiss and Marmar (1997). [c]Perrin et al. (2005).

Table 31

Chi-Square Test for Independence (with Yates Continuity Correction) for Level of Risk of Psychological Morbidity in Illness Perception Clusters

	Cluster 1		Cluster 2				
	n	%	*n*	%	χ^2	*p*	Φ
Psychological distress (K10)							
At risk	149	71.0%	161	93.1%	28.66	.000	.280
Not at risk	61	29.0%	12	6.9%			
Post-traumatic stress (IES-R)							
At risk	88	48.9%	106	84.1%	38.16	.000	.360
Not at risk	92	51.1%	20	15.9%			
Post-traumatic stress (CRIES)							
At risk	16	57.1%	45	82.2%	4.27	.039	.273
Not at risk	12	42.9%	8	17.8%			

Determining a cut-off score for identifying young people at risk. To facilitate examination of the discriminative validity of the PPIQ-C, a Total Negative Beliefs score was produced by summing the total scores of subscales that appeared most closely associated with poor psychological adjustment. This was based on the cluster analysis, which revealed higher levels of psychological morbidity among respondents who shared a pattern of illness perceptions represented by Cluster 2. On this basis, the Total Negative Beliefs score was comprised of scores on the *identity*, *emotional representations, consequences for personal development*, *consequences for family relationships*, *consequences for daily activities*, *unpredictability, chronicity, behavioural or environmental attributions*, and *emotional or psychological attributions* subscales. Used separately from the full PPIQ-C, the Total Negative Beliefs subscale consists of 48 items, and has a possible total minimum score of 48 and maximum score of 227.

ROC curves constructed using the Total Negative Beliefs score and the dichotomised level of risk for each measure of psychological morbidity (i.e., the K10, IES-R, and CRIES) are shown in Figure 5. Youden’s index was used to identify Total Negative Beliefs cut-off scores optimised for sensitivity and specificity; these are reported in Table 32, with AUCs and 95% confidence intervals. AUCs indicated moderate diagnostic accuracy of the Total Negative Beliefs cut-off score for identifying young people at risk of psychological distress (AUC = 0.829, 95% CI = 0.776-0.882, p = .000) and post-traumatic stress measured using the IES-R (AUC = 0.804, 95% CI = 0.752-0.857, p = .000) and CRIES (AUC = 0.731, 95% CI = 0.586-0.876, p = .002). Sensitivity and specificity values for alternative cut-off scores are reported in Appendix H.

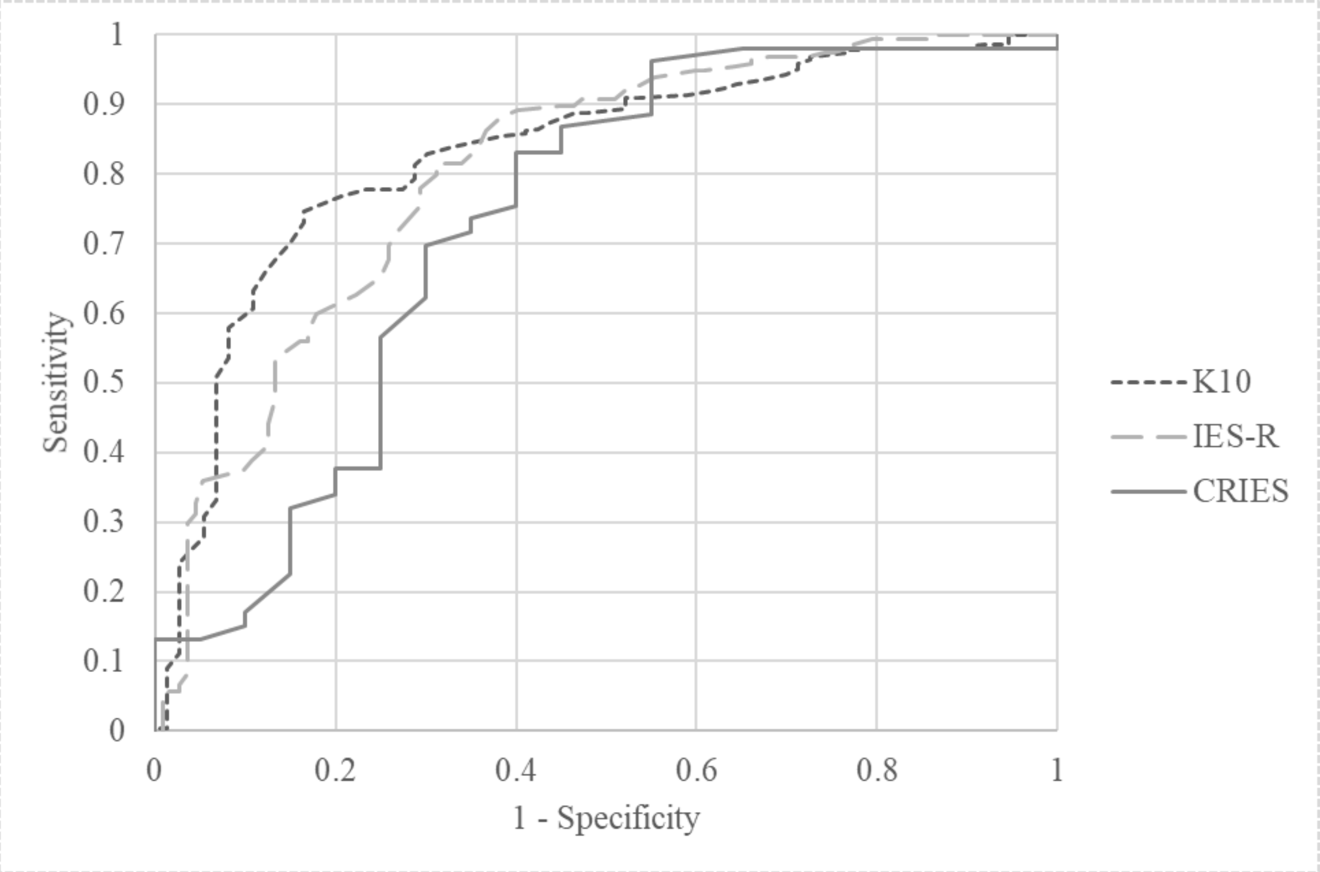


Figure 5. ROC Curves for Total Negative Beliefs Scores Predicting Psychological Morbidity Outcomes

Table 32

ROC Analysis for Screening Ability of the Total Negative Beliefs Subscale Scores for Identifying Psychological Morbidity

Outcome variable	Optimal cut-off score	Sensitivity	Specificity	Maximum Youden's index	AUC	95% CI	*p*
Psychological distress (K10)	137.5	0.746	0.836	0.582	0.829	0.776-0.882	.000
Post-traumatic stress (IES-R)	135.5	0.815	0.688	0.503	0.804	0.752-0.857	.000
Post-traumatic stress (CRIES)	140.5	0.830	0.60	0.430	0.731	0.586-0.876	.002

Comparing the predictive ability of the Total Negative Beliefs subscale and the *emotional representations* subscale. ROC curves constructed using the *emotional representations* subscale and the dichotomised level of risk for each measure of psychological morbidity are presented in Figure 6. ROC analysis indicated moderate diagnostic accuracy of the *emotional representations* subscale for identifying young people at risk of psychological distress (AUC = 0.712, 95% CI = 0.645-0.780, *p* = .000) and post-traumatic stress measured using the IES-R (AUC = 0.713, 95% CI = 0.653-0.773, *p* = .000) and CRIES (AUC = 0..805, 95% CI = 0.673-0.938, *p* = .000) (Table 33). Compared with the Total Negative Beliefs subscale, the *emotional representations* subscale demonstrated weaker diagnostic accuracy for identifying AYAs at risk of psychological distress (AUCs = 0.829 vs. 0.712) and young adults at risk of post-traumatic stress (AUCs = 0.804 vs. 0.713). In contrast, the *emotional representations* subscale demonstrated greater diagnostic accuracy (AUC = 0.805) and yielded better sensitivity (0.868) and specificity (0.70) than the Total Negative Beliefs subscale for identifying adolescents at risk of post-traumatic stress.

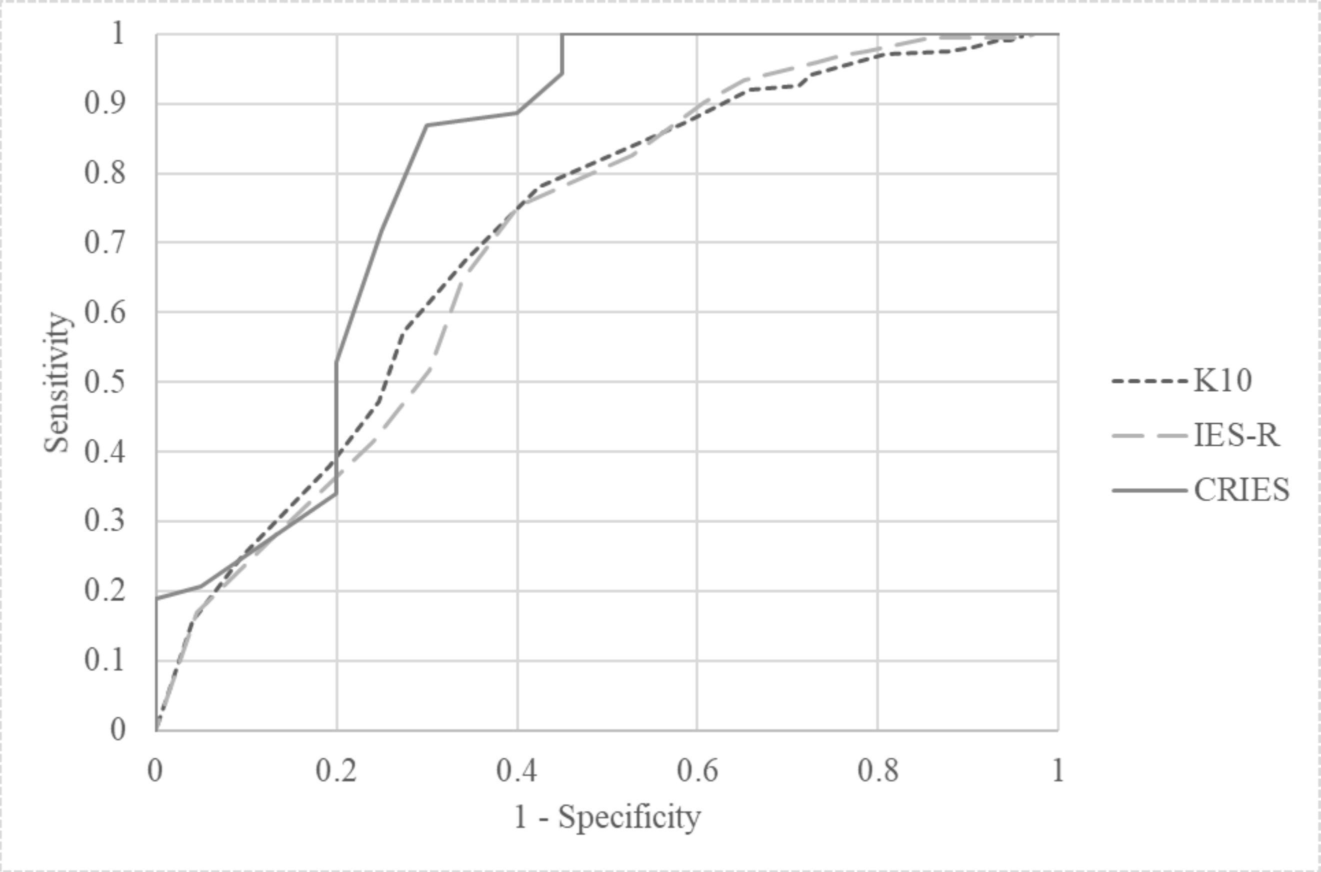


Figure 6. ROC Curves for *Emotional Representations* Scores Predicting Psychological Morbidity Outcomes

Table 33

ROC Analysis for Screening Ability of the Emotional Representations Subscale Scores for Identifying Psychological Morbidity

Outcome variable	Optimal cut-off score	Sensitivity	Specificity	Maximum Youden's index	AUC	95% CI	*p*
Psychological distress (K10)	23.5	0.781	0.575	0.357	0.712	0.645-0.780	.000
Post-traumatic stress (IES-R)	24.5	0.754	0.598	0.352	0.713	0.653-0.773	.000
Post-traumatic stress (CRIES)	23.5	0.868	0.70	0.568	0.805	0.673-0.938	.000

Discussion

The PPIQ-C is the first measure specifically developed to capture illness perceptions in young people who have a parent with cancer. Previously described findings indicate that certain domains of illness perceptions are more strongly associated with poor psychosocial outcomes; in particular, higher levels of psychological distress and post-traumatic stress were found to be associated with higher scores on the *identity, emotional representations*, *consequences for personal development, consequences for family relationships, consequences for daily activities*, *unpredictability*, and *chronicity* subscales of the PPIQ-C.[25] The aim of this study was to extend these findings by examining the utility the PPIQ-C as a screening tool for identifying psychological distress and post-traumatic stress in young people following their parent's cancer diagnosis.

The PPIQ-C assesses a range of beliefs that may either promote or hinder psychological adjustment to parental cancer. For example, illness perceptions within the *treatment control* subscale represent positive beliefs about the effectiveness of treatment in controlling the parent's cancer, whereas beliefs measured by the *identity* subscale represent perceptions of the negative impact of symptoms on the parent. Higher scores on subscales representing positive beliefs may not be associated with poorer psychosocial outcomes. Therefore, it was necessary to first identify a pattern of illness perceptions that was associated with higher levels of psychological distress and post-traumatic stress in young people affected by parental cancer.

Two distinct patterns of illness perceptions were identified. Cluster 1 appeared to describe a group of young people with more positive perceptions of their parent's cancer. They reported having a greater level of understanding of their parent's illness, stronger belief

---

[25] Reported in Chapter 5.

in their own ability and the ability of treatment to control their parent’s cancer, and stronger belief that their parent would recover. This group also had reduced belief in the severity of consequences and the unpredictable or chronic nature of their parent’s illness. They reported reduced belief in behavioural, environmental, or psychological causal attributions; instead they felt that their parent’s cancer was likely caused by chance or bad luck. Cluster 2 represented a group of young people who had stronger emotional responses to their parent’s cancer and perceived greater negative impacts of symptoms and side effects. They believed that their parent’s cancer was unpredictable, chronic, and caused greater disruption to their life. They also felt that their parent’s cancer had a specific cause; whether that was exposure to an environmental factor, engaging in risky health behaviours, or being stressed or worried.

Independent samples t-tests and chi-square analyses revealed that young people with an illness perception pattern represented by Cluster 2 experienced poorer psychosocial outcomes and were at greater risk of psychological distress and post-traumatic stress. This is consistent with previous research conducted among people diagnosed with cancer; McCorry et al. (2013) found that women reported higher levels of anxiety and depression when they perceived their breast cancer as having a more chronic and cyclical timeline, more severe consequences, and as being more symptomatic. They also found that women with this pattern of illness perceptions perceived lower personal and treatment control and reported a less coherent understanding of their illness (McCorry, Dempster, et al., 2013). Much of the research in this area has examined the predictive nature of individual domains of illness perceptions, rather than patterns or groupings of illness perception domains. Nonetheless, other research examining illness perceptions in people affected by cancer provides support for the PPIQ-C subscales that were identified to be most closely associated with psychological morbidity in the present study (Ashley, Marti, Jones, Velikova, & Wright, 2015; Dempster et al., 2010; Gibbons, Groarke, & Sweeney, 2016; E. Richardson et al., 2017; Traeger et al., 2009).

The Total Negative Beliefs score was calculated by summing scores on subscales representing this pattern of illness perceptions. When used separately from the full PPIQ-C, the Total Negative Beliefs subscale represents a relatively brief screening test, comprised of 48 items and with a possible total minimum score of 48 and maximum score of 227. The predictive ability of the Total Negative Beliefs subscale for identifying psychological morbidity in young people was examined using ROC curves. Significant and acceptable AUCs were found for each outcome measure (0.829 for psychological distress; 0.804 for post-traumatic stress in young adults; 0.731 for post-traumatic stress in adolescents), supporting the validity of the Total Negative Beliefs for discriminating between those at risk and not at risk of psychological morbidity.

The predictive ability of the Total Negative Beliefs subscale was compared with the *emotional representations* subscale to more closely examine its screening utility. The Total Negative Beliefs subscale was found to be a stronger predictor of risk of psychological distress in AYAs (AUCs = 0.829 vs. 0.712) and post-traumatic stress in young adults (AUCs = 0.804 vs. 0.713) than the *emotional representations* subscale. In contrast, the *emotional representations* subscale demonstrated slightly greater diagnostic accuracy for identifying adolescents at risk of post-traumatic stress (AUC = 0.805 vs. 0.731). This is consistent with previously reported findings that revealed differences in the strength of correlations between post-traumatic stress, emotional representations, and cognitive representations in adolescents and young adults.[26] For example, emotional representations were found to be more strongly correlated with post-traumatic stress (r = .556) than cognitive representations of parental

---

[26] See findings reported in Chapter 5.

cancer (correlation coefficients ranging from -.275 to .424) in adolescents.[27] Combined, these findings may be reflective of differences in how young people construct illness representations during adolescence and young adulthood, and further investigation is needed to examine the relative importance of emotional and cognitive representations in the development of psychological morbidity among adolescents and young adults.

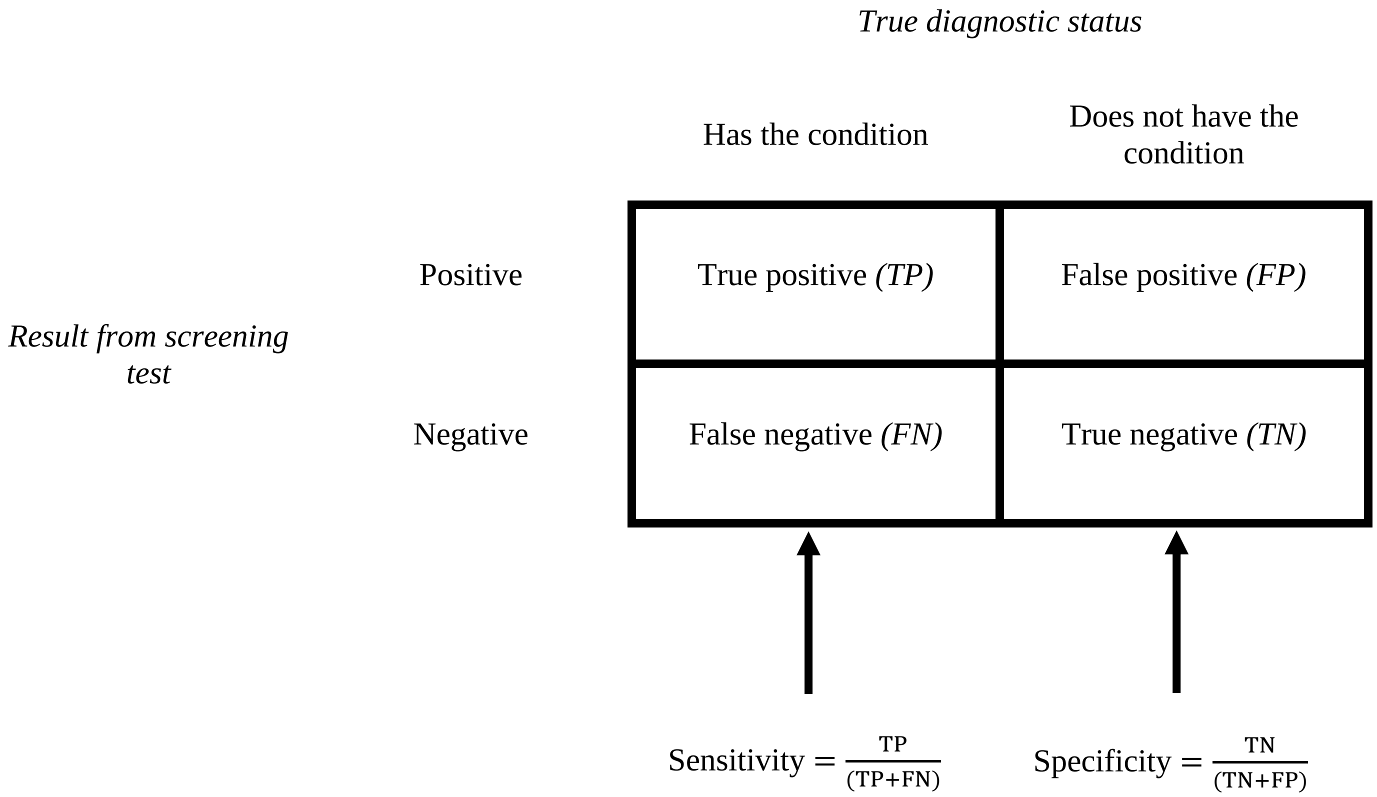
Youden's index was used to determine optimal cut-off scores on the Total Negative Beliefs subscale. A cut-off score of 137.5 was found to be optimal for identifying psychological distress, with a sensitivity of 0.746 and specificity of 0.836. The optimal cut-off score for identifying post-traumatic stress was found to be 135.5 for young adults (sensitivity = 0.815 and specificity = 0.688) and 140.5 for adolescents (sensitivity = 0.83 and specificity = 0.60). Although the Total Negative Beliefs subscale was found to have moderate diagnostic accuracy for identifying post-traumatic stress among adolescents, the *emotional representations* subscale yielded greater sensitivity (0.868) and specificity (0.70) when a cut-off score of 23.5 was applied.

An ideal screening test would have a positive result if and *only if* the individual has the condition, and a negative result if and *only if* the individual did not have the condition, but the reality is that most screening tests do not meet this standard (Maxim, Niebo, & Utell, 2014). The predictive ability of a screening test is impacted by the extent to which it produces "false positives" and "false negatives". The sensitivity of a screening test reflects the probability that it will be positive among those who have the condition (Akobeng, 2007), meaning that a highly sensitive test will capture almost all individuals who have the condition and will produce few false negative results (e.g., a test with a sensitivity of 0.90 will produce true positive results for 90% of individuals screened, but will also produce false negative

---

[27] Correlation coefficients reported in Chapter 5.

results for 10% of individuals screened). The specificity of a screening test reflects the probability that it will be negative among those who do not have the condition (Akobeng, 2007); a test that’s highly specific will screen out almost all individuals who *do not* have the condition and will produce few false positive results (e.g., a test with a specificity of 0.90 will produce true negative results for 90% of individuals screened, but will also produce false positive results for 10% of individuals screened). The relationships between screening test results, true diagnostic status (i.e., whether the individual has the condition), and the sensitivity and specificity of the screening test are illustrated in Figure 6. As can be inferred from the diagram, sensitivity and specificity exist in a state of balance; as sensitivity increases, specificity decreases (and vice versa) (R. Richardson et al., 2015). This trade-off between sensitivity and specificity is demonstrated by a ROC curve, where the rate of true positive results is plotted against the rate of false positive results for different possible cut-off scores (Loewy, Bearden, Johnson, Raine, & Cannon, 2005). When developing a screening test, it is important to consider what is an appropriate balance between sensitivity and specificity for the context in which the test is to be used (R. Richardson et al., 2015).



True diagnostic status

Result from screening test		Has the condition	Does not have the condition
	Positive	True positive *(TP)*	False positive *(FP)*
	Negative	False negative *(FN)*	True negative *(TN)*

Sensitivity $= \frac{\text{TP}}{(\text{TP}+\text{FN})}$ Specificity $= \frac{\text{TN}}{(\text{TN}+\text{FP})}$

Figure 7. Diagram Illustrating the Relationships between Screening Test Results, True Diagnostic Status, and the Sensitivity and Specificity of the Screening Test (adapted from Trevethan, 2017)

In the present study, Youden's index was used as an indicator for determining the optimal cut-off score for identifying psychological morbidity using the Total Negative Beliefs subscale. Youden's index is a common summary measure of the ROC curve and is defined as the sum of sensitivity and specificity minus one (Smits, 2010). It is worth noting that its use does pose some limitations; in particular, the index assumes false positives to be as undesirable as false negatives (Youden, 1950). Smits (2010) argues that this is a serious limitation of the index because "…it is doubtful whether this cost ratio represents the decision maker's true preferences in all cases".

In the case of a screening test – such as the Total Negative Beliefs subscale – the purpose is to identify individuals who are more likely to have a certain condition. Screening for a psychological disorder is the first stage of a two-stage diagnostic process; only those who screen positive will go through a more definitive diagnostic evaluation, which is generally more expensive and invasive than the initial screening procedure (Zimmerman & Guzman Holst, 2018). It is therefore imperative to strike the right balance between sensitivity and specificity when determining a cut-off score to be used in a clinical context. There are multiple factors to consider; first, the screening test should be broad enough to capture all individuals with the outcome of interest. Some researchers argue that, in order to capture as many individuals as possible, greater weighting should be given to the sensitivity of a test over specificity when its purpose is for screening (Loewy et al., 2005; Mazefsky, Kao, & Oswald, 2011; Pelizza et al., 2019).

A lower cut-off score may therefore be more appropriate for identifying young people at risk of psychological morbidity using the Total Negative Beliefs subscale. Youden's index indicated that a cut-off score of ≥137.5 is optimal for identifying young people experiencing psychological distress. With this cut-off score, a sensitivity of 0.746 and specificity of 0.836 is observed (Appendix H). This means that 74.6% of young people who are experiencing psychological distress (true positives) would be captured by the test and continue along the diagnostic pathway, but 25.4% of young people experiencing distress would be missed (false negatives). The proportion of true positives captured by the test can be increased by applying a lower cut-off score. For example, a cut-off score of ≥132.5 would yield a sensitivity of 0.814 and a specificity of 0.712 (Appendix H), meaning that 81.4% of true positives would be captured. To put this into perspective; by lowering the cut-off score from ≥137.5 to ≥132.5 an additional 7% of young people who are experiencing clinically significant levels of distress would undergo diagnostic evaluation and receive appropriate psychosocial support.

However, the capacity of psychosocial support services to be able to provide necessary support is limited. For a screening test to have practical utility, it is critical to also consider the economics of the screening procedure which will be undertaken in a resource-limited environment. Therefore, the cost of screening instruments, the efficiency of administration and scoring, and the availability of clinicians to conduct follow-up evaluations with individuals who screen positive are all factors that need to be considered (Lavigne, Feldman, & Meyers, 2016). For a psychological screening test, such as the Total Negative Beliefs subscale, follow-up testing (i.e., a diagnostic interview) is time-intensive both for clinicians and young people, as well as potentially distressing for the young person.

The cut-off scores reported in the present study may be considered optimal according to Youden's index, however they may not be optimal in terms of their practical utility within psychosocial support services – many of which are chronically under-resourced, understaffed and overstretched. Within this environment, a higher specificity may be more appropriate. Again, consider the cut-off score determined to be optimal for identifying young people experiencing psychological distress (≥137.5). This cut-off score yields a sensitivity of 0.746 with a specificity of 0.836, meaning that 74.6% of young people who are experiencing psychological distress (true positives) would be captured by the test and continue along the diagnostic pathway, while 83.6% of young people who are *not* experiencing psychological distress (true negatives) would be screened out and would not undergo further evaluation. The cut-off score determined for identifying young adults experiencing post-traumatic stress was ≥135.5, with a sensitivity of 0.815 and a specificity of 0.688. For this cut-off score, the specificity value is lower; 68.8% of young people who are *not* experiencing post-traumatic stress (true negatives) would be screened out, but 31.2% (false positives) would need to undergo further evaluation. If a cut-off score of ≥137.5 were applied, a sensitivity of 0.779 and specificity of 0.705 would be observed (Appendix H), reducing (albeit only slightly) the proportion of young people who would needlessly undergo further diagnostic evaluation. Similarly, for identifying adolescents experiencing post-traumatic stress, the determined cut-off score of ≥140.5 yielded a sensitivity of 0.830 and a low specificity of 0.6. If a cut-off score of ≥147.5 were applied, a sensitivity of 0.698 and specificity of 0.7 would be observed (Appendix H), meaning that 69.8% of true positives and 70% of true negatives would be captured. Keeping in mind that sensitivity and specificity exist in a state of balance; with this cut-off score, a greater proportion of false negatives are produced, and 30.2% of adolescents experiencing post-traumatic stress would not be captured.

A delicate balance exists between sensitivity and specificity – produced by the need to be able to capture as many young people as possible, whilst also acknowledging that there are limitations in service providers' capacity to carry out diagnostic evaluation and provide appropriate psychosocial intervention to those who screen positive. If the cut-off score applied has high sensitivity but low specificity, then resources would be wasted by providing services to young people who do not need them. However, if the cut-off score yields low sensitivity but high specificity, then a significant proportion of young people who are needing support would be missed. It is worth considering that the transition from adolescence to adulthood is a period of increased risk for onset of serious mental illness (Read, Roush, &

Downing, 2018). In young people, psychological disorders initially present in milder or subthreshold forms and increase in severity over time (Malla et al., 2016; McGorry et al., 2006). Early detection and intervention are key, not only for preventing the progression of serious mental illness, but also for reducing the risk of death by suicide or a lifetime of disability characterised by social isolation, poor functioning, and reduced engagement with educational and employment opportunities (Gibb, Fergusson, & Horwood, 2010; McGorry & Mei, 2018; Read et al., 2018). Early intervention may come at a cost to service providers and their respective funding bodies in the short term, but the cost of not providing appropriate early intervention may be greater to the individual and the mental health system at large in the long term (McGorry & Mei, 2018).

Use of the Total Negative Beliefs subscale can provide important insights into targets for supportive intervention, but it should be noted that the *emotional representations* subscale may be a better screening tool for identifying adolescents experiencing post-traumatic stress. Given the discussion on sensitivity and specificity, the *emotional representations* subscale yields a greater sensitivity (0.868) and specificity (0.70) when a cut-off score of 23.5 is applied. This would mean that 86.8% of adolescents experiencing post-traumatic stress would be captured by the test and 70% of those who are *not* experiencing post-traumatic stress would be screening out.

Further investigation of the Total Negative Beliefs subscale is needed. Van Stralen et al. (2009) suggest that the positive (PPV) and negative predictive values (NPV) may be more useful indicators of the practical utility of a screening test for daily clinical practice. The PPV indicates the proportion of individuals who are diagnosed with the disorder after receiving a positive screening test result, whereas the NPV is the proportion of individuals who are *not* diagnosed with the disorder after receiving a negative screening test result (R. Richardson et al., 2015). In order to calculate the PPV and NPV for a screening test, one needs to know the prevalence of the disorder as diagnosed among those who have been screened (both among those who screened positive as well as negative). The data needed to determine the PPV and NPV for the Total Negative Beliefs subscale were not collected as part of the present study. Smits (2010) suggests that in the early stages of test development, when the prevalence of the disorder among those screened remains unknown, the AUC is sufficient to assess the predictive utility of the test. The findings of the present study suggest that the Total Negative Beliefs subscale demonstrates moderate diagnostic accuracy for identifying young people experiencing psychological distress and post-traumatic stress (all AUCs >.70). Further research may extend these findings by determining the PPV and NPV for the Total Negative Beliefs subscale. Similarly, investigation of the PPV and NPV for the *emotional representations* subscale would be useful for examining the clinical utility of the scale for identifying post-traumatic stress in adolescents.

The findings of the present study are subject to the limitations described in the previous paper.[28] Most importantly, participants included young people whose parent had recently been diagnosed with cancer, was undergoing treatment, or had completed treatment or died. As a result, many participants responded to items on the PPIQ-C retrospectively. Longitudinal research should be conducted to examine whether scores on the Total Negative Beliefs subscale, measured within 6 months of their parent's diagnosis, predict future psychological morbidity. Moreover, the clinical utility of the Total Negative Beliefs subscale may be limited by its length (48 items). Further research may be conducted to develop and validate a shorter version of the scale, using an approach similar to that used by Broadbent and colleagues (2006) to construct the Brief IPQ. Further research should also be conducted to examine differences in how adolescents and young adults construct illness representations related to parental cancer. In particular, further research should aim to determine the relative

---

[28] See Chapter 5.

importance of emotional and cognitive representations in the development of psychological morbidity among adolescents and young adults. Furthermore, behavioural outcomes should be measured in order to test the full CSM and validity of the PPIQ-C for assessing illness perceptions among AYAs affected by parental cancer. This may be done by measuring internalising and externalising behaviours, for example with the use of the Strengths and Difficulties Questionnaire (R. Goodman, 2001), the Child Behavior Checklist (Achenbach & Ruffle, 2000), or the Youth Self-Report Questionnaire (Achenbach & Rescorla, 2001).

To conclude, this study provides the first insights into the pattern of illness perceptions that contribute to psychological morbidity in young people following their parent's cancer diagnosis. The Total Negative Beliefs subscale of the PPIQ-C demonstrates good discriminative validity and may be used as a screening tool for identifying young people at risk of psychological distress or post-traumatic stress. Youden's index was used to determine optimal cut-off scores to identifying young people experiencing psychological morbidity, however researchers and clinicians alike may consider alternative cut-off scores that yield sensitivity and specificity values that are appropriate for the context in which the screening test is to be used.

6.1. Chapter Summary and Future Directions

Chapter 6 used cross-sectional data collected as part of a previous study[29] to examine the discriminative validity and screening utility of the PPIQ-C for identifying young people at risk of psychological morbidity following their parent's cancer diagnosis. The pattern of PPIQ-C subscales most closely associated with psychological distress and post-traumatic stress symptoms was identified using cluster analysis, independent samples t-tests, and chi-square analyses. Based on this pattern, scores on the *identity*, *emotional representations*, *consequences for personal development*, *consequences for family relationships*, *consequences for daily activities*, *unpredictability*, *chronicity*, *behavioural or environmental attributions*, and *emotional or psychological attributions* subscales were summed to produce a Total Negative Beliefs subscale score.

The subscale consists of 48 items and has a possible total minimum score of 48 and maximum score of 227 (items included in Total Negative Beliefs subscale are presented in Appendix I). ROC curves were constructed in order to determine optimal cut-off scores for identifying young people at risk of psychological distress and post-traumatic stress. Results indicated optimal cut-off scores of 137.5 for identifying psychological distress in AYAs (sensitivity = 0.746; specificity = 0.836),135.5 for identifying post-traumatic stress in young adults (sensitivity = 0.815 and specificity = 0.688), and 140.5 for identifying post-traumatic stress in adolescents (sensitivity = 0.83 and specificity = 0.60). The subscale demonstrated moderate diagnostic accuracy for identifying young people experiencing psychological morbidity (AUC = 0.829 for psychological distress; 0.804 for post-traumatic stress in young adults; 0.731 for post-traumatic stress in adolescents).

The findings presented in Chapter 6 demonstrate that the Total Negative Beliefs subscale of the PPIQ-C is valid for the purpose of discriminating between young people at

---

[29] Reported in Chapter 5.

risk and not at risk of psychological distress and post-traumatic stress. Based on these findings, the subscale appears to be a suitable screening tool that may be used to identify young people in need of more definitive diagnostic evaluation. Use of the Total Negative Beliefs of the PPIQ-C may support service providers to direct their resources more efficiently by conducting diagnostic evaluation with only those who are most likely to experience psychological distress and post-traumatic stress. Future research should determine the PPV and NPV for the Total Negative Beliefs subscale to gain further insight into the practical utility of the subscale as a screening test for use in daily clinical practice.

CHAPTER SEVEN
Discussion and Thesis Conclusion

7.1. Overview

The aim of the research presented in this thesis was to adapt and validate an instrument to measure illness perceptions among adolescents and young adults (AYAs) with a parent diagnosed with cancer. The results from five studies were presented. The first study identified, assessed, and compared the psychometric properties of instruments designed to measure illness perceptions in family members of individuals with chronic physical illness. Only one of the identified instruments was designed for use in children with a parent with a chronic illness (the Perceptions of Parental Illness Questionnaire; PPIQ), suggesting that further research was needed to develop tools to enable measurement of illness perceptions in this population.

The subsequent four studies aimed to address the gap in research examining illness perceptions through the adaptation and psychometric evaluation of a cancer-specific version of the PPIQ (the Perceptions of Parental Illness Questionnaire-Cancer; PPIQ-C). The application of the Common-Sense Model of Self-Regulation (CSM) as a theoretical framework for describing AYAs' illness perceptions related to their parent's cancer was explored (Study 2) and data used to adapt the PPIQ. This initial version of the PPIQ-C was refined following conduct of cognitive interviews with AYAs impacted by parental cancer (Study 3). Data collected through a cross-sectional survey study were analysed to determine the dimensional structure of the PPIQ-C, and to assess the internal consistency, construct validity, and test-retest reliability of the instrument (Study 4). In the final study (Study 5), the utility of the PPIQ-C as a screening tool for identifying AYAs at risk of psychological distress and post-traumatic stress following their parent's cancer diagnosis was examined.

This final chapter reviews the findings of each study presented in this thesis, before examining how this research fits within the context of the wider literature and presenting implications for clinical practice and the provision of supportive intervention. Problems encountered throughout the conduct of this research and potential limitations of the presented studies are discussed, as well as the strengths. Lastly, recommendations for further research are provided and final conclusions drawn.

7.2. Review of Thesis Findings

7.2.1. Systematic Review

As described in Chapter 1, a large body of research demonstrates that the way that an individual thinks and feels about their illness influences their physical and psychological outcomes (Evers et al., 2001; Hagger et al., 2017; Lazarus, 1999; Leventhal et al., 2016a; Petrie & Weinman, 2006). These findings have also been noted for family members and informal carers of someone diagnosed with an illness, although the focus on these groups has received less attention in the literature (Beinke et al., 2016; Bogosian et al., 2014; Compas et al., 1996; Kazak et al., 2004; Sint Nicolaas et al., 2016; Sloper, 2000; Sultan et al., 2016). A systematic review was conducted to identify and compare instruments that were specifically designed or validated for the purpose of assessing illness perceptions in family members of individuals diagnosed with a chronic physical illness (reported in Chapter 2). Because various approaches have been used to conceptualise illness perceptions and describe their relationships with health outcomes, “illness perceptions” were broadly defined to reflect this diversity, and the instruments included in the review were not limited to those with a specific theoretical underpinning.

Of the nine instruments included in the review, only one had been designed and validated for use in children with a parent with a chronic illness (the PPIQ; Bogosian et al., 2014), highlighting that measurement of illness perceptions within this cohort has been overlooked. Generally, findings revealed the need for further psychometric testing of instruments and better reporting of studies describing their development and validation. Comparison of psychometric properties was restricted by the lack of published information on measurement error, test-retest reliability, responsiveness, floor and ceiling effects, and interpretability for most of the instruments. As a result of this lack of psychometric information (specifically measurement error, responsiveness, and interpretability), conclusions about which instruments were valid for the purpose of predicting individual differences in risk for poor psychological outcomes were limited.

The PPIQ was the only identified instrument that was adapted from the Illness Perception Questionnaire and Revised Illness Perception Questionnaire (IPQ/IPQ-R) – the most widely used measures of illness perceptions in individuals diagnosed with an illness (Moss-Morris et al., 2002; Weinman et al., 1996) – and therefore assessed the dimensions of illness perceptions outlined in the CSM (Leventhal et al., 2016a). Likely as a result of its theoretical underpinning, the PPIQ demonstrated good content and construct validity. Given that content validity is a prerequisite for other types of validity (Terwee et al., 2018; Zamanzadeh et al., 2015), evidence of good content validity was crucial when considering an appropriate instrument to adapt and validate for use in AYAs impacted by parental cancer. Moreover, the PPIQ was also the only identified instrument specifically designed for use in children with a chronically ill parent, providing further justification for adapting and validating a cancer-specific version of this instrument as part of this thesis.

7.2.2. Qualitative Study

Prior to adapting the PPIQ, it was necessary to examine whether the CSM was an appropriate framework with which to describe AYA's perceptions of their parent's cancer. Therefore, following the systematic review, a qualitative study was conducted to explore

AYA's perceptions of their parent's cancer and to examine how these perceptions corresponded with the dimensions of illness perceptions outlined in the CSM (reported in Chapter 3).

Eleven AYAs aged 15-24 years participated in interviews. Deductive thematic analysis revealed that all dimensions of illness perceptions described by the CSM were evident within the perspectives shared by the participants. The illness perceptions that participants shared were organised thematically according to the dimensions of the CSM: *emotional representations*, *identity*, *coherence*, *consequences*, *cure or control*, *timeline*, and *cause*. Additional themes that related to interpersonal aspects of the cancer experience (e.g., social support and communication about cancer) were also identified (using inductive thematic analysis), but not described as part of this thesis because they were unrelated to the thesis aims.

The experiences that participants outlined shared many commonalities, despite the differences in their personal circumstances. Young people described their emotional representations in response to their parent's cancer as feelings of fear, uncertainty, and a loss of control. They spoke about the illness identity (the type and stage of their parent's cancer) and the impact of common side effects on their parent's quality of life. Young people described a range of positive and negative consequences of their parent's cancer for them, other members of their family, and their parent. These included having to take on extra household responsibilities (e.g., preparing meals, cleaning the house, looking after pets, and babysitting younger siblings) and the impact of their parent's cancer on schooling (e.g., difficulties keeping up with schoolwork or finding the time to study). Many participants demonstrated an ability to reframe the experience as a positive one; they spoke about how their parent's cancer diagnosis had prompted them to re-evaluate their priorities, brought their family closer together, and taught them to be more appreciative of the time they had together.

Most participants demonstrated an understanding of their parent's cancer, their treatment, and what could be expected in terms of the progression of the disease. Importantly, they indicated that knowing as much information as possible about their parent's cancer helped them to cope. This was demonstrated by their desire to be informed about their parent's cancer and to be actively involved in their parent's cancer experience, by attending medical appointments, researching their parent's cancer, or talking with their parent about treatment options. In terms of the cause of their parent's cancer, many participants reported that cancer was something that "just happens" and others speculated about causes of cancer generally, while not attributing a specific cause to their parent's cancer.

Prior to the conduct of this study, there had been no recent research exploring the beliefs that young people form about their parent's cancer and the potential impact of these beliefs on their psychological well-being following their parent's diagnosis. These findings contributed significantly to knowledge about how young people perceive and make sense of their parent's cancer. Moreover, findings confirmed the relevance of the CSM for describing illness perceptions among young people impacted by parental cancer. The deduction of themes from a theoretical framework strengthened the findings of this study, and the decision to use the CSM as a framework for describing AYA's perceptions of parental cancer provided a suitable foundation from which the PPIQ could be adapted.

7.2.3. Content Validity of the Perceptions of Parental Illness Questionnaire-Cancer

The original PPIQ was developed to assess illness perceptions in adolescents with a parent with multiple sclerosis (Bogosian et al., 2014). Questionnaire items were constructed based on the dimensions of the CSM, items included in the IPQ/IPQ-R, and quotes generated from interviews with adolescents about their experiences with their parent's multiple sclerosis. The structure of the PPIQ differs from the original subscales of the IPQ/IPQ-R and dimensions of the CSM as it does not contain subscales assessing illness perceptions related to *identity, coherence*, and *treatment control*. Although initial drafts of the PPIQ did contain items assessing these dimensions, cognitive interviews and factor analysis revealed problems with these subscales and they were subsequently removed (Bogosian et al., 2014). To overcome this limitation, adaptation of the original PPIQ to produce a cancer-specific version was carried out in two stages. The first stage involved replacing the words 'multiple sclerosis' with 'cancer'. Next, items comprising subscales measuring *identity*, *coherence*, and *treatment control* aspects of the CSM were constructed using a combination of quotes from interviews conducted as part of the qualitative study (reported in Chapter 3) and items from the IPQ/IPQ-R.

Content validity of the initial version of the PPIQ-C was assessed through cognitive interviews with four young people impacted by parental cancer (reported in Chapter 4). Findings provided strong evidence supporting the content validity of the PPIQ-C. Overall, participants found the questionnaire simple to complete and the instructions easy to understand. They described the questionnaire content as highly relevant to their experiences with parental cancer and said the questionnaire items left them feeling acknowledged and understood.

Feedback provided by participants was used to revise the questionnaire and ultimately contributed to improving the relevance, comprehensiveness, and comprehension of questionnaire items. Cognitive interviews revealed several problems with the questionnaire content, the bulk of which centred around the *identity* and *cause* subscales. Participants also highlighted the sensitive nature of the topics covered by the questionnaire and the need for a content warning. Changes were made to the questionnaire on the basis of these findings; six items were reworded to address problems with comprehension, additional items were included to assess aspects of illness perceptions that had not been covered by the questionnaire, instructions for the *identity* subscale were revised to provide further direction and clarity, and instructions for the *cause* subscales were also revised to reduce potential for polarisation and alienation of participants. A brief content warning was also added to the beginning of the questionnaire.

