

Sleep Restriction Therapy as a Transdiagnostic Intervention for Insomnia and Anxiety in Middle Childhood

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Summary

Sleep problems occur in middle childhood. However, until recently, there has been very little research conducted into treatments for sleep problems in typically-developing children during the middle childhood years. The literature review presented in Chapter 2 reveals that, from 2011 – 2018, 9 published manuscripts reported on randomised controlled clinical trials evaluating cognitive-behavioural interventions for sleep problems in middle childhood, 2 published manuscripts reported on non-randomised controlled trials, and 3 published manuscripts reported on open trials or case reports. While these studies provide general support for the use of cognitive-behavioural sleep interventions in middle childhood, the heterogeneity of the treatment programs means that little can be inferred about the efficacy of individual treatment components. Furthermore, there has been a trend towards brief interventions for the treatment of sleep problems in both adults and adolescents, and it makes sense that this should also be extended to middle childhood.

In recent years, the field of clinical psychology has also seen a shift towards transdiagnostic interventions (which focus on psychological processes that are common among disorders, such as attentional bias), rather than diagnosis-specific interventions (e.g., cognitive behaviour therapy for insomnia). In light of the high comorbidity between sleep problems and anxiety during middle childhood, this was a logical area on which to focus research attention. Therefore, the randomised controlled trial described in Chapter 4 of the current thesis aimed to investigate the effect of a brief behavioural sleep intervention (i.e., sleep restriction or bedtime restriction therapy) on anxiety, during middle childhood. Both sleep restriction therapy and bedtime restriction therapy resulted in reduced total sleep time and increased evening sleepiness during the 2-week treatment period, as well as improved sleep onset latency and sleep efficiency, compared to a control group (who received bedtime regularisation). However, contrary to expectations, all three groups reported similar improvements in anxiety and worry, suggesting that there may be more factors contributing to this relationship.

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Implementation of any new treatment requires careful consideration of both the potential benefits and potential risks associated with its use. As described in Chapter 5, the present randomised controlled trial included measures of attention and cognitive performance, and also monitored occurrence of parasomnias during treatment, in order to assess for possible contraindications to the use of sleep restriction and bedtime restriction therapies in middle childhood. Somewhat surprisingly, results suggested that clinicians may not need to be overly concerned about the impacts of these therapies on daytime sleepiness, cognitive performance, or classroom attention, nor do they need to completely avoid using these techniques with school-aged children (6-14yrs) who have a history of parasomnias.

Overall, the findings of the present thesis suggest that sleep restriction and bedtime restriction therapies are effective, brief interventions that can improve the sleep of school-aged children with Chronic Insomnia Disorder. Further research is needed to investigate the mechanisms by which sleep restriction therapies may contribute to improvements in anxiety.

Declaration

I certify that this thesis does not incorporate without acknowledgement any material previously submitted for a degree or diploma in any university; and, 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.

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Glossary of Abbreviations

- AASM = American Academy of Sleep Medicine
- ADIS = Anxiety Disorders Interview Schedule
- APA = American Psychiatric Association
- ASD = Autism Spectrum Disorder
- ADHD = Attention Deficit Hyperactivity Disorder
- BRT = bedtime restriction therapy
- CBT = cognitive-behaviour therapy
- CBT-A = cognitive behaviour therapy for anxiety
- CBT-i = cognitive-behaviour therapy for insomnia
- DASS-21 = Depression Anxiety Stress Scales
- DSM-5 = Diagnostic and Statistical Manual for Mental Disorders, 5th Edition
- EEG = electroencephalogram
- 4wk FU = 4-week follow-up
- GP = general practitioner
- ICSD-2 = International Classification of Sleep Disorders, 2nd Edition
- ICSD-3 = International Classification of Sleep Disorders, 3rd Edition
- LMM = linear mixed model
- M = mean
- Mid = mid-treatment
- NES = Neuropsychological Evaluation System
- PDSS = Pediatric Daytime Sleepiness Scale
- Post= = post-treatment
- Pre = pre-treatment
- PSG = polysomnography
- RCT = randomised controlled trial