7.2.4. Psychometric Evaluation of the Perceptions of Parental Illness Questionnaire-Cancer

The next stage in the development and initial validation of the PPIQ-C aimed to determine the factor structure of the measure and evaluate its internal consistency, construct validity, and test-retest reliability (reported in Chapter 5). Survey responses were collected from 437 AYAs impacted by parental cancer, a subsample (*n* = 23) of which also completed a two-week retest of the PPIQ-C. To enable construct validity testing, AYAs also completed measures of psychological distress (K10; Kessler et al., 2002; Kessler et al., 2003), post-traumatic stress (IES-R and CRIES; Horowitz et al., 1979; Perrin et al., 2005; P. Smith et al., 2003; Weiss & Marmar, 1997), post-traumatic growth (PTGI-SF and PTGI-C-R; Cann et al., 2010; Kilmer et al., 2010; Tedeschi & Calhoun, 1996) and coping strategies (Brief COPE and KIDCOPE; Carver, 1997; Carver et al., 1989; Spirito et al., 1988).

As in the original IPQ/IPQ-R and PPIQ, the PPIQ-C is comprised of three sections (*identity* subscale, core subscales, and *cause* subscales), each with a separate factor structure. The *identity* subscale is unidimensional and did not undergo factor analysis, however factor analyses were carried out to determine the structure of the core and *cause* subscales. The final PPIQ-C is comprised of 67 items across 14 subscales. The *identity* subscale consists of 13 items, measuring AYAs' perceptions of the negative impact of their parent's cancer symptoms and side effects. Core items were factored into ten subscales: *emotional representations* (6 items), *coherence* (3 items), *consequences for personal development* (4 items), *consequences for family relationships* (4 items), *consequences for daily activities* (5 items), *personal control* (4 items), *treatment control* (2 items), *recovery* (4 items), *unpredictability* (5 items), and *chronicity* (3 items). Items measuring perceptions about the *cause* of cancer were factored into three subscales: *behavioural or environmental attributions* (5 items), *chance or luck attributions* (2 items), and *emotional or psychological attributions* (3 items). An additional four items that assessed causal attributions failed to load onto factors but were retained as part of the PPIQ-C because they represented medically substantiated causes of cancer or were endorsed by a moderate percentage of respondents as a possible cause of their parent's cancer. Differences between the original IPQ/IPQ-R, PPIQ, and final PPIQ-C are outlined in Appendix J.

Internal consistency of subscales was assessed using Cronbach's alpha; all subscales demonstrated acceptable internal consistency (α > .714), except *consequences for family relationships* (α = .589) and *chance or luck attributions* (α = .660). Intraclass correlation coefficients indicated good test-retest reliability over two weeks for most subscales (ICC > .715). Poor test-retest reliability was observed in subscales assessing beliefs about *recovery*, *unpredictability*, and *treatment control* (ICC = .603, .673, and .423, respectively). Possible explanations for this finding are discussed in Chapter 5. Young people who completed the survey were sharing their perspectives of their parent's cancer from a number of different post-diagnosis stages (i.e., recently diagnosed, undergoing curative treatment, completed treatment, receiving palliative care, or had died). Those who responded to the survey after their parent had completed treatment or had died were asked to select answers that best reflected their beliefs and perceptions at the time of their parent's diagnosis and treatment. This may have been particularly challenging for *recovery*, *unpredictability*, and *treatment control* subscales which assessed AYAs' beliefs about their parent's likelihood of recovery, the unpredictable nature of their parent's cancer, and the effectiveness of treatment in controlling their parent's cancer. It is possible that inconsistencies within the data arose from the diversity of experience and exposure to parental cancer within the sample. Post hoc analyses were conducted with the sample split based on the perspective from which young people were responding to the survey (i.e., concurrently or retrospectively). The majority of subscales demonstrated good test-retest reliability over two weeks in AYAs who responded to the questionnaire concurrently (i.e., their parents were recently diagnosed or undergoing treatment for cancer), including *recovery* (ICC = .891) and *unpredictability* (ICC = .787) subscales. However, the *treatment control* subscale did not demonstrate adequate test-retest reliability in either subsample, highlighting that poor test-retest reliability in this subscale is unlikely to arise from differences in the perspective from which respondents are completing the survey. Additional differences were observed for the *coherence* (concurrent response ICC = .426 vs. retrospective response ICC = .787) and *personal control* (concurrent response ICC = .621 vs. retrospective response ICC = .825) subscales, which demonstrated better test-retest reliability among AYAs who responded to the questionnaire retrospectively (i.e., their parent had completed treatment or had died).

Correlations between subscales and measures of psychological distress, post-traumatic stress, and post-traumatic growth provided support for construct validity. Higher levels of psychological distress were significantly associated with higher scores on *identity*, *emotional representations*, *consequences for family relationships*, *consequences for personal development*, *consequences for daily activities*, *unpredictability*, and *chronicity* subscales. Conversely, lower levels of psychological distress were significantly associated with higher scores on *coherence*, *treatment control*, and *recovery* subscales. Contrary to expectations, the correlation between scores on the *personal control* subscale and psychological distress was not significant.

As may be expected, findings for post-traumatic stress mirrored those for psychological distress. In young adults (*n* = 352), higher levels of post-traumatic stress were significantly associated with higher scores on *identity*, *emotional representations*, all *consequences*, *unpredictability*, and *chronicity* subscales. Lower levels of post-traumatic stress were associated with higher scores on *coherence*, *treatment control*, and *recovery* subscales. In adolescents (*n* = 85), higher levels of post-traumatic stress were significantly associated with higher scores on *identity*, *emotional representations*, *consequences for personal development* and *consequences for daily activities* subscales. Lower levels of post-traumatic stress were significantly associated with higher scores on the *treatment control* subscale.

Correlation analyses revealed some unexpected associations between subscale scores and post-traumatic growth. In young adults, higher levels of post-traumatic growth were significantly associated with higher scores on *identity, emotional representations, consequences for personal development, consequences for daily activities, personal control,* and *unpredictability* subscales. Lower levels of post-traumatic growth were significantly associated with higher scores on the *consequences for family relationships* and *treatment control* subscales. Similar associations were observed in adolescents; higher levels of post-traumatic growth were significantly associated with higher scores on *identity*, *emotional representations*, *consequences for personal development*, *personal control*, and *recovery* subscales. Lower levels of post-traumatic growth were significantly associated with higher scores on the *consequences for family relationships* subscale.

Collectively, these findings suggested that young people had poorer psychosocial outcomes following their parent’s cancer diagnosis if they had stronger emotional responses, stronger perceptions of the negative impacts of symptoms and side effects, perceptions that consequences were severe, and a stronger perception that their parent’s cancer was unpredictable and chronic. Moreover, many of the subscales that were associated with poorer psychosocial outcomes, were also associated with greater post-traumatic growth, indicating that an experience of trauma was necessary for the development of post-traumatic growth. This finding was consistent with research conducted among survivors of childhood and adolescent cancer (Turner et al., 2018). Greater perceived consequences for family relationships appeared to restrict post-traumatic growth, suggesting that social support particularly from family members is important for facilitating post-traumatic growth.

Overall, this study provided preliminary evidence that the PPIQ-C is a reliable, valid, and useful tool for assessing illness perceptions among young people impacted by parental cancer, and findings supported its use to identify those at increased risk of poorer psychosocial outcomes.

7.2.5. Screening Utility of the Perceptions of Parental Illness Questionnaire-Cancer

The final study presented in this thesis (Chapter 6) aimed to determine the discriminative validity and screening utility of the PPIQ-C and establish cut-off scores for identifying young people at risk of psychological distress and post-traumatic stress following their parent’s cancer diagnosis.

Data collected as part of the psychometric evaluation study (reported in Chapter 5) were analysed to identify patterns of illness perceptions within the sample. Hierarchical cluster analysis revealed two distinct clusters, with AYAs in Cluster 2 scoring higher, on average, on the *identity*, *emotional representations*, *consequences for family relationships*, *consequences for personal development*, *consequences for daily activities*, *unpredictability*, *chronicity*, *behavioural or environmental attributions*, and *emotional or psychological attributions* subscales. Chi-square tests confirmed that the proportion of AYAs in Cluster 2 who were at risk of psychological morbidity was significantly greater than in Cluster 1, both for psychological distress and post-traumatic stress outcomes. Based on this finding, scores on PPIQ-C subscales representing this pattern of illness perceptions were summed to produce a Total Negative Beliefs score. This subscale consisted of 48 items, with a possible total minimum score of 48 and maximum score of 227.

Optimal cut-off scores for identifying AYAs at risk of psychological distress and post-traumatic stress were determined using receiver operating characteristic (ROC) curves. The area under the curve (AUC) was interpreted to determine the diagnostic accuracy of the Total Negative Beliefs subscale for identifying young people experiencing psychological morbidity. Findings indicated that a cut-off score of 137.5 was optimal for identifying psychological distress (sensitivity = 0.746; specificity = 0.836; AUC = 0.829), and scores of 135.5 for young adults (sensitivity = 0.815; specificity = 0.688; AUC = 0.804) and 140.5 for adolescents (sensitivity = 0.830; specificity = 0.60; AUC = 0.731) were optimal for identifying AYAs experiencing post-traumatic stress. AUCs indicated moderate diagnostic accuracy of the Total Negative Beliefs score for identifying AYAs at risk of psychological morbidity (all AUCs > 0.7).

This study provided the first insights into the broader pattern of illness perceptions that contribute to psychological morbidity in young people following a parental cancer diagnosis. Importantly, findings demonstrated that the Total Negative Beliefs subscale of the PPIQ-C is valid for the purpose of identifying young people at risk of psychological distress and post-traumatic stress, and provides support for the use of the subscale as a screening tool to identify AYAs in need of more detailed diagnostic evaluation.

7.3. Application of the CSM to Parental Cancer

The CSM proposes that an individual’s thoughts and beliefs about an illness contribute to their coping behaviours and, ultimately, their illness outcomes (Hagger et al., 2017; Leventhal et al., 2016a). According to the model, individuals construct emotional and cognitive representations of illness, with cognitive representations organised into five dimensions: *identity*, *consequences*, *cure and control*, *timeline*, and *cause* (Leventhal et al., 2016a). The development of the IPQ and IPQ-R revealed that an individual’s *coherence* or understanding of their illness was another important aspect of illness perception (Moss-Morris et al., 2002; Weinman et al., 1996). This research also led to the reorganisation of the *cure and control* dimension into aspects of *personal control* and *treatment control* (Moss-Morris et al., 2002). Similarly, the *timeline* dimension was redefined as *acute/chronic* and *cyclical/unpredictable* (Moss-Morris et al., 2002).

The CSM was originally constructed to describe illness outcomes in those with the illness, but these relationships are also important for coping and psychological adjustment in family members and significant others. It might be expected that the dimensions of illness perception would be different for those experiencing the illness from an outside perspective; for example, as the child of a parent diagnosed with a chronic illness. The research presented in this thesis demonstrates that this is indeed the case for young people impacted by parental cancer. Based on the thesis findings, the CSM as applied to AYAs with a parent diagnosed with cancer is illustrated in Figure 7. The emotional and cognitive representations of illness constructed by AYAs following their parent’s cancer diagnosis, and their relationships with coping behaviours and psychosocial outcomes (as reported in Chapter 5), are summarised in Table 33.

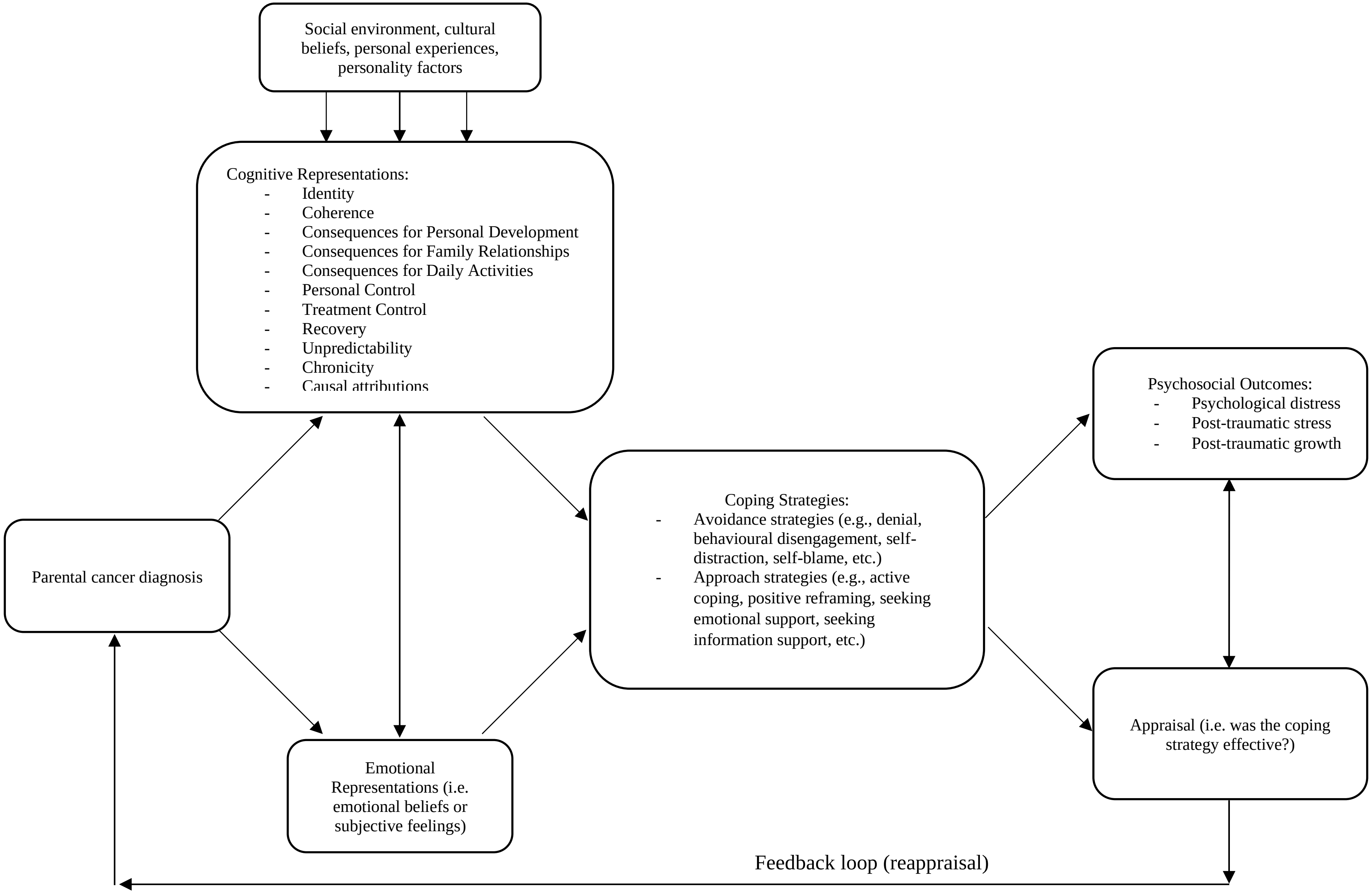


Figure 8. The Relationships between Illness Perceptions, Coping Strategies, and Psychosocial Outcomes in AYAs Impacted by Parental Cancer as Outlined by the Common-Sense Model of Self-Regulation

Table 34

Interpretation of PPIQ-C Subscale Scores and Summary of Relationships between Illness Perceptions, Coping Strategies, and Psychological Outcomes

			Direction of relationship				
PPIQ-C subscale	Definition of subscale	Meaning of a high subscale score	Psychological distress	Post-traumatic stress	Post-traumatic growth	Approach coping	Avoidance coping
Identity	Represents perceptions of parent's cancer symptoms and side effects (e.g., fatigue, pain, physical weakness, nausea).	Greater perceived negative impact of symptoms on parent	+	+[a] +[b]	+[a] +[b]	+[a] + (nonsig.)[b]	+[a] +[b]
Core subscales							
1. Emotional Representations	Represents emotional beliefs and subjective feelings about parent's cancer (e.g., worry, anger, overwhelm, stress, sadness).	More strongly held negative emotions associated with parent's cancer	+	+[a] +[b]	+[a] +[b]	+[a] + (nonsig.)[b]	+[a] +[b]
2. Coherence	Represents perceptions of personal understanding or comprehension of parent's cancer.	More coherent understanding of parent's cancer	-	-[a] - (nonsig.)[b]	- (nonsig.)[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]	-[a] -[b]
3. Consequences for Personal Development	Represents beliefs about the impact of parent's cancer on personal growth; for example, parent's cancer has made them more responsible or more understanding of other people.	Greater perceived positive consequences for personal growth	+	+[a] +[b]	+[a] +[b]	+[a] +[b]	+[a] +[b]
4. Consequences for Family Relationships	Represents perceptions of the consequences of parent's cancer for family relationships; for example, parent's cancer causes arguments in the family or puts a strain on the family.	Greater perceived negative consequences for family relationships	+	+[a] + (nonsig.)[b]	-[a] -[b]	-[a] -[b]	+[a] + (nonsig.)[b]
5. Consequences for Daily Activities	Represents perceptions of the consequences of parent's cancer on day-to-day activities; for example, housework, schoolwork, seeing friends, or other social activities.	Greater perceived negative consequences for daily activities	+	+[a] +[b]	+[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]	+[a] +[b]
6. Personal Control	Represents beliefs about personal ability to control parent's cancer symptoms and side effects by caring for parent or by modifying their behaviour around parent.	Positive beliefs about the effectiveness of their ability to control parent's symptoms	- (nonsig.)	- (nonsig.)[a] + (nonsig.)[b]	+[a] +[b]	+[a] +[b]	- (nonsig.)[a] +[b]
7. Treatment Control	Represents beliefs about the ability of treatment to control or cure parent's cancer.	Positive beliefs about the effectiveness of treatment to control parent's cancer	-	-[a] -[b]	-[a] - (nonsig.)[b]	- (nonsig.)[a] + (nonsig.)[b]	-[a] - (nonsig.)[b]
8. Recovery	Represents beliefs about parent's likelihood of recovering from cancer.	Positive beliefs about parent's likelihood of recovery from cancer	-	-[a] + (nonsig.)[b]	+ (nonsig.)[a] +[b]	+ (nonsig.)[a] + (nonsig.)[b]	-[a] + (nonsig.)[b]
9. Unpredictability	Represents perceptions that parent's cancer is unpredictable and that their symptoms vary day-to-day.	Perceptions that parent's cancer is unpredictable in nature	+	+[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]

PPIQ-C subscale	Definition of subscale	Meaning of a high subscale score	Direction of relationship				
			Psychological distress	Post-traumatic stress	Post-traumatic growth	Approach coping	Avoidance coping
10. Chronicity	Represents perceptions that parent's cancer is chronic and likely to last a long time.	Perceptions that parent's cancer is chronic in nature	+	+[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]	+[a] - (nonsig.)[b]	+[a] +[b]
Cause subscales							
1. Behavioural or Environmental Attributions	Represents beliefs that parent developed cancer because of behavioural or environmental factors (e.g., poor diet, lack of exercise, alcohol consumption, smoking, or chemicals or radiation in the environment).	Belief that parent's cancer was caused by behavioural or environmental factors	+	+[a] + (nonsig.)[b]	+ (nonsig.)[a] - (nonsig.)[b]	+ (nonsig.)[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]
2. Chance or Bad Luck	Represents beliefs that parent developed cancer by chance or because of bad luck	Beliefs that parent's cancer was caused by chance or bad luck	+	+ (nonsig.)[a] + (nonsig.)[b]	- (nonsig.)[a] + (nonsig.)[b]	- (nonsig.)[a] + (nonsig.)[b]	+ (nonsig.)[a] + (nonsig.)[b]
3. Emotional or Psychological Attributions	Represents beliefs that parent developed cancer because of stress or interpersonal problems.	Belief that parent's cancer was caused by emotional or psychological factors	+	+[a] + (nonsig.)[b]	+ (nonsig.)[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]	+[a] + (nonsig.)[b]

+ = positive correlation between scores; - = negative correlation between scores; [a]Young adult participants; [b]Adolescent participants.

7.4. Contribution to Knowledge

The systematic review presented in Chapter 2 highlighted the paucity of research examining illness perceptions in family members and carers and, importantly, identified that there is, to date, no reliable and valid quantitative instrument to assess illness perceptions in children impacted by parental cancer. A growing body of research demonstrates the relationships between illness perceptions, coping behaviours, and psychological outcomes among family members (Beinke et al., 2016; Bogosian et al., 2014; Compas et al., 1994; Compas et al., 1996; Kazak et al., 2004; Sint Nicolaas et al., 2016). Given the potential implications of negative perceptions for psychological morbidity within this cohort, further investigation was warranted.

The research presented in this thesis addresses this gap through the adaptation and validation of the PPIQ-C (described in Chapters 4, 5, and 6), and thus makes a significant contribution to research examining illness perceptions in family members and, more specifically, to knowledge about illness perceptions among AYAs with a parent with cancer. A notable strength of this research is the use of a theoretical framework to develop and validate a clinically useful measure of illness perceptions (the PPIQ-C) for use in this cohort.

Few studies have attempted to measure children's perceptions of their parent's cancer for the purpose of examining the relationships between illness perceptions and psychological adjustment following a parent's cancer diagnosis. Compas et al. (1994 and 1996) assessed children's cognitive appraisals of their parent's cancer using study-specific measures of perceived severity, seriousness, personal control, and external control. In their earlier paper, they reported that perceptions of the seriousness and stressfulness of their parent's cancer were significantly and positively associated with post-traumatic stress symptoms in young adults and adolescents, but not in younger children (Compas et al., 1994). Moreover, perceived stress was only significantly associated with anxiety and depression in adolescents, and not in young adults or preadolescent children. Their later paper replicated these findings (Compas et al., 1996); perceived seriousness was significantly associated with avoidance (of thoughts, feelings, situations, and ideas related to their parent's cancer) and perceived stressfulness was significantly associated with avoidance as well as symptoms of anxiety and depression. Compas et al. (1996) also examined children's perceptions of personal and external control, however correlation analyses revealed no significant relationships between perceived personal and external control, coping behaviours, or psychological outcomes.

Beyond this, there has been a concerning lack of research examining illness perceptions in the context of parental cancer. Although qualitative research has described the experiences of children who have a parent with cancer (Forrest et al., 2006; Karlsson et al., 2013; Kissil, Nino, Jacobs, Davey, & Tubbs, 2010; Thastum et al., 2008), none have done so with the explicit use of a theoretical framework. The qualitative study reported in Chapter 3 was the first to explore AYAs' perceptions of their parent's cancer using an existing model of illness perceptions and coping (i.e., the CSM). The benefit of this approach is that it facilitated the development of a measure based on the model that could be used to assess and identify the aspects of illness perceptions that AYAs hold most strongly (i.e., the PPIQ-C). Moreover, because the PPIQ-C assesses the aspects of illness perception outlined by the CSM, the relationships between illness perceptions, coping behaviours, and psychological outcomes may be interpreted with reference to their theoretical underpinning.

Furthermore, the PPIQ-C enables assessment of additional aspects of illness perception that contribute to the broader illness schema. Where the measures used by Compas et al. (1994 and 1996) assessed perceived seriousness, stressfulness, and controllability of parental cancer, the PPIQ-C measures AYA's emotional representations, coherence, causal attributions, and perceptions of the identity, consequences, controllability, likelihood of recovery, unpredictability, and chronicity of their parent’s cancer. The PPIQ-C also expands on the original PPIQ through the inclusion of subscales measuring perceived *identity*, *coherence*, and *treatment control*.

Use of the PPIQ-C has enabled exploration of the relationships between different aspects of illness perception and psychological outcomes and has facilitated identification of the overall pattern of illness perceptions that are most closely associated with psychological morbidity in AYAs following their parent’s cancer diagnosis. These findings are important contributions to understanding of how AYAs make sense of their parent’s cancer and how this is related to their coping and overall psychological well-being.

Another important contribution is the validation of the Total Negative Beliefs subscale of the PPIQ-C for the purpose of identifying AYAs at risk of psychological distress and post-traumatic stress following their parent’s cancer diagnosis. This has important implications for mental health screening programs and the delivery of psychosocial support targeted to AYAs impacted by parental cancer. These will be discussed further in the next section.

7.5. Broader Implications for Clinical Practice and Provision of Supportive Intervention

It should be noted that the cross-sectional design of the studies limits the conclusions that may be drawn regarding the causal relationships between illness perceptions and psychosocial outcomes in AYAs impacted by parental cancer. Nevertheless, the findings presented in this thesis do have some important implications for clinical practice and service provision.

First, the findings presented in Chapter 6 provide evidence for the discriminative validity of the Total Negative Beliefs subscale and support its use as a screening tool for identifying AYAs who are at risk of psychological distress or post-traumatic stress following their parent's cancer diagnosis. The primary purpose of screening is to improve the detection of psychological morbidity (Martin, Potter, Crocker, Wells, & Colman, 2016), often during an early or pre-symptomatic stage (Iragorri & Spackman, 2018). Given that delays in treatment are associated with a worse prognosis (Kraus, Kadriu, Lanzenberger, Zarate Jr., & Kasper, 2019; Patton et al., 2014; Rice, Eyre, Riglin, & Potter, 2017), appropriate screening and targeted intervention can potentially prevent the development of significant, on-going mental illness and improve psychosocial outcomes in those who are identified as being at risk. Clinical cut-off scores are suggested in Chapter 6; however, clinicians may consider alternative cut-offs to adjust the sensitivity and specificity of the test depending on the purpose and context of its use.

Second, the empirical findings may be used to guide the development of effective strategies to improve psychosocial outcomes and promote positive psychological growth in young people impacted by parental cancer. Because illness perceptions are constructed based on information gathered through social interactions, cultural beliefs, and personal experiences (Hagger et al., 2017; Leventhal et al., 2016a), they are subjective and are not necessarily scientifically or medically substantiated. This means that illness perceptions are malleable (Miceli et al., 2019), making them an appropriate target for personalised interventions that aim to challenge or modify harmful perceptions, promote beneficial perceptions, and improve understanding of the illness (A. Richardson & Broadbent, 2017).

In support of this, there is growing evidence in patient populations that psychosocial interventions can be effective in changing illness perceptions and improving illness-related behaviours (such as medication adherence; Petrie, Perry, Broadbent, & Weinman, 2012; Riaz & Jones Nielsen, 2019), physical outcomes (Glattacker, Heyduck, & Meffert, 2012; Siemonsma et al., 2013), and psychological outcomes (Fischer et al., 2012; Frolund Pedersen et al., 2019; A. Richardson, Tennant, Morton, & Broadbent, 2017). For example, Fischer et al. (2012) reported significant changes in illness perceptions and a reduction in distress in women diagnosed with breast cancer following participation in a psychoeducational group intervention. A significant reduction in emotional representations was observed from baseline to immediately post-intervention (Fischer et al., 2012). At one-year post-intervention, significant changes included a reduction in perceptions relating to illness identity and negative consequences, and an increase in illness coherence (Fischer et al., 2012). Decreases in emotional representations and perceptions relating to illness identity and the cyclical/unpredictable timeline of the cancer were significantly associated with lower levels of distress immediately post-intervention and after one year (Fischer et al., 2012). This has also been demonstrated in spouses of myocardial infarction patients; Broadbent et al. (2009a) found that a brief psychoeducational intervention based on the CSM and designed to change illness perceptions was effective in significantly reducing spouses' anxiety about the patients' condition.

Young people's illness perceptions could be specifically targeted using intervention strategies that provide education or employ therapeutic techniques that are known to be effective for treating depression, anxiety, and post-traumatic stress (such as Cognitive Behaviour Therapy; CBT). According to the findings reported in Chapter 6, young people are more likely to experience psychological morbidity if they have greater perceptions of the negative impact of their parent's cancer symptoms, more strongly held negative emotions about their parent's cancer, greater perceptions of serious consequences, and greater perceptions of the unpredictable and chronic nature of their parent's cancer, as well as greater endorsement of behavioural or emotional causes of their parent's cancer. Lower coherence or understanding of their parent's cancer, poorer perceptions of the controllability of their parent's cancer, and poorer perceptions of the likelihood of their parent's recovery were also observed in those at increased risk of psychological morbidity. Based on these findings, clinicians working with young people should be observant for these types of beliefs and aim to challenge or reframe them where appropriate.

There are a range of psychological and educational strategies that could be utilised to improve outcomes. For example, perceptions relating to *coherence* or *treatment control*, both of which are negatively correlated with psychological distress and post-traumatic stress, may be improved through psychoeducation specific to the parent’s cancer diagnosis (e.g., about the type of cancer and available treatments). The findings of the qualitative study (reported in Chapter 3) provide additional support for this; consistent with previous findings (Maynard et al., 2013), young people described a desire to know as much as possible about their parent’s cancer and felt that this helped them to cope with the experience. This type of intervention may also be appropriate for perceptions relating to *recovery*, *unpredictability, chronicity*, and causal attributions. As another example, supportive intervention strategies could address poor perceptions of *personal control* using cognitive restructuring or reframing techniques in order to improve self-efficacy, reduce psychological distress and post-traumatic stress, and facilitate post-traumatic growth.

It should be noted that not all aspects of illness perception are appropriate for intervention strategies designed to change unhelpful or harmful ways of thinking (as in CBT). For example, perceptions relating to the parent’s poor prognosis, or the *unpredictability* or *chronicity* of their cancer, all of which are related to poorer psychological outcomes, may in fact be accurate interpretations of the parent’s illness. In this instance, an intervention approach that promotes adjustment to, and acceptance of, challenging thoughts, beliefs, and emotions (such as Acceptance and Commitment Therapy; ACT) would be more appropriate.

Moreover, Karekla, Karademas, and Gloster (2019) point out that although the CSM has been used in combination with CBT-oriented approaches, these have often focused on cognitive representations, and generally have not incorporated aspects of emotional regulation (targeted to emotional representations). Karekla et al. (2019) argue that there is considerable overlap between the concepts of ACT and the CSM, and that, for this reason, ACT may have greater potential than other intervention strategies in translating the aspects of the CSM into intervention practice.

In addition to promoting acceptance of thoughts, beliefs, and emotions, ACT skills training cultivates an awareness of the present moment, while also clarifying and promoting behaviours that are in accordance with personal values (Hayes, Luoma, Bond, Masuda, & Lillis, 2006; Hayes, Strosahl, & Wilson, 2011; Karekla et al., 2019). The aim of this approach is to increase psychological flexibility and, in turn, reduce problematic responding to stressful situations (such as a parent's diagnosis with cancer) (Hayes et al., 2006). In practice, use of the PPIQ-C would enable clinicians to identify the negative emotions and maladaptive beliefs that young people hold most strongly. Using an ACT approach, clinicians could discuss the PPIQ-C results with young people and support them to understand the connections between their perceptions of their parent's cancer, their feelings, and their coping behaviours. For example, if the young person strongly believes that their parent's cancer will get worse, they may become angry and engage in coping strategies such as blaming others or rumination. According to ACT principles, this may result in the young person distancing themselves from valued activities in their life, such as social activities or spending quality time with their parent, and reduce their psychological well-being (Hayes et al., 2011; Karekla et al., 2019). In contrast to CBT-oriented approaches, an ACT intervention would not aim to challenge or reframe these types of illness perceptions. Instead, techniques such as cognitive defusion (i.e., observation of cognitions and emotions from a distance; "I am having the thought that my parent's cancer will get worse over time") or acceptance (i.e., allowing unpleasant thoughts, feelings, sensations, and evaluations to come and go without engaging with them or trying to suppress them) may be used (Karekla et al., 2019).

Overall, the availability of the PPIQ-C would allow clinicians to identify young people’s pattern of beliefs about their parent’s cancer. This knowledge may be used by clinicians to develop a nuanced therapeutic treatment plan that is targeted to the individual’s personal profile of illness perceptions.

Another important implication of the findings presented in this thesis is that the PPIQ-C may be helpful in facilitating communication about cancer. The findings presented in Chapters 3 and 4 suggest that being able to openly communicate about cancer and express their feelings about their experience is integral to young people’s psychological well-being. One young person who participated in the cognitive interview study (reported in Chapter 4) suggested that completing the PPIQ-C could help AYAs to identify and make sense of their thoughts and feelings about their parent’s cancer. This participant also suggested that the PPIQ-C could be used by clinicians to facilitate a conversation with AYAs about how they are coping with their parent’s cancer diagnosis in order to identify where extra support may be needed. Another participant said that the nature of the questionnaire content suggested health professionals had an understanding of the experiences and feelings that young people might have following their parent’s cancer diagnosis and made them feel acknowledged and understood. Based on this feedback, use of the PPIQ-C may also assist clinicians in building rapport with AYAs impacted by parental cancer.

7.6. Problems Encountered and Potential Limitations

Problems that were experienced during the conduct of the research and limitations in study design, measurement, and recruitment impact the interpretations and conclusions that may be drawn from the findings presented in this thesis. Many of these have already been discussed in earlier chapters but are considered here in the context of the broader research goals.

7.6.1. Study Design

The aim of the systematic review (reported in Chapter 2) was to identify and compare the psychometric properties of instruments developed and validated for measuring illness perceptions in family members and carers of someone diagnosed with a chronic illness. Diversity in how “illness perceptions” have been conceptualised within the literature likely impacted identification of instruments measuring this construct. The search string included terms relating specifically to illness cognitions, perceptions, and beliefs; however during the study selection process, a number of instruments were identified that measured perceptual constructs that were conceptually related to illness perceptions, although not specifically described as measures of illness perceptions by the study authors. These instruments were included in the review, but it is possible that others were missed. An effort was made to counter problems caused by the narrow scope of the search string, and ensure completeness of study selection, by supplementing the initial search using backward searching techniques (i.e., searching for papers that cited or were cited by the papers included in the review). In addition, the search string did not include terms describing spousal or grandparent-grandchild relationships. As a result, instruments validated for the purpose of measuring illness perceptions in these populations may not have been captured by the literature search. The review did not include any instruments measuring illness perceptions in spouses or grandparents. It is possible that this is related to limitations in the search string, or alternatively may be indicative of gaps in the literature.

The qualitative study reported in Chapter 3 used a predominantly deductive (top-down) approach to analyse and describe the themes evident within AYA’s perceptions of their parent’s cancer. A deductive method was chosen as it allowed themes to be identified and organised using the CSM as a theoretical framework and, given that the aim of the study was to examine how young people’s perceptions corresponded with the dimensions of the

CSM, this was considered most appropriate for the analysis. Use of this method is not without limitations; because deductive analysis is driven by theory, it can provide a detailed analysis of one aspect of the data (in this case, illness perceptions), but potentially produces a less rich description of the data than that offered by a fully inductive (bottom-up) approach (Braun & Clarke, 2006; Nowell, Norris, White, & Moules, 2017). In order to mitigate this limitation, inductive thematic analysis techniques were also employed to identify themes that were not related to the CSM and to ensure that data were analysed comprehensively. Another potential limitation of the deductive analysis is that it was conducted as groundwork for the development of the PPIQ-C. As a result, the researcher's expectations and hypotheses may have inadvertently influenced the analysis and findings. To ensure that themes were empirically grounded and that there was consensus on coding definitions and interpretation of data, a subset of transcripts were coded independently by a second researcher who was not a part of the research team. High levels of agreement were reached.

In the cognitive interview study reported in Chapter 4 a think-aloud approach was employed in combination with verbal probes to assess the content validity of the PPIQ-C by identifying problems that AYAs experienced while completing the questionnaire. Most of the problems identified were related to insufficient thinking aloud. Participants were provided with the questionnaire prior to the interview, and therefore were familiar with the questionnaire content. In hindsight, this potentially compromised the efficacy of the think-aloud method; however, direct verbal probes were used in addition to the think-aloud method and appeared effective in identifying problematic items that would have otherwise been overlooked as a result of insufficient thinking aloud. Another potential limitation of this study is that cognitive interviews were conducted using only the initial draft of the questionnaire; feedback on subsequent drafts may have provided further insight regarding the content validity of the PPIQ-C but was not feasible within candidature timeframe.

Chapters 5 and 6 report cross-sectional survey studies conducted to test the psychometric properties and screening utility of the PPIQ-C. The data that were collected to test the psychometric properties of the PPIQ-C were also analysed to evaluate the screening utility of the instrument. Ideally, separate samples would be used to determine factor structure and assess the discriminative validity of the PPIQ-C.

The survey in the current study was open to young people aged 12-24 years with a parent who had been diagnosed with or treated for cancer within the last five years. Consequently, those who completed the survey were sharing their perspectives of their parent's cancer from a number of different, post-diagnosis stages (i.e., recently diagnosed, undergoing curative treatment, completed treatment, receiving palliative care, or had died). This is likely to have contributed to inconsistencies within the data given the diversity of experience and exposure to parental cancer, potentially impacting the generalisability of the findings.

Some components of the PPIQ-C were less psychometrically robust than others. For example, as discussed in Section 7.2.4., several PPIQ-C subscales showed poor test-retest reliability (*recovery*, *unpredictability*, and *treatment control*). These subscales asked respondents to reflect on their beliefs about their parent's likelihood of recovery, the unpredictable nature of their parent's cancer, and the effectiveness of treatment in controlling their parent's cancer. It is possible that poor test-retest reliability may have resulted from inconsistencies within the data, given that a proportion of participants were responding to the questionnaire concurrently (i.e., soon after their parent's diagnosis or while their parent was undergoing treatment), while others were responding retrospectively. Post hoc analyses confirmed that this was the case for *recovery* and *unpredictability* subscales, which demonstrated better test-retest reliability among those completing the questionnaire concurrently. In contrast, *coherence* and *personal control* subscales demonstrated better test-retest reliability among those responding retrospectively. Moreover, the *treatment control* subscale did not demonstrate adequate test-retest reliability in either subsample. Another limitation of these findings is that test-retest reliability was only assessed among young adult participants.

Lastly, and critically, the cross-sectional design of these studies limits the conclusions that can be drawn from the findings. Although findings revealed associations between PPIQ-C subscales and psychosocial outcomes, because measurements were only collected at one time-point it is not possible to determine cause and effect relationships. Further work is needed to investigate whether certain types of illness beliefs predict psychosocial outcomes over time. This is discussed below in Section 7.8.

7.6.2. Measurement

Problems related to measurement of variables used to validate the PPIQ-C may have also impacted the findings presented in this thesis. The KIDCOPE was used to assess engagement with coping strategies among adolescent participants (aged 12-17 years) as part of the psychometric evaluation study (reported in Chapter 5). Internal reliability assessed with Cronbach's alpha revealed that internal consistency in the *approach strategies* subscale was low (Cronbach's $\alpha = .47$). As a result, findings related to associations between illness perceptions, approach coping strategies, and psychosocial outcomes in adolescent participants should be interpreted with caution.

Problems related to the sensitivity and specificity of the Total Negative Beliefs subscale of the PPIQ-C have been discussed in detail in Chapter 6. The subscale demonstrated good discriminative validity for all psychological outcomes (AUCs > 0.7), but specificity values were below thresholds recommended for screening tools (70%; Sheldrick et al., 2015) for identifying post-traumatic stress in young adults (sensitivity = 0.815 and specificity = 0.688) and adolescents (sensitivity = 0.830 and specificity = 0.60). It is recommended that cut-off scores are adjusted according to the context in which the screening test is to be used.

7.6.3. Recruitment

The generalisability of findings presented in this thesis is limited by the samples included in each of the studies. There was considerable difficulty recruiting young people for each of the studies and future studies could usefully enrol children in a prospective cohort that could be followed through time.

For the qualitative study, participants were recruited online through cancer support and advocacy groups, including Canteen,[30] which is the largest support and advocacy organisation for young people affected by cancer in Australia. Recruitment for this study was time and labour intensive. Young people who participated in the original qualitative study were invited to participate in the follow-up cognitive interview study and were asked to pass on information about the study to those within their social networks that met the eligibility criteria. Difficulties with recruitment contributed to small sample sizes for each of these studies (qualitative study, $n = 11$; cognitive interview study, $n = 4$), although data saturation was achieved (see findings reported in Chapter 3).

Based on the difficulties experienced, a wide range of recruitment strategies were employed to reach an extensive and broad sample of AYAs for the psychometric evaluation study. These included recruitment through cancer support and advocacy groups (Canteen, Breast Cancer Network Australia, Prostate Cancer Foundation Australia, Australian Melanoma Research Foundation, and Cancer Voices, among others), study-specific social media accounts (Facebook and Instagram), and an online platform for participant recruitment for research studies (Prolific). The sample recruited through Prolific included young people

---

[30] Canteen is an Australian support organisation for young people affected by cancer; including young people diagnosed with cancer, and young people who have a brother, sister, or parent diagnosed with cancer. See website for further information: https://www.canteen.org.au/

from Australia, as well as the UK and Ireland, the United States of America, Canada, and New Zealand. Even with such an intensive recruitment effort, full survey responses were collected from only 437 AYAs. This was an adequate sample size for the factor analysis that was conducted (i.e., 7x the number of items and ≥100 participants) and is comparable to other studies conducted among this population (McDonald et al., 2016; Patterson et al., 2013; Patterson et al., 2011). However, a larger sample may have been more representative of the broader population, which would have increased the generalisability of findings. The use of an international sample may limit the validity of the PPIQ-C in specific geographical and cultural contexts. For example, perceptions relating to *treatment control* are likely to vary depending on whether public healthcare is accessible. Additional testing is necessary to ensure the validity of the PPIQ-C for use in different geographical and cultural contexts. The findings presented in Chapters 5 and 6 are also limited in the conclusions that may be drawn because the same data was used to determine the factor structure and evaluate the discriminative validity of the PPIQ-C (as mentioned in Section 7.6.1.).

It should also be noted that samples in each of the studies had low representation of young people aged 12-17 years. This has significant implications for the validity of the PPIQ-C and the Total Negative Beliefs subscale for adolescents. It is possible that the low representation of young people aged 12-17 years was related to the requirement for them to obtain parental consent prior to participation.[31] Given the nature of the research topic (parental cancer), it is possible that needing to obtain parental consent discouraged young people from participating in the studies.

---

[31] Parental consent obtained for participants aged 12-17 years for qualitative study and cognitive interview study (reported in Chapters 3 and 4). For the psychometric evaluation and screening utility studies (reported in Chapter 5 and 6), this requirement for parental consent was modified so that only those younger than 16 years needed to obtain parental consent to participate in the study.

7.7. Ethical Issues

All human interactions have an ethical dimension – this includes human research, which can involve significant risks of harm. Given the sensitive nature of the topic explored, there were a number of ethical issues that impacted the conduct of the research presented in this thesis. Prior to commencement, each study was reviewed and approved by the Social and Behavioural Research Ethics Committee at Flinders University.[32]

The primary concern throughout the conduct of the research was the potential for participants to experience distress as a result of their participation. Every effort was made to mitigate the potential for this to occur. Individual interviews[33] were conducted with young adult participants prior to the inclusion of adolescents (aged 12-17 years). The purpose of this was to ensure that the research protocol did not pose any unforeseen risk of harm, discomfort, or inconvenience that could have a significant and damaging impact on the mental well-being of child participants. It was acknowledged that some participants might experience emotional discomfort during the interview. All recruitment materials clearly outlined that the interview was related to young people's perceptions of their parent's cancer. Participants were well-informed of the content of the discussion prior to participating in the interview; all participants received a copy of the interview schedule in the information pack that was sent to them, along with the information sheet and consent form, prior to deciding to participate in the interview.

Although it was anticipated that participants might experience some level of emotional discomfort while speaking about their experience with parental cancer, they were closely monitored during the interview to identify any signs of distress. A distress protocol was developed based on protocols by Braun and Clarke (2013), Haigh and Witham (2015),

---

[32] Project numbers 7436 (Study 2), 8022 (Study 3), and 8023 (Study 4 and Study 5).

[33] Conducted as part of Study 2; reported in Chapter 3.

and Draucker, Marthsolf, and Poole (2009). If a participant indicated that they were experiencing distress or exhibited behaviours indicative of high levels of distress (e.g., uncontrolled crying or shaking), they were offered immediate emotional support by the researcher. At this stage, the participant was offered a break from the interview and the audio-recorder was turned off. When the participant had calmed down, they were asked if they felt comfortable continuing the interview or if they would have preferred to withdraw from the interview and the study. If the participant decided to continue the interview, they were closely monitored for further signs of distress.

In practice, only one participant became distressed during the interview. This interview was conducted face-to-face. The participant broke down as she recalled having a panic attack during a maths exam; she was talking about how her mother's cancer had impacted her schooling and how she had felt that everything was out of her control. When the participant became distressed, she was asked if she would like to take a break, and the audio-recorder was turned off. The participant was provided with emotional support and asked whether she would like to continue the interview once she had calmed down. The participant agreed to continue the interview; the researcher closely monitored her for further signs of distress and offered additional breaks when behaviours indicative of distress were observed. After the interview, the participant stated that she wasn't used to talking about her mother's cancer in such "concentrated amounts" and that this had been the reason for her emotional reaction.

Young people who responded to the survey were informed prior to participation that the survey would ask questions about their thoughts and beliefs about their parent's cancer, their feelings about their parent's cancer, and their mood generally. The information sheet also advised participants that they were free to stop the survey at any time or to not answer certain questions, and provided a list of phone numbers for support services that they could call if they were feeling distressed.

Ethical issues relating to the consent process were also carefully considered. Although adolescent participants (aged 12-17 years) were required to have parental consent to participate in the interviews conducted at part of Study 2 and Study 3, the consent process was modified for Study 4 and Study 5 to exempt young people aged 16-17 years from needing a parent or guardian's consent to participate in the research. This decision was made following feedback from a young person informing that they were interested in completing the survey but were not comfortable asking their parent for permission because their parent's cancer was not something that was discussed within the family. It was felt that it was crucial for these young people to be represented within the sample because it is this group that would benefit most from the research. This exemption was consistent with guidelines outlined in paragraph 4.2.9 of the National Health and Medical Research Council's National Statement on Ethical Conduct in Human Research (2007, updated 2018), specifically that "it would be contrary to the best interests of the young person to seek consent from the parents, and provision is made to protect the young person's safety, security and well-being in the conduct of the research". Based on the feedback received, it was felt that requiring young people aged 16-17 years to obtain parental consent to participate in the research placed an unnecessary burden on them, caused them significant discomfort, and posed a risk to their mental well-being.

Care was taken to safeguard the confidentiality of the young people who participated in the research presented in this thesis. Prior to participation in an interview or completion of the survey, participants were given assurances that any information they shared would be kept confidential and that they would not be identified, or identifiable, in any resulting publications, presentations, or thesis. Data collected through the conduct of the research was stored securely on a password-protected computer that only the researcher was able to access. Identifying information was removed from the main dataset and stored separately.

7.8. Strengths

This thesis presents a series of studies that employed a methodologically rigorous and theoretically sound framework to address an important gap in research exploring the relationships between illness perceptions and psychological outcomes among family members (as identified by the systematic review reported in Chapter 2). The development and initial validation of the PPIQ-C has significant implications for the provision of psychosocial support to AYAs following their parent’s cancer diagnosis; it facilitates early detection of risk for psychological morbidity and informs the design and implementation of personalised therapeutic treatment.

It is important to acknowledge that the strength of the research reported in this thesis rests with its strong basis in well-supported theory and its systematic sequence of linked studies. Each study contributed to a narrative around young people’s experiences with parental cancer that was convincing and consistent with expectations based on theory. For example, qualitative exploration of perceptions of parental cancer using the CSM ensured that adaptation of a cancer-specific version of the PPIQ (which was identified as part of the systematic review) was theoretically grounded as well as relevant for the target population. Further development and validation of the PPIQ-C adhered, where feasible, to standards for good methodological quality for instrument development and evaluation of psychometric properties (see Appendix K for evaluation of PPIQ-C using COSMIN criteria; Mokkink et al., 2010; Prinsen et al., 2018; Terwee et al., 2007; Terwee et al., 2018). It is important to note that young people were involved in the adaptation of the PPIQ-C through participation in cognitive interviews and provision of feedback on the initial questionnaire draft, and this improved the relevance, comprehensiveness, and comprehension of the instrument. Consequently, the content validity of the PPIQ-C was optimised.

Psychometric evaluation provided evidence for the internal consistency, structural validity, and construct validity of the PPIQ-C, supporting the use of the CSM for examining the relationships between AYAs' perceptions of their parent's cancer, coping behaviours, and psychosocial outcomes. Another notable strength of this research is that the Total Negative Beliefs subscale was found to be valid for the purpose of identifying young people at risk of psychological morbidity (with AUCs indicating moderate diagnostic accuracy; findings presented in Chapter 6). Moreover, the subscales that were identified as being most closely associated with psychological morbidity were consistent with the conceptual framework of the CSM and previous research conducted in those personally diagnosed with cancer (Ashley et al., 2015; Dempster et al., 2010; Gibbons et al., 2016; E. Richardson et al., 2017; Traeger et al., 2009).

Overall, the findings presented in this thesis indicate that the PPIQ-C is a valid and highly relevant measure of perceptions of parental cancer among AYAs and could be used to identify those at risk of psychological morbidity.

7.9. Recommendations for Future Research

Although the research presented in this thesis reports important findings, further research is needed to examine whether these findings are consistent within larger samples. In particular, quantitative research using a larger sample size, in a prospective cohort study, should be conducted to ensure that findings may be generalised and replicated within the wider cancer populations and to highlight any differences in perceptions based on gender, age, family characteristics, demographic characteristics, treatment stage, and cancer type. In addition, given the low representation of young people aged 12-17 years in the presented studies, further research should be conducted to explore the validity of the PPIQ-C in this specific age group.

A prospective cohort study would describe any changes in young people's perceptions of their parent's cancer over time and how these changes relate to context as well as internal resources. This would provide further insight into the nature of the relationship between illness perceptions and psychosocial outcomes. If illness perceptions are found to predict psychosocial outcomes over time, research is needed to determine the critical time to measure illness perceptions; that is, are illness perceptions at the time of *diagnosis* most important for determining psychosocial outcomes at six months or is the relationship between illness perceptions upon *beginning treatment* and psychosocial outcomes at six months stronger? This knowledge is critical to the implementation of effective screening programs and the provision of timely intervention; for example, if AYAs' illness perceptions at the time of treatment commencement are the strongest predictor of psychosocial outcomes at six months, then it is pointless to screen AYAs for psychological morbidity based on their perceptions at the time of their parents' diagnosis.

Further research is also needed to determine where young people's coping responses fit in this model, and whether they mediate the relationships between illness perceptions and psychosocial outcomes, as proposed by the CSM. This has important implications for designing illness perception-based interventions (e.g., whether including a component that encourages specific coping behaviours would increase the efficacy of the intervention).

Because this research is the first attempt to apply the CSM to young people impacted by parental cancer, further intervention work could be conducted to test the model. The relationships between illness perceptions, coping behaviours, and psychological outcomes proposed by the CSM suggest that outcomes may be improved by modifying illness perceptions that are known to be associated with poorer outcomes (Leventhal et al., 2016a).

An illness perception-based intervention study would provide certainty about the direction of cause and effect.

The CSM proposes that an individual's illness perceptions are determined by their personal experience with illness, their personality traits, and the sociocultural context (Hagger & Orbell, 2003; Moss-Morris & Paterson, 1995). Personal attributes and processes such as mindfulness (Greco, Baer, & Smith, 2011), emotional intelligence (Ciarrochi, Chan, & Bajgar, 2001), thought suppression (Wegner & Zanakos, 1994), experiential avoidance (Hayes et al., 2004), and psychological inflexibility (Greco, Lambert, & Baer, 2008) may also contribute to young people's conceptualisation of their parent's cancer – as well as having important implications for illness perception-based interventions that use approaches such as ACT, mindfulness-based cognitive therapy, or mindfulness-based stress reduction.

Another personal attribute that may be worth considering is introspection. In terms of health behaviour, introspection is described as an important attribute that contributes to an individual's ability to identify and interpret their experience of physical symptoms (Hansell & Mechanic, 1986). Consistent with this, studies examining introspection among adolescents have found that those who are highly introspective report more physical symptoms and psychological distress than those who are less introspective (Hansell & Mechanic, 1985; Mechanic, 1983). In the context of parental cancer, it follows that AYAs who are more introspective spend more time thinking about the implications of their parent's cancer and reflecting on their feelings about their parent's diagnosis. Given this, it could be hypothesised that AYAs who are more introspective would experience higher levels of psychological distress than those who are less introspective. Similarly, empathy may also play a role in how an individual perceives the illness of someone close to them, by contributing to their ability to understand the experiences of another and to share their feelings.

The intersections between socioeconomic status, introspection, empathy, and illness perceptions (specifically the illness of another) are also worth investigation. The study of illness behaviour is based on the premise that illness experience is shaped by sociocultural and socio-psychological factors (Mechanic, 1986). Although research examining illness experience in family members has received less attention, it may be assumed that this applies not only to the individual diagnosed with the illness, but also to those who are exposed to illness in others. Previous research examining the relationships between socioeconomic status and emotion has found that individuals living in poorer socio-economic circumstances score more highly on measures of empathy (Manstead, 2018). This is possibly related to a tendency for these individuals to explain events in terms of external factors, which may heighten their sensitivity to the ways in which external events shape emotions, and increase their ability to identify emotion in others (Manstead, 2018). Given that illness perceptions are constructed based on subjective experience, and that the CSM proposes that the social environment contributes directly to how individuals perceive an illness, it is likely that socioeconomic status – and empathy by proxy – influence the mechanisms by which individuals construct representations of illness.

The relationship between young people’s perceptions of their parent’s cancer and the parent’s perceptions of their own cancer should also be examined, given that previous research suggests that the well-being of the individual with the illness and their carer may the impacted by each other’s understanding of the illness (i.e., their illness perceptions; Dempster et al., 2011a; Giannousi et al., 2016; A. Richardson et al., 2015, 2016a; Wu et al., 2013). As discussed in Chapter 1, some findings allude to a reciprocal relationship between the child’s emotional functioning and the parent’s physical and emotional functioning (Moore et al., 2015; Muriel et al., 2012; Visser et al., 2006). Research investigating the relationships between young people’s and parents’ illness perceptions may provide additional context for these findings. For example, Heyduck-Weides et al. (2019) emphasise the importance of examining illness perceptions and the broader self-regulation framework within social and interpersonal contexts – especially for young people, whose self-regulation always occurs in a social context.

As highlighted by the young people who participated in the qualitative study, communication about cancer appears to be another important factor related to coping and adjustment following a parent’s diagnosis. This is consistent with previous research (Huizinga, Visser, van der Graaf, Hoekstra, & Hoekstra-Weebers, 2005; Maynard et al., 2013; McDonald et al., 2016; Thastum et al., 2008), however there are no instruments available to measure parent-child communication about cancer, specifically. Further research should address this gap by developing an instrument to assess parent-child communication about cancer. This would enable further exploration of the predictive role of parent-child communication in young people’s coping and psychological adjustment following their parent’s cancer diagnosis. In general, the availability of a measure of parent-child communication about cancer would be clinically useful for service providers that support families impacted by parental cancer.

Finally, further research should be conducted to examine efficacy of the Total Negative Beliefs subscale for identifying young people at risk of psychological morbidity. This should be evaluated in terms of the positive (PPV) and negative (NPV) predictive values. The PPV indicates the proportion of individuals who are diagnosed with the disorder after receiving a positive screening result, whereas the NPV indicates the proportion of individuals who are confirmed to not have the disorder after receiving a negative screening test result (van Stralen et al., 2009). Other useful indicators of screening efficacy that could be utilised include the yield (i.e., proportion of newly detected cases) and efficiency (i.e., proportion of false positives) (Martin et al., 2016).

7.10. Conclusion

The research presented in this thesis extends previous research examining the factors that contribute to young people's psychological adjustment following a parent's diagnosis with cancer through the validation of an instrument to measure AYAs' perceptions of their parent's cancer.

Findings provide evidence that the PPIQ-C is a reliable and valid measure that may be used to identify patterns of illness perceptions among individuals that lead to poorer psychological outcomes. Development of the PPIQ-C was based on a well-established theoretical framework and involved the target population through participation in cognitive interviews, contributing to both its construct and content validity. Risk of psychological morbidity was found to be marked by stronger emotional reactions, greater perceptions of the negative impacts of their parent's symptoms and side effects, greater perceptions of the unpredictability and chronicity of their parent's cancer, perceptions of more severe consequences related to their parent's cancer, and causal attributions related to behavioural, environmental, emotional, or psychological factors. Findings support the discriminative validity of the Total Negative Beliefs subscale for the purpose of identifying individuals at risk of psychological morbidity. Implications for psychosocial support services have been discussed. Although further research is needed to clarify the direction of the relationships between illness perceptions and psychosocial outcomes, and to ascertain how coping behaviours fit into this model, this thesis has identified that illness perceptions show potential as effective targets for psychosocial interventions that aim to improve outcomes for young people impacted by parental cancer.

Above all, use of the PPIQ-C can contribute to better meeting the needs of young people following their parent's cancer diagnosis by supporting early detection of at-risk individuals, informing the design of psychosocial interventions that are personalised to the individual’s profile of illness perceptions, and promoting long-term psychological well-being and positive psychological growth.

References

Abubakari, A.-R., Jones, M. C., Lauder, W., Kirk, A., Devendra, D., & Anderson, J. R. (2012). Psychometric properties of the Revised Illness Perception Questionnaire: Factor structure and reliability among African-origin populations with type 2 diabetes. *International Journal of Nursing Studies, 49*(6), 672-681. doi:10.1016/j.ijnurstu.2011.11.008

Achenbach, T. M., & Rescorla, L. (2001). *Manual for the ASEBA school-age forms & profiles : an integrated system of multi-informant assessment*. Burlington, VT: ASEBA.

Achenbach, T. M., & Ruffle, T. M. (2000). The Child Behavior Checklist and related forms for assessing behavioral/emotional problems and competencies. *Pediatr Rev, 21*(8), 265-271. doi:10.1542/pir.21-8-265

Ajzen, I. (1985). From intentions to actions: A theory of planned behavior. In J. Kuhl & J. Beckmann (Eds.), *Action-control: From cognition to behavior*. Berlin, Heidelberg: Springer.

Akobeng, A. K. (2007). Understanding diagnostic tests 1: sensitivity, specificity and predictive values. *Acta Paediatrica, 96*(3), 338-341. doi:10.1111/j.1651-2227.2006.00180.x

Al Anbar, N. N., Dardennes, R. M., Prado-Netto, A., Kaye, K., & Contejean, Y. (2010). Treatment choices in autism spectrum disorder: The role of parental illness perceptions. *Research in Developmental Disabilities, 31*(3), 817-828. doi:10.1016/j.ridd.2010.02.007

Andrews, G., & Slade, T. (2001). Interpreting scores on the Kessler Psychological Distress Scale (K10). *Australiam and New Zealand Journal of Public Health, 25*(6), 494-497. doi:10.1111/j.1467-842x.2001.tb00310.x

Applebaum, A. J., & Breitbart, W. (2013). Care for the caregiver: a systematic review. *Palliative and Supportive Care, 11*(3), 231-252. doi:10.1017/S1478951512000594

Ashley, L., Marti, J., Jones, H., Velikova, G., & Wright, P. (2015). Illness perceptions within 6 months of cancer diagnosis are an independent prospective predictor of health-related quality of life 15 months post-diagnosis. *Psycho-Oncology, 24*(11), 1463-1470. doi:10.1002/pon.3812

Ashley, L., Smith, A. B., Keding, A., Jones, H., Velikova, G., & P., W. (2013). Psychometric evaluation of the Revised Illness Perception Questionnaire (IPQ-R) in cancer patients: Confirmatory factor analysis and Rasch analysis. *Journal of Psychosomatic Research, 75*(6), 556-562. doi:10.1016/j.jpsychores.2013.08.005

Assarroudi, A., Nabavi, F. H., Armat, M. R., Ebadi, A., & Vaismoradi, M. (2018). Directed qualitative content analysis: the description and elaboration of its underpinnng methods and data analysis process. *Journal of Research in Nursing, 23*(1), 42-55. doi:10.1177/1744987117741667

Aujla, N., Vedhara, K., Walker, M., & Sprigg, N. (2018). Evaluating a stroke-specific version of the Illness Perception Questionnaire-Revised, using the Think-Aloud method. *Journal of Health Psychology*. doi:10.1177/1359105318781942

Australian Bureau of Statistics. (2012). *4817.0.55.001 - Information Paper: Use of the Kessler Psychological Distress Scale in ABS Health Surveys, Australia, 2007-08* Retrieved from Canberra: https://www.abs.gov.au/ausstats/abs@.nsf/Lookup/4817.0.55.001Chapter92007-08

Australian Institute of Health and Welfare. (2011). *Young Australians: their health and wellbeing 2011* (Cat. no. PHE 140). Retrieved from Canberra:

Australian Institute of Health and Welfare. (2019). *Cancer in Australia: In brief 2019* (Cat. no: CAN 126). Retrieved from Canberra: https://www.aihw.gov.au/reports/cancer/cancer-in-australia-2019-in-brief

Australian Institute of Health and Welfare. (2020). *Cancer data in Australia* (Cat. no: CAN 122). Retrieved from Canberra: https://www.aihw.gov.au/reports/cancer/cancer-data-in-australia

Bara, T. S., Farias, A. C., Felden, E. P. G., & Cordeiro, M. L. (2017). Receiver operating characteristic curve analysis of the Child Behavior Checklist and Teacher's Report Form for assessing autism spectrum disorder in preschool-aged children. *Neuropsychiatric Disease and Treatment, 14*, 95-102. doi:10.2147/NDT.S151185

Barrington, A., & Shakespeare-Finch, J. (2013). Posttraumatic growth and posttraumatic depreciation as predictors of psychological adjustment. *Journal of Loss and Trauma, 18*(5), 429-443. doi:10.1080/15325024.2012.714210

Bassi, M., Falautano, M., Cilia, S., Goretti, B., Grobberio, M., Pattini, M., . . . Fave, A. D. (2016). Illness perception and well-being among persons with multiple sclerosis and their caregivers. *Journal of Clinical Psychology in Medical Settings, 23*(1), 33-52. doi:10.1007/s10880-015-9425-8

Beatty, L., & Kissane, D. (2017). Anxiety and depression in women with breast cancer. *Cancer Forum, 41*(1), 55-61.

Beinke, K., O'Callaghan, F. V., Morrissey, S., & Duregger, C. (2016). Illness perceptions and psychological adjustment of mothers of young adults with cystic fibrosis. *Cogent Psychology, 3*(1).

Belzer, F., Schmidt, S., Lucius-Hoene, G., Schneider, J. F., Orellana-Rios, C. L., & Sauer, S. (2013). Challenging the construct validity of mindfulness assessment - a cognitive interview study of the Freiburg Mindfulness Inventory. *Mindfulness, 4*(1), 33-44. doi:10.1007/s12671-012-0165-7

Bemister, T. B., Brooks, B. L., & Kirton, A. (2014). Development, reliability, and validity of the Alberta Perinatal Stroke Project Parental Outcome Measure. *Pediatric Neurology, 51*(1), 43-52. doi:10.1016/j.pediatrneurol.2014.01.052

Berjot, S., & Gillet, N. (2011). Stress and coping with discrimination and stigmatization. *Frontiers in Psychology, 2*(33). doi:10.3389/fpsyg.2011.00033

Berk, M., Berk, L., Dodd, S., Jacka, F. N., Fitzgerald, P. B., de Castella, A. R., . . . Stafford, L. (2012). Psychometric properties of a scale to measure investment in the sick role: the Illness Cognitions Scale. *Journal of Evaluation in Clinical Practice, 18*, 360-364. doi:10.1111/j.1365-2753.2010.01570.x

Biggs, A., Brough, P., & Drummond, S. (2017). Lazarus and Folkman's psychological stress and coping theory. In C. Cooper & J. C. Quick (Eds.), *The Handbook of Stress and Health: A Guide to Research and Practice* (pp. 351-364). USA: Wiley-Blackwell.

Bogosian, A., Moss-Morris, R., Bishop, F. L., & Hadwin, J. (2014). Development and initial validation of the Perceptions of Parental Illness Questionnaire. *Journal of Health Psychology, 19*(7), 931-942. doi:10.1177/1359105313482164

Bonner, M. J., Hardy, K. K., Guill, A. B., McLaughlin, C., Schweitzer, H., & Carter, K. (2006). Development and validation of the Parent Experience of Child Inness. *Journal of Pediatric Psychology, 31*(3), 310-321. doi:10.1093/jpepsy/jsj034

Bonner, M. J., Hardy, K. K., Willard, V. W., Hutchinson, K. C., & Guill, A. B. (2008). Further validation of the Parent Experience of Child Illness scale. *Children's Health Care, 37*(2), 145-157. doi:10.1080/02739610802006569

Bot, S. D. M., Terwee, C. B., van der Windt, D. A. W. M., Bouter, L. M., Dekker, J., & de Vet, H. C. W. (2004). Clinimetric evaluation of shoulder disability questionnaires: a systematic review of the literature. *Annals of the Rheumatic Diseases, 63*(4), 335-341. doi:10.1136/ard.2003.007724

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77-101. doi:10.1191/1478088706qp063oa

Braun, V., & Clarke, V. (2013). *Successful qualitative research: A practical guide for beginners*: SAGE Publications.

Breland, J. Y., Wong, J. J., & McAndrew, L. M. (2020). Are Common Sense Model constructs and self-efficacy simultaneously correlated with self-management behaviors and health outcomes: A systematic review. *Health Psychology Open, 7*(1). doi:10.1177/2055102919898846

Broadbent, E., Ellis, C. J., Thomas, J., Gamble, G., & Petrie, K. (2009a). Can an illness perception intervention reduce illness anxiety in spouses of myocardial infarction patients? A randomized controlled trial. *Journal of Psychosomatic Research, 67*(1), 11-15. doi:10.1016/j.jpsychores.2008.11.006

Broadbent, E., Ellis, C. J., Thomas, J., Gamble, G., & Petrie, K. (2009b). Further development of an illness perception intervention for myocardial infarction patients: A randomized controlled trial. *Journal of Psychosomatic Research, 67*(1), 17-23. doi:10.1016/j.jpsychores.2008.12.001

Broadbent, E., Petrie, K., Main, J., & Weinman, J. (2006). The brief illness perception questionnaire. *Journal of Psychosomatic Research, 60*(6), 631-637. doi:10.1016/j.jpsychores.2005.10.020

Brohan, E., Slade, M., Clement, S., & Thornicroft, G. (2010). Experiences of mental illness stigma, prejudice and discrimination: a review of measures. *BMC Health Services Research, 10*(80). doi:10.1186/1472-6963-10-80

Browning, K. K., Wewers, M. E., Ferketich, A. K., Otterson, G. A., & Reynolds, N. R. (2010). The Self-Regulation Model of Illness applied to smoking behavior in lung cancer. *Cancer Nursing, 32*(4), E15-E25. doi:10.1097/NCC.0b013e3181a0238f

Bultmann, J. C., Beierlein, V., Romer, G., Moller, B., Koch, U., & Bergelt, C. (2014). Parental cancer: Health-related quality of life and current psychosocial support needs of cancer survivors and their children. *International Journal of Cancer, 135*(11), 2668-2677. doi:10.1002/ijc.28905

Calhoun, L. G., & Tedeschi, R. G. (2006). The foundations of posttraumatic growth: an expanded framework. In L. G. Calhoun & R. G. Tedeschi (Eds.), *Handbook of Posttraumatic Growth: Research and Practice*. New York: Psychology Press.

Cameron, L. D., & Jago, L. (2010). Emotion regulation interventions: A common-sense model approach. *British Journal of Health Psychology, 13*(2), 215-221. doi:10.1348/135910708X288800

Cann, A., Calhoun, L. G., Tedeschi, R. G., Taku, K., Vishnevsky, T., Triplett, K. N., & Danhauer, S. C. (2010). A short form of the Post-traumatic Growth Inventory. *Anxiety, Stress, & Coping, 23*(2), 127-137. doi:10.1080/10615800903094273

Carson, D. K., Gravley, J. E., & Council, J. R. (1992). Children's prehospitalization conceptions of illness, cognitive development, and personal adjustment. *Children's Health Care, 21*(2), 103-110. doi:10.1207/s15326888chc2102_6

Carver, C. S. (1997). You want to measure coping but your protocol's too long: consider the brief COPE. *International Journal of Behavioral Medicine, 4*(1), 92-100.

Carver, C. S., Scheier, M. F., & Weintraub, J. K. (1989). Assessing coping strategies: a theoretically based approach. *Journal of Personality and Social Psychology, 56*(2), 267-283. doi:10.1037//0022-3514.56.2.267

Cassidy, T. (2012). Benefit finding through caring: The cancer caregiver experience. *Psychology and Health, 28*(3), 250-266. doi:10.1080/08870446.2012.717623

Cassidy, T., Giles, M., & McLaughlin, M. (2014). Benefit finding and resilience in child caregivers. *British Journal of Health Psychology, 19*(3), 606-618. doi:10.1111/bjhp.12059

Cassidy, T., McLaughlin, M., & Giles, M. (2014). Benefit finding in response to general life stress: measurement and correlates. *Health Psychology and Behavioral Medicine, 2*(1), 268-282. doi:10.1080/21642850.2014.889570

Chan, S. M., & Fung, T. C. T. (2014). Reliability and validity of K10 and K6 in screening depressive symptoms in Hong Kong adolescents. *Vulnerable Children and Youth Studies, 9*(1), 75-85. doi:10.1080/17450128.2013.861620

Chen, R., Regodón Wallin, A., Norén Selinus, E., Sjölander, A., Fall, K., Valdimarsdóttir, U., . . . Fang, F. (2018). Psychiatric disorders among children of parents with cancer: A Swedish register-based matched cohort study. *Psycho-Oncology, 27*(7), 1854-1860. doi:10.1002/pon.4738

Ciarrochi, J., Chan, A. Y. C., & Bajgar, J. (2001). Measuring emotional intelligence in adolescents. *Personality and Individual Differences, 31*(7), 1105-1119. doi:10.1016/S0191-8869(00)00207-5

Cleeland, C. S. (2007). Symptom burden: Multiple symptoms and their impact as patient-reported outcomes. *JNCI Monographs, 2007*(37), 16-21. doi:10.1093/jncimonographs/lgm005

Cohen, S., & Wills, T. A. (1985). Stress, social support, and the buffering hypothesis. *Psychological Bulletin, 98*(2), 310-357. doi:10.1037/0033-2909.98.2.310

Compas, B. E., Worsham, N. L., Epping-Jordan, J. E., Grant, K. E., Mireault, G., Howell, D. C., & Malcarne, V. L. (1994). When mom or dad has cancer: Markers of psychological distress in cancer patients, spouses, and children. *Health Psychology, 13*(6), 507-515. doi:10.1037/0278-6133.13.6.507

Compas, B. E., Worsham, N. L., Ey, S., & Howell, D. C. (1996). When mom or dad has cancer: II. Coping, cognitive appraisals, and psychological distress in children of cancer patients. *Health Psychology, 15*(3), 167-175. doi:10.1037/0278-6133.15.3.167

Cooper, C. (2014, November 3). Mind your language: 'Battling' cancer metaphors can make terminally ill patients worse. *The Independent*. Retrieved from https://www.independent.co.uk/life-style/health-and-families/health-news/mind-your-language-battling-cancer-metaphors-can-make-terminally-ill-patients-worse-9836322.html

Corbett, T., Groarke, A., Walsh, J. C., & McGuire, B. E. (2016). Cancer-related fatigue in post-treatment cancer survivors: Application of the common sense model of illness representations. *BMC Cancer, 16*(1), 919. doi:10.1186/s12885-016-2907-8

Davey, M., Askew, J., & Godette, K. (2003). Parental and adolescent responses to non-terminal parental cancer: A retrospective multiple-case pilot study. *Families, Systems, & Health, 21*(3), 245-258. doi:10.1037/1091-7527.21.3.245

Davey, M., Tubbs, C. Y., Kissil, K., & Nino, A. (2011). 'We are survivors too': African-American youths' experiences of coping with parental breast cancer. *Psycho-Oncology, 20*(1), 77-87. doi:10.1002/pon.1712

de Boer, M. R., Moll, A. C., de Vet, H. C. W., Terwee, C. B., Volker-Dieben, H. J. M., & van Rens, G. H. M. B. (2004). Psychometric properties of vision-related quality of life questionnaires: a systematic review. *Opthalmic and Physiological Optics, 24*(4), 257-273. doi:10.1111/j.1475-1313.2004.00187.x

de Leeuw, E., Borgers, N., & Smits, A. (2004). Pretesting questionnaires for children and adolescents. In J. M. R. Stanley Presser, Mick P. Couper, Judith T. Lessler, Elizabeth Martin, Jean Martin, and Eleanor Singer (Ed.), *Methods for Testing and Evaluating Survey Questionnaires*: John Wiley & Sons, Inc.

de Vet, K. A., & Ireys, H. T. (1998). Psychometric properties of the Maternal Worry Scale for children with chronic illness. *Journal of Pediatric Psychology, 23*(4), 257-266.

Del Castillo, A., Godoy-Izquierdo, D., Vazquez, M. L., & Godoy, J. F. (2011). Illness beliefs about cancer among healthy adults who have and have not lived with cancer patients. *International Journal of Behavioral Medicine, 18,* 342-351. doi:10.1007/s12529-010-9141-6

Dempster, M., & McCorry, N. K. (2012). The factor structure of the revised Illness Perception Questionnaire in a population of oesophageal cancer survivors. *Psycho-Oncology, 21*(5), 524-530. doi:10.1002/pon.1927

Dempster, M., McCorry, N. K., Brennan, E., Donnelly, M., Murray, L. J., & Johnston, B. T. (2010). Do changes in illness perceptions predict changes in psychological distress among oesophageal cancer survivors? *Journal of Health Psychology, 16*(3), 500-509. doi:10.1177/1359105310386633

Dempster, M., McCorry, N. K., Brennan, E., Donnelly, M., Murray, L. J., & Johnston, B. T. (2011a). Illness perceptions among carer-survivor dyads are related to psychological distress among oesophageal cancer survivors. *Journal of Psychosomatic Research, 70,* 432-439. doi:10.1016/j.jpsychores.2010.07.007

Dempster, M., McCorry, N. K., Brennan, E., Donnelly, M., Murray, L. J., & Johnston, B. T. (2011b). Psychological distress among family carers of oesophageal cancer survivors: The role of illness cognitions and coping. *Psycho-Oncology, 20*(7), 698-705. doi:10.1002/pon.1760

Diefenbach, M. A., & Leventhal, H. (1996). The Common-Sense Model of Illness Representation: Theoretical and practical considerations. *Journal of Social Distress and the Homeless, 5*(1), 11-37. doi:10.1007/BF02090456

Draucker, C. B., Martsolf, D. S., & Poole, C. (2009). Developing distress protocols for research on sensitive topics. *Arch Psychiatr Nurs, 23*(5), 343-350. doi:10.1016/j.apnu.2008.10.008

Drennan, J. (2003). Cognitive interviewing: Verbal data in the design and pretesting of questionnaires. *Journal of Advanced Nursing, 42*(1), 57-63. doi:10.1046/j.1365-2648.2003.02579.x

Edwards, L., Watson, M., St James-Roberts, I., Ashley, S., Tilney, C., Brougham, B., . . . Romer, G. (2008). Adolescent's stress responses and psychological functioning when a parent has early breast cancer. *Psycho-Oncology, 17*(10), 1039-1047. doi:10.1002/pon.1323

Ellis, S. J., Wakefield, C. E., Antill, G., Burns, M., & Patterson, P. (2016). Supporting children facing a parent's cancer diagnosis: a systematic review of children's psychosocial needs and existing interventions. *European Journal of Cancer Care, 26*(1). doi:10.1111/ecc.12432

Ernst, J., Gotze, H., Krauel, K., Romer, G., Bergelt, C., Hans-Henning, F., . . . von Klitzing, K. (2013). Psychological distress in cancer patients with underage children: gender-specific differences. *Psycho-Oncology, 22*(4), 823-828. doi:10.1002/pon.3070

Evers, A. W., Kraaimaat, F. W., van Lankveld, W., Jongen, P. J. H., Jacobs, J. W. G., & Bijlsma, J. W. J. (2001). Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *Journal of Consulting and Clinical Psychology, 69*(6), 1026-1036. doi:10.1037/0022-006X.69.6.1026

Faccio, F., Ferrari, F., & Pravettoni, G. (2018). When a parent has cancer: How does it impact on children's psychosocial functioning? A systematic review. *European Journal of Cancer Care, 27*(6), e12895.

Faulkner, R. A., & Davey, M. (2002). Children and adolescents of cancer patients: The impact of cancer on the family. *The American Journal of Family Therapy, 30*(1), 63-72. doi:10.1080/019261802753455651

Felnhofer, A., Goreis, A., Bussek, T., Kafka, J. X., König, D., Klier, C., . . . Kothgassner, O. D. (2019). Evaluating parents' and children's assessments of competence, health-related quality of life and illness perception. *Journal of Child and Family Studies*. doi:10.1007/s10826-019-01449-x

Ferlay, J., Colombet, M., Soerjomataram, I., Mathers, C., Parkin, D. M., Piñeros, M., . . . Bray, F. (2019). Estimating the global cancer incidence and mortality in 2018: GLOBOCAN sources and methods. *International Journal of Cancer, 144*(8), 1941-1953. doi:10.1002/ijc.31937

Fernandes, C., Muller, R., & Rodin, G. (2012). Predictors of parenting stress in patients with haematological cancer. *Journal of Psychosocial Oncology, 30*(1), 81-96. doi:10.1080/07347332.2011.633978

Fischer, M. J., Wiesenhaan, M. E., Does-den Heijer, A., Kleijn, W. C., Nortier, J. W. R., & Kaptein, A. A. (2012). From despair to hope: A longitudinal study of illness perceptions and coping in a psycho-educational group intervention for women with breast cancer. *British Journal of Health Psychology, 18*(3), 526-545. doi:10.1111/j.2044-8287.2012.02100.x

Fletcher, C., Wilson, C., Flight, I., Gunn, K., & Patterson, P. (2019). Illness cognitions among adolescents and young adults who have a parent with cancer: A qualitative exploration using the Common-Sense Model of Self-Regulation as a framework. *International Journal of Behavioral Medicine, 26*(5), 531-541. doi:10.1007/s12529-019-09793-4

Forrest, G., Plumb, C., Ziebland, S., & Stein, A. (2006). Breast cancer in the family - children's perceptions of their mother's cancer and its initial treatment: qualitative study. *BMJ, 332*(7548), 998-1003. doi:10.1136/bmj.38793.567801.AE

Fortune, D. G., Smith, J. V., & Garvey, K. (2011). Perceptions of psychosis, coping, appraisals, and psychological distress in the relatives of patients with schizophrenia: An exploration using self-regulation theory. *British Journal of Clinical Psychology, 44*(3). doi:10.1348/014466505X29198

French, D. P., Cooke, R., Mclean, N., Williams, M., & Sutton, S. (2007). What do people think about when they answer Theory of Planned Behaviour questionnaires? A 'think aloud' study. *Journal of Health Psychology, 12*, 672-687. doi:10.1177/1359105307078174

French, D. P., & Weinman, J. (2008). Assessing illness perceptions: Beyond the IPQ. *Psychology & Health, 23*(1), 5-9. doi:10.1080/08870440701616714

Frolund Pedersen, H., Holsting, A., Frostholm, L., Rask, C., Jensen, J. S., Hoeg, M. D., & Schroder, A. (2019). "Understand your illness and your needs": Assessment-informed patient education for people with multiple functional somatic syndromes. *Patient Education and Counseling, 102*(9), 1662-1671. doi:10.1016/j.pec.2019.04.016

Gatzoyia, D., Kotsis, K., Koullourou, I., Goulia, P., Carvalho, A. F., Soulis, S., & Hyphantis, T. (2014). The association of illness perceptions with depressive symptoms and general psychological distress in parents of an offspring with autism spectrum disorder. *Disability and Health Journal, 7*(2), 173-180. doi:10.1016/j.dhjo.2013.10.008

Gazendam-Donofrio, S. M., Hoekstra, H. J., van der Graaf, W. T., van de Wiel, H. B., Visser, A., Huizinga, G. A., & Hoekstra-Weebers, J. E. (2007). Family functioning and adolescents' emotional and behavioral problems: When a parent has cancer. *Annals of Oncology, 18*(12), P1951-1956. doi:10.1093/annonc/mdm373

Gazendam-Donofrio, S. M., Hoekstra, H. J., van der Graaf, W. T., van de Wiel, H. B., Visser, A., Huizinga, G. A., & Hoekstra-Weebers, J. E. (2011). Adolescents' emotional reactions to parental cancer: Effect on emotional and behavioral problems. *Journal of Pediatric Psychology, 36*(3), 346-359. doi:10.1093/jpepsy/jsq090

Giannousi, Z., Karademas, E. C., & Dimitraki, G. (2016). Illness representations and psychological adjustment of Greek couples dealing with a recently-diagnosed cancer: Dyadic, interaction and perception-dissimilarity effects. *Journal of Behavioral Medicine, 39*, 85-93. doi:10.1007/s10865-015-9664-z

Gibb, S. J., Fergusson, D. M., & Horwood, L. J. (2010). Burden of psychiatric disorder in young adulthood and life outcomes at age 30. *British Journal of Psychiatry, 197*(2), 122-127. doi:10.1192/bjp.bp.109.076570

Gibbons, A., Groarke, A., & Sweeney, K. (2016). Predicting general and cancer-related distress in women with newly diagnosed breast cancer. *BMC Cancer, 16*(1), 935. doi:10.1186/s12885-016-2964-z

Glattacker, M., Heyduck, K., & Meffert, C. (2012). Illness beliefs, treatment beliefs and information needs as starting points for patient information--evaluation of an intervention for patients with chronic back pain. *Patient Education and Counseling, 86*(3), 378-389. doi:10.1016/j.pec.2011.05.028

Gobel, A., Henning, A., Moller, C., & Aschersleben, G. (2016). The relationship between emotion comprehension and internalizing and externalizing behavior in 7-to 10-year-old children. *Frontiers in Psychology, 7*, 1917. doi:10.3389/fpsyg.2016.01917

Goh, Y. W., Sawang, S., & Oei, T. P. S. (2010). The Revised Transactional Model (RTM) of Occupational Stress and Coping: An improved process approach. *The Australasian Journal of Organisational Psychology, 3*(1), 13-20. doi:10.1375/ajop.3.1.13

Goodman, R. (2001). Psychometric Properties of the Strengths and Difficulties Questionnaire. *Journal of the American Academy of Child & Adolescent Psychiatry, 40*(11), 1337-1345. doi:10.1097/00004583-200111000-00015

Goodman, V. D. (2011). *Qualitative Research and the Modern Library*: Chandos Publishing.

Götze, H., Ernst, J., Brähler, E., Romer, G., & von Klitzing, K. (2015). Predictors of quality of life of cancer patients, their children, and partners. *Psycho-Oncology, 24*(7), 787-795. doi:10.1002/pon.3725

Gough, B. (2003). Deconstructing reflexivity. In L. Finlay & B. Gough (Eds.), *Reflexivity: A practical guide for researchers in health and social sciences* (pp. 21-35). Oxford: Blackwell Science.

Grabiak, B. R., Bender, C. M., & Puskar, K. R. (2007). The impact of parental cancer on the adolescent: an analysis of the literature. *Psycho-Oncology, 16*(2), 127-137. doi:10.1002/pon.1083

Graham, L., Dempster, M., McCorry, N. K., Donnelly, M., & Johnston, B. T. (2016). Change in psychological distress in longer-term oesophageal cancer carers: are clusters of illness perception change a useful determinant? *Psycho-Oncology, 25*(6), 663-669. doi:10.1002/pon.3993

Granger, K. (2014, April 25). Having cancer is not a fight or a battle. *The Guardian*. Retrieved from https://www.theguardian.com/society/2014/apr/25/having-cancer-not-fight-or-battle

Grant, K. E., & Compas, B. E. (1995). Stress and axious-depressed symptoms among adolescents: Searching for mechanisms of risk. *Journal of Consulting and Clinical Psychology, 63*(6), 1015-1021. doi:10.1037//0022-006x.63.6.1015

Gray, S. E., & Rutter, D. R. (2007). Illness representations in young people with Chronic Fatigue Syndrome. *Psychology and Health, 22*(2), 1-16. doi:10.1080/14768320600774595

Greco, L. A., Baer, R. A., & Smith, G. T. (2011). Assessing mindfulness in children and adolescents: Development and validation of the Child and Adolescent Mindfulness Measure (CAMM). *Psychological Assessment*, *23*(3), 606-614. doi:10.1037/a0022819

Greco, L. A., Lambert, W., & Baer, R. A. (2008). Psychological inflexibility in childhood and adolescence: Development and evaluation of the Avoidance and Fusion Questionnaire for Youth. *Psychological Assessment, 20*(2), 93-102. doi:10.1037/1040-3590.20.2.93

Green, J., & Thorogood, N. (2018). *Qualitative Methods for Health Research*. London: SAGE Publications.

Hagger, M. S., Koch, S., Chatzisarantis, N. L. D., & Orbell, S. (2017). The common sense model of self-regulation: Meta-analysis and test of a process model. *Psychological Bulletin, 143*(11), 1117-1154. doi:10.1037/bul0000118

Hagger, M. S., & Orbell, S. (2003). A meta-analytic review of the Common-Sense Model of Illness Representations. *Psychology and Health, 18*(2), 141-184. doi:10.1080/088704403100081321

Haigh, C., & Witham, G. (2015). Distress protocol for qualitative data collection. In: Manchester Metropolitan University.

Hale, E. D., Treharne, G. J., & Kitas, G. D. (2007). The Common-Sense Model of self-regulation of health and illness: How can we use it to understand and respond to our patients' needs? *Rheumatology, 46*(6), 904-906. doi:10.1093/rheumatology/kem060

Hansell, S., & Mechanic, D. (1985). Introspectiveness and adolescent symptom reporting. *Journal of Human Stress, 11*(4), 165-176. doi:10.1080/0097840X.1985.9936755

Hansell, S., & Mechanic, D. (1986). The Socialization of Introspection and Illness Behaviour. In S. McHugh & T. M. Vallis (Eds.), *Illness Behavior*. Boston, MA: Springer.

Harris, C. A., & Zakowski, S. G. (2003). Comparisons of distress in adolescents of cancer patients and controls. *Psycho-Oncology, 12*(2), 173-182. doi:10.1002/pon.631

Hasson-Ohayon, I., & Braun, M. (2011). Being a parent and coping with cancer: Intervention development. *Palliative and Supportive Care, 9*(2), 149-152. doi:10.1017/S1478951511000174

Hawkins, Y., Ussher, J., Gilbert, E., Perz, J., Sandoval, M., & Sundquist, K. (2009). Changes in sexuality and intimacy after the diagnosis and treatment of cancer: The experience of partners in a sexual relationship with a person with cancer. *Cancer Nursing, 32*(4), 271-280. doi:10.1097/NCC.0b013e31819b5a93

Hayes, S. C., Luoma, J. B., Bond, F. W., Masuda, A., & Lillis, J. (2006). Acceptance and commitment therapy: Model, processes and outcomes. *Behaviour Research and Therapy, 44*(1), 1-25. doi:10.1016/j.brat.2005.06.006

Hayes, S. C., Strosahl, K. D., & Wilson, K. G. (2011). *Acceptance and commitment therapy: The process and practice of mindful change*. New York, NY: The Guilford Press.

Hayes, S. C., Strosahl, K. D., Wilson, K. G., Bissett, R. T., Pistorello, J., Toarmino, D., . . . McCurry, S. M. (2004). Measuring experiential avoidance: A preliminary test of a working model. *The Psychological Record, 54*, 553-578. doi:10.1007/BF03395492

Heffernan, E., Coulson, N. S., Henshaw, H., Barry, J. G., & Ferguson, M. A. (2016). Understanding the psychosocial experiences of adults with mild-moderate hearing loss: An application of Leventhal's self-regulatory model. *International Journal of Audiology, 55*(Suppl 3), S3-S12. doi:10.3109/14992027.2015.1117663

Heiney, S. P., Bryant, L. H., Walker, S., Parrish, R. S., Provenzano, F. J., & Kelly, K. E. (1997). Impact of parental anxiety on child emotional adjustment when a parent has cancer. *Oncology Nursing Forum, 24*(4), 655-661.