- REM = rapid eye movement
- SCAS = Spence Children's Anxiety Scale
- SD = standard deviation
- SE = sleep efficiency
- SNS = sympathetic nervous system
- SOL = sleep onset latency
- SPSS = Statistical Package for the Social Sciences
- SRT = sleep restriction therapy
- SSQ-R = School Situations Questionnaire Revised
- SWS = slow wave sleep
- TIB = time in bed
- TST = total sleep time
- WASO = wake after sleep onset
- WL = waitlist
- WSC = Worry Scale for Children
- WUT = wake-up time

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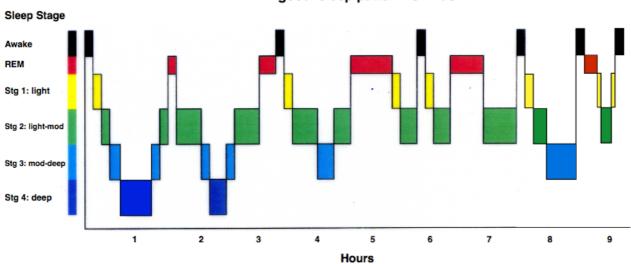
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Chapter 1: Introduction to Sleep and Insomnia in Middle Childhood

1.1 What is Sleep?

Many people believe that "good" sleep should be like a deep valley of unconsciousness across the night (Bruck, Dolan, & Lack, 2015). However, the real structure of sleep is more like a rollercoaster, with the individual moving through different stages of light sleep (stages 1 and 2), deep sleep (stages 3 and 4), and rapid eye movement (REM) sleep during the night (Dement & Kleitman, 1957). These stages of sleep are defined by distinct patterns of cortical activity, muscle tension, and eye movements, and are clustered into sleep cycles of approximately 90-mins that repeat across the night (Dement & Kleitman, 1957; Figure 1.1).



A 'good' sleep pattern for kids

Figure 1.1. An example of a good night's sleep, showing possible movement through stages of light sleep, deep sleep, and REM sleep, in approx. 90-min sleep cycles.

Note: Image developed by Child & Adolescent Sleep Clinic, Flinders University, Australia.

During light sleep (i.e., stages 1 and 2), the individual is easily aroused from sleep and may lack awareness that they have even fallen asleep (Goodenough et al., 1965; Williams et al., 1964). In contrast, it is much more difficult to arouse from deep sleep (i.e., stages 3 and 4) and, if an individual is woken from deep sleep, they are likely to feel very drowsy (Goodenough et al., 1965; Williams et al., 1964). During REM sleep the individual experiences increased cortical activity, characteristic eye movements, and the subjective experience of dreaming (Dement & Kleitman, 1957; Goodenough et al., 1965). As seen in Figure 1.1, deep sleep usually occurs more in the first half of the sleep period, and REM sleep occurs more in the second half of the sleep period (Dement & Kleitman, 1957; Williams et al., 1964). Brief awakenings can also be a normal part of a healthy sleep pattern for individuals of all ages.

Figure 1.2. has been removed due to copyright restrictions.

Broad changes in sleep architecture and overall sleep duration occur throughout the lifespan (Figure 1.2). Beginning in infancy, there is a steady decline in overall sleep duration with increasing age (Crabtree & Williams, 2009; Ohayon, Carskadon, Guilleminault, & Vitiello, 2004; Scholle et al., 2011), which continues even through adulthood (Ohayon et al., 2004). With a particular focus on childhood and adolescence, there is an age-related decrease in deep sleep and a corresponding

increase in stage 2 sleep (Crabtree & Williams, 2009; Ohayon et al., 2004; Scholle et al., 2011), with relative stability for stage 1 sleep (Scholle et al., 2011). Results for REM sleep have been inconsistent, with some reporting age-related increases in REM (Ohayon et al., 2004) and others reporting decreases in REM sleep during childhood and adolescence (Scholle et al., 2011).

1.2 What is Middle Childhood?

Middle childhood is a unique period of development that is nestled between early childhood (0-5 yrs) and adolescence (approx. 13 yrs and above), and thus typically includes children approx. 6-12 yrs (Eccles, 1999). In particular, children enter a crucial stage of cognitive development at approx. 6 yrs of age, which coincides with the commencement of formal schooling in many cultures (Eccles, 1999). The onset of adolescence is typically defined by physical changes associated with puberty, which may begin between 8-13 yrs for girls and 9-14 yrs for boys (with a mean of 10-11 yrs for girls and 11-12 yrs for boys; Kail & Cavanaugh, 2010; Mensah et al., 2013; Warren & Yu, 2016); thus the end of the "middle childhood" period may vary between individuals.

As described in later sections of the present thesis, sleep problems in middle childhood are typically characterised by difficulties with sleep onset or maintenance that may be associated with night-time fears and worries, a desire for parental presence, and/or inappropriate parental limit-setting at bedtime (American Academy of Sleep Medicine [AASM], 2005; AASM, 2014). In contrast, adolescent sleep is typically characterised by increasingly later sleep onset times and an increasing discrepancy between weekday (i.e., school day) and weekend wake-up times, reflecting a tendency to sleep-in on weekend mornings to compensate for lost sleep during the school week (Carskadon & Acebo, 2002; Laberge, Petit, Simard, Vitaro, Tremblay, & Montplaisir, 2001). For the purposes of the present thesis, the middle childhood period will be defined as including school-aged children approximately 6-12 yrs, excluding individuals who experience delayed sleep timing characteristic of adolescence.

Chapter 1: Introduction

1.3 "Sleep Need" in Middle Childhood

An individual's "sleep need" is broadly defined as the amount of sleep required for optimal health and daytime functioning (Jenni, 2013) and during middle childhood experts agree that ~9-12 hours sleep is ideal (Hirshkowitz et al., 2015; Paruthi et al., 2016). Obtaining the recommended hours of sleep on a regular basis is associated with improved outcomes in a number of domains, such as attention, behaviour, learning, memory, emotion regulation, mental health, physical health, and overall quality of life (Paruthi et al., 2016). Likewise, short sleep duration in children has been associated with poorer school performance (for a review, see Dewald, Meijer, Oort, Kerkhof, & Bogels, 2010), difficulties with attention, impulse control, behaviour regulation, and cognitive functioning (for a review, see Beebe, 2011), and increased risk of overweight and obesity (for a review, see Fatima, Doi, & Mamun, 2015).

There is large variation in "normal" sleep duration for children (Jenni, 2013; Matricciani, Blunden, Rigney, Williams, & Olds, 2013; Price et al., 2014) and it is recognised that there are individual differences in sleep need for children, just like there are individual differences in other physical attributes such as height and weight. While several research groups have produced percentile charts to reflect optimal sleep durations for children and adolescents (e.g., Price et al., 2014; Williams, Zimmerman, & Bell, 2013), this information can only reassure parents that their child's sleep duration lies within the typical range for their child's age, but it cannot confirm that the individual child is obtaining adequate sleep (Jenni, 2013). Instead, factors such as whether or not the child wakes spontaneously in the morning, their overall daytime alertness, behaviour, and emotional functioning all provide clues about whether the individual child is obtaining sufficient sleep (Jenni, 2013).

In line with this inter-individual variability, not all studies have found significantly different outcomes for children based on sleep duration alone (e.g., Price, Quach, Wake, Bittman, & Hiscock, 2015). This has led to an increasing focus on the importance of sleep quality, sleep timing, and sleep regularity, in addition to sleep duration (Paruthi et al., 2016). A combination of internal

(i.e., biological) and external (i.e., behavioural, environmental) factors are thought to affect these sleep variables during middle childhood.