Helms, R. L., O'Hea, E. L., & Corso, M. (2008). Body image issues in women with breast cancer. *Psychology, Health & Medicine, 13*(3). doi:10.1080/13548500701405509

Henoch, I., & Danielson, E. (2008). Existential concerns among patients with cancer and interventions to meet them: an integrative literature review. *Psycho-Oncology, 18*(3), 225-236. doi:10.1002/pon.1424

Henry, J. D., & Crawford, J. R. (2005). The short-form version of the Depression Anxiety Stress Scales (DASS-21): Construct validity and normative data in a large non-clinical sample. *British Journal of Clinical Psychology, 44*(2), 227-239. doi:10.1348/014466505X29657

Heyduck-Weides, K., Bengel, J., Farin, E., & Glattacker, M. (2019). Measuring illness perceptions in the family context: Psychometric properties of the IPQ-R adapted for adolescent-caregiver dyads. *Psychology & Health, 34*(1), 1-23. doi:10.1080/08870446.2018.1494830

Hilton, B. A., & Elfert, H. (1996). Children's experiences with mothers' early breast cancer. *Cancer Practice, 4*(2), 96-104.

Hodges, L. J., Humphris, G. M., & Macfarlane, G. (2005). A meta-analytic investigation of the relationship between the psychological distress of cancer patients and their carers. *Social Science & Medicine, 60*(1), 1-12. doi:10.1016/j.socscimed.2004.04.018

Hoke, L. A. (2001). Psychosocial adjustment in children of mothers with breast cancer. *Psycho-Oncology, 10*(5), 361-369. doi:10.1002/pon.515

Horowitz, M., Wilner, N., & Alvarez, W. (1979). Impact of Event Scale: a measure of subjective stress. *Psychosomatic Medicine, 41*(3), 209-218.

Hoving, J. L., van der Meer, M., Volkova, A. Y., & Frings-Dresen, M. H. W. (2010). Illness preceptions and work participation: A systematic review. *International Archives of Occupational and Environmental Health, 83*(6), 595-605. doi:10.1007/s00420-010-0506-6

Howlader, N., Noone, A. M., Krapcho, M., Miller, D., Brest, A., Yu, M., . . . Cronin, K. A. (2019). *SEER Cancer Statistics Review, 1975-2016*. Retrieved from Bethesda, MD: https://seer.cancer.gov/csr/1975_2016/

Huizinga, G. A., van der Graaf, W. T., Visser, A., Dijkstra, J. S., & Hoekstra-Weebers, J. E. (2003). Psychosocial consequences for children of a parent with cancer: A pilot study. *Cancer Nursing, 26*(3), 195-202. doi:10.1097/00002820-200306000-00004

Huizinga, G. A., Visser, A., van der Graaf, W. T., Hoekstra, H. J., Gazendam-Donofrio, S. M., & Hoekstra-Weebers, J. E. (2010). Stress response symptoms in adolescents during the first year after a parent's cancer diagnosis. *Supportive Care in Cancer, 18*(11), 1421-1428. doi:10.1007/s00520-009-0764-6

Huizinga, G. A., Visser, A., van der Graaf, W. T., Hoekstra, H. J., & Hoekstra-Weebers, J. E. (2005). The quality of communication between parents and adolescent children in the case of parental cancer. *Annals of Oncology, 16*(12), 1956-1961. doi:10.1093/annonc/mdi395

Huizinga, G. A., Visser, A., van der Graaf, W. T., Hoekstra, H. J., Klip, E. C., Pras, E., & Hoekstra-Weebers, J. E. (2005). Stress response symptoms in adolescent and young adult children of parents diagnosed with cancer. *European Journal of Cancer, 41*(2), 288-295. doi:10.1016/j.ejca.2004.10.005

Huizinga, G. A., Visser, A., van der Graaf, W. T., Hoekstra, H. J., Stewart, R. E., & Hoekstra-Weebers, J. E. (2011). Family-oriented multilevel study on the psychological functioning of adolescent children having a mother with cancer. *Psycho-Oncology, 20*(7), 730-737. doi:10.1002/pon.1779

Huizinga, G. A., Visser, A., Zelders-Steyn, Y. E., Teule, J. A., Reijneveld, S. A., & Roodbol, P. F. (2011). Psychological impact of having a parent with cancer. *European Journal of Cancer, 47*(Suppl 3), 239-246. doi:10.1016/S0959-8049(11)70170-8

Hulbert-Williams, N. J., Morrison, V., Wilkinson, C., & Neal, R. D. (2013). Investigating the cognitive precursors of emotional response to cancer stress: Re-testing Lazarus's transactional model. *British Journal of Health Psychology, 18*(1), 97-121. doi:10.1111/j.2044-8287.2012.02082.x

Huston, S. A., & Houk, C. P. (2011). Common sense model of illness in youth with type 1 diabetes or sickle cell disease. *Journal of Pediatric Pharmacology and Therapeutics, 16*(4), 270-280. doi:10.5863/1551-6776-16.4.270

Hymovich, D. P. (1993). Child-rearing concerns of parents with cancer. *Oncology Nursing Forum, 20*(9), 1355-1360.

Inbar, C., Ety, B., Ayala, H., & Tamer, P. (2013). The mental health of breast cancer survivors and their adolescent daughters. *Psycho-Oncology, 22*(6), 1236-1241. doi:10.1002/pon.3127

Inhestern, L., Bultmann, J. C., Beierlein, V., Moller, B., Romer, G., Muriel, A. C., . . . Bergelt, C. (2016). Psychometric properties of the Parenting Concerns Questionnaire in cancer survivors with minor and young adult children. *Psycho-Oncology, 25*(9), 1092-1098. doi:10.1002/pon.4049

Iragorri, N., & Spackman, E. (2018). Assessing the value of screening tools: reviewing the challenges and opportunities of cost-effectiveness analysis. *Public Health Reviews, 39*(17). doi:10.1186/s40985-018-0093-8

James, D. (2018, May 18). It's time to stop seeing cancer as an enemy we can wage war against - you can't fight a battle that's not fair. *The Sun*. Retrieved from https://www.thesun.co.uk/fabulous/6317663/cancer-isnt-fight-battle-deborah-james/

Jeppesen, E., Bjelland, I., Fosså, S. D., Loge, J. H., & Dahl, A. A. (2016). Health-related quality of life in teenagers with a parent with cancer. *European Journal of Oncology Nursing, 22*, 46-53. doi:10.1016/j.ejon.2016.03.004

Johnson, M. D., Anderson, J. R., Wilcox, A., Walker, A., Robbins, D. C., & Lewis, V. L. (2013). Relationship beliefs and illness appraisals are differentially associated with specific ways of coping in spouses of patients with type 2 diabetes. *Psychology & Health, 28*(12), 1442-1459. doi:10.1080/08870446.2013.822499

Joice, S. (2012). Self-management following stroke. *Nursing Standard, 26*(22), 39-46. doi:10.7748/ns2012.02.26.22.39.c8919

Jones, C. J., Smith, H. E., & Llewellyn, C. D. (2016). A systematic review of the effectiveness of interventions using the Common Sense Self-Regulatory Model to improve adherence behaviours. *Journal of Health Psychology, 21*(11), 2709-2724. doi:10.1177/1359105315583372

Jootun, D., McGhee, G., & Marland, G. R. (2009). Reflexivity: promoting rigour in qualitative research. *Nursing Standard (through 2013), 23*(23), 42-46.

Jurkovic, G. J. (1997). *Lost Childhoods: The Plight of the Parentified Child*. Philadelphia, PA: Brunner/Mazel.

Karademas, E. C., Dimitraki, G., Thomadakis, C., & Giannousi, Z. (2019). The relation of spouse illness representations to patient representations and coping behaviour: A study in couples dealing with a newly diagnosed cancer. *Journal of Psychosocial Oncology, 37*(2), 145-159. doi:10.1080/07347332.2018.1508534

Karademas, E. C., Zarogiannos, A., & Karamvakalis, N. (2010). Cardiac patient-spouse dissimilarities in illness perception: Associations with patient self-rated health and coping strategies. *Psychology and Health, 25*(4), 451-463. doi:10.1080/08870440802652089

Karekla, M., Karademas, E. C., & Gloster, A. T. (2019). The Common Sense Model of Self-Regulation and Acceptance and Commitment Therapy: Integrating strategies to guide interventions for chronic illness. *Health Psychology Review, 13*(4), 490-503. doi:10.1080/17437199.2018.1437550

Karlsson, E., Andersson, K., & Ahlstrom, B. H. (2013). Loneliness despite the presence of others - Adolescents' experiences of having a parent who becomes ill with cancer. *European Journal of Oncology Nursing, 17*(6), 697-703. doi:10.1016/j.ejon.2013.09.005

Karnilowicz, W. (2011). Identity and psychological ownership in chronic illness and disease state. *European Journal of Cancer Care, 20*(2), 276-282. doi:10.1111/j.1365-2354.2010.01220.x

Kazak, A. E., McClure, K. S., Alderfer, M. A., Hwang, W. T., Crump, T. A., Le, L. T., . . . Rourke, M. T. (2004). Cancer-related parental beliefs: the Family Illness Beliefs Inventory (FIBI). *Journal of Pediatric Psychology, 29*(7), 531-542. doi:10.1093/jpepsy/jsh055

Kemp, S., Morley, S., & Anderson, E. (1999). Coping with epilepsy: Do illness representations play a role? *British Journal of Clinical Psychology, 38*(1), 43-58. doi:10.1348/014466599162656

Kennedy, V. L., & Lloyd-Williams, M. (2009). How children cope when a parent has advanced cancer. *Psycho-Oncology, 18*(8), 886-892. doi:10.1002/pon.1455

Kessler, R. C., Andrews, G., Colpe, L. J., Hiripi, E., Mroczek, D. K., Normand, S. L., . . . Zaslavsky, A. M. (2002). Short screening scales to monitor population prevalences and trends in non-specific psychological distress. *Psychological Medicine, 32*(6), 959-976.

Kessler, R. C., Barker, P. R., Colpe, L. J., Epstein, J. F., Gfroerer, J. C., Hiripi, E., . . . Zaslavsky, A. M. (2003). Screening for serious mental illness in the general population. *Archives of General Psychiatry, 60*(2), 184-189.

Kilmer, R. P., Gil-Rivas, V., Tedeschi, R. G., Cann, A., Calhoun, L. G., Buchanan, T., & Taku, K. (2010). Use of the Revised Post-traumatic Growth Inventory for Children. *Journal of Traumatic Stress, 22*(3), 248-253. doi:10.1002/jts.20410

Kimberlin, C. L., & Winterstein, A. G. (2008). Validity and reliability of measurement instruments used in research. *American Journal of Health-System Pharmacy, 65*(23), 2276-2284. doi:10.2146/ajhp070364

Kissil, K., Nino, A., Jacobs, S., Davey, M., & Tubbs, C. Y. (2010). "It has been a good growing experience for me": Growth experiences among African American youth coping with parental cancer. *Families, Systems, & Health, 28*(3), 274-289. doi:10.1037/a0020001

Klein Woolthuis, E. P., de Grauw, W. J. C., Cardol, M., van Weel, C., Metsemakers, J. F. M., & Biermans, M. C. J. (2013). Patients' and partners' illness perceptions in screen-detected versus clinically diagnosed type 2 diabetes: Partners matter! *Family Practice, 30*(4), 418-425. doi:10.1093/fampra/cmt003

Konings, S., McDonald, F. E., & Patterson, P. (2020). Supporting parents impacted by cancer: Development of an informational booklet for parents with cancer who have adolescent and young adult children. *Psycho-Oncology, Advance online publication*. doi:10.1002/pon.5475

Koo, T. K., & Li, M. Y. (2016). A guideline of selecting and reporting intraclass correlation coefficients for reliability research. *Journal of Chiropractic Medicine, 15*(2), 155-163. doi:10.1016/j.jcm.2016.02.012

Krattenmacher, T., Kuhne, F., Ernst, J., Bergelt, C., Romer, G., & Moller, B. (2012). Parental cancer: factors associated with children's psychosocial adjustment - a systematic review. *Journal of Psychosomatic Research, 72*(5), 344-356. doi:10.1016/j.jpsychores.2012.01.011

Krattenmacher, T., Kuhne, F., Fuhrer, D., Beierlein, V., Brahler, E., Resch, F., . . . Moller, B. (2013). Coping skills and mental health status in adolescents when a parent has cancer: a multicenter and multi-perspective study. *Journal of Psychosomatic Research, 74*(3), 252-259. doi:10.1016/j.jpsychores.2012.10.003

Krauel, K., Simon, A., Krause-Hebecker, N., Czimbalmos, A., Bottomley, A., & Flechtner, H. (2012). When a parent has cancer: Challenges to patients, their families and health providers. *Expert Review of Pharmacoeconomics and Outcomes Research, 12*(6), 795-808. doi:10.1586/erp.12.62

Kraus, C., Kadriu, B., Lanzenberger, R., Zarate Jr., C. A., & Kasper, S. (2019). Prognosis and improved outcomes in major depression: A review. *Translational Psychiatry, 9*(127). doi:10.1038/s41398-019-0460-3

Kristjanson, L. J., Chalmers, K. I., & Woodgate, R. (2004). Information and support needs of adolescent children of women with breast cancer. *Oncology Nursing Forum, 31*(1), 111-119. doi:10.1188/04.ONF.111-119

Küçükoğlu, S., & Çelebioğlu, A. (2013). Identification of psychological symptoms and associated factors in adolescents who have a parent with cancer in Turkey. *European Journal of Oncology Nursing, 17*(1), P75-80. doi:10.1016/j.ejon.2011.10.008

Kühne, F., Krattenmacher, T., Bergelt, C., Ernst, J., Flechtner, H., Führer, D., . . . Möller, B. (2012). Parental palliative cancer: Psychosocial adjustment and health-related quality of life in adolescents participating in a German family counselling service. *BMC Palliative Care, 11*(21). doi:10.1186/1472-684X-11-21

Kuipers, E., Watson, P., Onwumere, J., Bebbington, P., Dunn, G., Weinman, J., . . . Garety, P. (2007). Discrepant illness perceptions, affect and expressed emotion in people with psychosis and their carers. *Social Psychiatry and Psychiatric Epidemiology, 42*(277-83). doi:10.1007/s00127-007-0165-4

Lau, R. R., & Hartman, K. A. (1983). Common sense representations of common illnesses. *Health Psychology, 2*(2), 167-185. doi:10.1037/0278-6133.2.2.167

Lavigne, J. V., Feldman, M., & Meyers, K. M. (2016). Screening for mental health problems: Addressing the base rate fallacy for a sustainable screening program in integrated primary care. *Journal of Pediatric Psychology, 41*(10), 1081-1090. doi:10.1093/jpepsy/jsw048

Lazarus, R. S. (1991). *Emotion and adaptation*. Oxford, UK: Oxford University Press.

Lazarus, R. S. (1993). From psychological stress to the emotions: A history of changing outlooks. *Annual Review of Psychology, 44*, 1-21. doi:10.1146/annurev.ps.44.020193.000245

Lazarus, R. S. (1999). *Stress and emotion: A new synthesis*. New York, NY: Springer.

Lazarus, R. S., & Folkman, S. (1984). *Stress, appraisal, and coping*. New York, NY, US: Springer.

Leventhal, H., Brissette, I., & Leventhal, E. A. (2003). The common-sense model of self-regulation of health and illness. In L. D. Cameron & H. Leventhal (Eds.), *The Self-Regulation of Health and Illness Behaviour* (pp. 42-65). London: Routledge.

Leventhal, H., Diefenbach, M. A., & Leventhal, E. A. (1992). Illness cognition: using common sense to understand treatment adherence and affect cognition interactions. *Cognitive Therapy and Research, 16*(2), 143-163. doi:10.1007/BF01173486

Leventhal, H., Meyer, D., & Nerenz, D. (1980). The common sense representation of illness danger. In S. Rachman (Ed.), *Contributions to medical psychology* (Vol. II, pp. 7-30). New York: Pergamon Press.

Leventhal, H., & Nerenz, D. (1985). The assessment of illness cognition. In P. Karoly (Ed.), *Measurement Strategies in Health Psychology* (pp. 517-555). New York: Wiley.

Leventhal, H., & Niles, P. (1965). Persistence of influence for varying durations of exposure to threat stimuli. *Psychological Reports, 16*(1), 223-233. doi:10.2466/pr0.1965.16.1.223

Leventhal, H., Phillips, L. A., & Burns, E. (2016a). The Common-Sense Model of Self-Regulation (CSM): A dynamic framework for understanding illness self-management. *Journal of Behavioral Medicine, 39*(6), 935-946. doi:10.1007/s10865-016-9782-2

Leventhal, H., Phillips, L. A., & Burns, E. (2016b). Modelling management of chronic illness in everyday life: A common-sense approach. *Psychological Topics, 25*(1), 1-18.

Levesque, J. V., & Maybery, D. (2012). Parental cancer: Catalyst for positive growth and change. *Qualitative Health Research, 22*(3), 397-408. doi:10.1177/1049732311421617

Lewis, F. M., Ellison, E. S., & Woods, N. F. (1985). The impact of breast cancer on the family. *Seminars in Oncology Nursing, 1*(3), 206-213. doi:10.1016/S0749-2081(85)80010-3

Li, Q. P., Mak, Y. W., & Loke, A. Y. (2012). Spouses' experience of caregiving for cancer patients: A literature review. *International Nursing Review, 60*(2), 178-187. doi:10.1111/inr.12000

Lindqvist, B., Schmitt, F., Santalahti, P., Romer, G., & Piha, J. (2007). Factors associated with the mental health of adolescents when a parent has cancer. *Scandinavian Journal of Psychology, 48*(4), 345-351. doi:10.1111/j.1467-9450.2007.00573.x

Liu, J., Chen, X., & Lewis, G. (2017). Childhood internalizing behaviour: Analysis and implications. *Journal of Psychiatric Mental Health Nursing, 18*(10), 884-894. doi:10.1111/j.1365-2850.2011.01743.x

Loewy, R. L., Bearden, C. E., Johnson, J. K., Raine, A., & Cannon, T. D. (2005). The prodromal questionnaire (PQ): Preliminary validation of a self-report screening measure for prodromal and psychotic syndromes. *Schizophrenia Research, 79*(1), 117-125. doi:10.1016/j.schres.2005.03.007

Maddams, J., Utley, M., & Moller, H. (2012). Projections of cancer prevalence in the United Kingdom, 2010-2040. *British Journal of Cancer, 107*(7), 1195-1202. doi:10.1038/bjc.2012.366

Malla, A., Iyer, S., McGorry, P. D., Cannon, M., Coughlan, H., Singh, S., . . . Joober, R. (2016). From early intervention in psychosis to youth mental health reform: A review of the evolution and transformation of mental health services for young people. *Social Psychiatry and Psychiatric Epidemiology, 51*(3), 319-326. doi:10.1007/s00127-015-1165-4

Manne, S. L., Kashy, D. A., Virtue, S., Criswell, K. R., Kissane, D., Ozga, M., . . . Rodriguez, L. (2018). Acceptance, social support, benefit-finding, and depression in women with gynecological cancer. *Quality of Life Research, 27,* 2991-3002. doi:10.1007/s11136-018-1953-x

Manstead, A. S. R. (2018). The psychology of social class: How socioeconomic status impacts thought, feelings, and behaviour. *British Journal of Social Psychology, 57,* 267-291. doi:10.1111/bjso.12251

Martin-Cook, K., Remakel-Davis, B., Svetlik, D., Hynan, L. S., & Weiner, M. F. (2003). Caregiver attribution and resentment in demetia care. *American Journal of Alzheimer's Disease and Other Demetias, 18*(6), 366-374. doi:10.1177/153331750301800606

Martin, M. S., Potter, B. K., Crocker, A. G., Wells, G. A., & Colman, I. (2016). Yield and efficiency of mental health screening: A comparison of screening protocols at intake to prison. *PLoS One, 11*(5), e0154106. doi:10.1371/journal.pone.0154106

Matthews, A., Lenz, K. R., Peugh, J., Copps, E. C., & Peterson, C. M. (2018). Caregiver burden and illness perceptions in caregivers of medically hospitalized youth with anorexia nervosa. *Eating Behaviors, 29*, 14-18. doi:10.1016/j.eatbeh.2018.01.003

Maxim, L. D., Niebo, R., & Utell, M. J. (2014). Screening tests: A review with examples. *Inhalation Toxicology, 26*(13), 811-828. doi:10.3109/08958378.2014.955932

Maynard, A., Patterson, P., McDonald, F. E., & Stevens, G. (2013). What is helpful to adolescents who have a parent diagnosed with cancer? *Journal of Psychosocial Oncology, 31*(6), 675-697. doi:10.1080/07347332.2013.835021

Mazefsky, C. A., Kao, J., & Oswald, D. P. (2011). Preliminary evidence suggesting caution in the use of psychiatric self-report measures with adolescents with high-functioning autism spectrum disorders. *Research in Autism Spectrum Disorders, 5*(1), 164-174. doi:10.1016/j.rasd.2010.03.006

McAndrew, L. M., Musumeci-Szabó, T. J., Mora, P. A., Vileikyte, L., Burns, E., Halm, E. A., . . . Leventhal, H. (2010). Using the common sense model to design interventions for the prevention and management of chronic illness threats: From description to process. *British Journal of Health Psychology, 13*(2), 195-204. doi:10.1348/135910708X295604

McCorry, N. K., Dempster, M., Quinn, J., Hogg, A., Newell, J., Moore, M., . . . Kirk, S. J. (2013). Illness perception clusters at diagnosis predict psychological distress among women with breast cancer at 6 months post diagnosis. *Psycho-Oncology, 22*(3), 692-698. doi:10.1002/pon.3054

McCorry, N. K., Scullion, L., McMurray, C. M., Houghton, R., & Dempster, M. (2013). Content validity of the illness perceptions questionnaire - revised among people with type 2 diabetes: A think-aloud study. *Psychology & Health, 28*(6), 675-685. doi:10.1080/08870446.2012.746690

McDonald, F. E., Patterson, P., White, K. J., Butow, P. N., Costa, D. S., & Kerridge, I. (2016). Correlates of unmet needs and psychological distress in adolescent and young adults who have a parent diagnosed with cancer. *Psycho-Oncology, 25*(4), 447-454. doi:10.1002/pon.3942

McGorry, P. D., Hickie, I. B., Yung, A. R., Pantelis, C., & Jackson, H. J. (2006). Clinical staging of psychiatric disorders: A heuristic framework for choosing earlier, safer and more effective interventions. *Australiam and New Zealand Journal of Psychiatry, 40*(8), 616-622. doi:10.1080/j.1440-1614.2006.01860.x

McGorry, P. D., & Mei, C. (2018). Early intervention in youth mental health: Progress and future directions. *Evidence-Based Mental Health, 21*(4), 182-184. doi:10.1136/ebmental-2018-300060

Mechanic, D. (1983). Adolescent health and illness behavior: Review of the literature and a new hypothesis for the study of stress. *Journal of Human Stress, 9*(2). doi:10.1080/0097840X.1983.9936119

Mechanic, D. (1986). The concept of illness behaviour: Culture, situation and personal predisposition. *Psychological Medicine, 16*(1), 1-7. doi:10.1017/S0033291700002476

Meeus, W. (2016). Adolescent psychosocial development: A review of longitudinal models and research. *Developmental Psychology, 52*(12), 1969-1993. doi:10.1037/dev0000243

Mehnert, A. (2011). Employment and work-related issues in cancer survivors. *Critical Reviews in Oncology/Hematology, 77*(2), 109-130. doi:10.1016/j.critrevonc.2010.01.004

Metcalf, C. A., Arch, J. J., & Greer, J. A. (2017). Anxiety and its correlates among young adults with a history of parental cancer. *Journal of Psychosocial Oncology, 35*(5), 597-613. doi:10.1080/07347332.2017.1307895

Meyer, D., Leventhal, H., & Gutmann, M. (1985). Common-sense models of illness: The example of hypertension. *Health Psychology, 4*(2), 115-135.

Mi, E., Mi, E., Ewing, G., Mahadeva, R., Gardener, A. C., Butcher, H. H., . . . Farquhar, M. (2017). Associations between the psychological health of patients and carers in advanced COPD. *International Journal of Chronic Obstructive Pulmonary Disease, 12*, 2813-2821. doi:10.2147/COPD.S139188

Miceli, J., Geller, D., Tsung, A., Hecht, C. L., Wang, Y., Pathak, R., . . . Steel, J. (2019). Illness perceptions and perceived stress in patients with advanced gastrointestinal cancer. *Psycho-Oncology, 28*(7), 1513-1519. doi:10.1002/pon.5108

Miller, J. M. (2008). *Parental cancer: The parent-child bond at risk.* (Unpublished doctoral dissertation), University of North Carolina, Greensboro.

Miller, R. S. (2010). Speak Up: 8 words and phrases to ban in oncology! *Oncology Times, 32*(12), 20. doi:10.1097/01.COT.0000383777.50536.b2

Mishel, M. H. (1981). The measurement of uncertainty in illness. *Nursing Research, 30*(5), 258-263. doi:10.1097/00006199-198109000-00002

Mishel, M. H. (1983). Parents' perception of uncertainty concerning their hospitalized child. *Nursing Research, 32*(6), 324-330. doi:10.1097/00006199-198311000-00002

Mitchell, A. J., Ferguson, D. W., Gill, J., Paul, J., & Symonds, P. (2013). Depression and anxiety in long-term cancer survivors compared with spouses and healthy controls: a systematic review and meta-analysis. *The Lancet, 14*(8), 721-732. doi:10.1016/S1470-2045(13)70244-4

Mokkink, L. B., de Vet, H. C. W., Prinsen, C. A. C., Patrick, D. L., Alonso, J., Bouter, L. M., & Terwee, C. B. (2018). COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome Measures. *Quality of Life Research, 27*(5), 1171-1179. doi:10.1007/s11136-017-1765-4

Mokkink, L. B., Prinsen, C. A. C., Bouter, L. M., de Vet, H. C. W., & Terwee, C. B. (2016). The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) and how to select an outcome measurement instrument. *Brazillian Journal of Physical Therapy, 20*(2), 105-113. doi:10.1590/bjpt-rbf.2014.0143

Mokkink, L. B., Terwee, C. B., Patrick, D. L., Alonso, J., Stratford, P. W., Knol, D. L., . . . de Vet, H. C. W. (2010). The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement instruments: an international Delphi study. *Quality of Life Research, 19*, 539-549. doi:10.1007/s11136-010-9606-8

Moller, B., Barkmann, C., Krattenmacher, T., Kuhne, F., Bergelt, C., Beierlein, V., . . . Romer, G. (2014). Children of cancer patients: Prevalence and predictors of emotional and behavioral problems. *Cancer, 120*(15), 2361-2370. doi:10.1002/cncr.28644

Molzon, E. S., Brannon, E. E., Fedele, D. A., Grant, D. M., Suorsa, K. I., & Mullins, L. L. (2014). Factor structure of the parent perception of uncertainty scale in parents of children with cancer. *Psycho-Oncology, 23*(12), 1435-1438. doi:10.1002/pon.3574

Moore, C. W., Rauch, P. K., Baer, L., Pirl, W. F., & Muriel, A. C. (2015). Parenting changes in adults with cancer. *Cancer, 121*(19), 3551-3557. doi:10.1002/cncr.29525

Moore, C. W., Shea, S., Russell, K., Convery, M. S., & Rauch, P. K. (2018). Communicating with children about parental cancer: The devil in the details. *Journal of Clinical Oncology, 36*(7). doi:10.1200/JCO.2018.36.7_suppl.81

Morris, J. N., Martini, A., & Preen, D. (2016). The well-being of children impacted by a parent with cancer: an integrative review. *Supportive Care in Cancer, 24*(7), 3235-3251. doi:10.1007/s00520-016-3214-2

Morris, J. N., Turnbull, D., Martini, A., Preen, D., & Zajac, I. (2020). Coping and its relationship to post-traumatic growth, emotion, and resilience among adolescents and young adults impacted by parental cancer. *Journal of Psychosocial Oncology, 38*(1), 73-88. doi:10.1080/07347332.2019.1637384

Morris, J. N., Turnbull, D., Preen, D., Zajac, I., & Martini, A. (2018). The psychological, social, and behavioural impact of a parent's cancer on adolescent and young adult offspring aged 10-24 at time of diagnosis: A systematic review. *Journal of Adolescence, 65*, 61-71. doi:10.1016/j.adolescence.2018.03.001

Moss-Morris, R., & Paterson, J. (1995). Understanding children's concepts of health and illness: Implications for developmental therapists. *Physical and Occupational Therapy in Pediatrics, 14*(3-4), 95-108. doi:10.1080/J006v14n03_05

Moss-Morris, R., Petrie, K. J., & Weinman, J. (1996). Functioning in chronic fatigue syndrome: Do illness perceptions play a regulatory role? *British Journal of Health Psychology, 1*(1), 15-25. doi:10.1111/j.2044-8287.1996.tb00488.x

Moss-Morris, R., Weinman, J., Petrie, K. J., Horne, R., Cameron, L. D., & Buick, D. (2002). The revised Illness Perception Questionnaire (IPQ-R). *Psychology and Health, 17*(1), 1-16. doi:10.1080/08870440290001494

Muriel, A. C., Moore, C. W., Baer, L., Park, E. R., Kornblith, A. B., Pirl, W., . . . Rauch, P. K. (2012). Measuring psychosocial distress and parenting concerns among adults with cancer: the Parenting Concerns Questionnaire. *Cancer, 118*(22), 567108. doi:10.1002/cncr.27572

Murray, S. A., Kendall, M., Boyd, K., Grant, L., Highet, G., & Sheikh, A. (2010). Archetypal trajectories of social, psychological, and spiritual well-being and distress in family caregivers of patients with lung cancer: secondary analysis of serial qualitative interviews. *BMJ, 340*. doi:10.1136/bmj.c2581

National Health and Medical Research Council. (2007, updated 2018). *National Statement on Ethical Conduct in Human Research*. Retrieved from Canberra:

Nelson, E., Sloper, P., Charlton, A., & While, D. (1994). Children who have a parent with cancer: A pilot study. *Journal of Cancer Education, 9*(1), 30-36. doi:10.1080/08858199409528262

Nelson, E., & While, D. (2002). Children's adjustment during the first year of a parent's cancer diagnosis. *Journal of Psychosocial Oncology, 20*(1), 15-36. doi:10.1300/J077v20n01_02

Nelson, S., Slusar, M. B., Albert, J. M., Liu, Y., & Riedy, C. A. (2016). Psychometric properties of a caregiver illness perception measure for caries in children under 6 years old. *Journal of Psychosomatic Research, 81*, 46-53. doi:10.1016/j.jpsychores.2016.01.002

Newton, B. J., Rothlingova, Z., Gutteridge, R., LeMarchand, K., & Raphael, J. H. (2011). No room for reflexivity? Critical reflections following a systematic review of qualitative research. *Journal of Health Psychology, 17*(6), 866-885. doi:10.1177/1359105311427615

Nowell, L. S., Norris, J. M., White, D. E., & Moules, N. J. (2017). Thematic analysis: Striving to meet the trustworthiness criteria. *International Journal of Qualitative Methods, 16*(1), 1-13. doi:10.1177/1609406917733847

Nozawa, K., Shimizu, C., Kakimoto, M., Mizota, Y., Yamamoto, S., Takahashi, Y., . . . Fujiwara, Y. (2013). Quantitative assessment of appearance changes and related distress in cancer patients. *Psycho-Oncology, 22*(9), 2140-2147. doi:10.1002/pon.3268

O'Neill, C., O'Neill, C. S., & Semple, C. (2020). Children navigating parental cancer: Outcomes of a psychosocial intervention. *Comprehensive Child and Adolescent Nursing, 43*(2), 111-127. doi:10.1080/24694193.2019.1582727

Osborn, T. (2007). The psychosocial impact of parental cancer on children and adolescents: a systematic review. *Psycho-Oncology, 16*(2), 101-126. doi:10.1002/pon.1113

Park, E. M., Deal, A. M., Check, D. K., Hanson, L. C., Reeder-Hayes, K. E., Mayer, D. K., . . . Rosenstein, D. L. (2016). Parenting concerns, quality of life, and psychological distress in patients with advanced cancer. *Psycho-Oncology, 25*(8), 942-948. doi:10.1002/pon.3935

Park, T., Reilly-Spong, M., & Gross, C. R. (2013). Mindfulness: A systematic review of instruments to measure an emergent patient-reported outcome (PRO). *Quality of Life Research, 22*(10), 2639-2659. doi:10.1007/s11136-013-0395-8

Patterson, P., McDonald, F. E., Butow, P. N., White, K. J., Costa, D. S., Pearce, A., & Bell, M. L. (2013). Psychometric evaluation of the Offspring Cancer Needs Instrument (OCNI): An instrument to assess the psychosocial unmet needs of young people who have a parent with cancer. *Supportive Care in Cancer, 21*(7), 1927-1938. doi:10.1007/s00520-013-1749-z

Patterson, P., McDonald, F. E., & Orchard, P. (2014). A new Australian online and phone mental health support service for young people living with cancer. *Australasian Psychiatry, 22*(2), 165-169. doi:10.1177/1039856213519144

Patterson, P., McDonald, F. E., White, K. J., Walczak, A., & Butow, P. N. (2017). Levels of unmet needs and distress amongst adolescents and young adults (AYAs) impacted by familial cancer. *Psycho-Oncology, 26*(9), 1285-1292. doi:10.1002/pon.4421

Patterson, P., Pearce, A., & Slawitschka, E. (2011). The initial development of an instrument to assess the psychosocial needs and unmet needs of young people who have a parent with cancer: piloting the Offspring Cancer Needs Instrument (OCNI). *Supportive Care in Cancer, 19*(8), 1165-1174. doi:10.1007/s00520-010-0933-7

Patton, G. C., Coffey, C., Romaniuk, H., Mackinnon, A., Carlin, J. B., Degenhardt, L., . . . Moran, P. (2014). The prognosis of common mental disorders in adolescents: A 14-year prospective cohort study. *The Lancet, 383*(9926), P1404-1411. doi:10.1016/S0140-6736(13)62116-9

Pederson, S., & Revenson, T. A. (2005). Parental illness, family functioning, and adolescent well-being: a family ecology framework to guide research. *Journal of Family Psychology, 19*(3), 404-419. doi:10.1037/0893-3200.19.3.404

Pelizza, L., Azzali, S., Paterlini, F., Garlassi, S., Scazza, I., Chiri, L. R., . . . Raballo, A. (2019). Screening for psychosis risk among help-seeking adolescents: Application of the Italian version of the 16-item prodromal questionnaire (iPQ-16) in child and adolescent neuropsychiatry services. *Early Intervention in Psychiatry, 13*(4), 752-760. doi:10.1111/eip.12554

Perrin, S., Meiser-Stedman, R., & Smith, P. (2005). The Children's Revised Impact of Event Scale (CRIES): Validity as a Screening Instrument for PTSD. *Behavioural and Cognitive Psychotherapy, 33*(4), 487-498. doi:10.1017/S1352465805002419

Peterson, C. H., Peterson, N. A., & Powell, K. G. (2017). Cognitive interviewing for item development: Validity evidence based on content and response processes. *Measurement and Evaluation in Counseling and Development, 50*(4), 217-223. doi:10.1080/07481756.2017.1339564

Petrie, K., Perry, K., Broadbent, E., & Weinman, J. (2012). A text message programme designed to modify patients' illness and treatment beliefs improves self-reported adherence to asthma preventer medication. *British Journal of Health Psychology, 17*(1), 74-84. doi:10.1111/j.2044-8287.2011.02033.x

Petrie, K., & Weinman, J. (2006). Why illness perceptions matter. *Clinical Medicine, 6*(6), 536-539. doi:10.7861/clinmedicine.6-6-536

Phillips, F., & Lewis, F. M. (2015). The adolescent's experience when a parent has advanced cancer: A qualitative inquiry. *Palliative Medicine, 29*(9), 851-858. doi:10.1177/0269216315578989

Piaget, J., & Inhelder, B. (1969). *The Psychology of the Child*. New York: Basic Books.

Prinsen, C. A., Mokkink, L. B., Bouter, L. M., Alonso, J., Patrick, D. L., de Vet, H. C., & Terwee, C. B. (2018). COSMIN guideline for systematic reviews of patient-reported outcome measures. *Quality of Life Research, 27*(5), 1147-1157. doi:10.1007/s11136-018-1798-3

Quine, L., & Pahl, J. (1991). Stress and coping in mothers caring for a child with severe learning difficulties: A test of Lazarus' transactional model of coping. *Journal of Community & Applied Social Psychology, 1*(1), 57-70. doi:10.1002/casp.2450010109

Quinn, C., Rees Jones, I., & Clare, L. (2017). Illness representations in caregivers of people with dementia. *Aging & Mental Health, 21*(5), 553-561. doi:10.1080/13607863.2015.1128882

Rainville, F., Dumont, S., Simard, S., & Savard, M.-H. (2012). Psychological distress among adolescents living with a parent with advanced cancer. *Journal of Psychosocial Oncology, 30*(5), 519-534. doi:10.1080/07347332.2012.703765

Rashi, C., Wittman, T., Tsimicalis, A., & Loiselle, C. G. (2015). Balancing illness and parental demands: Coping with cancer while raising minor children. *Oncology Nursing Forum, 42*(4), 337-344. doi:10.1188/15.ONF.337-344

Read, H., Roush, S., & Downing, D. (2018). Early intervention in mental health in adolescents and young adults: a systematic review. *The American Journal of Occupational Therapy, 72*(5), 7205190040p7205190041-7205190040p7205190048. doi:10.5014/ajot.2018.033118

Riaz, S., & Jones Nielsen, J. D. (2019). A pilot study of a text messaging intervention to modify illness and medication beliefs among patients diagnosed with inflammatory bowel disease. *Journal of Technology in Behavioral Science, 4*, 42-52. doi:10.1007/s41347-018-0083-1

Rice, F., Eyre, O., Riglin, L., & Potter, R. (2017). Adolescent depression and the treatment gap. *The Lancet, 4*(2), P86-87. doi:10.1016/S2215-0366(17)30004-4

Richardson, A., & Broadbent, E. (2017). Illness-Related Cognition. *Oxford Research Encyclopedia of Psychology*. Retrieved from https://oxfordre.com/psychology/view/10.1093/acrefore/9780190236557.001.0001/acrefore-9780190236557-e-120

Richardson, A., Morton, R., & Broadbent, E. (2015). Caregivers' illness perceptions contribute to quality of life in head and neck cancer patients at diagnosis. *Journal of Psychosocial Oncology, 33*(4), 414-432. doi:10.1080/07347332.2015.1046011

Richardson, A., Morton, R., & Broadbent, E. (2016a). Changes over time in head and neck cancer patients' and caregivers' illness perceptions and relationships with quality of life. *Psychology and Health, 31*(10), 1203-1219. doi:10.1080/08870446.2016.1203686

Richardson, A., Morton, R., & Broadbent, E. (2016b). Illness perceptions and coping predict post-traumatic stress in caregivers of patients with head and neck cancer. *Supportive Care in Cancer, 24*(10), 4443-4450. doi:10.1007/s00520-016-3285-0

Richardson, A., Tennant, G., Morton, R., & Broadbent, E. (2017). A self-regulatory intervention for patients with head and neck cancer: Pilot randomized trial. *Annals of Behavioral Medicine, 51*(5), 629-641. doi:10.1007/s12160-017-9885-1

Richardson, E., Schuz, N., Sanderson, K., Scott, J., & Schuz, B. (2017). Illness representations, coping, and illness outcomes in people with cancer: a systematic review and meta-analysis. *Psycho-Oncology, 26*(6), 724-737. doi:10.1002/pon.4213

Richardson, R., Trepel, D., Perry, A., Ali, S., Duffy, S., Gabe, R., . . . McMillan, D. (2015). Screening for psychological and mental health difficulties in young people who offend: a systematic review and decision model. *Health Technology Assessment, 19*(1), 1-128. doi:10.3310/hta19010

Ringlever, L., Otten, R., van Schayck, O. C. P., & Engels, R. C. M. E. (2012). The role of maternal illness perceptions in perceived asthma symptoms in school-aged children. *Journal of Asthma, 49*(10), 1030-1036. doi:10.3109/02770903.2012.726305

Rodriguez, L. (2019). Selective sharing: The impact of patterns of communication in young people experiencing maternal cancer. *Cancer Reports, 2*(1), e1138. doi:10.1002/cnr2.1138

Romeo, A., Di Tella, M., Ghiggia, A., Tesio, V., Gasparetto, E., Stanizzo, M. R., . . . Castelli, L. (2019). The traumatic experience of breast cancer: which factors can relate to the post-traumatic outcomes? *Frontiers in Psychology, 10*, 891. doi:10.3389/fpsyg.2019.00891

Rosenkoetter, U., & Tate, R. L. (2018). Assessing features of psychometric assessment instruments: A comparison of the COSMIN Checklist with other critical appraisal tools. *Brain Impairment, 19*(1), 103-118. doi:10.1017/BrImp.2017.29

Rosenstock, I. M. (1974). The Health Belief Model and preventive health behavior. *Health Education Monographs, 2*(4), 354-386. doi:10.1177/109019817400200405

Rosenstock, I. M., Hochbaum, G. M., & Leventhal, H. (1960). *The impact of Asian influenza on community life: A study in five cities* (DHEW Publication No. 766). Retrieved from Washington, DC:

Rosenstock, I. M., Strecher, V. J., & Becker, M. H. (1988). Social learning theory and the Health Belief Model. *Health Education Quarterly, 15*(2), 175-183. doi:10.1177/109019818801500203

Santacroce, S. (2001). Measuring parental uncertainty during the diagnosis phase of serious illness in a child. *Journal of Pediatric Nursing, 16*(1), 3-12. doi:10.1053/jpdn.2001.20547

Sawyer, S. M., Azzopardi, P. S., Wickremarathne, D., & Patton, G. C. (2018). The age of adolescence. *The Lancet Child and Adolescent Health, 2*(3), 223-228. doi:10.1016/S2352-4642(18)30022-1

Scerri, J., Saliba, T., Saliba, G., Scerri, C. A., & Camilleri, L. (2019a). Illness perceptions, depression and anxiety in informal carers of persons with depression: A cross-sectional survey. *Quality of Life Research, 28*, 451-460. doi:10.1007/s11136-018-2009-y

Scerri, J., Saliba, T., Saliba, G., Scerri, C. A., & Camilleri, L. (2019b). Illness perceptions, depression and anxiety in informal carers of persons with depression: a cross-sectional survey. *Quality of Life Research, 28*(2), 451-460. doi:10.1007/s11136-018-2009-y

Schiaffino, K. M., & Cea, C. D. (1995). Assessing chronic illness representations: the Implicit Models of Illness Questionnaire. *Journal of Behavioral Medicine, 18*(6), 531-548. doi:10.1007/BF01857894

Schiena, E., Hocking, A., Joubert, L., Wiseman, F., & Blaschke, S. (2019). An exploratory needs analysis of parents diagnosed with cancer. *Australian Social Work, 72*(3), 325-335. doi:10.1080/0312407X.2019.1577472

Schmitt, F., Piha, J., Helenius, H., Baldus, C., Kienbacher, C., Steck, B., . . . Romer, G. (2008). Multinational study of cancer patients and their children: Factors associated with family functioning. *Journal of Clinical Oncology, 26*(36), 5877-5883. doi:10.1200/JCO.2007.12.8132

Searle, A., Norman, P., Thompson, R., & Vedhara, K. (2007). Illness representations among patients with type 2 diabetes and their partners: Relationships with self-management behaviors. *Journal of Psychosomatic Research*, *63*(2), 175-184. doi:10.1016/j.jpsychores.2007.02.006

Semino, E. (2015). Whether you 'battle' cancer or experience a 'journey' is an individual choice. Retrieved from https://theconversation.com/whether-you-battle-cancer-or-experience-a-journey-is-an-individual-choice-39142

Semino, E., Demjen, Z., Demmen, J., Koller, V., Payne, S., Hardie, A., & Rayson, P. (2015). The online use of Violence and Journey metaphors by patients with cancer, as compared with health professionals: a mixed methods study. *BMJ Supportive and Palliative Care*, *7*(1), 60-66. doi:10.1136/bmjspcare-2014-000785

Semple, C., & McCance, T. (2010a). Experience of parents with head and neck cancer who are caring for young children. *Journal of Advanced Nursing*, *66*(6), 1280-1290. doi:10.1111/j.1365-2648.2010.05311.x

Semple, C., & McCance, T. (2010b). Parents' experience of cancer who have young children: A literature review. *Cancer Nursing*, *33*(2), 110-118. doi:10.1097/NCC.0b013e3181c024bb

Semple, C., & McCaughan, E. (2013). Family life when a parent is diagnosed with cancer: Impact of a psychosocial intervention for young children. *European Journal of Cancer Care*, *22*(2), 219-231. doi:10.1111/ecc.12018

Shakespeare-Finch, J., Obst, P., & Rogers, E. (2019). The influence of giving and receiving social support on the psychological outcomes of two trauma-exposed samples. *Journal of Loss and Trauma*, *24*(8), 766-781. doi:10.1080/15325024.2019.1652407

Shand, L. K., Brooker, J. E., Burney, S., Fletcher, J., & Ricciardelli, L. A. (2018). Psychosocial factors associated with posttraumatic stress and growth in Australian women with ovarian cancer. *Journal of Psychosocial Oncology*, *36*(4), 470-483. doi:10.1080/07347332.2018.1461728

Shands, M. E., Lewis, F. M., & Zahlis, E. H. (2000). Mother and child interactions about the mother's breast cancer: An interview study. *Oncology Nursing Forum*, *27*(1), 77-85.