1.4 Basic Sleep Biology

1.4.1 Process S and Process C

There are two biological processes that determine when we sleep (Borbély, 1982). Firstly, the homeostatic sleep drive (Process S; Borbély, 1982) results in a gradual build-up of sleep pressure during wakefulness (see Figure 1.3), which is dissipated during sleep. This increase in sleep pressure is experienced subjectively via an increased feeling of sleepiness, and objectively via decreased sleep onset latency (Taylor, Jenni, Acebo, & Carskadon, 2005) and increased delta wave activity (Achermann & Borbély, 2000). Sleep pressure first appears in infants by 2 months of age (Jenni, Borbély, & Achermann, 2004). During childhood, there is a gradual slowing in the rate of sleep pressure accumulation with increasing age, allowing children to sustain wakefulness for longer periods and eliminating the need for daytime naps before children start school (Figure 1.3; Jenni & LeBourgeois, 2006). This corresponds with a gradual decrease in "sleep need" across childhood (e.g., Price et al., 2014) as overnight sleep duration appears relatively constant even when daytime sleep decreases (Jenni & LeBourgeois, 2006). There is a further slowing of sleep pressure accumulation at the onset of puberty, with multiple studies finding a significant difference between pre-pubertal (approx. 11yrs) and pubertally-mature adolescents (approx. 14yrs; Jenni, Achermann, & Carskadon, 2005; Taylor et al., 2005). In particular, studies using polysomnography (PSG) have consistently reported reduced spectral power in the delta bands of the sleep electroencephalogram (EEG) during early adolescence, which suggests reduced homeostatic sleep drive (Campbell, Darchia, Khaw, Higgins, & Feinberg, 2005; Feinberg & Campbell, 2010; Gaudreau, Carrier, & Montplaisir, 2001; Jenni & Carskadon, 2004; Tarokh & Carskadon, 2010).

Figure 1.3. has been removed due to copyright restrictions.

In addition to the homeostatic sleep drive, sleep timing is also influenced by the circadian regulation system (Process C; Borbély, 1982). This explains natural fluctuations in alertness and sleepiness across the 24-hour day that are closely tied with the light/dark cycle and independent of changes in homeostatic sleep pressure (Figure 1.4; Dijk & Czeisler, 1994). Importantly, a circadian peak in alertness is thought to counteract increasing homeostatic sleep pressure late in the day, allowing the maintenance of wakefulness in the face of increasing sleep pressure, and a circadian trough assists with the maintenance of sleep despite lower sleep pressure in the second half of the sleep period (Dijk & Czeisler, 1994). While the circadian regulation system is primarily influenced by light exposure, there are individual differences in specific timing of the circadian "clock", with some individuals classified as morning-type (i.e., preferring early bedtimes and rise times) and others classified as evening-type (i.e., preferring later bedtimes and rise times; Roenneberg, Daan, & Merrow, 2003). The circadian rhythm for alertness and sleepiness, as depicted in Figure 1.4, is thought to develop in infants by 6 months of age, allowing the major sleep period to become consolidated at night (Jenni, Deboer, & Achermann, 2006; Weinraub et al., 2012). Furthermore, significant changes to the circadian system also occur at the onset of puberty and the interaction of the circadian regulation system and the homeostatic sleep drive at this time commonly results in a

pattern of delayed sleep timing (i.e., evening chronotype) for adolescents (Carskadon, Vieira, & Acebo, 1993; Crowley, Acebo, & Carskadon, 2007).

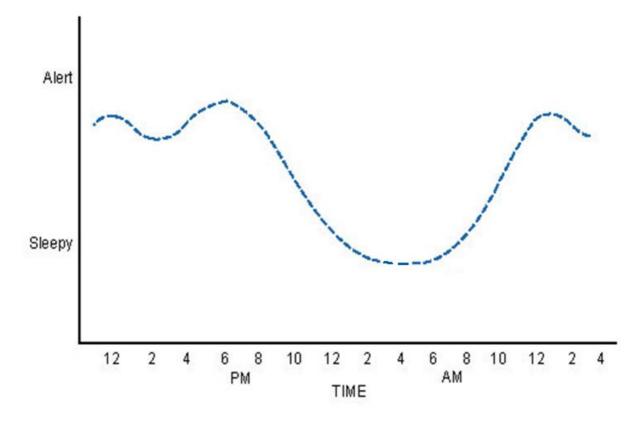


Figure 1.4. Circadian fluctuations in alertness and sleepiness across the 24-hour day. Note: Image developed by Child & Adolescent Sleep Clinic, Flinders University, Australia.

1.4.2 Sympathetic Nervous System

Two key components of the autonomic nervous system play a critical role in regulating human physiological arousal. The sympathetic nervous system (SNS) works to prepare the body for vigorous activity (e.g., increased heart rate, rapid breathing, constriction of blood vessels, decreased digestive activity) and is linked to the "fight or flight" response (Jansen, Nguyen, Karpitskiy, Mettenleiter, & Loewy, 1995). This system is activated in response to perceived threats in the environment in order to preserve safety. In contrast, the parasympathetic nervous system facilitates the opposite responses (e.g., decreased heart rate, slower breathing, increased digestive activity). When sleep onset naturally occurs, there is withdrawal of the SNS (Freedman & Sattler, 1982; Gradisar, Lack, Wright, Harris, & Brooks, 2006; Monroe, 1967). For individuals with insomnia, however, there is evidence of lack of withdrawal of the SNS due to hyperarousal during the pre-sleep period (for a review, see Bonnet & Arand, 2010). Interestingly, children with anxiety disorders have been found to have higher levels of cortisol during the 2-hour period prior to sleep onset, compared to children with depression or children with no history of psychiatric disorder (Forbes et al., 2006). This suggests that children who experience anxiety during the pre-sleep period may also experience elevated SNS activity prior to sleep onset (Forbes et al., 2006).

The relationship between sleep and anxiety in middle childhood will be discussed further in Section 1.9.1 of the current thesis. However, of relevance to basic sleep biology, it may be important to consider that anxiety may "mask" the measurement of processes C and S, as multiple internal and external variables may mask the endogenous circadian sleep rhythm (Rietveld, Minors, & Waterhouse, 1993).

1.5 Behavioural and Environmental Factors Influencing Sleep in Middle Childhood

In addition to the biological factors affecting sleep quality, duration, and timing, there are also many behavioural and environmental contributing factors. Across the middle childhood years, dramatic changes occur in children's social environments, in addition to the considerable physical and cognitive growth that occurs during this period (Eccles, 1999). Children learn to be increasingly independent from their parents and forge relationships outside the home, as well as developing their own sense of personal identity, primarily through their experiences at school and participation in extra-curricular activities (Eccles, 1999). However, compared to adolescence, parents still maintain considerable influence over their children's lives.

1.5.1 Parental influences

During middle childhood, parents play an important role in influencing child sleep. Research suggests that greater parental knowledge about child sleep is associated with better sleep habits for children (e.g., regular bedtime and wake time, falling asleep without parental presence; McDowall, Galland, Campbell, & Elder, 2017; Owens & Jones, 2011; Owens, Jones, & Nash; 2011), although not necessarily longer sleep duration (McDowall et al., 2017; Owens & Jones, 2011). Overall, however, parental knowledge about child sleep tends to be poor (McDowall et al., 2017). According to Harvey's (2002) cognitive model of adult insomnia, dysfunctional beliefs about sleep play a key role in the maintenance of sleep problems. While the application of this model to childhood insomnia is unknown, recent findings suggest that both children's own dysfunctional beliefs about sleep (Gregory, Cox, Crawford, Holland, & Harvey, 2009; Ng, Dodd, Gamble, & Hudson, 2013), and mothers' beliefs about child sleep (Ng et al., 2013), may be associated with child sleep problems.

During middle childhood, parents have an important role in setting and enforcing rules related to bedtime schedules and routines, and thus influencing the child's sleep timing. Perhaps not surprisingly, the presence and enforcement of rules about bedtime for school-aged children and adolescents (5-17yrs) has been associated with longer sleep duration on school nights (Buxton et al., 2015; Pyper, Harrington, & Manson, 2017) and better sleep quality (Buxton et al., 2015). Similarly, children who decide their own bedtime have been found to have less time in bed (TIB) on school nights (Meijer, Habekothe, & van den Wittenboer, 2001), parents of children with inconsistent bedtimes are more likely to report that their child has a 'sleep problem' (Uebergang, Arnup, Hiscock, Care, & Quach, 2017), and children with "excellent" sleep quality are more likely to have bedtimes and wake-up times that are "about the same every day" (Buxton et al., 2015). Therefore, it appears that having parental rules about bedtime that are enforced with regular bedtimes and wake-up times may provide the best context for good sleep in middle childhood.