Sheldrick, R. C., Benneyan, J. C., Kiss, I. G., Briggs-Gowan, M. J., Copeland, W., & Carter, A. S. (2015). Thresholds and accuracy in screening tools for early detection of psychopathology. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, *56*(9), 936-948. doi:10.1111/jcpp.12442

Siegel, K., Karus, D., & Raveis, V. H. (1996). Adjustment of children facing the death of a parent due to cancer. *Journal of the American Academy of Child and Adolescent Psychiatry*, *35*(4), 442-450. doi:10.1097/00004583-199604000-00010

Siemonsma, P. C., Stuive, I., Roorda, L. D., Vollebregt, J. A., Walker, M. F., Lankhorst, G. J., & Lettinga, A. T. (2013). Cognitive treatment of illness perceptions in patients with chronic low back pain: A randomized controlled trial. *Physical Therapy*, *93*(4), 435-448. doi:10.2522/ptj.20110150

Sigal, J. J., Perry, J. C., Robbins, J. M., Gagne, M.-A., & Nassif, E. (2003). Maternal preoccupation and parenting as predictors of emotional and behavioral problems in children of women with breast cancer. *Journal of Clinical Oncology*, *21*(6), 1155-1160. doi:10.1200/JCO.2003.03.031

Sim, L., & Matthews, A. (2012). The role of maternal illness perceptions in family functioning in adolescent girls with anorexia nervosa. *Journal of Child and Family Studies*, *22*, 541-550. doi:https://doi.org/10.1007/s10826-012-9607-z

Simons, G., Mason, A., Falahee, M., Kumar, K., Mallen, C. D., Raza, K., & Stack, R. J. (2017). Qualitative exploration of illness perceptions of rheumatoid arthritis in the general public. *Musculoskeletal Care*, *15*(1), 13-22. doi:10.1002/msc.1135

Singer, S. (2014). Psychosocial Impact of Cancer. In U. Goerling (Ed.), *Psycho-Oncology. Recent Results in Cancer Research* (Vol. 197). Berlin, Heidelberg: Springer.

Sint Nicolaas, S. M., Schepers, S. A., van den Bergh, E. M., Evers, A. W., Hoogerbrugge, P. M., Grootenhuis, M. A., & Verhaak, C. M. (2016). Illness cognitions and family adjustment: psychometric properties of the Illness Cognition Questionnaire for parents of a child with cancer. *Supportive Care in Cancer, 24*(2), 529-537. doi:10.1007/s00520-015-2795-5

Sloper, P. (2000). Predictors of distress in parents of children with cancer: a prospective study. *Journal of Pediatric Psychology, 25*(2), 79-91. doi:10.1093/jpepsy/25.2.79

Smith, P., Perrin, S., Dyregrov, A., & Yule, W. (2003). Principal components analysis of the impact of event scale with children in war. *Personality and Individual Differences, 34*(2), 315-322. doi:10.1016/S0191-8869(02)00047-8

Smith, T. F., Russell, H. F., Kelly, E. H., Mulcahey, M. J., Betz, R. R., & Vogel, L. C. (2013). Examination and measurement of coping among adolescents with spinal cord injury. *Spinal Cord, 51*(9), 710-714. doi:10.1038/sc.2013.65

Smits, N. (2010). A note on Youden's J and its cost ratio. *BMC Medical Research Methodology, 10*(89). doi:10.1186/1471-2288-10-89

Smout, M. F. (2019). The factor structure and predictive validity of the Kessler Psychological Distress Scale (K10) in children and adolescents. *Australian Psychologist, 54*(2), 102-113. doi:10.1111/ap.12376

Sodergren, S. C., Husson, O., Rohde, G. E., Tomaszewska, I. M., Vivat, B., Yarom, N., . . . on behalf of the European Organization for Research and Treatment of Cancer Quality of Life Group. (2018). A life put on pause: A exploration of the health-related quality of life issues relevant to adolescents and young adults with cancer. *Journal of Adolescent and Young Adult Oncology, 7*(4), 453-464. doi:10.1089/jayao.2017.0110

Spira, M., & Kenemore, E. (2000). Adolescent daughters of mothers with breast cancer: Impact of implications. *Clinical Social Work Journal, 28,* 183-195. doi:10.1023/A:1005106301713

Spirito, A. (1996). Commentary: Pitfalls in the use of brief screening measures of coping. *Journal of Pediatric Psychology, 21*(4), 573-575. doi:10.1093/jpepsy/21.4.573

Spirito, A., Stark, L. J., & Williams, C. (1988). Development of a brief coping checklist for use with pediatric populations. *Journal of Pediatric Psychology, 13*(4), 555-574. doi:10.1093/jpepsy/13.4.555

Stenhammar, C., Isaksson, J., Granstrom, B., Laurell, G., & Tiblom Ehrsson, Y. (2017). Changes in intimate relationships following treatment for head and neck cancer - A qualitative study. *Journal of Psychosocial Oncology, 35*(5), 614-630. doi:10.1080/07347332.2017.1339224

Su, Y. H., & Ryan-Wenger, N. A. (2007). Children's adjustment to parental cancer: a theoretical model development. *Cancer Nursing, 30*(5), 362-381; quiz 382-363. doi:10.1097/01.NCC.0000290817.37442.e6

Sultan, S., Leclair, T., Rondeau, E., Burns, W., & Abate, C. (2016). A systematic review on factors and consequences of parental distress as related to childhood cancer. *European Journal of Cancer Care, 25*(4). doi:10.1111/ecc.12361

Szabó, M. (2010). The short version of the Depression Anxiety Stress Scales (DASS-21): Factor structure in a young adolescent sample. *Journal of Adolescence, 33*(1), 1-8. doi:10.1016/j.adolescence.2009.05.014

Szentes, A., Kökönyei, G., Békési, A., Bokrétás, I., & Török, S. (2017). Differences in illness perception between children with cancer and other chronic diseases and their parents. *Clinical Child Psychology and Psychiatry, 23*(3), 365-380. doi:10.1177/1359104517731899

Tedeschi, R. G., & Calhoun, L. G. (1996). The Posttraumatic Growth Inventory: measuring the positive legacy of trauma. *Journal of Traumatic Stress, 9*(3), 455-471. doi:10.1002/jts.2490090305

Teixeira, R. J., Applebaum, A. J., Bhatia, S., & Brandao, T. (2018). The impact of coping strategies of cancer caregivers on psychophysiological outcomes: an integrative review. *Psychology Research and Behavior Management, 24*(11), 207-215. doi:10.2147/PRBM.S164946

Terwee, C. B., Bot, S. D., de Boer, M. R., van der Windt, D. A., Knol, D. L., Dekker, J., . . . de Vet, H. C. (2007). Quality criteria were proposed for measurement properties of health status questionnaires. *Journal of Clinical Epidemiology, 60*, 34-42. doi:10.1016/j.jclinepi.2006.03.012

Terwee, C. B., Prinsen, C. A. C., Chiarotto, A., Westerman, M. J., Patrick, D. L., Alonso, J., . . . Mokkink, L. B. (2018). COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Quality of Life Research, 27*(5), 1159-1170. doi:10.1007/s11136-018-1829-0

Thastum, M., Johansen, M. B., Gubba, L., Olesen, L. B., & Romer, G. (2008). Coping, social relations, and communication: A qualitative exploratory study of children of parents with cancer. *Clinical Child Psychology and Psychiatry, 13*(1), 123-138. doi:10.1177/1359104507086345

Thastum, M., Watson, M., Kienbacher, C., Piha, J., Steck, B., Zachariae, R., . . . Romer, G. (2009). Prevalence and predictors of emotional and behavioural functioning of children where a parent has cancer: A multinational study. *Cancer, 115*(17), 4030-4039. doi:10.1002/cncr.24449

Thomson, F. E., Bergman, P. B., & Hay, M. (2018). Psychological well-being of carers of children with type 1 diabetes: a comparison with carers of children without a chronic condition. *Journal of Social Health and Diabetes, 6*(2), 96-103. doi:10.1055/s-0038-1673741

Timmons, A., Gooberman-Hill, R., & Sharp, L. (2013). "It's at a time in your life when you are most vulnerable": A qualitative exploration of the financial impact of a cancer diagnosis and implications for financial protection in health. *PLoS One, 8*(11), e77549. doi:10.1371/journal.pone.0077549

Tomasetti, C., Li, L., & Vogelstein, B. (2017). Stem cell divisions, somatic mutations, cancer etiology, and cancer prevention. *Science, 355*(6331), 1330-1334. doi:10.1126/science.aaf9011

Torp, S., Thoresen, L., Gronningsaeter, A. B., Grov, E. K., & Gustavsen, K. (2013). Financial and social effects on children and adolescents when a parent is diagnosed with cancer. *Child and Adolescent Social Work Journal, 30*, 293-310. doi:10.1007/s10560-012-0290-0

Tourangeau, R. (1984). Cognitive science and survey methods: A cognitive perspective. In *Cognitive aspects of survey design: Building a bridge between disciplines* (pp. 73-100). Washington, DC: National Academy Press.

Tourangeau, R., & Rasinki, K. A. (1988). Cognitive processes underlying context effects in attitude measurement. *Psychological Bulletin, 103*(3), 299-314. doi:10.1037/0033-2909.103.3.299

Traeger, L., Penedo, F. J., Gonzalez, J. S., Dahn, J. R., Lechner, S. C., Schneiderman, N., & Antoni, M. H. (2009). Illness perceptions and emotional well-being in men treated for localized prostate cancer. *Journal of Psychosomatic Research, 67*(5), 389-397. doi:10.1016/j.jpsychores.2009.03.013

Trevethan, R. (2017). Sensitivity, specificity, and predictive values: Foundations, pliabilities, and pitfalls in research and practice. *Frontiers in Public Health, 5*, 307. doi:10.3389/fpubh.2017.00307

Turk, D. C., Rudy, T. E., & Salovey, P. (1986). Implicit models of illness. *Journal of Behavioral Medicine, 9*, 453-474. doi:10.1007/BF00845133

Turner, J. K., Hutchinson, A., & Wilson, C. (2018). Correlates of post-traumatic growth following childhood and adolescent cancer: a systematic review and meta-analysis. *Psycho-Oncology, 27*(4), 1100-1109. doi:doi.org/10.1002/pon.4577

United Nations Educational Scientific and Cultural Organisation. (2017). What do we mean by "youth"? Retrieved from http://www.unesco.org/new/en/social-and-human-sciences/themes/youth/youth-definition/

van Oort, L., Schroder, C., & French, D. P. (2011). What do people think about when they answer the Brief Illness Perception Questionnaire? A 'think-aloud' study. *British Journal of Health Psychology, 16*(Pt 2), 231-245. doi:10.1348/135910710X500819

van Stralen, K. J., Stel, V. S., Reitsma, J. B., Dekker, F. W., Zoccali, C., & Jager, K. J. (2009). Diagnostic methods I: sensitivity, specificity, and other measures of accuracy. *Kidney International, 75*(12), 1257-1263. doi:10.1038/ki.2009.92

Vannatta, K., Ramsey, R. R., Noll, R. B., & Gerhardt, C. A. (2010). Associations of child adjustment with parent and family functioning: Comparison of families of women with and without breast cancer. *Journal of Developmental and Behavioral Pediatrics, 31*(1), 9-16. doi:10.1097/DBP.0b013e3181c82a44

Visser, A., B., G., & Vingerhoets, A. (2009). Spirituality and well-being in cancer patients: A review. *Psycho-Oncology, 19*(6), 565-572. doi:10.1002/pon.1626

Visser, A., Huizinga, G. A., Hoekstra, H. J., van der Graaf, W. T., Gazendam-Donofrio, S. M., & Hoekstra-Weebers, J. E. (2007). Emotional and behavioral problems in children of parents recently diagnosed with cancer: A longitudinal study. *Acta Oncologica, 46*(1), 67-76. doi:10.1080/02841860600949560

Visser, A., Huizinga, G. A., Hoekstra, H. J., van der Graaf, W. T., & Hoekstra-Weebers, J. E. (2006). Parental cancer: characteristics of parents as predictors for child functioning. *Cancer, 106*(5), 1178-1187. doi:10.1002/cncr.21691

Visser, A., Huizinga, G. A., Hoekstra, H. J., van der Graaf, W. T., & Hoekstra-Weebers, J. E. (2007). Temperament as a predictor of internalising and externalising problems in adolescent children of parents diagnosed with cancer. *Supportive Care in Cancer, 15*(4), 395-403. doi:10.1007/s00520-006-0117-7

Visser, A., Huizinga, G. A., Hoekstra, H. J., van der Graaf, W. T., Klip, E. C., Pras, E., & Hoekstra-Weebers, J. E. (2005). Emotional and behavioural functioning of children of a parent diagnosed with cancer: A cross-informant perspective. *Psycho-Oncology, 14*(9), 746-758. doi:10.1002/pon.902

Visser, A., Huizinga, G. A., van der Graaf, W. T., Hoekstra, H. J., & Hoekstra-Weebers, J. E. (2004). The impact of parental cancer on children and the family: a review of the literature. *Cancer Treatment Reviews, 30*(8), 683-694. doi:10.1016/j.ctrv.2004.06.001

Walczak, A., McDonald, F. E., Patterson, P., Dobinson, K., & Allison, K. (2018). How does parental cancer affect adolescent and young adult offspring? A systematic review. *International Journal of Nursing Studies, 77*, 54-80. doi:10.1016/j.ijnurstu.2017.08.017

Waldron, E. A., Janke, E. A., Bechtel, C. F., Ramirez, M., & Cohen, A. (2013). A systematic review of psychosocial interventions to improve cancer caregiver quality of life. *Psycho-Oncology, 22*(6), 1200-1207. doi:10.1002/pon.3118

Waller, N. G., Compas, B. E., Hollon, S. D., & Beckjord, E. (2005). Measurement of depressive symptoms in women with breast cancer and women with clinical depression: A differential item functioning analysis. *Journal of Clinical Psychology in Medical Settings, 12*(2), 127-141. doi:10.1007/s10880-005-3273-x

Wang, Y.-P., & Gorenstein, C. (2013). Assessment of depression in medical patients: A systematic review of the utility of the Beck Depression Inventory-II. *Clinics, 68*(9), 1274-1287. doi:10.6061/clinics/2013(09)15

Ward Jr., J. H. (1963). Hierarchical grouping to optimize an objective function. *Journal of the American Statistical Association, 58*(301), 236-244. doi:10.1080/01621459.1963.10500845

Wearden, A., & Peters, S. (2008). Therapeutic techniques for interventions based on Leventhal's common sense model. *British Journal of Health Psychology, 13*(Pt 2), 189-193. doi:10.1348/135910708X295613

Weaver, K. E., Rowland, J. H., Alfano, C. M., & McNeel, T. S. (2010). Parental cancer and the family: A population-based estimate of the number of US cancer survivors residing with their minor children. *Cancer, 116*(18), 4395-4401. doi:10.1002/cncr.25368

Weeks, N., McDonald, F. E., Patterson, P., Konings, S., & Coad, J. (2019). A summary of high quality online information resources for parents with cancer who have adolescent and young adult children: A scoping review. *Psycho-Oncology, 28*(12), 2323-2335. doi:10.1002/pon.5274

Wegner, D. M., & Zanakos, S. (1994). Chronic thought suppression. *Journal of Personality, 62*(4), 615-640. doi:10.1111/j.1467-6494.1994.tb00311.x

Weinman, J., Petrie, K. J., Moss-Morris, R., & Horne, R. (1996). The Illness Perception Questionnaire: A new method for assessing the cognitive representation of illness. *Psychology and Health, 11*(3), 431-445. doi:10.1080/08870449608400270

Weiss, D. S., & Marmar, C. R. (1997). The Impact of Event Scale-Revised. In J. P. Wilson & T. M. Keane (Eds.), *Assessing psychological trauma and PTSD* (pp. 399-411): Guilford Press.

Welch, A. S., Wadsworth, M. E., & Compas, B. E. (1996). Adjustment of children and adolescents to parental cancer. Parents' and children's perspectives. *Cancer, 77*(7), 1409-1418. doi:10.1002/(SICI)1097-0142(19960401)77:7<1409::AID-CNCR28>3.0.CO;2-4

Wethington, E., Glanz, K., & Schwartz, M. D. (2008). Stress, coping, and health behavior. In K. Glanz, B. K. Rimer, & K. Viswanath (Eds.), *Health Behavior and Health Education: Theory, Research, and Practice* (pp. 223-242). San Francisco, CA: Jossey-Bass.

Whitehead, K., Stone, J., Norman, P., Sharpe, M., & Reuber, M. (2015). Differences in relatives' and patients' illness perceptions in functional neurological symptom disorders compared with neurological diseases. *Epilepsy & Behavior, 42*, 159-164. doi:10.1016/j.yebeh.2014.10.031

Wild, C. P., Weiderpass, E., & Stewart, B. W. (2020). *World Cancer Report: Cancer Research for Cancer Prevention*. Retrieved from Lyon, France:

Willis, G. B. (2005). *Cognitive Interviewing: A Tool for Improving Questionnaire Design*. Beverly Hills, California: SAGE Publications.

Windle, G., Bennett, K. M., & Noyes, J. (2011). A methodological review of resilience measurement scales. *Health & Quality of Life Outcomes, 9*(8). doi: 10.1186/1477-7525-9-8

Wong, M. L., Cavanaugh, C. E., Macleamy, J. B., Sojourner-Nelson, A., & Koopman, C. (2009). Post-traumatic growth and adverse long-term effects of parental cancer in children. *Families, systems & health: the journal of collaborative family healthcare, 27*(1), 53-63. doi:10.1037/a0014771

World Health Organization. (2018). *Noncommunicable diseases country profiles 2018*. Retrieved from Geneva:

World Health Organization. (2020). *WHO report on cancer: setting priorities, investing wisely and providing care for all* (Licence: CC BY-NC-SA 3.0 IGO). Retrieved from Geneva:

Wozniak, K., & Izycki, D. (2014). Cancer: A family at risk. *Przeglad Menopauzalny, 13*(4), 253-261. doi:10.5114/pm.2014.45002

Wu, L. M., Mohamed, N. E., Winkel, G., & Diefenbach, M. A. (2013). Patient and spouse illness beliefs and quality of life in prostate cancer patients. *Psychology and Health, 28*(4), 355-368. doi:10.1080/08870446.2012.722219

Ye, Z. J., Liu, M. L., Zhang, Z., Liao, K. L., Peng, C. F., Huang, H., . . . Qiu, H. Z. (2017). Psychometric properties of the Chinese version of the Parent Perception of Uncertainty Scale (PPUS) among parents of children with cancer diagnosis. *International Journal of Nursing Sciences, 4*(3), 278-284. doi:10.1016/j.ijnss.2017.05.004

Youden, W. J. (1950). Index for rating diagnostic tests. *Cancer, 3*(1), 32-35. doi:10.1002/1097-0142(1950)3:1<32::aid-cncr2820030106>3.0.co;2-3

Yu, H., Lingler, J. H., Sereika, S. M., & Erlen, J. A. (2017). Characteristics and correlates of caregivers' perceptions of their family members' memory loss. *Nursing Research, 66*(3), 240-245. doi:10.1097/NNR.0000000000000220

Zamanzadeh, V., Ghahramanian, A., Rassouli, M., Abbaszadeh, A., Alavi-Majd, H., & Nikanfar, A.-R. (2015). Design and Implementation Content Validity Study: development of an instrument for measuring Patient-Centered Communication. *Journal of Caring Sciences, 4*(2), 165-178. doi:10.15171/jcs.2015.017

Zhang, Y., & Wildemuth, B. M. (2009). Qualitative analysis of content. In B. Wildemuth (Ed.), *Applications of social research methods to questions in information and library* (pp. 308-319). Westport, Connecticut: Libraries Unlimited.

Zieba, M., Wiechec, K., Bieganska-Banas, J., & Mieleszczenko-Kowszewicz, W. (2019). Coexistence of post-traumatic growth and post-traumatic depreciation in the aftermath of trauma: Qualitative and quantitative narrative analysis. *Frontiers in Psychology, 29*(10), 687. doi:10.3389/fpsyg.2019.00687

Zimmerman, M., & Guzman Holst, C. (2018). Screening for psychiatric disorders with self-administered questionnaires. *Psychiatry Research, 270*, 1068-1073. doi:10.1016/j.psychres.2018.05.022

Appendices

Appendix A

Co-author approval forms for published studies

Content removed for privacy reasons

Content removed for privacy reasons

Content removed for privacy reasons

Appendix B

Published manuscript for Study 1

Scandinavian Journal of
Caring Sciences
REVIEW ARTICLE
doi: 10.1111/scs.12822

Comparative systematic review of the psychometric properties of measures of illness perceptions in family members of individuals diagnosed with a chronic physical illness

Chloe Fletcher BSc, BHlthSc (Hons) (PhD Candidate)[1], **Ingrid Flight** BA, MPH, PhD (Research Fellow)[1], **Kate Gunn** BPsych (Hons), MPsych (Clin), PhD (Research Fellow)[1,2], **Pandora Patterson** BSocSc (Hons), PhD (General Manager and Adjunct Associate Professor)[3,4] and **Carlene Wilson** BA (Hons), MBA, PhD (Professor)[1,5,6]

[1]Flinders Centre for Innovation in Cancer, College of Medicine and Public Health, Flinders University, Bedford Park, SA, Australia, [2]University of South Australia Cancer Research Institute, University of South Australia, Adelaide, SA, Australia, [3]CanTeen Australia, Newtown, NSW, Australia, [4]Cancer Nursing Research Unit, University of Sydney, Camperdown, NSW, Australia, [5]Olivia Newton-John Cancer Wellness and Research Centre, Heidelberg, VIC, Australia and [6]School of Psychology and Public Health, La Trobe University, Bundoora, VIC, Australia

---

Scand J Caring Sci; 2020

Comparative systematic review of the psychometric properties of measures of illness perceptions in family members of individuals diagnosed with a chronic physical illness

Although illness perceptions have significant implications for psychological morbidity in those diagnosed with a physical illness, the strength of this relationship in their family members remains understudied. The validity of findings is dependent on the quality of the instruments used; therefore, it is essential that psychometrically robust measures of illness perceptions are available. The purpose of this systematic review was to identify, assess and compare the psychometric properties of instruments designed to measure illness perceptions in family members of individuals with chronic physical illnesses. A systematic search was conducted using MEDLINE, PubMed, CINAHL, Scopus and PsycINFO databases, and supplemented with forward and backward searches. Studies were included in the review if they described the development, adaptation or psychometric evaluation of an instrument designed to measure illness perceptions in family members of an individual with a chronic physical illness. The methodological quality of included studies was assessed using the COSMIN Risk of Bias checklist. The psychometric quality of instruments was evaluated using published quality assessment criteria. Eleven articles describing nine different instruments were included in the review. Almost all instruments were designed for parents of a child with a chronic illness. There was wide variation in the quality of methods used to develop, adapt or evaluate the instruments, and missing information restricted the evaluation of psychometric properties. Further validation is needed for all instruments before meaningful conclusions can be drawn. Findings indicate that measurement of illness perceptions in children or siblings of an individual with a chronic physical illness has been largely ignored. Future research addressing this gap would be an important addition to the current body of work examining illness perceptions in family members.

Keywords: illness perceptions, self-report instruments, psychometric properties, psychometric evaluation, reliability and validity.

Submitted 17 July 2019, Accepted 10 January 2020

Introduction

The impacts of chronic illness are far-reaching, extending beyond the person diagnosed with the illness to their parents, siblings and children. An emerging body of evidence highlights the high levels of distress and psychological morbidity experienced by family members and carers of someone diagnosed with a chronic illness e.g. (1) and (2–5), with some findings indicating that levels of anxiety and depression experienced by informal carers

---

Correspondence to:
Chloe Fletcher, Flinders Centre for Innovation in Cancer, College of Medicine and Public Health, Flinders University, Sturt Road, Bedford Park, SA 5042, Australia.
E-mail: chloe.fletcher@flinders.edu.au

This is an original manuscript that has not been submitted elsewhere for publication or previously published, presented, or otherwise disseminated in any form.

are comparable to those with the illness and significantly greater than healthy controls (5,6).

When an individual experiences a health threat, either through identifying a physical symptom or receiving a clinical diagnosis, they form beliefs about the illness that are based on their perception, interpretation and understanding of the illness and its treatment (7–9). These beliefs play an important mediating role between their physical experience of the illness and their subsequent coping and well-being. Differences in physical and psychological outcomes between individuals diagnosed with the same illness can be, at least partly, explained by differences in the way that they perceive and think about their illness (8–12).

It is likely that the mechanisms by which illness perceptions influence coping and psychological adjustment are also implicated in outcomes for family members and carers of those with the illness. For example, Beinke et al. (13) found that perceptions of the cyclical nature of their child's illness were associated with distress in mothers of young adults with cystic fibrosis. Similar findings have been found in parents who have a child with cancer; parental distress has been linked with perceptions of the stressfulness and severity of the illness, the impact of the illness on the family, the intensity of treatment and the potential for the illness to be life-threatening (14–16). Some findings indicate that this association is independent of the child's treatment and illness status (15). Likewise, children's perceptions of the stressfulness of their parent's cancer have been shown to be associated with anxiety and depression (17). In adolescents with a parent with multiple sclerosis, beliefs about the impact of the illness on the family environment and the unpredictable course of the illness were found to be strongly correlated with emotional and behavioural difficulties (18).

A range of theoretical models have been utilised to describe the relationships between illness perceptions and health outcomes (e.g. Lazarus and Folkman's Transactional Model of Stress and Coping (19), the Health Belief Model (20) and the Theory of Planned Behaviour (21)). Diversity in approaches used to conceptualise illness perceptions and describe their relationships with health outcomes has led to the development of a number of multidimensional instruments designed to measure this construct. These include the Illness Cognitions Scale (22), the Illness Cognition Questionnaire (10), the Illness Perception Questionnaire (23) and the revised Illness Perception Questionnaire (24), all of which were originally developed to measure illness perceptions amongst people experiencing ill health. Since then, the Illness Perception Questionnaire and the Illness Cognition Questionnaire have been adapted to measure illness perceptions in adolescents who have a parent with multiple sclerosis (18) and parents who have a child with cancer (25), respectively. However, neither has been validated for other illnesses and no one instrument has been consistently used across illness populations to assess illness perceptions in family members of an individual with a chronic illness.

To date, a systematic review of instruments developed and validated to measure illness perceptions in family members of an individual with a chronic illness has not been undertaken. The ability to identify illness perceptions in family members of people with a chronic illness is essential, given the significant impact on carers and supporters and the growing body of research indicating that these beliefs may be important predictors or mediators of psychological outcomes in this cohort. Moreover, the validity of findings in this area is dependent on the quality of the instruments used; it is essential that psychometrically robust measures of illness perceptions are available for use with family members, particularly where these instruments are used to screen for potential psychological morbidity. Therefore, the purpose of this review was to:

1 identify instruments designed to measure illness perceptions in family members and carers of individuals with chronic physical illnesses;
2 assess and compare the psychometric properties of identified instruments using published quality assessment criteria (26); and
3 assess the methodological quality of studies describing the development and/or evaluation of identified instruments using the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) Risk of Bias checklist (27–29).

Given the heterogeneous nature of research exploring illness perceptions and that the relationship between illness perceptions and psychological outcomes in family members of a person with a chronic illness has attracted little hypothesis testing, this review was not limited to instruments developed with a specific theoretical underpinning. 'Illness perceptions' were broadly defined to encompass a diverse range of conceptualisations. Consequently, we identified relevant papers as describing measures that capture an individual's cognitive and affective representation of a family member's illness.

Method

The review protocol was registered at the PROSPERO international prospective register of systematic reviews (registration number: CRD42017072845).

Search strategy

Databases MEDLINE (from 1946-), PubMed (excluding MEDLINE; from 1946-), Cumulative Index to Nursing and Allied Health Literature (CINAHL), Scopus (Social Sciences and Humanities subject areas; from 1823-) and PsycINFO (from 1806-) were searched using a combination of terms relating to illness cognitions, perceptions or beliefs, measurement, family members or carers, and chronic illness or disease. Searches were limited to studies conducted with human participants and published in English. The full search string run in MEDLINE is outlined in Table 1. The search was performed on 27 July 2017.

Following systematic screening procedures, forward and backward searching techniques were employed to identify relevant studies that cited, or were cited by, the studies that were included. Google Scholar was used to identify studies that had cited included studies, and the reference lists of included studies were hand searched. When an included study described the adaptation or evaluation of an existing instrument, the reference list was searched to identify the original development study.

Inclusion criteria

Studies were included if they were published in peer-reviewed journals; they described the development, adaptation or psychometric evaluation of an instrument; the instrument was designed to measure illness perceptions; the instrument was designed for use in family members or carers of an individual; the individual had been diagnosed with a chronic physical illness; and there was a hypothesised predictive relationship between the construct being measured and psychological outcomes in the respondent.

Exclusion criteria

Studies were excluded if the instrument was designed for use in the individual diagnosed with the illness or measured illness perceptions relating to mental illness. Studies not published in English were also excluded if no translation was readily available.

Study selection

In total, 6360 articles were retrieved from initial searches on MEDLINE (n = 2092), PubMed (excluding MEDLINE; n = 2246), CINAHL (excluding MEDLINE; n = 1650), Scopus (n = 347) and PsycINFO (n = 25; see Fig. 1 for PRISMA flow diagram of study selection process). Citations that were imported into reference management software EndNote and duplicates (n = 623) were removed. Titles and abstracts were screened by the first author for relevance according to the inclusion and exclusion criteria resulting in a further 5683 citations being excluded. A subset (10%) of abstracts were screened independently by a second reviewer, with excellent agreement (95%) observed between reviewers (Kappa = 0.877, $p < 0.001$). Following title and abstract screening, 54 full-text articles were retrieved and considered for inclusion. A second reviewer independently screened a subset (10%) of full-text articles, with 100% agreement observed between reviewers. Fifty articles were excluded for the reasons outlined in Fig. 1, and the remaining four were included in the review. Forward and backward searches were conducted for each of the included studies, and an additional seven articles were identified and included in the review.

Data extraction

Descriptive information was extracted for each of the included studies. Information relating to characteristics of

Table 1 Advanced search string run in MEDLINE

1	(illness cognition* or illness perception* or illness belief* or cognition* or perception* or belief*).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
2	(scale or instrument or measure or survey or questionnaire or assessment or checklist).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
3	(family member or parent or mother or father or m*m or dad or child or children or adolescent or young adult).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
4	(chronic illness or chronic disease or palliative or terminal or cancer or diabetes or multiple sclerosis or heart disease or stroke or asthma or arthritis or osteoporosis or chronic obstructive pulmonary disease or chronic kidney disease).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
5	1 and 2 and 3 and 4
6	Limit 5 to (English language and humans)
7	exp "Surveys and Questionnaires"/st [Standards]
8	health care surveys/or questionnaires/
9	7 or 8
10	6 and 9

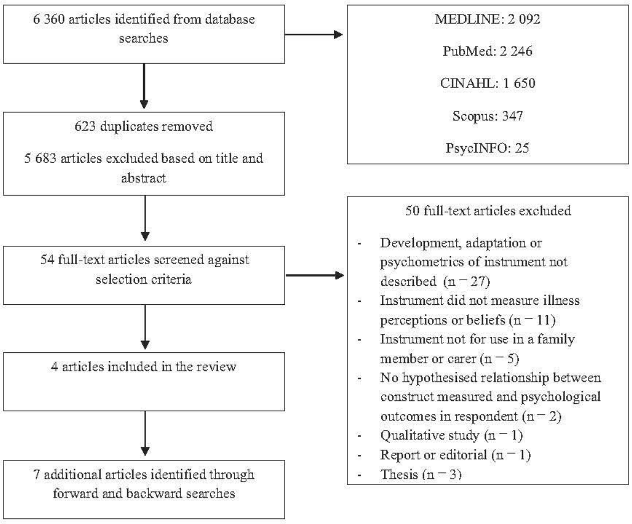


Figure 1 Flow diagram of study selection process. CINAHL, Cumulative Index to Nursing and Allied Health Literature; n, number of articles.

the study (type of study, sample type, sample size, mean age, gender distribution and the country where the study was conducted) and characteristics of the instrument (instrument name, construct measured, the population the instrument was developed or adapted for, number of subscales, number of items, scale type, response options, sample items, information about instrument development and the available languages of the instrument) was extracted. Psychometric data addressing the relevant quality criteria (described below and in Table 2) were also extracted.

Quality assessment

The psychometric properties of each instrument were evaluated using published quality assessment criteria originally developed for the purpose of evaluating the psychometric properties of health status measures (26). The criteria address content validity, structural validity, internal consistency, criterion validity, construct validity, measurement error, test–retest reliability, responsiveness, floor and ceiling effects, and interpretability. Each study was assessed individually, with findings rated as positive (+), negative (−) or indeterminate (?) for each property (see Table 2 for detailed quality criteria). Only properties that were reported in the original study were assessed.

The methodological quality of included studies was evaluated using the COSMIN Risk of Bias checklist (27–29). The COSMIN checklist is a standardised tool for evaluating the methodological quality of studies describing measurement properties. It was developed in an international Delphi study involving 57 researchers with backgrounds in epidemiology, psychometrics, qualitative research, health care, and development and evaluation of outcome measurement instruments (30,31).

The checklist contains ten boxes used to assess whether a study meets standards for good methodological quality for testing psychometric properties. Where reported in the original study, methods used to evaluate structural validity, internal consistency, cross-cultural validity, reliability, measurement error, criterion validity, construct validity and responsiveness were assessed using the designated COSMIN boxes. Methods were rated as very good, adequate, doubtful or inadequate according to the criteria outlined in each box. As recommended in the COSMIN guidelines (27–29), the 'worst score counts' method was used to determine the overall rating for the quality of methods used to evaluate each psychometric property. This meant that the lowest rating of any of the items in the designated box for each psychometric property was taken as the overall score for that particular property. Boxes for properties that were not evaluated in the study did not receive a score at all. Most studies reported very little information on the methods used to develop the instrument and assess content validity, meaning that the boxes for instrument development and content validity could not be completed. Instead, the development of the instrument and any cognitive interview studies or other pilot studies were qualitatively described and outlined in the results (e.g. instrument development and content validity testing of the

Table 2 Quality criteria for assessment of psychometric properties (adapted from Terwee et al. (26))

	Property	*Definition*	*Quality criteria*^{a,b}
1	Content validity	The extent to which the domain of interest is comprehensively sampled by the items in the questionnaire	+ A clear description is provided of the measurement aim, the target population, the concepts that are being measure and the item selection AND target population and (investigators OR experts) were involved in item selection; ? A clear description of above-mentioned aspects is lacking OR only target population involved OR doubtful design or method; − No target population involvement; 0 No information found on target population involvement
2	Structural validity	The extent to which scores on a particular questionnaire are an adequate reflection of the dimensionality of the construct to be measured	+ Factors analyses performed on adequate sample size (7 * # items and ≥100); ? No factor analysis OR doubtful design or method; 0 No information found on structural validity
3	Internal consistency	The extent to which items in a (sub)scale are intercorrelated, thus measuring the same construct	+ Cronbach's alpha(s) calculated per dimension AND Cronbach's alpha(s) between 0.70 and 0.95; ? Cronbach's alpha(s) NOT calculated for each dimension OR doubtful design or method; − Cronbach's alpha(s) <0.70 or >0.95, despite adequate design and method; 0 No information found on internal consistency
4	Criterion validity	The extent to which scores on a particular questionnaire relate to a gold standard	+ Convincing arguments that gold standard is 'gold' AND correlation with gold standard ≥0.70; ? No convincing arguments that gold standard is 'gold' OR doubtful design or method; − Correlation with gold standard <0.70, despite adequate design and method; 0 No information found on criterion validity
5	Construct validity	The extent to which scores on a particular questionnaire relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are being measured	+ Specific hypotheses were formulated AND at least 75% of the results are in accordance with these hypotheses; ? Doubt design or method (e.g. no hypotheses); − Less than 75% of hypotheses were confirmed, despite adequate design and methods; 0 No information found on construct validity
6	Measurement error	The extent to which scores on repeated measures are close to each other (absolute measurement error)	+ SDC < MIC OR MIC outside the LOA OR convincing arguments that agreement is acceptable; ? Doubtful design or method OR (MIC not defined AND no convincing arguments that agreement is acceptable); − SDC ≥ MIC OR MIC equals or inside LOA, despite adequate design and method; 0 No information found on agreement
7	Reliability	The extent to which respondents can be distinguished from each other, despite measurement errors (relative measurement error)	+ ICC or weighted Kappa ≥ 0.70; ? Doubtful design or method (e.g. time interval not mentioned); − ICC or weighted Kappa < 0.70, despite adequate design and method; 0 No information found on reliability
7	Responsiveness	The ability of a questionnaire to detect clinically important changes over time	+ SDC or SDC < MIC OR MIC outside the LOA OR RR > 1.96 OR AUC ≥ 0.70; ? Doubtful design or method; − SDC or SDC ≥ MIC OR MIC equals or inside LOA OR RR ≤ 1.96 OR AUC < 0.70, despite adequate design and methods; 0 No information found on responsiveness
8	Floor and ceiling effects	The number of respondents who achieved the lowest or highest possible score	+ ≤15% of the respondents achieved the highest or lowest possible scores; ? Doubtful design or method; − >15% of the respondents achieved the highest or lower possible scores, despite adequate design and methods; 0 No information found on floor and ceiling effects

Table 2 (Continued)

	Property	Definition	Quality criteria[a,b]
9	Interpretability	The degree to which one can assign qualitative meaning to quantitative scores	+ Mean and SD scores presented of at least four relevant subgroups of respondents and MIC defined; ? Doubtful design or method OR less than four subgroups OR no MIC defined; 0 No information found on interpretation

Abbreviations: AUC, area under the curve; ICC, intraclass correlation; LOA, limits of agreement; MIC, minimal important change; RR, responsiveness ratio; SD, standard deviation; SDC, smallest detectable change.

[a]+ = positive rating; ? = indeterminate rating; − = negative rating; 0 = no information available.

[b]Doubtful design or method = lacking of a clear description of the design or methods of the study, sample size smaller than 50 participants (should be at least 50 in every (subgroup) analysis) or any important methodological weakness in the design of the study.

Perceptions of Parental Illness Questionnaire is described in Table 4 as follows: *Questionnaire items were developed based on Leventhal's Common-Sense Model of Self-Regulation and interviews conducted with adolescents with a parent with multiple sclerosis. Questionnaire items were refined based on cognitive interviews conducted with six adolescents. Two subscales were removed and item wording was modified based on feedback.*)

Assessments were conducted by the first author, with a subset (10%) of studies independently assessed by a second reviewer (assessed the quality of psychometric properties of instruments and IF assessed the methodological quality of included studies). At each stage of assessment, disagreement in ratings was discussed amongst reviewers until consensus was reached.

It should be noted that although the quality of methodology used to evaluate content validity could not be properly assessed, following the criteria developed by Terwee et al. (26), instruments were given a positive rating for content validity if the development involved input from the target population.

Results

Instruments identified

The search string was developed to identify studies that used specific terms relating to illness cognitions, perceptions or beliefs. During the screening process, we identified a number of instruments that had been developed to measure perceptual constructs that were conceptually related, although not specifically described as illness cognitions, perceptions or beliefs by the study authors. These constructs were worry, uncertainty, chronic sorrow, perceived emotional resources and perceived impact of illness.

Worry was defined by the study authors as 'thoughts and images that relate to possible negative or threatening outcomes', and they predicted a clear relationship between worry, coping and psychological outcomes in the respondent (32). Mishel (33) proposed uncertainty as 'a major perceptual variable influencing parents' experiences during their child's illness' that affects appraisal of the illness and the ability to cope. Bonner et al. (34) sought to develop a measure of the important aspects of psychological adjustment that encompassed constructs of uncertainty, chronic sorrow (defined as 'prolonged feelings of grief, disappointment, and fear in response to the continual losses experienced over the trajectory of an illness'), and perceived emotional resources (defined as 'cognitive appraisals of internal resources and perceived self-efficacy'). Bemister (35) developed a measure of perceived psychosocial impact of illness that captured parents' sense of guilt and blame in relation to their child's diagnosis. Because each of these constructs captures a cognitive or affective representation of a family member's illness, and a clear relationship between each construct and coping in the face of a health threat was predicted, these instruments were included in the review. For this reason, a majority of the included articles were identified through forward and backward searching techniques, rather than through the initial database search.

Table 3 describes the eleven articles included in the review. Five articles described the development and initial psychometric properties of an original instrument. An additional four articles described the validation of adapted (three articles) or translated (one article) instruments. The remaining two articles provided further validation of an existing instrument. Instruments were primarily developed to be applicable for any chronic illness, although three instruments were developed for specific use in family members of an individual with cancer (Family Illness Beliefs Inventory; (14), multiple sclerosis (Perceptions of Parental Illness Questionnaire; (18) and perinatal stroke (Parental Outcome Measure; (35) (see Table 4). All of the instruments were designed for parents with a child with a chronic illness, except for the Perceptions of Parental Illness Questionnaire (18).

Table 3 Characteristics of included studies

Instrument/Study	*Type of study*	*Sample type*	*Sample size*	*Age, mean (SD)*	*Female (%)*	*Country*
Parental Outcome Measure						
(35)	Development and validation	Parents of children with perinatal stroke	Time 1: 110, time 2: 58	39.5 (7.4) years	Time 1: 74.5	Canada
Perceptions of Parental Illness Questionnaire						
(18)	Development and validation	Adolescents with a parent with multiple sclerosis	Development: 15, cognitive interviews: 6, validation: 104	Validation: 15.4 (1.97) years	59.6	UK
Parental Experience of Child Illness						
(34)	Development and initial validation	Parents of children with brain tumours	Pilot testing: 6, validation: 148	39.5 (5.9) years	82.6	USA
(38)	Validation	Parents of children diagnosed with cancer	Time 1: 125, time 2: 75	39.6 years	Time 1: 80.8	USA
Maternal Worry Scale						
(32)	Development and psychometric evaluation	Mothers of children with juvenile rheumatoid arthritis, sickle cell anaemia or diabetes	140	-	100	USA
Family Illness Beliefs Inventory						
(14)	Development and validation	Parents of children with cancer	119 mothers, 56 fathers	-	68	USA
Parental Perception of Uncertainty Scale						
(33)	Adaptation and validation	Parents of children hospitalised with an illness	272	-	80.1	USA
(37)	Validation	Parents of children with cancer	183	-	86.9	USA
Parental Perception of Uncertainty Scale-Diagnosis						
(39)	Adaptation and validation	Mothers of infants who are HIV seropositive	Content validity testing: 11, preliminary psychometric evaluation: 25	29 years	100	USA
Parental Perception of Uncertainty Scale-Chinese						
(36)	Adaptation and validation of translated version	Parents of children diagnosed with cancer	377	-	-	China
Illness Cognition Questionnaire-Parent version						
(25)	Adaptation and validation	Parents of children with cancer	242	39.98 (6.30) years	76	The Netherlands

Psychometric properties of instruments

Table 5 summarises the quality of psychometric properties of instruments evaluated in included studies.

Content validity. Most instruments were comprised of items generated by the researchers, informed by literature reviews and/or input from clinical experts, and involved the target population in item selection through cognitive interview studies or other pilot testing. Both strategies resulted in a positive score for content validation. The Parental Perception of Uncertainty Scale and the Illness Cognition Questionnaire-Parent Version were both adapted from existing measures that were originally developed to measure uncertainty and illness cognitions in the individual with the illness. Neither Mishel (33) nor Sint Nicolaas et al. (25) described including the new target population (i.e. parents) during the adaptation process to ensure content validity. Similarly, the new target population was not involved during the translation of the Parental Perception of Uncertainty Scale to Chinese (36). The remaining studies both involved further validation of an existing measure and did not include additional assessment of content validity.

Table 4 Characteristics of the included instruments

Instrument	*Construct*	*Target Population*	*Dimensions (number of items)*	*Total number of items*	*Response options (range)*	*Instrument development and content validity testing*	*Language*
Parental Outcome Measure (35)	Perceived Impact of Illness, Guilt and Blame	Parents of children with perinatal stroke	Three dimensions – Psychosocial Impact (17) Guilt (4) Blame (5)	31 items, revised to 26 items based on exploratory factor analysis	5-point scale (0 = strongly disagree to 4 = strongly agree)	Questionnaire items were developed by a multidisciplinary team based on their experience with perinatal stroke and the literature on caring for children with disabilities. A draft 34-item questionnaire was piloted amongst 10 healthcare professionals and six parents of children with perinatal stroke to collect feedback on the content and face validity and wording of the questionnaire. The questionnaire was modified based on the feedback. The resulting questionnaire consisted of 31 items. This was later revised to 26 items based on exploratory factor analysis.	English
Perceptions of Parental Illness Questionnaire (18)	Illness Beliefs	Adolescents with a parent with multiple sclerosis	11 dimensions – Internal items: Emotional representation (5) Adolescents' control (4) Negative consequences for family (3) Positive consequences for adolescents (4) Negative consequences for adolescents (3) Chronic timeline (4) Unpredictable timeline (2) Causal items: Psychological attributions (2) Central nervous system (2) External/environmental attributions (2) Hereditary/chance (2)	33 items	Not described	Questionnaire items were developed based on Leventhal's Common-Sense Model of Self-Regulation and interviews conducted with adolescents with a parent with multiple sclerosis. Questionnaire items were refined based on cognitive interviews conducted with six adolescents. Two subscales were removed, and item wording was modified based on feedback.	English

Table 4 (Continued)

Instrument	*Construct*	*Target Population*	*Dimensions (number of items)*	*Total number of items*	*Response options (range)*	*Instrument development and content validity testing*	*Language*
Parental Experience of Child Illness (34,38)	Chronic Sorrow, Uncertainty and Emotional Resources	Parents of children with a chronic illness	Four dimensions – Emotional Resources (3) Guilt and Worry (11) Unresolved Sorrow and Anger (7) Long-term Uncertainty (4)	25 items	5-point scale (0 = never to 4 = always)	A preliminary list of items reflecting issues relevant to parenting a child with chronic illness was developed by a multidisciplinary team. The initial 60-item draft covered theoretical domains derived from the literature. Discussion amongst research team resulted in removal of 24 items that were redundant, ambiguous or had low relevance to theoretical constructs. The remaining 36 items were reviewed by six parents of children diagnosed with cancer. A further 11 items were removed following feedback provided by the parents.	English
Maternal Worry Scale (32)	Worry	Mothers of children with a chronic illness	One dimension – Maternal worry (11)	11 items	4-point scale (1 = not at all to 4 = most of the time)	Questionnaire items were developed through discussions with five mothers of young adults who had arthritis since childhood. Mothers were participants in a parent support intervention. The theme of 'worrying about your kid' emerged repeatedly throughout group discussions. The content of worries was recorded over a six-month period and was used to develop an initial set of 30 items. Additional items were included based on previous research and clinical experience. Items were worded to apply to children with any ongoing physical illness. The initial set of items was piloted amongst five mothers of children with arthritis. Items that were redundant or related to infrequently mentioned worries were removed or combined with other items to produce the final 11-item questionnaire.	English

Table 4 (Continued)

Instrument	*Construct*	*Target Population*	*Dimensions (number of items)*	*Total number of items*	*Response options (range)*	*Instrument development and content validity testing*	*Language*
Family Illness Beliefs Inventory (14)	Illness Beliefs	Parents of children with cancer	Five dimensions – Treatment-related Suffering (11) Death and Devastation (8) Caregiver Competence (9) Connection (8) Finding Meaning (5)	41 items	Card-sort method, with cards sorted into one of four piles: 1 = not at all true for me, 2 = just a little bit true for me, 3 = pretty much true for me and 4 = very true for me	A set of 75 items was developed by a multidisciplinary team based on clinical experience and the literature. Items were structured in six important areas: affect, finding meaning, optimism, self-efficacy, connection and illness-specific beliefs. Items were worded to reflect growth-promoting and growth-inhibiting beliefs. Items were reviewed by five parents of paediatric oncology patients. One item relating to fear of relapse was added based on their feedback.	English
Parental Perception of Uncertainty Scale (33,37)	Uncertainty	Parents of children hospitalised with a serious illness	One dimension – Global uncertainty (25)	25 items in final structure proposed by Molzon et al. (37)	5-point scale (1 = strongly disagree to 5 = strongly agree)	The original Mishel Uncertainty in Illness Scale (49) was modified to produce a measure of perceived uncertainty in parents. Items were modified from the original format and reviewed by a group of paediatric nurses for content validity. Items were evaluated to assess relevance to the paediatric hospitalisation experience and parental concerns.	English
Parental Perception of Uncertainty Scale-Diagnosis (39)	Uncertainty	Parents of children undergoing diagnosis of a serious illness	One dimension – Global parental uncertainty (24)	32 items in original questionnaire, revised to 24 items after psychometric analysis	As above	The original Parental Perception of Uncertainty Scale was modified to produce a measure of parental uncertainty during diagnosis. Content validity was assessed by five mothers of seropositive infants undergoing diagnosis for perinatally acquired HIV infection and six advanced practice nurses. Mothers and nurses were asked to provide feedback on the face validity of the questionnaire, comment on the clarity of items, indicate items that seemed redundant or unrelated and suggest additional items or alternative wording.	English

Table 4 (Continued)

Instrument	*Construct*	*Target Population*	*Dimensions (number of items)*	*Total number of items*	*Response options (range)*	*Instrument development and content validity testing*	*Language*
Parental Perception of Uncertainty Scale-Chinese (36)	Uncertainty	Parents of children with cancer	Two dimensions – Ambiguity (9) Comprehension (5)	19 items in original questionnaire, revised to 14 items after psychometric analysis	As above	The Parental Perception of Uncertainty Scale was translated into Chinese by two researchers. The questionnaire was revised by removing redundant or irrelevant items through discussion amongst six psychology professors and nine clinical nursing professors. Content validity was assessed using the content validity index.	Chinese
Illness Cognition Questionnaire-Parent version (25)	Illness Beliefs	Parents of children with a chronic illness	Three dimensions – Helplessness (6) Acceptance (6) Perceived Benefits (6)	18 items	4-point scale (1 = not at all to 4 = completely)	The original Illness Cognition Questionnaire was modified to measure illness cognitions in parents with a child with cancer. No information available on content validity.	Full questionnaire published in English, but validated in Dutch

Structural validity. In a large proportion of the included studies, factor analyses were conducted using an inadequate sample size (i.e. if the sample size was less than 7× the number of items OR less than 100) (14,18,34,35,37). These studies were given an indeterminate rating. Bonner et al. (38) and Santacroce (39) did not conduct factor analysis and were also given indeterminate ratings.

Internal consistency. Cronbach's alphas were reported to be ≥0.70 for all subscales in a number of studies (14,25,32–38). Bogosian et al. (18) reported a Cronbach's alpha of 0.64 for the Chronic Timeline subscale of the Perceptions of Parental Illness Questionnaire. Santacroce (39) did not report separate Cronbach's alphas for each unidimensional subscale of the Parental Perception of Uncertainty Scale-Diagnosis.

Criterion validity. There is no apparent 'gold standard' for validation of illness perceptions in family members of an individual with a chronic illness. Although several studies reported that they had assessed criterion validity, many used a comparator measure that may have been more appropriate to assess construct validity (18,35,36). These studies were assessed using criteria for construct validity.

Construct validity. Construct validity was assessed for all of the instruments; however, only seven studies received a positive rating (14,18,25,32,35,36,39). The remaining studies were given an indeterminate rating because they did not outline specific theoretically derived hypotheses (33,34,37,38). Construct validity was assessed primarily by examining convergent validity, specifically, the extent to which scores on the instrument correlated with measures of psychological outcomes, including the Hospital Anxiety and Depression Scale (25,35), the Profile of Mood States (25), the Strengths and Difficulties Questionnaire (18), the Brief Symptom Inventory (14,34,37), the Impact of Event Scale (14,34,37), the Beck Depression Inventory (32) and the Psychiatric Symptom Index (32).

Measurement error. The standard error of measurement was not calculated for any of the instruments.

Reliability. Test–retest reliability was assessed for four of the instruments (32,35,36,38), all of which assessed test–retest reliability using a Pearson or Spearman correlation coefficient. According to the criteria developed by Terwee et al. (26), this is not considered to be an adequate reliability parameter because systematic differences are not taken into account. Terwee et al. (26) recommend that test–retest reliability is assessed using the intraclass correlation coefficient (with a cut-off of >0.70) and a sample size of >50. Despite the study authors reporting large

Table 5 Quality of psychometric properties of instruments assessed in included studies[a]

Instrument/ study	*Content validity*	*Structural validity*	*Internal consistency*	*Construct validity*	*Criterion validity*	*Measurement error*	*Reliability*	*Responsiveness*	*Floor or ceiling effect*	*Interpretability*
Parental Outcome Measure										
(35)	+	?	+	+	0	0	?	0	+	0
Perceptions of Parental Illness Questionnaire										
(18)	+	?	–	+	0	0	0	?	0	0
Parental Experience of Child Illness										
(34)	+	?	+	?	0	0	0	0	0	0
(38)	0	?	+	?	0	0	?	0	0	0
Maternal Worry Scale										
(32)	+	+	+	+	0	0	?	0	+	0
Family Illness Beliefs Inventory										
(14)	+	?	+	+	0	0	0	0	0	?
Parental Perception of Uncertainty Scale										
(33)	–	+	+	?	0	0	0	0	0	?
(37)	0	?	+	?	0	0	0	0	0	0
Parental Perception of Uncertainty Scale–Chinese										
(36)	–	+	+	+	0	0	?	0	0	?
Parental Perception of Uncertainty Scale–Diagnosis										
(39)	+	?	?	+	0	0	0	0	+	0
Illness Cognition Questionnaire–Parent version										
(25)	–	+	+	+	0	0	0	0	0	?

[a]+ = positive rating; ? = indeterminate rating; – = negative rating; 0 = no information available.

Pearson correlation coefficients, following Terwee's criteria, indeterminate ratings were given for the Parental Outcome Measure (r = 0.87; 35), the Parental Experience of Child Illness (r = 0.83 - 0.86 across the four subscales; 38), the Maternal Worry Scale (r = 0.84; 32) and the Parental Perception of Uncertainty Scale-Chinese version (r = 0.88; 36).

Responsiveness. Bogosian et al. (18) reported that they assessed the relationship between illness perceptions at baseline and adjustment at 6 months follow-up; however, insufficient information was provided to give a positive or negative rating.

Floor/ceiling effects. Potential floor and ceiling effects were examined for the Parental Outcome Measure (35), the Maternal Worry Scale (32) and the Parental Perception of Uncertainty Scale-Diagnosis (39). None of these instruments showed evidence of a floor or ceiling effect.

Interpretability. Three studies were given an indeterminate rating for interpretability because they did not report mean scores and standard deviations for at least four subgroups (14,25,33). Ye et al. (36) did not provide descriptive statistics for subgroups, but did recommend a cut-off score for the Parental Perception of Uncertainty Scale-Chinese version, suggesting that parents who scored higher than 42.5 may require further attention.

Methodological quality of studies

Table 6 summarises the methodological quality of studies for each psychometric property. None of the studies assessed or reported on the full range of properties recommended in the COSMIN guidelines. There was some variation in the quality of methods used to assess each of the properties across the studies. Almost all of the studies involved assessment of structural validity; however, the methodology was rated as inadequate or doubtful in several studies because the sample size was not large enough for the number of items included in the factor analysis or because there were other minor methodological flaws (e.g. the type of factor analysis performed was not described). Overall, most studies were rated as very good for methodology used to assess internal consistency. Santacroce (39) was rated as inadequate because Cronbach's alphas were not reported for all unidimensional subscales. Only one study reported cross-cultural adaptation of an existing measure (36); however, the cross-cultural validity of the translated measure was not assessed. Reliability was assessed in four of the studies; the methodology was rated as doubtful in all of the studies because they assessed reliability using a Pearson or Spearman correlation coefficient. None of the studies assessed measurement error or criterion validity. Ratings for hypotheses testing for construct validity were varied, mostly because of a lack of information on comparator

Table 6 Methodological quality of included studies

Instrument/ study	*Structural validity*	*Internal consistency*	*Cross-cultural validity*	*Reliability*	*Measurement error*	*Criterion validity*	*Hypotheses testing*	*Responsiveness*
Parental Outcome Measure								
(35)	Inadequate[a]	Very good		Doubtful[f]			Adequate[g]	
Perceptions of Parental Illness Questionnaire								
(18)	Inadequate[a]	Very good					Adequate[g]	Very Good
Parental Experience of Child Illness								
(34)	Adequate[b]	Very good					Adequate[g]	
(38)		Very good		Doubtful[f]			Adequate[g]	
Maternal Worry Scale								
(32)	Doubtful[c]	Very good		Doubtful[f]			Adequate[g]	
Family Illness Beliefs Inventory								
(14)	Inadequate[a]	Very good					Adequate[g]	
Parental Perception of Uncertainty Scale								
(33)	Adequate[d]	Very good					Inadequate[h]	
(37)	Adequate[b]	Very good					Doubtful[i]	
Parental Perception of Uncertainty Scale-Chinese								
(36)	Very good	Very good		Doubtful[f]			Doubtful[i]	
Parental Perception of Uncertainty Scale-Diagnosis								
(39)		Inadequate[e]					Doubtful[i]	
Illness Cognition Questionnaire-Parent version								
(25)	Adequate[d]	Very good					Adequate[g]	

[a] Sample size <5 times the number of items.

[b] Sample size at least 5 times the number of items and >100.

[c] Factor analysis and rotation method not described.

[d] Confirmatory factor analysis was not performed.

[e] Internal consistency was not assessed for each unidimensional subscale.

[f] Pearson or Spearman correlation coefficient calculated without evidence of no systematic change.

[g] Assumable that statistical methods adequate.

[h] No information on measurement properties of comparator instruments.

[i] Insufficient information on measurement properties of comparator instruments.

instruments. Only one study assessed responsiveness (18); the methods used were rated as very good.

Discussion

Measurement of illness perceptions in family members of an individual with a chronic illness has the potential to help identify those at risk of poor psychological outcomes and inform the development of interventions to target unhelpful beliefs and perceptions. However, this requires the availability of instruments that have been psychometrically validated as appropriate for predicting these outcomes in the population in which they are to be used. The purpose of this review was to identify, assess and compare the psychometric properties of instruments designed to measure illness perceptions in family members and carers of individuals with chronic physical illness with the goal of identifying instruments that could predict individual differences in risk for poor psychological outcomes. The review identified nine instruments that assessed aspects of illness perception and were developed or validated for use in family members and carers. Most instruments were developed to assess illness perceptions in parents of a child with a chronic illness; only the Perceptions of Parental Illness Questionnaire was validated in children of a parent with a chronic illness. Eight instruments were developed in English; the Illness Cognition Questionnaire-Parent version was developed and validated in Dutch. Of the eight instruments developed in English, only one was translated into a different language (i.e. the Parental Perception of Uncertainty Scale-Chinese).

The findings of this review suggest that (i) further validation is needed for all of the instruments assessed, and (ii) reporting of instrument development and validation needs to be improved. Information on measurement error, test–retest reliability (assessed using the intraclass correlation coefficient), responsiveness, floor and ceiling effects, and interpretability was missing for most of the instruments. Many studies also failed to use adequate methodology or provide adequate information on content, structural and construct validity. At a minimum, adequate information should be provided on content validity, structural validity, internal consistency and construct validity to demonstrate that the instrument measures a construct that is theoretically sound, that items within subscales are adequately correlated (to indicate that they are measuring the same construct) and that analyses will be able to discriminate between and/or predict the relevant outcome variables (40). Additional information about measurement error and reliability is required depending on whether the instrument is intended for evaluative or discriminative purposes (26). Moreover, information about responsiveness and interpretability – specifically, information about what cut-off or change in score would be clinically meaningful – is imperative for instruments intended for clinical use (26). Inclusion of this information in development and validation studies will also enable researchers to make informed decisions about which instrument is most appropriate to address the aims of their study.

The most widely used measures of illness perceptions in individuals diagnosed with a chronic physical illness are the Illness Perception Questionnaire (23) and the revised Illness Perception Questionnaire (24). Only one of the instruments included in the present review, the Perceptions of Parental Illness Questionnaire (18), was adapted from the Illness Perception Questionnaire. Although the Illness Perception Questionnaire is commonly used to assess illness perceptions in spouses of an individual diagnosed with an illness (41–43), limited data on the validity of its application in this population exist. Psychometric evidence is relative to the target population, and measures of illness perceptions need to be appropriately adapted and validated for the groups in which they are intended to be used. Based on the findings of the present review, it is recommended that research should be conducted to examine the psychometric properties of the Illness Perception Questionnaire when adapted for use in family members in a range of illness groups, age groups, cultural groups and for different genders.

It should also be noted that only one of the instruments examined in the present review was designed for use in children, under the age of 18 years, who have a parent with a chronic illness. Furthermore, none of the reviewed instruments were validated for use in children under the age of 12 years. This highlights a huge gap in research examining illness perceptions and is concerning given that children's perceptions of their parent's illness may have significant implications for their affective development. The only study that validated a measure of illness perceptions in young people with a parent with a chronic illness was conducted by Bogosian et al. (18). They found that beliefs about the impact of the illness on the family environment and the unpredictable course of the illness were strongly correlated with emotional and behavioural difficulties in adolescents with a parent with multiple sclerosis (18). Future research may potentially be conducted to validate this measure for use with other illness groups. The availability of an evidence-based and psychometrically robust measure of illness perceptions that has been validated for use in children who have a parent with a diverse range of chronic illnesses would facilitate a more thorough examination of how children think about their parent's illnesses and how illness perceptions impact psychological adjustment following their parent's diagnosis.

The findings of the present review should be considered in light of potential limitations. First, the heterogeneous nature of research exploring illness perceptions and differences in conceptualisations of 'illness perceptions' posed significant limitations. Although the search string was developed to identify studies that used specific terms relating to illness cognitions, perceptions or beliefs, a number of instruments that measured perceptual constructs that were associated with coping and psychological outcomes were included in the review. For the purposes of the review, illness perceptions were broadly defined as an individual's cognitive and affective characterisation of a family member's illness. This definition was used to guide decisions about whether an instrument should be included in the review and led to the inclusion of some instruments not specifically described as measures of illness perceptions by the study authors. To address limitations posed by the search string not including terms relating to perceived impact of illness, worry and uncertainty, and hence ensure completeness of study selection, the initial search was supplemented using forward and backward searching techniques that involved searching for articles that cited or were cited by the articles included in the review.

Second, the psychometric properties of many instruments were only described in one of the included studies, although two had been evaluated in more than one study. Previous systematic reviews that have assessed and compared the psychometric properties of instruments measuring the same (or similar) construct(s) have determined the overall quality of properties by reference to results reported across multiple studies. For example, Park et al. (44) determined an overall rating for each of the instruments in their study by using a level of evidence approach that considered the number of studies that had evaluated the instrument, their methodological quality and the consistency of their findings. Furthermore, the COSMIN guidelines recommend using the GRADE approach to summarise evidence of the quality of each psychometric property of an instrument based on the quality of methodology used to assess the property (27–29). In the present study, only two of the instruments (i.e. the Parental Experience of Child Illness questionnaire and the Parental Perception of Uncertainty

Scale) had been assessed in more than one study and neither had been assessed in sufficient studies for a level of evidence approach to support meaningful conclusions. The methodological quality of studies included in the review was assessed primarily for descriptive purposes; however, the assessments may be used by researchers and clinicians to guide instrument selection based on the quality of psychometric properties of an instrument and the quality of methodology used to assess psychometric properties in studies that have evaluated the instrument.

Third, a lack of published psychometric information limited the evaluation of psychometric properties for each of the instruments. Missing information or indeterminate ratings are not necessarily indicative of poor instrument quality, but instead may mean that the properties have not yet been evaluated or that the instrument is newly developed (26). It is worth noting that the initial search excluded articles that were not published in English, even though studies that described translated instruments were included in the review. It is possible that additional studies describing the development or psychometric evaluation of translated measures were published in languages other than English, in which case they would have been excluded from this review.

Finally, there are no standardised criteria to evaluate the quality of self-report instruments. However, the criteria published by Terwee et al. (26) were developed as a first attempt to establish a standardised assessment tool for the psychometric properties of measurement instruments (45). These criteria, or variations thereof, have been used in systematic reviews to identify the strengths and weaknesses of a range of self-report instruments, including those that measure health-related patient-reported outcomes (46,47), psychological constructs (40,44) and social constructs (48).

Despite these limitations, the present review provides important insights into the quality of psychometric properties of instruments designed to measure illness perceptions in family members and carers of an individual with a chronic physical illness. This information may be utilised by researchers and clinicians concerned with identifying individuals at risk for psychological morbidity following a family member's diagnosis. In general, findings highlight that further psychometric evaluation of instruments is needed and reporting of development and validation studies should be improved. Future research should aim to report as much information on psychometric properties and the methodology used to evaluate psychometric properties as possible, as a minimum information on content, structural and construct validity and internal consistency is needed to enable researchers to make informed decisions when choosing an instrument to use in their studies. Furthermore, almost all of the instruments were designed for parents with a child with a chronic illness. Research that aims to develop instruments to measure illness perceptions in children who have a parent with a chronic illness, or to validate the existing Perceptions of Parental Illness Questionnaire for use in different illness populations, is a much-needed addition to fill this gap in the current body of work examining the relationship between illness perceptions and psychological morbidity in family members of a person diagnosed with a chronic illness.

Conflict of interest

The authors have no real or apparent conflicts of interest to disclose.

Author contributions

Chloe Fletcher generated the initial research question; developed the research protocol and defined the parameters of the review; conducted the literature search; selected the papers for inclusion based on the eligibility criteria; extracted descriptive information from the included studies; conducted the evaluation of methodological quality of the included studies; conducted the evaluation of quality of psychometric properties of the included instruments; and wrote the manuscript. Ingrid Flight provided guidance throughout the review process; contributed as a second reviewer to selection of papers for inclusion based on the eligibility criteria; contributed as a second reviewer to evaluation of methodological quality of the included studies; and provided feedback on drafts of the manuscript. Kate Gunn provided guidance throughout the review process; contributed as a second reviewer to selection of papers for inclusion based on the eligibility criteria; and provided feedback on drafts of the manuscript. Pandora Patterson provided guidance throughout the review process and feedback on drafts of the manuscript. Carlene Wilson provided guidance throughout the review process; contributed to evaluation of quality of psychometric properties of the included instruments; and provided feedback on drafts of the manuscript.

Funding information

The authors did not receive any specific funding for this research.

References

1 Thomson FE, Bergman PB, Hay M. Psychological well-being of carers of children with type 1 diabetes: a comparison with carers of children without a chronic condition. *J Soc Health Diabet* 2018; 6: 96–103.

2 Mi E, Mi E, Ewing G, Mahadeva R, Gardener AC, Butcher HH, Booth S, Farquhar M. Associations between the psychological health of patients and carers in advanced COPD. *Int J Chron Obstruct Pulmon Dis* 2017; 12: 2813–21.

3 Li QP, Mak YW, Loke AY. Spouses' experience of caregiving for cancer patients: a literature review. *Int Nurs Rev* 2012; 60: 178–87.

4 Murray SA, Kendall M, Boyd K, Grant L, Highet G, Sheikh A. Archetypal trajectories of social, psychological, and spiritual well-being and distress in family caregivers of patients with lung cancer: secondary analysis of serial qualitative interviews. *BMJ* 2010; 340: c2581.

5 Patterson P, McDonald F, White K, Walczak A, Butow P. Levels of unmet needs and distress amongst adolescents and young adults (AYAs) impacted by familial cancer. *Psycho-Oncol* 2017; 26: 1285–92.

6 Mitchell AJ, Ferguson DW, Gill J, Paul J, Symonds P. Depression and anxiety in long-term cancer survivors compared with spouses and healthy controls: a systematic review and meta-analysis. *Lancet.* 2013; 14: 721–32.

7 Diefenbach MA, Leventhal H. The common-sense model of illness representation: theoretical and practical considerations. *J Soc Distress Homeless* 1996; 5: 11–37.

8 Hagger MS, Orbell S. A meta-analytic review of the Common-Sense Model of Illness Representations. *Psychol Health* 2003; 18: 141–84.

9 Petrie KJ, Weinman J. Why illness perceptions matter. *Clin Med* 2006; 6: 536–9.

10 Evers AW, Kraaimaat FW, van Lankveld W, Jongen PJH, Jacobs JWG, Bijlsma JWJ. Beyond unfavourable thinking: the Illness Cognition Questionnaire for chronic diseases. *J Consult Clin Psychol* 2001; 69: 1026–36.

11 Lazarus RS. *Stress and Emotion: A New Synthesis.* 1999, Springer, New York, NY, USA.

12 Leventhal H, Diefenbach MA, Leventhal EA. Illness cognition: using common sense to understand treatment adherence and affect cognition interactions. *Cognit Ther Res* 1992; 16: 143–63.

13 Beinke K, O'Callaghan FV, Morrissey S, Duregger C. Illness perceptions and psychological adjustment of mothers of young adults with cystic fibrosis. *Cogent Psychol* 2016; 3: 1207324.

14 Kazak AE, McClure KS, Alderfer MA, Hwang WT, Crump TA, Le LT et al Cancer-related parental beliefs: the Family Illness Beliefs Inventory (FIBI). *J Pediatr Psychol* 2004; 29: 531–42.

15 Sloper P. Predictors of distress in parents of children with cancer: a prospective study. *J Pediatr Psychol* 2000; 25: 79–91.

16 Sultan S, Leclair T, Rondeau E, Burns W. Abate C. A systematic review on factors and consequences of parental distress as related to childhood cancer. *Eur J Cancer Care* 2016; 25: 616–37.

17 Compas BE, Worsham NL, Ey S, Howell DC. When mom or dad has cancer: II. Coping, cognitive appraisals, and psychological distress in children of cancer patients. *Health Psychol* 1996; 15: 167–75.

18 Bogosian A, Moss-Morris R, Bishop FL, Hadwin J. Development and initial validation of the Perceptions of Parental Illness Questionnaire. *J Health Psychol* 2014; 19: 931–42.

19 Lazarus RS, Folkman S. *Stress, Appraisal, and Coping.* 1984, Springer, New York, NY, USA.

20 Rosenstock IM. The Health Belief Model and preventive health behavior. *Health Educ Monographs* 1974; 2: 324–508.

21 Ajzen I. From intentions to actions: a theory of planned behavior. In *Action Control* (Kuhl J, Beckmann J eds), 1985, Springer, Berlin, Heidelberg, 11-39.

22 Berk M, Berk L, Dodd S, Jacka FN, Fitzgerald PB, de Castella AR et al Psychometric properties of a scale to measure investment in the sick role: the Illness Cognitions Scale. *J Eval Clin Pract* 2012; 18: 360–4.

23 Weinman J, Petrie KJ, Moss-Morris R, Horne R. The Illness Perception Questionnaire: a new method for assessing the cognitive representation of illness. *Psychol Health* 1996; 11: 431–45.

24 Moss-Morris R, Weinman J, Petrie KJ, Horne R, Cameron LD, Buick D. The revised Illness Perception Questionnaire (IPQ-R). *Psychol Health* 2002; 17: 1–16.

25 Sint Nicolaas SM, Schepers SA, van den Bergh EM, Evers AW, Hoogerbrugge PM, Grootenhuis MA et al Illness cognitions and family adjustment: psychometric properties of the Illness Cognition Questionnaire for parents of a child with cancer. *Supp Care Cancer* 2016; 24: 529–37.

26 Terwee CB, Bot SDM, de Boer MR, van der Windt DAWM, Knol DL, Dekker J et al Quality criteria were proposed for measurement properties of health status questionnaires. *J Clin Epidemiol* 2007; 60: 34–42.

27 Mokkink LB, de Vet HCW, Prinsen CAC, Patrick DL, Alonso J, Bouter LM et al COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome Measures. *Qual Life Res* 2018; 27: 1171–9.

28 Prinsen CAC, Mokkink LB, Bouter LM, Alonso J, Patrick DL, de Vet HCW et al COSMIN guideline for systematic reviews of patient-reported outcome measures. *Qual Life Res* 2018; 27: 1147–57.

29 Terwee CB, Prinsen CAC, Chiarotto A, Westerman MJ, Patrick DL, Alonso J et al COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Qual Life Res* 2018; 27: 1159–70.

30 Mokkink LB, Prinsen CAC, Bouter LM, De Vet HCW, Terwee CB. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) and how to select an outcome measurement instrument. *Braz J Phys Ther* 2016; 20: 105–13.

31 Mokkink LB, Terwee CB, Patrick DL, Alonso J, Stratford PW, Knol DL et al The COSMIN checklist for assessing the methodological quality of studies on measurement properties of health status measurement

instruments: an international Delphi study. *Qual Life Res* 2010; 19: 539–49.

32 DeVet KA, Ireys HT. Psychometric properties of the Maternal Worry Scale for children with chronic illness. *J Pediatr Psychol* 1998; 23: 257–66.

33 Mishel MH. Parents' perception of uncertainty concerning their hospitalized child. *Nurs Res* 1983; 32: 324–30.

34 Bonner MJ, Hardy KK, Guill AB, McLaughlin C, Schweitzer H, Carter K. Development and validation of the parent experience of child inness. *J Pediatr Psychol* 2006; 31: 310–21.

35 Bemister TB, Brooks BL, Kirton A. Development, reliability, and validity of the Alberta Perinatal Stroke Project Parental Outcome Measure. *Pediatr Neurol* 2014; 51: 43–52.

36 Ye ZJ, Liu ML, Zhang Z, Liao KL, Peng CF, Huang H et al Psychometric properties of the Chinese version of the Parent Perception of Uncertainty Scale (PPUS) among parents of children with cancer diagnosis. *Int J Nurs Sci* 2017; 4: 278–84.

37 Molzon ES, Brannon EE, Fedele DA, Grant DM, Suorsa KI, Mullins LL. Factor structure of the parent perception of uncertainty scale in parents of children with cancer. *Psycho-Oncol* 2014; 23: 1435–8.

38 Bonner MJ, Hardy KK, Willard VW, Hutchinson KC, Guill AB. Further validation of the Parent Experience of Child Illness scale. *Child Health Care* 2008; 37: 145–57.

39 Santacroce S. Measuring parental uncertainty during the diagnosis phase of serious illness in a child. *J Pediatr Nurs* 2001; 16: 3–12.

40 Windle G, Bennett KM, Noyes J. A methodological review of resilience measurement scales. *Health Qual Life Outcomes* 2011; 9: 8.

41 Karademas EC, Zarogiannos A, Karamvakalis N. Cardiac patient-spouse dissimilarities in illness perception: Associations with patient self-rated health and coping strategies. *Psychol Health* 2010; 25: 451–63.

42 Wu LM, Mohamed NE, Winkel G, Diefenbach MA. Patient and spouse illness beliefs and quality of life in prostate cancer patients. *Psychol Health* 2012; 28: 355–68.

43 Searle A, Norman P, Thompson R, Vedhara K. Illness representations among patients with type 2 diabetes and their partners: Relationships with self-management behaviors. *J Psychosom Res* 2007; 63: 175–84.

44 Park T, Reilly-Spong M, Gross CR. Mindfulness: a systematic review of instruments to measure an emergent patient-reported outcome (PRO). *Qual Life Res* 2013; 22: 2639–59.

45 Rosenkoetter U, Tate RL. Assessing features of psychometric assessment instruments: A comparison of the COSMIN Checklist with other critical appraisal tools. *Brain Impairment* 2018; 19: 103–18.

46 Bot SDM, Terwee CB, van der Windt DAWM, Bouter LM, Dekker J, de Vet HCW. Clinimetric evaluation of shoulder disability questionnaires: a systematic review of the literature. *Ann Rheum Dis* 2004; 63: 335–41.

47 de Boer MR, Moll AC, de Vet HCW, Terwee CB, Volker-Dieben HJM, van Rens GHMB. Psychometric properties of vision-related quality of life questionnaires: a systematic review. *Opthalmic Physiol Opt* 2004; 24: 257–73.

48 Brohan E, Slade M, Clement S, Thornicroft G. Experiences of mental illness stigma, prejudice and discrimination: a review of measures. *BMC Health Serv Res* 2010; 10: 80.

49 Mishel MH. The measurement of uncertainty in illness. *Nurs Res* 1981; 30: 258–63.

Appendix C

Published manuscript for Study 2

International Journal of Behavioral Medicine (2019) 26:531–541
https://doi.org/10.1007/s12529-019-09793-4

Illness Cognitions Among Adolescents and Young Adults Who Have a Parent with Cancer: a Qualitative Exploration Using the Common-Sense Model of Self-regulation as a Framework

Chloe Fletcher[1] · Carlene Wilson[1,2,3] · Ingrid Flight[1] · Kate Gunn[1,4] · Pandora Patterson[5,6]

Abstract

Background Individuals construct beliefs about an illness based on their own perceptions, interpretation, and understanding of the illness and its treatment. These beliefs (collectively referred to as "illness cognitions" or "representations") can have implications for psychological outcomes in family members and carers of an individual with an illness. The aim of this study was to explore young people's perceptions of their parent's cancer using the Common-Sense Model of Self-Regulation as a theoretical framework.
Methods Semi-structured, one-on-one interviews were conducted with young people who had a parent diagnosed with cancer. Interview transcripts were analysed using deductive thematic analysis techniques.
Results Eleven young people aged 15–24 years participated in the study. Major themes aligned with the dimensions of the Common-Sense Model of Self-Regulation. Young people described their experiences with parental cancer with reference to cognitive representations (beliefs about the illness identity, their understanding or coherence of the illness, and consequences, curability or controllability, timeline, and cause of the illness) and emotional representations (emotional beliefs and subjective feelings about the illness).
Conclusions Findings indicate that young people's perceptions of their parent's cancer can be usefully described within the framework of the Common-Sense Model of Self-Regulation. Future research should investigate the relationships between young people's illness cognitions, coping strategies, and psychological adjustment following their parent's cancer diagnosis. This will provide valuable insights for the development of interventions that target specific types of illness cognitions associated with maladaptive coping strategies and poor adjustment.

Keywords Adolescent and young adult · Common-sense model of self-regulation · Illness cognitions · Parental cancer · Oncology · Qualitative research

Introduction

A parent's diagnosis of cancer can have a significant impact on the family and can be especially distressing for dependent children. Adolescents and young adults, who are at an age where they have the capacity to understand the potential implications of cancer for themselves and their family [1, 2], have been identified as experiencing higher levels of distress than younger children [3–5]. In Australia, it is estimated that 21,000 young people aged 12–25 years have a parent diagnosed with cancer each year [6], and more than half experience clinically-elevated levels of distress [7].

A range of factors are known to moderate young people's psychological adjustment following a parent's cancer diagnosis, including their age and gender, their parent's gender, time since parent's diagnosis, having unmet needs, and poor family functioning [3, 8–11]. Some findings also

---

✉ Chloe Fletcher
chloe.fletcher@flinders.edu.au

[1] Flinders Centre for Innovation in Cancer, College of Medicine and Public Health, Flinders University, Sturt Road, Bedford Park, SA 5042, Australia

[2] Olivia Newton-John Cancer Wellness and Research Centre, 145 Studley Road, Heidelberg, VIC 3084, Australia

[3] School of Psychology and Public Health, La Trobe University, Bundoora, VIC 3086, Australia

[4] University of South Australia Cancer Research Institute, University of South Australia, North Terrace, Adelaide, SA 5000, Australia

[5] CanTeen Australia, 75 King Street, Newtown, NSW 2042, Australia

[6] Cancer Nursing Research Unit, University of Sydney, Camperdown, NSW 2006, Australia

suggest that the way that children think about their parent's cancer is associated with their ability to cope with, and adjust to, the illness. For example, Compas et al. [5] found that children's perception of the stressfulness of their parent's cancer was associated with the development of anxiety and depression, and that this had a more significant impact than the stage of their parent's disease and their prognosis. Furthermore, they found that children generally perceived low levels of personal control over their parent's cancer, which may potentially prevent them from using problem-focused approaches to coping [5]. Despite these important findings, to the best of our knowledge, there has been no recent research exploring the beliefs that young people form about their parent's cancer and the potential impact of these beliefs on their psychological adjustment to the illness.

Leventhal's Common-Sense Model of Self-Regulation proposes an explicit causal link between an individual's beliefs about an illness, the coping strategies that they employ in response to the illness, and their physical health and psychological outcomes (see Fig. 1) [14–16]. An individual constructs beliefs about an illness based on their own perceptions, interpretation, and understanding of the illness and its treatment [15, 17, 18]. These beliefs are collectively defined as "illness cognitions" or "representations", and are not necessarily scientifically or medically substantiated [19]. They are formulated based on information provided by three broad domains of information: previous social interactions, personal experiences, and cultural knowledge of the illness; external social influences from perceived important others or authoritative figures; and current experiences with the illness [15]. Illness cognitions are complex and dynamic [20], evolving over time in response to internal and external stimuli, for example; emerging side effects of treatment or public opinion about the causes of the illness [19].

The Common-Sense Model of Self-Regulation is conceptualized as a parallel processing framework, whereby an individual simultaneously constructs cognitive and emotional representations of an illness [15]. Cognitive representations are organized around five central themes or dimensions: identity (the label used to describe the illness and perceptions of symptoms associated with the illness), consequences (beliefs about the anticipated or experienced impact of the illness on quality of life or physical, cognitive, or social capacity), cure or control (beliefs about the efficacy of treatment or the extent to which the individual has personal control over the illness), timeline (beliefs about the duration or progression of the illness), and cause (beliefs about the factors responsible for causing the illness). These dimensions are considered to be the "basic building blocks" of investigations into how individuals construct beliefs about illness [15]. The Common-Sense Model of Self-Regulation reflects an ongoing interplay between empirical data and theoretical concepts [21]. As a result, the original model has been extended through the inclusion of an additional cognitive representation dimension of coherence (personal understanding or comprehension of the illness) [22]. Other changes have included incorporating a cyclical timeline dimension, and splitting the cure or control dimension into separate dimensions of treatment control and personal control [22].

In response to an illness or health threat, an individual is thought to employ coping strategies that are consistent with their own representation of the illness or threat. For example, research indicates that perceptions of serious consequences are associated with avoidance and denial coping strategies, such as behavioural disengagement [23]. Furthermore, perceptions that the illness is uncontrollable, chronic, and highly symptomatic have also been associated with avoidance and denial coping strategies [15]. In contrast, perceptions that the illness is controllable have been associated with active coping strategies, including problem-focused coping [24]. This

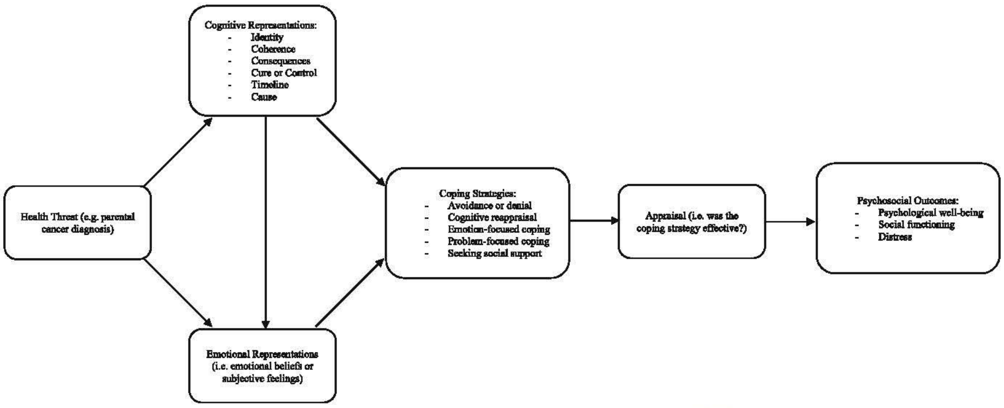


Fig. 1 Leventhal's Common-Sense Model of Self-Regulation applied to parental cancer [adapted from 12, 13]

research highlights the importance of understanding these associations; by identifying and modifying beliefs that are associated with maladaptive coping strategies, an individual's distress may be reduced, potentially resulting in better psychological adjustment [15].

Although the model was originally developed to describe "lay" understanding of illness experiences in individuals diagnosed with an illness [25], it has also been used to explore illness cognitions in family members and significant others caring for someone experiencing ill-health [26–33]. Bogosian et al. [29] used the model to describe illness cognitions among adolescents who have a parent with multiple sclerosis. Using a quantitative measure, developed for specific use in this cohort, they found that stronger subjective beliefs about the negative consequences of their parent's illness and a perception that it was chronic and unpredictable were associated with poorer psychological adjustment, independent of parental report of illness severity [29].

Based on these findings, the Common-Sense Model of Self-Regulation is proposed as an appropriate theoretical framework for exploring illness cognitions in young people who have a parent with cancer. The aim of this study was to explore young people's perceptions of their parent's cancer and examine how these perceptions correspond with the dimensions of illness cognitions as understood within the Common-Sense Model of Self-Regulation.

Methods

Design

Semi-structured, one-on-one interviews were conducted with young people who had a parent diagnosed with cancer. Interviews explored young people's experiences following their parent's diagnosis and their cognitive and emotional representations of their parent's cancer. Open-ended questions included in the interview schedule (Table 1) were based on the Common-Sense Model of Self-Regulation.

Participants

Participants were recruited through advertisements distributed online through cancer support and advocacy groups, including CanTeen[1] and Australian-based cancer support groups on social media. All advertisements contained information about the study and interested young people (or their parents) were asked to contact the researcher via telephone or email if they (or their child) were interested in participating in the study.

Table 1 Interview schedule outlining relevant dimension of the Common-Sense Model of Self-Regulation (questions asked in past tense for bereaved participants and participants whose parent had finished active treatment)

1. Could you tell me about your experience with parental cancer? (illness identity, treatment control)
a. Which of your parents was diagnosed with cancer?
b. What type of cancer were they diagnosed with?
c. What type of treatment have they had?
2. What was it like when your parent was first diagnosed with cancer? (emotional representations)
a. How did you react when you first found out?
b. How did your family deal with your parent's diagnosis?
3. In general, how do you feel when you think about your parent's cancer? (emotional representations)
4. How has your parent's cancer impacted your life? (consequences)
a. In what way has your parent's cancer negatively impacted your life?
b. Has your parent's cancer had a positive impact on your life in any way?
5. How has cancer affected your parent? (illness identity)
6. What symptoms and side effects has your parent experienced as a result of their cancer? (illness identity)
7. How do you think your parent's cancer might affect them in the long term?[a] (timeline, treatment control)
8. Can you tell me about any changes you have noticed in the way you act around your parent since their diagnosis? (personal control)
a. What sort of things do you do that you feel make a difference to their illness or how they are feeling?
9. Can you tell me about the sort of things that you might talk about with your parent or with your family about your parent's cancer? (coherence)
10. Can you tell me about what you think of the information that has been shared with you about your parent's cancer? (coherence)
a. Do you feel like you know enough about your parent's cancer?
b. Is there anything that you might like to know more about?
11. Do you think there is anything your parent could have done to avoid cancer? (cause)
a. Do you have any thoughts on what might have caused your parent's cancer?
12. Is there anything else you would like to add to what has already been said?

[a] Bereaved participants were not asked this question

Young people aged 12–24 years were eligible to participate if they had a parent who had been diagnosed with any type or stage of cancer, were aware of their parent's cancer diagnosis, and were living at home at the time of their parent's diagnosis and treatment. This included young people who had a parent who had been diagnosed with curable, metastatic, or advanced cancer, and young people who had a parent who had died from cancer. Although there are various definitions of 'youth' (e.g. 15–24 years as defined by the United Nations [34] and 10–24 years as defined within the scientific literature [35]), the age range of 12–24 years was chosen because it aligns with

---

[1] CanTeen is an Australian support organization for young people affected by cancer, including young people diagnosed with cancer, and young people who have a brother, sister, or parent diagnosed with cancer.

definitions provided by the Australian Institute of Health and Welfare [36].