In a broader family context, family involvement in daily activities (e.g., eating meals together, being at home together after school) has been associated with longer sleep duration for children (Adam, Snell, & Pendry, 2007; Ray & Roos, 2012). However, increased parental involvement is not universally beneficial, as parental presence at sleep onset has been associated with more night wakings (Mindell, Meltzer, Carskadon, & Chervin, 2009) and poor parental sleep

habits (in particular, irregular sleep schedules and late sleep timing) have been associated with increased sleep problems in children (Komada et al., 2009).

1.5.2 Pre-bedtime activities and routines

Consistency in pre-bedtime routines has been associated with both good sleep quality (Henderson & Jordan, 2010) and longer sleep duration (Mindell et al., 2009). Similarly, disruptions in pre-bedtime routines have been associated with increased night waking (Fiese, Winter, Sliwinski, & Anbar, 2007). Henderson and Jordan (2010) also found differences between children with good and poor sleep quality in the types of activities completed in the hour before bedtime. In particular, good sleepers had more "adaptive" pre-bedtime activities (e.g., shower/bath, brushing teeth, putting on pyjamas, going to the toilet, having a cuddle/story, being tucked-in by parents) and poor sleepers had more "maladaptive" pre-bedtime activities (e.g., active play, video games, TV, having a snack/drink; Henderson & Jordan, 2010). Thus, it seems that both the type of activities completed before bedtime, and the consistency of the pre-bedtime routine, have an important impact on subsequent sleep.

1.5.3 Screen time

In recent years, the potential influence of screen-based media on sleep in children has been studied extensively (for reviews, see Cain & Gradisar, 2010; Hale & Guan, 2015; LeBourgeois et al., 2017). The relationship between poor sleep and use of screen-based media is complex and bidirectional, with some longitudinal studies suggesting that increased use of screen-based media is associated with later sleep difficulties (e.g., later bedtimes, shorter total sleep time [TST]; Nuutinen, Ray, & Roos, 2013). In contrast, other studies suggest that shorter TST at baseline predicts longer screen time at follow-up (Bartlett, Gentile, Bartlett, Eisenmann, & Walsh, 2012; Magee, Lee, & Vella, 2014). Some studies have also found different results for different media types (e.g., Magee et al., 2014), which adds to the complexity of this relationship.

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1.5.4 Cultural factors

Research suggests that cultural and racial differences exist in sleep practices and sleep patterns for children. Children from predominantly Asian countries have been found to have significantly later bedtimes and shorter night-time sleep (Liu, Liu, Owens, & Kaplan, 2005; Mindell, Sadeh, Wiegand, How, & Goh, 2010; Mindell, Sadeh, Kwon, & Goh, 2013), increased bed-sharing and room-sharing (Mindell et al., 2010; Mindell et al., 2013), and increased parental perception of sleep problems (Liu et al., 2005; Mindell et al., 2010; Mindell et al., 2013; Sadeh, Mindell, & Rivera, 2011) compared with those from predominantly Caucasian countries. During the pre-school years (3-6yrs), differences in daytime napping also occur, with increased daytime napping for children from predominantly Asian countries compared to predominantly Caucasian countries (Mindell et al., 2013). Interestingly, similar observations have been made between different racial groups within the United States (Patrick, Millet, & Mindell, 2016) and napping persists into middle childhood (up to 8 yrs) for a greater percentage of black American children compared to white American children (Crosby, LeBourgeois, & Harsh, 2005). Overall, though, there are no differences in total sleep duration over 24 hours for these different cultural or racial groups, suggesting no difference in overall sleep need (Crosby et al, 2005; Mindell et al., 2013). Therefore, the differences are more likely to be related to cultural differences in parental beliefs about sleep and sleep practices (e.g., attitude towards napping, parent-set bedtimes; Crosby et al., 2005 Patrick et al., 2016; Short et al., 2013) and cultural differences in daytime activities (e.g., school start time, extra-curricular activities; Short et al., 2013).