Eleven young people from ten families participated in the interviews (age range = 15–24 years; $M = 19.1$ years; female = 7). Of those, two participants had both parents who had been diagnosed with cancer, two participants were bereaved, and two had a parent with advanced or metastatic cancer, one had a parent who was still undergoing treatment for a curable form of cancer, and four had a parent who had finished active treatment. One participant's father had been diagnosed with several unrelated types of cancer. Time since initial diagnosis ranged from 6 months to 8 years. Parental cancer diagnoses included non-Hodgkin's lymphoma, lung, bowel, pancreatic, kidney (including collecting duct carcinoma), testicular, ovarian, cervical, and breast cancer. Participants described various household structures; two participants lived alone with a single parent, three participants lived with a single parent and other relatives (two with siblings and one with grandparents), and six participants lived with both parents (four with siblings and one with an aunt). Of those that lived with both parents, two participants lived with parents who were separated but had moved in together in order to fulfil a caregiver role.

Interviews

Interviews were conducted by the first author (CF). CF is a PhD researcher with a background in social health sciences and previous research experience in cancer support. The researcher had no contact with participants prior to the study commencement. Interviews were conducted either face-to-face, via telephone, or via a web-conferencing programme. Face-to-face interviews were conducted at the CanTeen Division office in Sydney, Australia. Interviews ranged from 9 to 37 min in length ($M = 23$ min). Interviews were audio-recorded and transcribed verbatim using a professional transcription service. The interview schedule was used flexibly to explore participants' cognitive and emotional representations of their parent's cancer in relation to the dimensions of the Common-Sense Model of Self-Regulation. All participants were asked each of the questions outlined in the interview schedule, with the exception of bereaved participants who were not asked about their thoughts on how their parent's cancer might affect them long term. Probing questions were used to obtain more specific or in-depth information on emerging themes. Similar themes were apparent within all interviews and recruitment was finalized when no new themes emerged in three consecutive interviews.

Ethical Considerations

The study was reviewed and approved by the Social and Behavioural Research Ethics Committee at Flinders University (project number 7436). All participants received a research pack containing written information about the study, formal written consent forms, and a questionnaire to collect participant demographic and parent cancer-related medical information. A copy of the interview schedule was also included to enable potential participants to make a fully informed decision about participating in the study. Parental consent was obtained for participants younger than 18 years. Participants returned signed consent forms and completed demographics questionnaires to the researcher prior to the interview. Participants were given assurances regarding the confidentiality and anonymity of the information they shared during the interview. Participants were informed that they were able to withdraw from the study at any time.

Analysis

Following previous qualitative research exploring illness perceptions using the Common-Sense Model of Self-Regulation as a framework [25, 29, 37], thematic analysis methods were used to analyse the data. Thematic analysis involves identifying themes or patterns of meaning across a qualitative data set [38]. It is a flexible method that does not prescribe methods for data collection, theoretical positions, or epistemological or ontological frameworks [38]. In the present study, a combination of deductive (theory-driven) and inductive (data-driven) approaches were employed to identify and organize themes using the Common-Sense Model of Self-Regulation as a framework, while also allowing themes outside of the framework to be identified to ensure that the data were analysed comprehensively.

Braun and Clarke's [38, 39] steps for coding and analysing qualitative data were followed to conduct the thematic analysis. Interview transcripts were verified for accuracy prior to analysis and were read-through several times for familiarization. Interview transcripts were initially coded by the first author (CF) according to the dimensions of the model and the remaining text was coded according to additional themes derived from the data. Themes were reviewed to ensure that data within themes were coherent, and that there were clear and identifiable distinctions between themes. Following this, a codebook was developed by the first author to outline definitions of codes. A sub-set of transcripts (30%) was coded independently by a second researcher with a background in psychology and qualitative research (see Acknowledgements), who was not part of the research team, through reference to the codebook. Discrepancies in coding were resolved through discussion until consensus on definition of codes and interpretation of data was reached.

This paper describes the themes that related to the Common-Sense Model of Self-Regulation; additional themes that were identified were related to interpersonal aspects of the cancer experience (e.g. social support and communication about cancer), rather than young people's cognitive and emotional representations of their parent's cancer.

Results

In general, participants expressed appreciation for the opportunity to share their experience and they hoped that this would, in some way, help other young people in a similar situation. Most young people spoke openly during the interview, with very minimal prompting. Two participants required more direct prompting with probing questions; these interviews were the two shortest in length (9 and 14 min). One of these participants had a parent with terminal cancer, possibly contributing to her difficulty in answering some of the questions. The other participant's parent had a recent diagnosis, and hence was at the beginning of the cancer experience, also potentially contributing to his short responses, as some of the questions asked may not have been relevant to his individual experience. Analyses confirmed that the Common-Sense Model of Self-Regulation could be utilized to describe young people's experience of parental cancer and that these experiences were varied. Definitions and illustrative quotes for themes and subthemes are presented in Table 2.

Emotional Representations

Young people described a mixture of initial feelings after learning of their parent's cancer diagnosis: shock (e.g. "it felt like all the breath had been knocked out of me"), confusion (e.g. "I didn't really understand what, the magnitude of what it was, or like, how to feel or what to do"), numbness (e.g. "the immediate reaction, it was for me, it was a bit muted"), a loss of control (e.g. "It felt like I had no control back over my life"), and, in contrast, a level of acceptance (e.g. "I guess I was prepared for it to be cancer"). Participants described their experiences since their parent's diagnosis as "surreal", "a bumpy ride" and "very full on, very confronting". In general, young people felt fear over the possibility of losing their parent and uncertainty about the future, despite whether their parent was in remission, undergoing curative treatment, or receiving palliative care.

Identity

All participants were able to describe the type of cancer that their parent had been diagnosed with; many were also able to describe the stage of cancer. Participants spoke about the impact of common side effects including fatigue, hair loss, nausea, weight gain, weight loss, and problems with memory or concentration. Some participants spoke about the high cost of symptoms like fatigue or problems with memory or concentration; in particular for participants who had a parent with terminal cancer. This was noted by one participant as she recounted the last few months spent with her dad: "…we wanted to spend time with him but he just, I don't know, couldn't – didn't have the thinking capacity to do it.". This experience was similar to another participant who described the impact of her mother's fatigue:

> "…in the last few months, probably just her being really exhausted all the time – having to not just rest sort of once a day or occasionally but being – becoming, I guess, less … around and available in some ways and probably … energy wise, just not being able to sort of talk with people for a longer period of time."

Young people also spoke about the emotional side effects of cancer; cancer did not just impact their parent physically, it was also a "mental game". One participant described the changes in her mother since her diagnosis with ovarian cancer: "…emotionally she's a lot more anxious about things and she cries a lot and gets down a fair bit". Some participants felt that although their parent was likely experiencing a number of physical symptoms, this was not always expressed; for example, two participants described their parent's pain as being something that was not really "complained about" or "overemphasised", despite being ever-present.

Coherence

Young people described a desire to be informed about their parent's cancer, and they achieved this by accompanying their parent to doctor's appointments, speaking with their parent's treating doctors, researching the disease in their own time, and having open communication with their parent about their cancer. One participant described checking in with his mother to "see how it's all going, how the treatment's going … [and to] see if she's alright." Another participant commented that he found knowing as much as he could about his mother's cancer beneficial:

> "…I just tried to get as much information as possible about it, and I think that helped me to accept that, helped me to really, like reduce the anxiety just knowing this, like percentages of chance of it coming back, and all that, I think, because that was one of the ways I coped by like, information and reading about it, and knowing as much as I could about it."

In contrast, some participants felt that knowing too much would have been overwhelming:

> "I wanted to know enough information to get me by, but not enough information to know every detail. …because

Table 2 Themes and sub-themes identified from the deductive analysis

Main theme (dimension of Common-Sense Model)	Definition	Sub-theme	Illustrative quotes from young peoples' interviews
1. Identity	Young peoples' perceptions of the side effects and symptoms that their parent experiences due to their cancer		"She did not really eat, she was very nauseous and she spent a lot of time sleeping."
2. Emotional representations	Young peoples' emotional beliefs and subjective feelings about their parent's cancer		"I think, shock and confusion, just because I did not really understand what, the magnitude of what it was, or like, how to feel or what to do."
3. Coherence	Young peoples' understanding or comprehension of their parent's cancer		"I wanted to know enough information to get me by, but not enough information to know every detail."
4. Consequences	Young peoples' perceptions of the impact their parent's cancer has had on their daily life	1. Young person	"I've just become a lot more anxious kind of person, yeah. It's really changed my perspective on things and there's been like actually a bit of role reversal, so because my mum's a single parent, yeah, so it's actually like I am kind of mothering her in a sense."
	Young peoples' perceptions of the impact their parent's cancer has had on their family life	2. Family	"…it's caused a lot of tension in our house as well because this is so much stress all of the time."
	Young peoples' perceptions of the impact their parent's cancer has had on their parent's life	3. Parent	"…she's also figured out who her true friends are, and emotionally that has really helped her, she feels a lot lighter, she even says 'That whole experience, I know who my true friends are, because they were the ones that offered to come to a round of chemo, or offered to take me to radiation' …"
5. Cure or control	Young peoples' perceptions of the personal control they have over their parent's cancer symptoms, side effects, and distress	1. Personal Control — Relieving Symptoms and Distress	"…you had to sort of have a calming presence, you could not be loud and yelling or anything like that, because if it stressed her out … she would feel sicker and you did not want to her feel unhappy and unwell."
	Young peoples' perceptions of the personal control they have over maintaining a normal life separate from their parent's cancer	2. Personal Control — Maintaining Normality	"…when they are sick and that sort of idea of someone's helpless, I guess, is kind of pretty prevalent in society in general. So, I think just making sure that you do not replicate that as much as possible at home and treating family members as human…"
	Young peoples' perceptions of the control treatment can have over their parent's cancer	3. Treatment control	"…she's got like medically it's stage 4 breast cancer, secondary breast cancer so it's caused by the production of the hormones but it's coming from her bones, so it's in her bones so it's like it's inoperable. So essentially it's pretty much just keeping it under control but it's not going to go away."
6. Timeline	Young peoples' perceptions of the expected duration and progression of their parent's cancer		"…that's all pretty uncertain, they have sort of said to us, this is incurable and we know that the end is coming soon but they will not give us a time frame … it's kind of like switched between denial and … she'll be fine and then thinking that she's going to die like tomorrow."
7. Cause	Young peoples' perceptions of the causes of their parent's cancer	1. Chance	"…to me it just feels like it picks a random person in the world there's no actual reason for it, because there's no history of it going through our family either so."
		2. Environmental or external attributions	"I've heard that people have said that, other bad people in their lives can cause cancer, …"
		3. Genetic/hereditary attributions	"…cancer has also been a fairly genetic sort of disease going through – I mean her father passed away when she was fifteen or something of cancer and he had a melanoma in the eye or something and she had a few other relatives that've passed away early because of cancer. So whether or not there is some sort of gene in your DNA that has something to do with the likelihood of her developing cancer, …"
		4. Lifestyle attributions	"…she eats everything I eat, she drinks whereas I do not, but drinking usually is a liver issue not just any random cancer issue. She smokes but it wasn't lung cancer it wasn't anything that could have linked to that. So I worry more now that she's still smoking because well I feel like that might cause it to come back sooner or things like that."
		5. Psychological attributions	"…I think she told me that it was stress."

> it almost made it too real, and I kind of wanted to keep this fantasy land that we were all okay."

A similar sentiment was shared by another participant who commented that "it's good to be a little bit oblivious".

Consequences

Young people described a range of consequences that their parent's cancer had for them, their family, and their parent. For many, the most prominent consequence for themselves was having to take on extra household responsibilities:

> "…I would often come home from school and I'd cook dinner and I'd clean the house and I found myself taking on a lot more responsibilities just because my mum wasn't able to…"

Many young people also accompanied their parent to treatment and other medical appointments and provided emotional support. Two participants likened this to fulfilling a parental role, with one stating that: "… it's actually like I am kind of mothering her in a sense." Taking on extra responsibilities had a flow-on effect on schooling:

> "…my results went down the drain, because I didn't have time to study for exams on top of looking after mum, and looking after my little brother after he got home from school."

One participant spoke about how she had considered dropping out of school to work full-time and care for her mother. Some described this not necessarily as a negative consequence of their parent's cancer, but as a shift in their priorities. In particular, many participants spoke about how the experience brought their family closer together and taught them to be more appreciative of the time they had together. One participant described it in this way:

> "…we care more about going to things and it's important to go watch someone in the football grand final, because you don't know what could happen the next day. So I think everything has become more important to us."

Young people also spoke about the consequences for their parent. In particular, two participants described how breast cancer had negatively impacted their mothers' self-esteem. One participant reflected on her mother losing her hair after chemotherapy: "I went from seeing my mum being such a strong, beautiful woman to someone who felt so insecure." Another participant stated that her mother no longer wore swimsuits because of the visible scars on her breasts.

Young people also highlighted the positive consequences for their parent. One participant stated that his mother was "pretty proud of herself that she's made it". Several participants spoke about how cancer had improved their parent's relationships by helping them to "figure out who [their] true friends are" and had given them "a new fresh start … a start again, and at a new place, with different people".

Cure or Control

Young people were very well-informed about what could be done medically to treat their parent's cancer. One participant, whose mother was diagnosed with metastatic breast cancer, described the difference between treating to cure and treating to control: "…essentially it's pretty much just keeping it under control, but it's not going to go away." Another participant spoke about how chemotherapy reduced her mother's risk of cancer recurrence, although she acknowledged that the risk was not completely eliminated. Frustration was expressed by another participant who stated that he had been "stonewalled" by an oncologist who deemed his mother too unwell to try an experimental treatment for her rare form of cancer.

Young people gained a sense of personal control over the cancer experience by doing anything that they could to "make [their parent's] life easier or less stressful". One participant said that she felt that this "made it just so much easier for [her mother] because she didn't have to worry about those things, she could concentrate on the things that she felt were most important to her." Another participant described how changing the way he behaved around his mother could help to make her feel better:

> "…you had to sort of have a calming presence, you couldn't be loud and yelling or anything like that, because if it stressed her out … she would feel sicker and you didn't want to her feel unhappy and unwell."

For some participants, this meant shielding their parent from their emotional reactions to their cancer. One participant stated that she "hid away [her] feelings because it – she [mother] was really upset and we just needed to get things done…" Another participant stated that she "didn't want her [mother] to know that it was affecting me in such a great way until I knew she would be able to cope with the fact that, it was obviously affecting me as well as her."

Personal control over the cancer experience was as much about what could be done to support the parent or relieve their symptoms and distress as it was about preventing cancer from becoming the parent's identity; one participant stated that it was important to make sure that she did not make her mother feel like she was helpless just because she was sick; as much as possible, she wanted to avoid her mother "becoming the cancer patient … in every facet of life". There was also a desire to maintain a normal life outside of the cancer experience; that although cancer was a big aspect of their family's lives, "it [didn't] have to be the focus".

Timeline

Young people spoke about cancer as being a "battle". One participant expressed this through her belief that "my mum's not going to die, my mum's going to win this battle." For others, their parent's future was much less certain. One participant reflected on the unpredictable timeline of his father's cancer: "it's a battle for him, and it's touch and go, whether or not he lives or dies". Similarly, other young people who had a parent with terminal cancer expressed a lot of uncertainty over their parent's future and a desire to know exactly how much time they had left with their parent:

> "…that's all pretty uncertain, they've sort of said to us, this is incurable and we know that the end is coming soon but they won't give us a time frame … it's kind of like switched between denial and … she'll be fine and then thinking that she's going to die like tomorrow."

Although this uncertainty was a continuous stress for those with a parent with terminal cancer, young people showed resilience through their ability to adapt to the situation. One participant commented that she and her family had learned to take it "day by day … and well we're kind of figuring out like a pattern now."

Those who had a parent in remission spoke about their fears that the cancer might come back in the future, although one participant commented that these fears "have slowly dissipated and gone away" over time. Another participant was able to rationalize her fears that her mother's cancer would return: "…I figured with the amount of chemicals and treatment they gave her, at the moment, for the next couple of years, I know that we're okay and it's not going to come back."

Cause

Although they were able to offer a range of potential causes for their parent's cancer, including biological, environmental, lifestyle, or psychological causes, many participants felt that cancer was something that "just happens". One participant felt that in her mother's case "[cancer] picks a random person in the world, there's no actual reason for it, because there's no history of it going through our family".

Young people also talked about their fears about their own risk of developing cancer in the future (e.g. "I get worried of is it going to be me in five years or ten years or however long…") and how their attitude towards cancer prevention behaviours has changed in response to their parent's cancer (e.g. "Even things like putting sun block on, I'm so crazy, obsessive about putting it on now…").

Discussion

These findings extend previous research examining young people's experiences with parental cancer by providing an insight into young people's perceptions of, and emotional reactions to, their parent's cancer. Results confirmed the relevance of the dimensions of the Common-Sense Model of Self-Regulation for describing illness cognitions in young people following their parent's diagnosis with cancer.

The 11 young people who participated in interviews were a diverse sample; but despite this, there were common themes in the experiences they reported. Participants described negative emotional reactions, such as feelings of fear, uncertainty, and a loss of control, in response to their parent's cancer. These feelings were not just limited to participants who had a parent with terminal cancer or who were reflecting on the time before their parent had died; they were shared by participants who had a parent in remission. Many still feared that the cancer could return.

Participants described having an active involvement in helping their parent during their cancer experience; through attending their parent's appointments, researching their parent's cancer in their own time, or talking with their parent about treatment options. They had a coherent understanding of their parent's cancer, their treatment, and what could be expected in terms of the progression of the disease, and indicated that knowing as much information as possible about their parent's cancer was beneficial. This is consistent with findings reported by Maynard et al. [40], where adolescent participants described that having information about their parent's cancer and its treatment was important in helping them cope. Using a problem-focused coping strategy, whereby young people play an active role in supporting their parent through their illness, may improve their sense of control over the situation. This sentiment was not shared by all participants however; one participant felt that knowing too much about her parent's cancer would erase the illusion that everything was okay, suggesting a personal preference for a more emotion-focused coping strategy, such as denial or avoidance.

Participants described taking on extra responsibilities that would have otherwise been fulfilled by their parent, following their parent's diagnosis. These included preparing meals, cleaning the house, looking after pets, and babysitting younger siblings. Although almost all participants spoke about taking on these extra responsibilities, this was particularly notable for those with a single parent. Many participants described how having these responsibilities impacted their schooling. For some, this meant considering whether or not to drop out of school to find full-time work and care for their parent. Others felt that this shift in their priorities was a positive experience, because it made them focus on what was important in their lives — their family. Benefit finding, defined as "the process of deriving positive growth from adversity" [41], has been shown to lead to positive outcomes among family caregivers [42, 43], and may be an important aspect of coping for young people following a parent's cancer diagnosis.

Several participants described their parent's cancer as a "battle". The use of violence-related metaphors for the cancer experience has been described previously both in peer-reviewed literature [44, 45] and in the media [46–49] as potentially detrimental for people diagnosed with cancer. These kinds of metaphors position cancer as an opponent — an enemy to defeat — and may contribute to feelings of helplessness and anxiety. In particular for people with terminal cancer, this can cause feelings of failure or guilt for losing the "fight" against cancer [49]. For young people who have a parent with terminal cancer, this may generate feelings of resentment towards their parent for not "fighting hard enough" and lead to difficulties accepting their parent's diagnosis and eventual death. Instead it may be more helpful to view cancer as a natural process — as something that "just happens" — as several participants described when reflecting on possible causes of their parent's cancer. This belief is substantiated by research showing that two-thirds of all cancers are caused by unavoidable random errors that occur during DNA replication [50]. Changing the rhetoric around the cancer experience by avoiding the use of violence-related metaphors that place blame on the person with the diagnosis and emphasizing the random nature of the disease may be helpful for young people to reach an acceptance of their parent's cancer and prognosis.

Facing the potential death of a parent is not an experience that is normal in adolescence or young adulthood [51]. Despite their difficult and unusual circumstances, many participants did what they could to maintain a sense of normality in their life. For some, this involved ensuring that family life did not revolve around their parent's cancer, by drawing a distinction between their parent as a mother or father versus a "cancer patient". This attempt to separate the disease from the family has been previously described as adaptive. In particular, a family identity that revolves around the parent with cancer for an extended period of time may negatively affect the family's ability to meet the developmental needs of all family members, particularly dependent children [2]. Although taking ownership of the disease appears to be an important aspect of perceived control for the person with cancer [52], maintaining a sense of normality — an identity separate from the cancer — may be an important coping strategy for young people when their parent has cancer.

The findings of this study should be considered in light of some potential limitations. Participants were recruited primarily through an organization aimed at supporting young people through a cancer experience. Furthermore, participants were self-selected. As a result, the findings may not be representative of the experiences of young people outside of the organization. The sample size was quite small, however, this is a small and difficult-to-recruit population. Talking about parental cancer can be uncomfortable and potentially distressing, and it is possible that many young people may have not wanted to participate in the study for this reason. The young people who participated in the study had varied experiences with parental cancer and varied in the length of time since their parent's initial diagnosis. Despite this, there were common themes among the experiences that young people reported and they were easily able to recall and recount their experiences in great detail.

The findings of this research indicate that young people's perceptions of their parent's cancer can be usefully described within the framework of the Common-Sense Model of Self-Regulation. It should be noted that it was not the aim of this research to make conclusions about the relationships between specific types of illness cognitions, coping strategies, and psychological morbidity, but instead to examine how young people's perceptions of their parent's cancer correspond with the dimensions of the Common-Sense Model of Self-Regulation. The present study extends previous research by exploring young people's perceptions of their parent's cancer in the context of a theoretical framework. This enables the opportunity for measures based on the model to be developed and used in further research, and potentially clinical practice, to identify those at risk of poor psychological adjustment following a parent's cancer diagnosis. Hence, this work may provide a basis for the development of quantitative measures of illness cognitions in young people who have a parent with cancer.

Future quantitative research should expand on these findings by determining the relationships between young people's illness cognitions, coping strategies, and psychological adjustment following their parent's cancer diagnosis. Quantitative research should be conducted, using a larger sample size, to examine differences in illness cognitions among young people of different ages, and to ensure that the findings may be generalized and replicated within the wider cancer population. Future research may also explore how young people's perceptions of their parent's cancer impact their own coping with personal cancer symptoms as an adult, using a longitudinal design. This research may provide valuable insight for the development of interventions that target harmful beliefs and promote positive coping strategies to support young people through the cancer experience.

Acknowledgements The authors would like to thank Paige Fletcher, who is a research assistant at Flinders University, for her assistance with coding interview transcripts. The authors especially thank the young people who participated in this research for their willingness to share their experiences.

Funding This study was funded by the Flinders Foundation, with the support of its donors and partners.

Compliance with Ethical Standards

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

Conflict of Interest The authors declare that they have no conflict of interest.

References

1. Gazendam-Donofrio SM, Hoekstra HJ, van der Graaf WT, et al. Adolescents' emotional reactions to parental cancer: effect on emotional and behavioral problems. J Pediatr Psychol. 2011;36(3):346–59. https://doi.org/10.1093/jpepsy/jsq090.
2. Pederson S, Revenson TA. Parental illness, family functioning, and adolescent well-being: a family ecology framework to guide research. J Fam Psychol. 2005;19(3):404–19. https://doi.org/10.1037/0893-3200.19.3.404.
3. Visser A, Huizinga GA, van der Graaf WT, Hoekstra HJ, Hoekstra-Weebers JE. The impact of parental cancer on children and the family: a review of the literature. Cancer Treat Rev. 2004;30(8):683–94. https://doi.org/10.1016/j.ctrv.2004.06.001.
4. Huizinga GA, Visser A, Zelders-Steyn YE, Teule JA, Reijneveld SA, Roodbol PF. Psychological impact of having a parent with cancer. Eur J Cancer. 2011;47(Suppl 3):239–46. https://doi.org/10.1016/S0959-8049(11)70170-8.
5. Compas BE, Worsham NL, Ey S, Howell DC. When mom or dad has cancer: II. Coping, cognitive appraisals, and psychological distress in children of cancer patients. Health Psychol. 1996;15(3):167–75. https://doi.org/10.1037/0278-6133.15.3.167.
6. Patterson P, McDonald FEJ, Orchard P. A new Australian online and phone mental health support service for young people living with cancer. Aust Psychiatry. 2014;22(2):165–9. https://doi.org/10.1177/1039856213519144.
7. Patterson P, McDonald FEJ, White KJ, Butow PN, Walczak A. Levels of unmet needs and distress in AYAs impacted by familial cancer. Psycho-Oncology. 2016;26(9):1285–92. https://doi.org/10.1002/pon.4421.
8. Krattenmacher T, Kuhne F, Ernst J, Bergelt C, Romer G, Moller B. Parental cancer: factors associated with children's psychosocial adjustment - a systematic review. J Psychosom Res. 2012;72(5):344–56. https://doi.org/10.1016/j.jpsychores.2012.01.011.
9. Su YH, Ryan-Wenger NA. Children's adjustment to parental cancer: a theoretical model development. Cancer Nurs. 2007;30(5):362–81; quiz 82-3. https://doi.org/10.1097/01.NCC.0000290817.37442.e6.
10. Ellis SJ, Wakefield CE, Antill G, Burns M, Patterson P. Supporting children facing a parent's cancer diagnosis: a systematic review of children's psychosocial needs and existing interventions. Eur J Cancer Care (Engl). 2016;26(1). https://doi.org/10.1111/ecc.12432.
11. McDonald FEJ, Patterson P, White KJ, Butow PN, Costa DSJ, Kerridge I. Correlates of unmet needs and psychological distress in adolescent and young adults who have a parent diagnosed with cancer. Psycho-Oncology. 2016;25(4):447–54. https://doi.org/10.1002/pon.3942.
12. Hagger MS, Koch S, Chatzisarantis NLD, Orbell S. The common sense model of self-regulation: meta-analysis and test of a process model. Psychol Bull. 2017;143(11):1117–54. https://doi.org/10.1037/bul0000118.
13. Joice S. Self-management following stroke. Nurs Stand. 2012;26(22):39–46. https://doi.org/10.7748/ns2012.02.26.22.39.c8919.
14. Gray SE, Rutter DR. Illness representations in young people with chronic fatigue syndrome. Psychol Health. 2007;22(2):1–16. https://doi.org/10.1080/14768320600774595.
15. Hagger MS, Orbell S. A meta-analytic review of the common-sense model of illness representations. Psychol Health. 2003;18(2):141–84. https://doi.org/10.1080/088704403100081321.
16. Hoving JL, van der Meer M, Volkova AY, Frings-Dresen MHW. Illness preceptions and work participation: a systematic review. Int Arch Occup Environ Health. 2010;83(6):595–605. https://doi.org/10.1007/s00420-010-0506-6.
17. Petrie KJ, Weinman J. Why illness perceptions matter. Clin Med. 2006;6(6):536–9. https://doi.org/10.7861/clinmedicine.6-6-536.
18. Diefenbach MA, Leventhal H. The common-sense model of illness representation: theoretical and practical considerations. J Soc Distress Homeless. 1996;5(1):11–37. https://doi.org/10.1007/BF02090456.
19. Browning KK, Wewers ME, Ferketich AK, Otterson GA, Reynolds NR. The self-regulation model of illness applied to smoking behavior in lung cancer. Cancer Nurs. 2010;32(4):E15–25. https://doi.org/10.1097/NCC.0b013e3181a0238f.
20. Leventhal H, Phillips LA, Burns E. The common-sense model of self-regulation (CSM): a dynamic framework for understanding illness self-management. J Behav Med. 2016;39(6):935–46. https://doi.org/10.1007/s10865-016-9782-2.
21. Leventhal H, Phillips LA, Burns E. Modelling management of chronic illness in everyday life: a common-sense approach. Psychol Top. 2016;25(1):1–18.
22. Moss-Morris R, Weinman J, Petrie KJ, Horne R, Cameron LD, Buick D. The revised Illness Perception Questionnaire (IPQ-R). Psychol Health. 2002;17(1):1–16. https://doi.org/10.1080/08870440290001494.
23. Moss-Morris R, Petrie KJ, Weinman J. Functioning in chronic fatigue syndrome: do illness perceptions play a regulatory role? Br J Health Psychol. 1996;1(1):15–25. https://doi.org/10.1111/j.2044-8287.1996.tb00488.x.
24. Kemp S, Morley S, Anderson E. Coping with epilepsy: do illness representations play a role? Br J Clin Psychol. 1999;38(1):43–58. https://doi.org/10.1348/014466599162656.
25. Corbett T, Groarke A, Walsh JC, McGuire BE. Cancer-related fatigue in post-treatment cancer survivors: application of the common sense model of illness representations. BMC Cancer. 2016;16(1):919. https://doi.org/10.1186/s12885-016-2907-8.
26. Del Castillo A, Godoy-Izquierdo D, Vazquez ML, Godoy JF. Illness beliefs about cancer among healthy adults who have and have not lived with cancer patients. Int J Behav Med. 2011;18:342–51. https://doi.org/10.1007/s12529-010-9141-6.
27. Dempster M, McCorry NK, Brennan E, Donnelly M, Murray LJ, Johnston BT. Illness perceptions among carer-survivor dyads are related to psychological distress among oesophageal cancer survivors. J Psychosom Res. 2011;70:432–9. https://doi.org/10.1016/j.jpsychores.2010.07.007.

28. Fortune DG, Smith JV, Garvey K. Perceptions of psychosis, coping, appraisals, and psychological distress in the relatives of patients with schizophrenia: an exploration using self-regulation theory. Br J Clin Psychol. 2011;44(3):319–31. https://doi.org/10.1348/014466505X29198.
29. Bogosian A, Moss-Morris R, Bishop FL, Hadwin J. Development and initial validation of the perceptions of parental illness questionnaire. J Health Psychol. 2014;19(7):931–42. https://doi.org/10.1177/1359105313482164.
30. Yu H, Lingler JH, Sereika SM, Erlen JA. Characteristics and correlates of caregivers' perceptions of their family members' memory loss. Nurs Res. 2017;66(3):240–5. https://doi.org/10.1097/NNR.0000000000000220.
31. Scerri J, Saliba T, Saliba G, Scerri CA, Camilleri L. Illness perceptions, depression and anxiety in informal carers of persons with depression: a cross-sectional survey. Qual Life Res. 2019;28(2):451–60. https://doi.org/10.1007/s11136-018-2009-y.
32. Graham L, Dempster M, McCorry NK, Donnelly M, Johnston BT. Change in psychological distress in longer-term oesophageal cancer carers: are clusters of illness perception change a useful determinant? Psycho-Oncology. 2016;25(6):663–9. https://doi.org/10.1002/pon.3993.
33. Gatzoyia D, Kotsis K, Koullourou I, Goulia P, Carvalho AF, Soulis S, et al. The association of illness perceptions with depressive symptoms and general psychological distress in parents of an offspring with autism spectrum disorder. Disabil Health J. 2014;7(2):173–80. https://doi.org/10.1016/j.dhjo.2013.10.008.
34. United Nations Educational Scientific and Cultural Organisation. What do we mean by “youth”? 2017. http://www.unesco.org/new/en/social-and-human-sciences/themes/youth/youth-definition/. Accessed 21 Oct 2018.
35. Sawyer SM, Azzopardi PS, Wickremarathne D, Patton GC. The age of adolescence. Lancet Child Adolesc Health. 2018;2(3):223–8. https://doi.org/10.1016/S2352-4642(18)30022-1.
36. Australian Institute of Health and Welfare. Young Australians: their health and wellbeing 2011. Canberra: Australian Institute of Health and Welfare; 2011. Report No.: Cat. no. PHE 140
37. Heffernan E, Coulson NS, Henshaw H, Barry JG, Ferguson MA. Understanding the psychosocial experiences of adults with mild-moderate hearing loss: an application of Leventhal’s self-regulatory model. Int J Audiol. 2016;55(Suppl 3):S3–S12. https://doi.org/10.3109/14992027.2015.1117663.
38. Braun V, Clarke V. Successful qualitative research: a practical guide for beginners. Thousand Oaks: SAGE Publications; 2013.
39. Braun V, Clarke V. Using thematic analysis in psychology. Qual Res Psychol. 2006;3(2):77–101. https://doi.org/10.1191/1478088706qp063oa.
40. Maynard A, Patterson P, McDonald FEJ, Stevens G. What is helpful to adolescents who have a parent diagnosed with cancer? J Psychosoc Oncol. 2013;31(6):675–97. https://doi.org/10.1080/07347332.2013.835021.
41. Cassidy T, McLaughlin M, Giles M. Benefit finding in response to general life stress: measurement and correlates. Health Psychol Behav Med. 2014;2(1):268–82. https://doi.org/10.1080/21642850.2014.889570.
42. Cassidy T. Benefit finding through caring: the cancer caregiver experience. Psychol Health. 2012;28(3):250–66. https://doi.org/10.1080/08870446.2012.717623.
43. Cassidy T, Giles M, McLaughlin M. Benefit finding and resilience in child caregivers. Br J Health Psychol. 2014;19(3):606–18. https://doi.org/10.1111/bjhp.12059.
44. Semino E, Demjen Z, Demmen J, et al. The online use of violence and journey metaphors by patients with cancer, as compared with health professionals: a mixed methods study. BMJ Support Palliat Care. 2015;7(1):60–6. https://doi.org/10.1136/bmjspcare-2014-000785.
45. Miller RS. Speak up: 8 words and phrases to ban in oncology! Oncol Times. 2010;32(12):20. https://doi.org/10.1097/01.COT.0000383777.50536.b2.
46. Granger K. Having cancer is not a fight or a battle. Kings Place: The Guardian; 2014.
47. James D. It’s time to stop seeing cancer as an enemy we can wage war against - you can't fight a battle that's not fair. London: The Sun; 2018.
48. Cooper C. Mind your language: 'Battling' cancer metaphors can make terminally ill patients worse. London: The Independent; 2014.
49. Semino E. Whether you 'battle' cancer or experience a ‘journey’ is an individual choice. The Conversation. 2015. https://theconversation.com/whether-you-battle-cancer-or-experience-a-journey-is-an-individual-choice-39142. Accessed 8 Oct 2018.
50. Tomasetti C, Li L, Vogelstein B. Stem cell divisions, somatic mutations, cancer etiology, and cancer prevention. Science. 2017;355(6331):1330–4. https://doi.org/10.1126/science.aaf9011.
51. Phillips F, Lewis FM. The adolescent’s experience when a parent has advanced cancer: a qualitative inquiry. Palliat Med. 2015;29(9):851–8. https://doi.org/10.1177/0269216315578989.
52. Karnilowicz W. Identity and psychological ownership in chronic illness and disease state. Eur J Cancer Care (Engl). 2011;20(2):276–82. https://doi.org/10.1111/j.1365-2354.2010.01220.x.

Appendix D

Initial draft of PPIQ-C following adaptation of original PPIQ[34]

Having a parent diagnosed with cancer can have a big impact on the whole family. There are a range of reactions that young people might have to their parent's diagnosis. Everyone's experiences are different and there are no right or wrong responses. We are interested in your own personal views of how you see your parent's cancer. Below is a list of symptoms or side effects that your parent might have experienced as part of their cancer and treatment. Please select the answer that best reflects overall how much of an impact you think each symptom and side effect has had on your parent. It isn't necessary to ask your parent about their experiences, we are interested in what you believe.[35]

	No Impact	*Some Impact*	*Quite A Bit of An Impact*	*A Very Big Impact*
Identity				
1. Fatigue (tiredness)				
2. Pain				
3. Weakness				
4. Nausea (feeling sick in the stomach)				
5. Vomiting				
6. Hair loss				
7. Weight gain				
8. Weight loss				
9. Skin irritation				
10. Loss of appetite				
11. Depression				
12. Problems with concentration, thinking, or memory (chemo brain)				

---

[34] The content validity of this version of the PPIQ-C was assessed in the cognitive interview study reported in Chapter 4.

[35] Alternative instructions were provided for respondents completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *Thinking back to when your parent was going through treatment for cancer, please select the answer that best reflects overall how much of an impact you think each symptom or side effect had on your parent. It isn't necessary to ask your parent about their experiences, we are interested in what you believe.*

Below is a list of statements that describe thoughts or beliefs that young people might have about their parent’s cancer. Please select an answer to show how much you agree or disagree with each statement.[36]

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Emotional Representations					
1. My parent’s cancer symptoms are confusing to me					
2. When I think about my parent’s cancer I get upset					
3. My parent’s cancer makes me feel angry					
4. My parent’s cancer worries me					
5. My parent having cancer makes me feel stressed					
6. I try not to think about my parent’s cancer					
7. I feel overwhelmed when I think about my parent’s cancer					
8. I try to hide my feelings about my parent’s cancer					
9. I feel numb when I think about my parent’s cancer					
Coherence					
10. I have a good understanding of my parent’s cancer					
11. I have enough information about my parent’s cancer					
12. I would like to know more about my parent’s cancer					
13. I would rather not know details about my parent’s cancer					
Personal Control					
14. I can help my parent to manage their symptoms/side effects by looking after them					
15. My parent’s cancer symptoms get better when I do not stress them out (e.g., staying out later, arguing with brother or sister)					
16. If I’m not playing up, I can make my parent’s symptoms get better					
17. My parent not being stressed or worried can make their symptoms get better					
18. There isn’t anything I can do to make my parent’s symptoms get better					
19. I do what I can to help make my parent’s life easier					

---

[36] Additional instructions were provided for respondents completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *We would like you to think back to how you felt when your parent was going through treatment as you answer this section.*

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Treatment Control					
20. There isn’t a lot that can be done to treat my parent’s cancer					
21. My parent’s treatment will be effective in curing their cancer					
22. My parent’s treatment can control their cancer					
23. There is nothing that can be done to treat my parent’s cancer					
Consequences					
24. My parent’s cancer causes arguments in the family					
25. My parent’s cancer puts strain on the family					
26. My parent’s cancer makes it more difficult to do family activities					
27. My parent’s cancer has made me more responsible					
28. My parent’s cancer has made me more independent					
29. My parent’s cancer has made me grow up quicker					
30. My parent’s cancer has made me more understanding of other people					
31. My parent’s cancer brought me closer to my family					
32. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports)					
33. Because of my parent’s cancer, I spend more time doing housework					
34. Because of my parent’s cancer, I spend less time with my friends					
35. Because of my parent’s cancer, I spend less time doing schoolwork					
Timeline (acute/chronic)					
36. My parent’s cancer will get worse					
37. My parent’s cancer suddenly got worse and never got better					
38. I expect my parent to have cancer for the rest of their life					
39. My parent’s cancer will stay the same					
40. My parent’s cancer will improve in time					
41. My parent will recover from their cancer					
Timeline (cyclical/unpredictable)					
42. The severity of my parent’s cancer symptoms/side effects change a great deal from day to day					

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
43. The number of my parent’s symptoms/side effects change a great deal from day to day					
44. My parent’s symptoms/side effects come and go in cycles					
45. I never know how my parent will be feeling one day to the next					
46. My parent goes through cycles where their cancer symptoms/side effects get better or worse					
47. My parent’s cancer is very unpredictable					

We are also interested in your thoughts about what might have caused your parent’s cancer. As people are very different, there are no right or wrong answers. We are really interested in your own views about what caused your parent’s cancer.

Below is a list of statements describing possible causes of your parent’s cancer. Please select an answer to show how much you agree or disagree with each statement.[37]

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Cause					
1. My parent’s cancer was caused by stress or worry					
2. My parent’s cancer was caused by family problems or worries					
3. My parent’s cancer was caused by a germ or virus					
4. My parent’s cancer is hereditary – it runs in the family					
5. My parent’s cancer was caused by chance or bad luck					
6. My parent’s cancer was caused by poor diet					
7. My parent’s cancer was caused by sun exposure					
8. My parent’s cancer was caused by lack of exercise					
9. My parent’s cancer was caused by smoking					
10. My parent’s cancer was caused by drinking alcohol					
11. My parent’s cancer was caused by chemicals or radiation in the environment					
12. My parent’s cancer was caused by bad or toxic people in their life					

---

[37] Additional instructions were provided for respondents completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *We would like you to think back to how you felt when your parent was going through treatment as you answer this section.*

Appendix E

Draft of PPIQ-C following modifications informed by cognitive interviews conducted in Study 3[38]

Your Perceptions of Your Parent's Cancer

The following questions ask about your thoughts and beliefs about your parent’s cancer. Having a parent diagnosed with cancer can have a big impact on the whole family. There are a range of reactions that young people might have to their parent’s diagnosis. Everyone’s experiences are different and there are no right or wrong responses.

You may find that some of the questions are upsetting or difficult to think about and answer. Please answer as many questions as you feel able to.

---

[38] This version of the PPIQ-C was subjected to psychometric testing. Findings reported in Chapter 5.

Section 1: Perceptions of your parent's cancer symptoms and side effects

Below is a list of symptoms and side effects that your parent might have experienced as part of their cancer and treatment. Please tick the answer that best reflects overall how much you believe each symptom and side effect has **negatively** impacted your parent. You don't need to ask your parent about their experiences, we are interested in your own views of your parent's cancer.[39]

	No Impact	*Some Impact*	*Quite A Big Impact*	*A Very Big Impact*
Identity				
1. Fatigue (tiredness)				
2. Pain				
3. Physical weakness (loss of strength)				
4. Nausea (feeling sick in the stomach)				
5. Vomiting				
6. Hair loss				
7. Weight gain				
8. Weight loss				
9. Skin irritation (dry skin, rashes, or itchiness)				
10. Loss of appetite				
11. Depression				
12. Anxiety				
13. Problems with concentration, thinking, or memory (chemo brain)				

---

[39] Alternative instructions were provided for respondents completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *Below is a list of symptoms and side effects that your parent might have experienced as part of their cancer and treatment. Thinking back to when your parent was going through treatment for cancer, please tick the answer that best reflects overall how much you believe each symptom and side effect negatively impacted your parent. You don't need to ask your parent about their experiences, we are interested in your own views of your parent's cancer.*

Section 2: Thoughts and beliefs about your parent's cancer

Below is a list of statements that describe thoughts and beliefs that young people might have about their parent's cancer. Please tick an answer to show how much you agree or disagree with each statement. Some of the statements in this section might seem really similar, but they are saying different things. It's important that you answer as many as you are able to.[40]

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Emotional Representations					
1. When I think about my parent's cancer I get upset					
2. My parent's cancer makes me feel angry					
3. My parent's cancer worries me					
4. My parent having cancer makes me feel stressed					
5. My parent having cancer makes me feel worried about the future					
6. I try not to think about my parent's cancer					
7. I feel overwhelmed when I think about my parent's cancer					
8. I try to hide my feelings about my parent's cancer					
9. I feel numb when I think about my parent's cancer					
Coherence					
10. My parent's cancer is confusing to me					
11. I have a good understanding of my parent's cancer					
12. I have enough information about my parent's cancer					
13. I would like to know more about my parent's cancer					
14. I would rather not know details about my parent's cancer					
Personal Control					
15. I can help my parent to manage their symptoms/side effects by looking after them					

---

[40] Alternative instructions were provided for respondents completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *Below is a list of statements that describe thoughts and beliefs that young people might have about their parent's cancer. Please tick an answer to show how much you agree or disagree with each statement. We would like you to think back to how you felt when your parent was going through treatment as you answer this section. Some of the statements in this section might seem really similar, but they are saying different things. It's important that you answer as many as you are able to.*

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
16. My parent’s cancer symptoms/side effects get better when I do not stress them out (e.g., staying out later, arguing with brother or sister)					
17. If I’m not misbehaving, I can make my parent’s symptoms/side effects get better					
18. My parent not being stressed or worried can make their symptoms/side effects get better					
19. There isn’t anything I can do to make my parent’s symptoms/side effects get better					
20. I do what I can to help make my parent’s life easier					
Treatment Control					
21. There isn’t a lot that can be done to treat my parent’s cancer					
22. My parent’s treatment will be effective in curing their cancer					
23. My parent’s treatment can control their cancer					
24. There is nothing that can be done to treat my parent’s cancer					
Consequences					
25. My parent’s cancer causes arguments in the family					
26. My parent’s cancer puts strain on the family					
27. My parent’s cancer makes it more difficult to do family activities					
28. My parent’s cancer has made me more responsible					
29. My parent’s cancer has made me more independent					
30. My parent’s cancer has made me grow up quicker					
31. My parent’s cancer has made me more understanding of other people					
32. My parent’s cancer brought me closer to my family					
33. My parent’s cancer brought me closer to my parent					
34. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports)					
35. Because of my parent’s cancer, I spend more time doing housework					
36. Because of my parent’s cancer, I spend less time with my friends					
37. Because of my parent’s cancer, I spend less time doing schoolwork					
Timeline (acute/chronic)					
38. My parent’s cancer will get worse					
39. My parent’s cancer suddenly got worse and never got better					

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
40. I expect my parent to have cancer for the rest of their life					
41. My parent’s cancer will stay the same					
42. My parent’s cancer will improve in time					
43. My parent will recover from their cancer					
Timeline (cyclical/unpredictable)					
44. The intensity of my parent’s symptoms/side effects change a great deal from day to day					
45. The number of my parent’s symptoms/side effects change a great deal from day to day					
46. My parent’s symptoms/side effects come and go in cycles					
47. I never know how my parent will be feeling one day to the next					
48. My parent goes through cycles where their symptoms/side effects get better or worse					
49. My parent’s symptoms/side effects are very unpredictable					

Section 3: Beliefs about the cause of your parent’s cancer

Below is a list of statements that describe things that young people commonly believe might have caused their parent’s cancer. Please tick an answer to show how much you agree or disagree with each statement. As people are very different, there are no right or wrong answers. We are really interested in your own views about what caused your parent’s cancer.

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Cause					
1. My parent’s cancer was caused by stress or worry					
2. My parent’s cancer was caused by chance					
3. My parent’s cancer was caused by smoking					
4. My parent’s cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)					
5. My parent’s cancer was caused by a germ or virus					
6. My parent’s cancer was caused by family problems or worries					
7. My parent’s cancer was caused by bad luck					
8. My parent’s cancer was caused by poor diet					
9. My parent’s cancer was caused by bad or toxic people in their life					
10. My parent’s cancer was caused by sun exposure					
11. My parent’s cancer was caused by a higher power (e.g., it was an act of God)					
12. My parent’s cancer was caused by lack of exercise					
13. My parent’s cancer is hereditary (it runs in the family)					
14. My parent’s cancer was caused by drinking alcohol					

Appendix F

Two-week test-retest reliabilities for PPIQ-C subscales in concurrent and retrospective responses

	Concurrent[a]		Retrospective[b]	
PPIQ-C subscale	ICC	*n*	ICC	*n*
1. Identity	.884*	4	.743**	15
2. Emotional Representations	.703*	6	.573**	17
3. Coherence	.426	6	.787**	17
4. Consequences for Personal Development	.772*	6	.503*	16
5. Consequences for Family Relationships	.894**	6	.559**	17
6. Consequences for Daily Activities	.705	5	.732**	16
7. Personal Control	.621	6	.825**	17
8. Treatment Control	.082	6	.482*	17
9. Recovery	.891**	6	.290	16
10. Unpredictability	.787*	6	.639**	16
11. Chronicity	.962**	6	.727**	15
12. Behavioural or Environmental Attributions	.724*	6	.558*	16
13. Chance or Luck Attributions	.690*	6	.640**	17
14. Emotional or Psychological Attributions	.651	6	.756**	17

**p < .01. *p < .05. [a]Parent was recently diagnosed, undergoing treatment, or receiving palliative care. [b]Parent had completed treatment or died.

Appendix G

Final PPIQ-C following item reduction informed by psychometric evaluation conducted in Study 4

Your Perceptions of Your Parent's Cancer

The following questions ask about your thoughts and beliefs about your parent’s cancer. Having a parent diagnosed with cancer can have a big impact on the whole family. There are a range of reactions that young people might have to their parent’s diagnosis. Everyone’s experiences are different and there are no right or wrong responses.

You may find that some of the questions are upsetting or difficult to think about and answer. Please answer as many questions as you feel able to.

Section 1: Perceptions of your parent's cancer symptoms and side effects

Below is a list of symptoms and side effects that your parent might have experienced as part of their cancer and treatment. Please tick the answer that best reflects overall how much you believe each symptom and side effect has **negatively** impacted your parent. You don't need to ask your parent about their experiences, we are interested in your own views of your parent's cancer.[41]

	No Impact	*Some Impact*	*Quite A Big Impact*	*A Very Big Impact*
Identity				
1. Fatigue (tiredness)				
2. Pain				
3. Physical weakness (loss of strength)				
4. Nausea (feeling sick in the stomach)				
5. Vomiting				
6. Hair loss				
7. Weight gain				
8. Weight loss				
9. Skin irritation (dry skin, rashes, or itchiness)				
10. Loss of appetite				
11. Depression				
12. Anxiety				
13. Problems with concentration, thinking, or memory (chemo brain)				

---

[41] Alternative instructions for AYAs completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *Below is a list of symptoms and side effects that your parent might have experienced as part of their cancer and treatment. Thinking back to when your parent was going through treatment for cancer, please tick the answer that best reflects overall how much you believe each symptom and side effect negatively impacted your parent. You don't need to ask your parent about their experiences, we are interested in your own views of your parent's cancer.*

Section 2: Thoughts and beliefs about your parent’s cancer

Below is a list of statements that describe thoughts and beliefs that young people might have about their parent’s cancer. Please tick an answer to show how much you agree or disagree with each statement. Some of the statements in this section might seem really similar, but they are saying different things. It’s important that you answer as many as you are able to.[42]

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Emotional Representations					
1. When I think about my parent’s cancer I get upset					
2. My parent’s cancer makes me feel angry					
3. My parent’s cancer worries me					
4. My parent having cancer makes me feel stressed					
5. My parent having cancer makes me feel worried about the future					
6. I feel overwhelmed when I think about my parent’s cancer					
Coherence					
7. My parent’s cancer is confusing to me (r)					
8. I have a good understanding of my parent’s cancer					
9. I have enough information about my parent’s cancer					
Consequences for Personal Development					
10. My parent’s cancer has made me more responsible					
11. My parent’s cancer has made me more independent					
12. My parent’s cancer has made me grow up quicker					
13. My parent’s cancer has made me more understanding of other people					
Consequences for Family Relationships					
14. My parent’s cancer causes arguments in the family					
15. My parent’s cancer puts strain on the family					
16. My parent’s cancer brought me closer to my family (r)					

---

[42] Alternative instructions for AYAs completing questionnaire with reference to a past experience of parental cancer (i.e., retrospectively): *Below is a list of statements that describe thoughts and beliefs that young people might have about their parent’s cancer. Please tick an answer to show how much you agree or disagree with each statement. We would like you to think back to how you felt when your parent was going through treatment as you answer this section. Some of the statements in this section might seem really similar, but they are saying different things. It’s important that you answer as many as you are able to.*

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
17. My parent’s cancer brought me closer to my parent (r)					
Consequences for Daily Activities					
18. My parent’s cancer makes it more difficult to do family activities					
19. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports)					
20. Because of my parent’s cancer, I spend more time doing housework					
21. Because of my parent’s cancer, I spend less time with my friends					
22. Because of my parent’s cancer, I spend less time doing schoolwork					
Personal Control					
23. My parent’s cancer symptoms/side effects get better when I do not stress them out (e.g., staying out later, arguing with brother or sister)					
24. If I’m not misbehaving, I can make my parent’s symptoms/side effects get better					
25. My parent not being stressed or worried can make their symptoms/side effects get better					
26. There isn’t anything I can do to make my parent’s symptoms/side effects get better (r)					
Treatment Control					
27. There isn’t a lot that can be done to treat my parent’s cancer (r)					
28. There is nothing that can be done to treat my parent’s cancer (r)					
Recovery					
29. My parent’s treatment will be effective in curing their cancer					
30. My parent’s treatment can control their cancer					
31. My parent’s cancer will improve in time					
32. My parent will recover from their cancer					
Unpredictability					
33. The intensity of my parent’s symptoms/side effects change a great deal from day to day					
34. My parent’s symptoms/side effects come and go in cycles					
35. I never know how my parent will be feeling one day to the next					
36. My parent goes through cycles where their symptoms/side effects get better or worse					

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
37. My parent’s symptoms/side effects are very unpredictable					
Chronicity					
38. My parent’s cancer will get worse					
39. I expect my parent to have cancer for the rest of their life					
40. My parent’s cancer will stay the same					

(r) = item reverse-scored

Section 3: Beliefs about the cause of your parent's cancer

Below is a list of statements that describe things that young people commonly believe might have caused their parent's cancer. Please tick an answer to show how much you agree or disagree with each statement. As people are very different, there are no right or wrong answers. We are really interested in your own views about what caused your parent's cancer.

	Strongly Disagree	*Disagree*	*Neither Agree nor Disagree*	*Agree*	*Strongly Agree*
Behavioural or Environmental Attributions					
1. My parent's cancer was caused by poor diet					
2. My parent's cancer was caused by lack of exercise					
3. My parent's cancer was caused by drinking alcohol					
4. My parent's cancer was caused by smoking					
5. My parent's cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)					
Chance or Luck Attributions					
6. My parent's cancer was caused by chance					
7. My parent's cancer was caused by bad luck					
Emotional or Psychological Attributions					
8. My parent's cancer was caused by stress or worry					
9. My parent's cancer was caused by family problems or worries					
10. My parent's cancer was caused by bad or toxic people in their life					
Other items[a]					
11. My parent's cancer was caused by sun exposure					
12. My parent's cancer was caused by a higher power (e.g., it was an act of God)					
13. My parent's cancer was caused by a germ or virus					
14. My parent's cancer is hereditary (it runs in the family)					

[a] Items that did not load onto separate coherent factor during factor analysis.

Appendix H

Sensitivity, specificity, and Youden's index for Total Negative Beliefs cut-off scores used to identify psychological distress

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden's index
71.0000	1.000	0.000	0.000
76.0000	1.000	0.014	0.014
81.0000	1.000	0.027	0.027
82.5000	1.000	0.041	0.041
85.0000	1.000	0.055	0.055
87.5000	0.997	0.055	0.052
88.5000	0.994	0.055	0.048
90.5000	0.990	0.055	0.045
92.5000	0.987	0.055	0.042
93.5000	0.987	0.082	0.069
95.0000	0.984	0.096	0.080
96.5000	0.981	0.096	0.077
97.5000	0.981	0.123	0.104
98.5000	0.981	0.151	0.131
99.5000	0.981	0.164	0.145
101.5000	0.981	0.192	0.172
103.5000	0.981	0.205	0.186
104.5000	0.981	0.219	0.200
105.5000	0.977	0.219	0.197
107.0000	0.977	0.233	0.210
109.0000	0.974	0.233	0.207
110.5000	0.968	0.274	0.242
111.5000	0.965	0.274	0.239
112.5000	0.958	0.288	0.246
113.5000	0.952	0.288	0.239
114.5000	0.942	0.301	0.243
115.5000	0.936	0.329	0.264
116.5000	0.929	0.356	0.285
117.5000	0.923	0.370	0.293
118.5000	0.920	0.384	0.303
119.5000	0.913	0.411	0.324
120.5000	0.910	0.466	0.376
121.5000	0.910	0.479	0.389
122.5000	0.894	0.479	0.373
123.5000	0.887	0.521	0.408
124.5000	0.887	0.534	0.422
125.5000	0.875	0.562	0.436
126.5000	0.865	0.575	0.440

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden’s index
127.5000	0.862	0.589	0.451
128.5000	0.859	0.589	0.448
129.5000	0.855	0.616	0.472
130.5000	0.839	0.671	0.510
131.5000	0.830	0.699	0.528
132.5000	0.814	0.712	0.526
133.5000	0.794	0.712	0.507
134.5000	0.778	0.726	0.504
135.5000	0.778	0.767	0.545
136.5000	0.768	0.795	0.563
137.5000	0.746	0.836	0.582
138.5000	0.730	0.836	0.566
139.5000	0.701	0.849	0.550
140.5000	0.682	0.863	0.545
141.5000	0.659	0.877	0.536
142.5000	0.633	0.890	0.524
143.5000	0.608	0.890	0.498
144.5000	0.579	0.918	0.497
145.5000	0.559	0.918	0.477
146.5000	0.547	0.918	0.464
147.5000	0.537	0.918	0.455
148.5000	0.508	0.932	0.440
149.5000	0.489	0.932	0.420
150.5000	0.466	0.932	0.398
151.5000	0.441	0.932	0.372
152.5000	0.408	0.932	0.340
153.5000	0.379	0.932	0.311
154.5000	0.360	0.932	0.292
155.5000	0.331	0.932	0.263
156.5000	0.309	0.945	0.254
157.5000	0.299	0.945	0.244
158.5000	0.280	0.945	0.225
159.5000	0.260	0.959	0.219
160.5000	0.241	0.973	0.214
161.5000	0.228	0.973	0.201
162.5000	0.206	0.973	0.178
163.5000	0.196	0.973	0.169
164.5000	0.177	0.973	0.149
165.5000	0.164	0.973	0.137
166.5000	0.145	0.973	0.117
167.5000	0.132	0.973	0.104
168.5000	0.113	0.973	0.085
169.5000	0.090	0.986	0.076
170.5000	0.080	0.986	0.067

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden’s index
171.5000	0.071	0.986	0.057
172.5000	0.064	0.986	0.051
173.5000	0.061	0.986	0.047
175.0000	0.048	0.986	0.035
176.5000	0.035	0.986	0.022
178.5000	0.029	0.986	0.015
180.5000	0.019	0.986	0.006
181.5000	0.016	0.986	0.002
183.0000	0.013	0.986	-0.001
184.5000	0.006	0.986	-0.007
185.5000	0.003	0.986	-0.010
186.5000	0.003	1.000	0.003
188.0000	0.000	1.000	0.000

Sensitivity, specificity, and Youden's index for Total Negative Beliefs cut-off scores used to identify post-traumatic stress in young adults

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden's index
71.0000	1.000	0.000	0.000
76.0000	1.000	0.009	0.009
81.0000	1.000	0.018	0.018
82.5000	1.000	0.027	0.027
85.0000	1.000	0.036	0.036
87.5000	1.000	0.045	0.045
90.0000	1.000	0.054	0.054
92.5000	1.000	0.063	0.063
93.5000	1.000	0.080	0.080
95.0000	1.000	0.098	0.098
96.5000	1.000	0.107	0.107
97.5000	1.000	0.125	0.125
98.5000	1.000	0.143	0.143
99.5000	0.995	0.143	0.138
101.5000	0.995	0.161	0.156
103.5000	0.995	0.170	0.165
104.5000	0.995	0.179	0.173
105.5000	0.995	0.188	0.182
107.0000	0.995	0.196	0.191
109.0000	0.995	0.205	0.200
110.5000	0.985	0.232	0.217
111.5000	0.979	0.232	0.212
112.5000	0.974	0.250	0.224
113.5000	0.974	0.259	0.233
114.5000	0.969	0.277	0.246
115.5000	0.969	0.295	0.264
116.5000	0.969	0.321	0.291
117.5000	0.964	0.339	0.303
118.5000	0.959	0.339	0.298
119.5000	0.954	0.357	0.311
120.5000	0.949	0.393	0.342
121.5000	0.949	0.402	0.351
122.5000	0.938	0.446	0.385
123.5000	0.928	0.464	0.392
124.5000	0.928	0.473	0.401
125.5000	0.908	0.491	0.399
126.5000	0.908	0.527	0.434
127.5000	0.897	0.536	0.433
128.5000	0.897	0.545	0.442
129.5000	0.897	0.554	0.451
130.5000	0.892	0.598	0.491

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden’s index
131.5000	0.882	0.616	0.498
132.5000	0.862	0.634	0.495
133.5000	0.836	0.643	0.479
134.5000	0.815	0.661	0.476
135.5000	0.815	0.688	0.503
136.5000	0.800	0.688	0.488
137.5000	0.779	0.705	0.485
138.5000	0.754	0.705	0.459
139.5000	0.718	0.732	0.450
140.5000	0.697	0.741	0.439
141.5000	0.677	0.741	0.418
142.5000	0.651	0.750	0.401
143.5000	0.626	0.777	0.402
144.5000	0.600	0.821	0.421
145.5000	0.574	0.830	0.405
146.5000	0.559	0.830	0.389
147.5000	0.559	0.839	0.398
148.5000	0.533	0.866	0.399
149.5000	0.508	0.866	0.374
150.5000	0.477	0.866	0.343
151.5000	0.441	0.875	0.316
152.5000	0.410	0.875	0.285
153.5000	0.390	0.893	0.283
154.5000	0.374	0.902	0.276
155.5000	0.359	0.946	0.305
156.5000	0.328	0.955	0.284
157.5000	0.313	0.955	0.268
158.5000	0.297	0.964	0.262
159.5000	0.267	0.964	0.231
160.5000	0.231	0.964	0.195
161.5000	0.210	0.964	0.175
162.5000	0.174	0.964	0.139
163.5000	0.164	0.964	0.128
164.5000	0.154	0.964	0.118
165.5000	0.144	0.964	0.108
166.5000	0.128	0.964	0.092
167.5000	0.113	0.964	0.077
168.5000	0.082	0.964	0.046
169.5000	0.067	0.973	0.040
170.5000	0.056	0.973	0.030
171.5000	0.056	0.982	0.039
172.5000	0.056	0.991	0.047
173.5000	0.051	0.991	0.042
175.0000	0.041	0.991	0.032

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden’s index
178.0000	0.026	0.991	0.017
180.5000	0.010	0.991	0.001
182.5000	0.005	0.991	-0.004
185.0000	0.000	0.991	-0.009
187.0000	0.000	1.000	0.000

Sensitivity, specificity, and Youden's index for Total Negative Beliefs cut-off scores used to identify post-traumatic stress in adolescents

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden's index
113.0000	1.000	0.000	0.000
114.5000	0.981	0.000	-0.019
115.5000	0.981	0.100	0.081
117.0000	0.981	0.150	0.131
118.5000	0.981	0.200	0.181
121.0000	0.981	0.250	0.231
126.0000	0.981	0.300	0.281
129.5000	0.981	0.350	0.331
130.5000	0.962	0.450	0.412
133.5000	0.943	0.450	0.393
136.5000	0.887	0.450	0.337
137.5000	0.868	0.550	0.418
138.5000	0.849	0.550	0.399
139.5000	0.830	0.550	0.380
140.5000	0.830	0.600	0.430
141.5000	0.755	0.600	0.355
143.0000	0.736	0.650	0.386
145.5000	0.717	0.650	0.367
147.5000	0.698	0.700	0.398
148.5000	0.660	0.700	0.360
149.5000	0.642	0.700	0.342
151.0000	0.623	0.700	0.323
152.5000	0.566	0.750	0.316
153.5000	0.528	0.750	0.278
154.5000	0.491	0.750	0.241
155.5000	0.453	0.750	0.203
157.0000	0.434	0.750	0.184
158.5000	0.396	0.750	0.146
159.5000	0.377	0.750	0.127
161.5000	0.377	0.800	0.177
163.5000	0.340	0.800	0.140
164.5000	0.321	0.850	0.171
165.5000	0.283	0.850	0.133
167.5000	0.226	0.850	0.076
169.5000	0.170	0.900	0.070
170.5000	0.151	0.900	0.051
171.5000	0.132	0.950	0.082
173.0000	0.132	1.000	0.132
175.5000	0.094	1.000	0.094
180.5000	0.057	1.000	0.057
184.5000	0.038	1.000	0.038

Positive result if Total Negative Beliefs score ≥	Sensitivity	Specificity	Youden’s index
186.0000	0.019	1.000	0.019
188.0000	0.000	1.000	0.000

Appendix I

Items included in Total Negative Beliefs subscale of the PPIQ-C

Full PPIQ-C subscale/item
Identity
1. Fatigue (tiredness)
2. Pain
3. Physical weakness (loss of strength)
4. Nausea (feeling sick in the stomach)
5. Vomiting
6. Hair loss
7. Weight gain
8. Weight loss
9. Skin irritation (dry skin, rashes, or itchiness)
10. Loss of appetite
11. Depression
12. Anxiety
13. Problems with concentration, thinking, or memory (chemo brain)
Emotional Representations
14. When I think about my parent’s cancer I get upset
15. My parent’s cancer makes me feel angry
16. My parent’s cancer worries me
17. My parent having cancer makes me feel stressed
18. My parent having cancer makes me feel worried about the future
19. I feel overwhelmed when I think about my parent’s cancer
Consequences for Personal Development
20. My parent’s cancer has made me more responsible
21. My parent’s cancer has made me more independent
22. My parent’s cancer has made me grow up quicker
23. My parent’s cancer has made me more understanding of other people
Consequences for Family Relationships
24. My parent’s cancer causes arguments in the family
25. My parent’s cancer puts strain on the family
26. My parent’s cancer brought me closer to my family*
27. My parent’s cancer brought me closer to my parent*
Consequences for Daily Activities
28. My parent’s cancer makes it more difficult to do family activities
29. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports)
30. Because of my parent’s cancer, I spend more time doing housework
31. Because of my parent’s cancer, I spend less time with my friends
32. Because of my parent’s cancer, I spend less time doing schoolwork

Full PPIQ-C subscale/item
Unpredictability
33. The intensity of my parent’s symptoms/side effects change a great deal from day to day
34. My parent’s symptoms/side effects come and go in cycles
35. I never know how my parent will be feeling one day to the next
36. My parent goes through cycles where their symptoms/side effects get better or worse
37. My parent’s symptoms/side effects are very unpredictable
Chronicity
38. My parent’s cancer will get worse
39. I expect my parent to have cancer for the rest of their life
40. My parent’s cancer will stay the same
Behavioural or Environmental Attributions
41. My parent’s cancer was caused by poor diet
42. My parent’s cancer was caused by lack of exercise
43. My parent’s cancer was caused by drinking alcohol
44. My parent’s cancer was caused by smoking
45. My parent’s cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology)
Emotional or Psychological Attributions
46. My parent’s cancer was caused by stress or worry
47. My parent’s cancer was caused by family problems or worries
48. My parent’s cancer was caused by bad or toxic people in their life

Appendix J

Differences between IPQ-R, PPIQ, and PPIQ-C questionnaires

	IPQ-R	PPIQ	PPIQ-C
Identity subscale	*Please rate whether or not you have experienced each symptom since your illness, then rate whether or not you believe the symptom is specifically related to your illness (Rated: yes/no)* 1. Pain 2. Nausea 3. Breathlessness 4. Weight loss 5. Fatigue 6. Stiff joints 7. Sore eyes 8. Headaches 9. Upset stomach 10. Sleep difficulties 11. Dizziness 12. Loss of strength 13. Sore throat 14. Wheeziness	*Identity* subscale not included	*Below is a list of symptoms and side effects that your parent might have experienced as part of their cancer and treatment. Please tick the answer that best reflects overall how much you believe each symptom and side effect has negatively impacted your parent. You don't need to ask your parent about their experiences, we are interested in your own views of your parent's cancer. (Rated: no impact, some impact, quite a big impact, a very big impact)* 1. Fatigue (tiredness) 2. Pain 3. Physical weakness (loss of strength) 4. Nausea (feeling sick in the stomach) 5. Vomiting 6. Hair loss 7. Weight gain 8. Weight loss 9. Skin irritation (dry skin, rashes, or itchiness) 10. Loss of appetite 11. Depression 12. Anxiety 13. Problems with concentration, thinking, or memory (chemo brain)
Core subscales	*We are interested in your own personal views of how you now see your illness. Please indicate how much you agree or disagree with the following statements about your illness. (Rated:*	*We are interested in your own personal views of how you NOW see your parent's MS. Please indicate how*	*Below is a list of statements that describe thoughts and beliefs that young people might have about their parent's cancer. Please tick an answer to show how much you agree or*

	IPQ-R	PPIQ	PPIQ-C
	strongly disagree, disagree, neither agree nor disagree, agree, strongly agree) *Timeline (acute/chronic)* 1. My illness will last a short time 2. My illness is likely to be permanent rather than temporary 3. My illness will last for a long time 4. This illness will pass quickly 5. I expect to have this illness for the rest of my life 6. My illness will improve in time *Timeline (cyclical)* 7. The symptoms of my illness change a great deal from day to day 8. My symptoms come and go in cycles 9. My illness is very unpredictable 10. I go through cycles in which my illness gets better and worse *Consequences* 11. My illness is a serious condition 12. My illness has major consequences on my life 13. My illness does not have much effect on my life 14. My illness strongly affects the way others see me 15. My illness has serious economic and financial consequences 16. My illness causes difficulties for those who are close to me *Personal Control* 17. There is a lot which I can do to control my symptoms	*much you agree or disagree with the following statements about your dad's illness by ticking the appropriate (Rated: strongly disagree, disagree, neither agree nor disagree, agree, strongly agree)* *Chronic timeline* 1. My parent's MS will get worse 2. My parent's MS suddenly got worse and never got better 3. I expect my parent to have MS for the rest of their life 4. My parent's MS will stay the same *Unpredictable timeline* 5. The severity of my parent's MS symptoms change a great deal from day to day 6. The number of my parent's symptoms change a great deal from day to day *Negative consequences for family* 7. My parent's MS causes arguments in the family 8. My parent's MS puts strain on the family 9. My parent's MS makes it more difficult to do family activities *Positive consequences for adolescents* 10. My parent's MS has made me more responsible 11. My parent's MS has me more independent 12. My parent's MS has made more understanding of other people 13. My parent's MS brought my closer to my family *Negative consequences for adolescents*	*disagree with each statement. Some of the statements in this section might seem really similar, but they are saying different things. It's important that you answer as many as you are able to. (Rated: strongly disagree, disagree, neither agree nor disagree, agree, strongly agree)* *Chronicity* 1. My parent's cancer will get worse 2. I expect my parent to have cancer for the rest of their life 3. My parent's cancer will stay the same *Unpredictability* 4. The intensity of my parent's symptoms/side effects change a great deal from day to day 5. My parent's symptoms/side effects come and go in cycles 6. I never know how my parent will be feeling one day to the next 7. My parent goes through cycles where their symptoms/side effects get better or worse 8. My parent's symptoms/side effects are very unpredictable *Consequences for Personal Development* 9. My parent's cancer has made me more responsible 10. My parent's cancer has made me more independent 11. My parent's cancer has made me grow up quicker 12. My parent's cancer has made me more understanding of other people *Consequences for Family Relationships*

	IPQ-R	PPIQ	PPIQ-C
	18. What I do can determine whether my illness gets better or worse 19. The course of my illness depends on me 20. Nothing I do will affect my illness 21. I have the power to influence my illness 22. My actions will have no effect on the outcome of my illness *Treatment Control* 23. There is very little that can be done to improve my illness 24. My treatment will be effective in curing my illness 25. The negative effects of my illness can be prevented (avoided) by my treatment 26. My treatment can control my illness 27. There is nothing which can help my condition *Illness Coherence* 28. The symptoms of my condition are puzzling to me 29. My illness is a mystery to me 30. I don’t understand my illness 31. My illness doesn’t make any sense to me 32. I have a clear picture or understanding of my condition *Emotional representations* 33. I get depressed when I think about my illness 34. When I think about my illness I get upset 35. My illness makes me feel angry 36. My illness does not worry me 37. Having this illness makes me feel anxious 38. My illness makes me feel afraid	14. Because of my parent’s MS, I spend less time doing social activities (e.g. hobbies, sports) 15. Because of my parent’s MS, I spend more time doing housework 16. Because of my parent’s MS, I spend less time with my friends *Adolescents’ control* 17. I can help my parent manage their symptoms by looking after them 18. My parent’s MS symptoms get better when I do not stress them out (e.g. staying out late, arguing with brother or sister) 19. If I’m not playing up, I can make my parent’s symptoms get better 20. My parent not being stressed or worried can make their symptoms get better *Emotional representations* 21. My parent’s MS symptoms are confusing to me 22. When I think about my parent’s MS I get upset 23. My parent’s MS makes me feel angry 24. My parent’s MS worries me 25. My parent having MS makes me feel stressed Note: subscales for *treatment control* and *illness coherence* not included	13. My parent’s cancer causes arguments in the family 14. My parent’s cancer puts strain on the family 15. My parent’s cancer brought me closer to my family 16. My parent’s cancer brought me closer to my parent *Consequences for Daily Activities* 17. My parent’s cancer makes it more difficult to do family activities 18. Because of my parent’s cancer, I spend less time doing social activities (e.g., hobbies, sports) 19. Because of my parent’s cancer, I spend more time doing housework 20. Because of my parent’s cancer, I spend less time with my friends 21. Because of my parent’s cancer, I spend less time doing schoolwork *Personal Control* 22. My parent’s cancer symptoms/side effects get better when I do not stress them out (e.g., staying out later, arguing with brother or sister) 23. If I’m not misbehaving, I can make my parent’s symptoms/side effects get better 24. My parent not being stressed or worried can make their symptoms/side effects get better 25. There isn’t anything I can do to make my parent’s symptoms/side effects get better *Treatment Control* 26. There isn’t a lot that can be done to treat my parent’s cancer 27. There is nothing that can be done to treat my parent’s cancer

	IPQ-R	PPIQ	PPIQ-C
			Recovery 28. My parent’s treatment will be effective in curing their cancer 29. My parent’s treatment can control their cancer 30. My parent’s cancer will improve in time 31. My parent will recover from their cancer *Coherence* 32. My parent’s cancer is confusing to me 33. I have a good understanding of my parent’s cancer 34. I have enough information about my parent’s cancer *Emotional Representations* 35. When I think about my parent’s cancer I get upset 36. My parent’s cancer makes me feel angry 37. My parent’s cancer worries me 38. My parent having cancer makes me feel stressed 39. My parent having cancer makes me feel worried about the future 40. I feel overwhelmed when I think about my parent’s cancer
Cause subscales	*Psychological attributions* 1. Stress or worry 2. My mental attitude (e.g. thinking about life negatively) 3. Family problems or worries caused my illness 4. Overwork 5. My emotional state (e.g. feeling down, lonely, anxious, empty) a 6. My personality *Risk factor attributions*	*We are interested in what you think may have been the cause of your parent’s MS. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that caused your parent’s illness rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your parent’s MS. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box. (Rated: strongly disagree,*	*Below is a list of statements that describe things that young people commonly believe might have caused their parent’s cancer. Please tick an answer to show how much you agree or disagree with each statement. As people are very different, there are no right or wrong answers. We are really interested in your own views about what caused your parent’s cancer. (Rated: strongly disagree, disagree, neither agree nor disagree, agree, strongly agree)*

	IPQ-R	PPIQ	PPIQ-C
	7. Hereditary – it runs in my family 8. Diet or eating habits 9. Poor medical care in my past 10. My own behaviour 11. Ageing 12. Smoking 13. Alcohol *Immune attributions* 14. A germ or virus 15. Pollution in the environment 16. Altered immunity *Chance attributions* 17. Chance or bad luck 18. Accident or injury	*disagree, neither agree nor disagree, agree, strongly agree)* *Psychological attributions* 1. Stress or worry 2. Family problems or worries *Central nervous system* 3. Scars on the spine 4. Nerve damage *External/environmental attributions* 5. A germ or virus 6. Environmental changes *Hereditary/chance* 7. Hereditary – it runs in the family 8. Chance or bad luck	*Emotional or Psychological Attributions* 1. My parent's cancer was caused by stress or worry 2. My parent's cancer was caused by family problems or worries 3. My parent's cancer was caused by bad or toxic people in their life *Behavioural or Environmental Attributions* 4. My parent's cancer was caused by poor diet 5. My parent's cancer was caused by lack of exercise 6. My parent's cancer was caused by drinking alcohol 7. My parent's cancer was caused by smoking 8. My parent's cancer was caused by chemicals or radiation in the environment (e.g., pollution or radiation caused by modern technology) *Chance or Luck Attributions* 9. My parent's cancer was caused by chance 10. My parent's cancer was caused by bad luck Other items[a] 11. My parent's cancer was caused by sun exposure 12. My parent's cancer was caused by a higher power (e.g., it was an act of God) 13. My parent's cancer was caused by a germ or virus 14. My parent's cancer is hereditary (it runs in the family)

[a] Items that did not load onto separate coherent factor during factor analysis.

Appendix K

Evaluation of PPIQ-C using COSMIN criteria for measurement development

Measurement properties	Criteria	Were criteria addressed?
Content validity		
PROM development	**1a. PROM design**	
	General design requirements	
	1. Is a clear description provided of the construct to be measured?	Yes – PPIQ-C assesses young people's perceptions of their parent's cancer.
	2. Is the origin of the construct clear: was a theory, conceptual framework or disease model used or clear rationale provided to define the construct to be measured?	Yes – measure developed based on dimensions outlined within CSM.
	3. Is a clear description provided of the target population for which the PROM was developed?	Yes – adolescents and young adults aged 12-24 years who have a parent diagnosed with cancer.
	4. Is a clear description provided of the context of use?	Yes – PPIQ-C can be used for evaluative purposes to gain insight into young people's perceptions of their parent's cancer (e.g., to assist with development of supportive intervention). Total Negative Beliefs subscale can be used for discriminative purposes as a screening tool to identify young people at risk of psychological morbidity.
	5. Was the PROM development study performed in a sample representing the target population for which the PROM was developed	Convenience sample used – young people recruited through cancer support and advocacy groups. Sample not representative of the broader population of young people impacted by parental cancer.

Measurement properties	Criteria	Were criteria addressed?
	Concept elicitation (relevance and comprehensiveness)	
	6. Was an appropriate qualitative data collection method used to identify relevant items for a new PROM?	Yes – semi-structured, one-on-one interviews conducted with young people who had a parent diagnosed with cancer. Open-ended questions used to explore young people's experiences following their parent's cancer diagnosis and their cognitive and emotional representations of their parent's cancer.
	7. Were skilled group moderators/interviewers used?	Interviews were conducted by the student researcher/author of the thesis. The student researcher has a background in social health sciences and previous research experience in cancer support.
	8. Were the group meetings or interviews based on an appropriate topic or interview guide?	Yes – questions included in the interview schedule were based on the CSM (the theoretical framework underpinning the research).
	9. Were the group meetings or interviews recorded and transcribed verbatim?	Yes – interviews were audio-recorded and transcribed verbatim.
	10. Was an appropriate approach used to analyse the data?	Yes – a deductive (theory-driven) thematic analysis approach was employed to identify and organise themes using the CSM as a framework.
	11. Was at least part of the data coded independently?	Yes – a sub-set (30%) of transcripts were coded independently by a second researcher with a background in psychology and qualitative research.
	12. Was data collection continued until saturation was reached?	Yes – recruitment was finalised when no new themes emerged in three consecutive interviews (i.e., data saturation was reached).
	1b. Cognitive interview study or other pilot test	
	13. Was a cognitive interview study or other pilot test conducted?	Yes – cognitive interview study described in Chapter 4.
	General design requirements	
	14. Was the cognitive interview study or other pilot test performed in a sample representing the target population?	Convenience sample used – young people recruited through cancer support and advocacy groups. Sample not representative of the broader population of young people impacted by parental cancer. No male participants.

Measurement properties	Criteria	Were criteria addressed?
	Comprehensibility	
	15. Were patients asked about the comprehensibility of the PROM?	Yes – questions asked to assess comprehension and interpretation of items.
	16. Were all items tested in their final form?	No – items were not tested in their final form due to time restraints.
	17. Was an appropriate qualitative method used to assess the comprehensibility of the PROM instructions, items, response options, and recall period?	Yes – a think-aloud approach was used in combination verbal probes to assess relevance, comprehensiveness, comprehension, and accurate interpretation of items, and to identify any other problems with recall, the response format, and the instructions.
	18. Was each item tested in an appropriate number of patients?	Adequate – four young people participated in cognitive interviews.
	19. Were skilled interviewers used?	Cognitive interviews were conducted by the student researcher/author of the thesis. The student researcher has a background in social health sciences and previous research experience in cancer support.
	20. Were the interviews based on an appropriate interview guide?	Yes – cognitive interview schedule developed in accordance with De Leeuw and colleagues (2004) and previous think-aloud studies (Aujla, Vedhara, Walker, & Sprigg, 2018; McCorry, Scullion, et al., 2013; van Oort et al., 2011).
	21. Were the interviews recorded and transcribed verbatim?	Yes – interviews were audio-recorded and transcribed verbatim.
	22. Was an appropriate approach used to analyse the data?	Yes – during initial stage of analysis, participants’ thinking processes were coded according to the type of problem (or no problem) they experienced as they responded to each item. Codes defined based on specific categories used in previous think-aloud studies (Aujla, Vedhara, Walker, & Sprigg, 2018; McCorry, Scullion, et al., 2013; van Oort et al., 2011). Following this, participants’ responses to verbal probes were coded according to the source of confusion of any problems identified. Codes developed based on Peterson et al. (2017).
	23. Were at least two researchers involved in the analysis?	No – data were analysed by the student researcher only, due to time restraints and limitations to staff capacity.

Measurement properties	Criteria	Were criteria addressed?
	24. Were problems regarding the comprehensibility of the PROM instructions, items, response options, and recall period appropriately addressed by adapting the PROM?	Yes – changes were made to the wording of six items, instructions for *identity* subscale modified for clarity, instructions for *cause* subscale modified for clarity, response format modified for clarity and conciseness.
	Comprehensiveness	
	25. Were patients asked about the comprehensiveness of the PROM?	Yes – questions asked to assess relevance and comprehensiveness of items.
	26. Was the final set of items tested?	See item 16
	27. Was an appropriate qualitative method used to assess the comprehensiveness of the PROM?	See item 17
	28. Was each item tested in an appropriate number of patients?	See item 18
	29. Were skilled interviewers used?	See item 19
	30. Were the interviews based on an appropriate interview guide?	See item 20
	31. Were the interviews recorded and transcribed verbatim?	See item 21
	32. Was an appropriate approach used to analyse the data?	See item 22
	33. Were at least two researchers involved in the analysis?	See item 23
	34. Were problems regarding the comprehensiveness of the PROM appropriately addressed by adapting the PROM?	Yes – changes were made to include four additional items.
Content validity	**2a. Asking patients about relevance**	
	Design requirements	
	35. Was an appropriate method used to ask patients whether each item is relevant for their experience with the condition?	See item 17
	36. Was each item tested in an appropriate number of patients?	See item 18

Measurement properties	Criteria	Were criteria addressed?
	37. Were skilled group moderators/interviewers used?	See item 19
	38. Were the group meetings or interviews based on an appropriate interview guide?	See item 20
	39. Were the group meetings or interviews recorded and transcribed verbatim?	See item 21
	Analyses	
	40. Was an appropriate approach used to analyse the data?	See item 22
	41. Were at least two researchers involved in the analysis?	See item 23
	2b. Asking patients about comprehensiveness	
	Design requirements	
	42. Was an appropriate method used for assessing the comprehensiveness of the PROM?	See item 17
	43. Was each item tested in an appropriate number of patients?	See item 18
	44. Were skilled group moderators/interviewers used?	See item 19
	45. Were the group meetings or interviews based on an appropriate interview guide?	See item 20
	46. Were the group meetings or interviews recorded and transcribed verbatim?	See item 21
	Analyses	
	47. Was an appropriate approach used to analyse the data?	See item 22
	48. Were at least two researchers involved in the analysis?	See item 23

Measurement properties	Criteria	Were criteria addressed?
	2c. Asking patients about comprehensibility	
	Design requirements	
	49. Was an appropriate qualitative method used for assessing the comprehensibility of the PROM instructions, items, response options, and recall period?	See item 17
	50. Was each item tested in an appropriate number of patients?	See item 18
	51. Were skilled group moderators/interviewers used?	See item 19
	52. Were the group meetings or interviews based on an appropriate interview guide?	See item 20
	53. Were the group meetings or interviews recorded and transcribed verbatim?	See item 21
	Analyses	
	54. Was an appropriate approach used to analyse the data?	See item 22
	55. Were at least two researchers involved in the analysis?	See item 23
	2d. Asking professionals about relevance	PPIQ-C items not tested among professionals due to time constraints
	Design requirements	
	56. Was an appropriate method used to ask professionals whether each item is relevant for the construct of interest?	-
	57. Were professionals from all relevant disciplines included?	-
	58. Was each item tested in an appropriate number of professionals?	-

Measurement properties	Criteria	Were criteria addressed?
	Analyses	
	59. Was an appropriate approach used to analyse the data?	-
	60. Were at least two researchers involved in the analysis?	-
	2e. Asking professionals about comprehensiveness	PPIQ-C items not tested among professionals due to time constraints
	Design requirements	
	61. Was an appropriate method used for assessing the comprehensiveness of the PROM?	-
	62. Were professionals from all relevant disciplines included?	-
	63. Was each item tested in an appropriate number of professionals?	-
	Analyses	
	64. Was an appropriate approach used to analyse the data?	-
	65. Were at least two researchers involved in the analysis?	-
Internal structure		
Structural validity	*Statistical methods*	
	66. Was exploratory or confirmatory factor analysis performed?	Both – initial confirmatory factor analysis conducted to determine whether structure of PPIQ-C was consistent with original PPIQ.
	67. Was the sample size included in the analysis adequate?	Yes – sample size was > 7x the number of items and ≥100 participants.
	Other	
	68. Were there any other important flaws in the design or statistical methods of the study?	No – appropriate rotation method used and described – oblique rotation method (direct oblimin) employed to allow for correlation between factors.

Measurement properties	Criteria	Were criteria addressed?
Internal consistency	*Design requirements*	
	69. Was an internal consistency statistic calculated for each unidimensional scale or subscale separately?	Yes – Cronbach's alpha calculated for each subscale.
	Statistical methods	
	70. Was Cronbach's alpha or omega calculated?	Yes – Cronbach's alpha calculated for each subscale.
	Other	
	71. Were there any other important flaws in the design or statistical methods of the study?	No
Remaining measurement properties		
Test-retest reliability	*Design requirements*	
	72. Were the patients stable in the interim period on the construct to be measured?	Yes – assumed to be stable in interim period.
	73. Was the time interval appropriate?	Yes – two-week retest.
	74. Were the test conditions similar for the measurements (e.g., type of administration, environment, instructions)?	Yes – survey completed under same test conditions (online, at a location of participants' choosing) and using same instructions.
	Statistical methods	
	75. Was an intraclass correlation coefficient (ICC) calculated?	Yes

Measurement properties	Criteria	Were criteria addressed?
	Other	
	76. Were there any other important flaws in the design or statistical methods of the study?	Participants were comprised of young people who had a parent who had been diagnosed with, or received treatment for, any type or stage of cancer, within the last five years. This included participants whose parent had completed treatment or had died; these participants were asked to respond to items on the PPIQ-C based on how they would have felt at the time of their parent's diagnosis and treatment (i.e., retrospectively). This may have led to inaccuracies or inconsistencies in the data.
Hypotheses testing for construct validity	**6a. Comparison with other outcome measurement instruments (convergent validity)**	
	Design requirements	
	77. Is it clear what the comparator instrument(s) measure(s)?	Yes – outlined on pp. 182-6. Comparator instruments were the K10 (measure of psychological distress), IES-R (measure of post-traumatic stress symptoms in young adult participants), CRIES (measure of post-traumatic stress responses in adolescent participants), PTGI-SF (measure of post-traumatic growth in young adult participants), PTGI-C-R (measure of post-traumatic growth in adolescent participants), Brief COPE (measure of coping strategies in young adult participants), KIDCOPE (measure of coping strategies in adolescent participants).
	78. Were the measurement properties of the comparator instrument(s) sufficient?	Yes, all comparator instruments demonstrated excellent or good internal consistency in the sample under study, except for the KIDCOPE (avoidance strategies subscale demonstrated acceptable internal consistency, Cronbach's α = .72; approach strategies subscale demonstrated low internal consistency, Cronbach's α = .47).

Measurement properties	Criteria	Were criteria addressed?
	Statistical methods	
	79. Was the statistical method appropriate for the hypotheses to be tested?	Yes – Spearman rank-order correlation analysis conducted to examine correlations between
	Other	
	80. Were there any other important flaws in the design or statistical methods of the study?	PPIQ-C subscales and measures of psychological distress (K10), post-traumatic stress symptoms (IES-R and CRIES), post-traumatic growth (PTGI-SF and PTGI-C-R), and coping strategies (Brief COPE and KIDCOPE)
	6b. Comparison between subgroups (discriminative or known-groups validity)	
	Design requirements	
	81. Was an adequate description provided of important characteristics of the subgroups?	Yes – at risk vs. not at risk for psychological distress and post-traumatic stress.
	Statistical methods	
	82. Was the statistical method appropriate for the hypotheses to be tested?	Yes – receiver operating characteristic analysis.
	Other	
	83. Were there any other important flaws in the design or statistical methods of the study?	As described above for item 76, participants were comprised of young people who had a parent who had been diagnosed with, or received treatment for, any type or stage of cancer, within the last five years. This included participants whose parent had completed treatment or had died; these participants were asked to respond to items on the PPIQ-C based on how they would have felt at the time of their parent's diagnosis and treatment (i.e., retrospectively). This may have led to inaccuracies or inconsistencies in the data.

Measurement properties	Criteria	Were criteria addressed?
Responsiveness	**7. Construct approach (i.e. hypotheses testing: comparison between subgroups)**	
	Design requirements	
	84. Was an adequate description provided of important characteristics of the subgroups?	See item 81
	Statistical methods	
	85. Was the statistical method appropriate for the hypotheses to be tested?	See item 82
	Other	
	86. Were there any other important flaws in the design or statistical methods of the study?	See item 83