In summary, there are many behavioural and environmental factors influencing sleep in middle childhood, including parental knowledge and beliefs about child sleep, parental sleep habits, household rules about sleep, pre-bedtime activities and routine, and daily activities (e.g., screen time). While all children experience occasional nights of poor sleep, the complex interaction of these environmental factors (e.g., parental beliefs and rules about sleep) with the child's unique

biology (e.g., short sleep need) may contribute to the development of a more persistent "sleep problem" for some children.

1.6 Common Sleep Problems in Middle Childhood

1.6.1 Insomnia

A recent survey of Australian children at school entry level found that almost 40% of parents reported that their child had a "sleep problem", including difficulty falling asleep, difficulty sleeping alone, nocturnal wakefulness, and morning tiredness (Quach, Hiscock, & Wake, 2012). Night-time fears and anxiety are common in middle childhood, with recent studies suggesting that 80-85% of children 7-12yrs experience night-time fears (Gordon, King, Gullone, Muris, & Ollendick, 2007; Muris, Merckelbach, Ollendick, King, & Bogie, 2001). The content of children's worry changes with age, with younger children worrying more about imaginary creatures, and older children worrying more about personal security and environmental threats (Gordon et al., 2007). Furthermore, children as young as 8 years old have been found to catastrophise about poor sleep and its consequences (Gregory, Noone, Eley, & Harvey, 2010), which is a common characteristic of adult insomnia (Harvey & Greenall, 2003). Children whose sleep difficulties are persistent and associated with daytime impairments may thus meet criteria for a diagnosis of insomnia (AASM, 2014).

According to the International Classification of Sleep Disorders, 3rd Edition (ICSD-3; AASM, 2014), insomnia is defined as a persistent difficulty with initiation, maintenance, consolidation, or quality of sleep, which occurs despite adequate opportunity and circumstances, and results in one or more negative consequences during the day (e.g., fatigue, mood disturbance, behavioural problems). For a child to obtain a diagnosis of Chronic Insomnia Disorder, the nighttime sleep difficulties and daytime impairments must be present at least 3 days/nights per week for a period of >3 months (for full criteria see Appendix A.1; AASM, 2014). Individuals presenting with insomnia symptoms present for <3 months may be given a diagnosis of Short-Term Insomnia Disorder (AASM, 2014). A diagnosis of insomnia requires a comprehensive clinical interview, including detailed information about medical, psychiatric, and specific sleep history (Littner et al., 2003) and a diagnosis may be given regardless of comorbid psychological or medical conditions, because even in these cases the sleep problem often deserves treatment in its own right (American Psychiatric Association [APA], 2013). Insomnia can occur at any point across the lifespan, and has been acknowledged to occur in middle childhood (AASM, 2014).

In children, the night-time sleep difficulties associated with insomnia may present as bedtime resistance or inability to fall asleep without parental assistance (AASM, 2014). In line with this, a child's sleep difficulties may appear to "resolve" when their desired conditions for sleep are met (e.g., when a parent is present) but may "reappear" if the child is forced to sleep alone. Prior to 2014, this was described as Behavioural Insomnia of Childhood (for full criteria see Appendix A.2; AASM, 2005). It is estimated that approximately 10-30% of children experience this form of insomnia (AASM, 2014).

According to the International Classification of Sleep Disorders, 2nd Edition (ICSD-2; AASM, 2005), there were two distinct subtypes of Behavioural Insomnia of Childhood. The *sleeponset association type* was characterised by reliance on specific stimuli (e.g., people, objects, or settings) for initiating sleep or returning to sleep after waking, and an inability to sleep without these stimuli (AASM, 2005). The *limit-setting type* was characterised by bedtime resistance or refusal related to parental difficulties with limit-setting and behaviour management (AASM, 2005). Children could also meet criteria for a *combined type* if they met criteria for both subtypes for the specified time period. While this distinction is no-longer made under the current diagnostic system (AASM, 2014), it may be noted that both subtypes are relevant to middle childhood. For example, children with night-time fears and worries who experience difficulty falling asleep without parental presence may develop sleep-onset associations (e.g., sleeping in parents' bed or bedroom, parent sleeping in child's bedroom) and this may result in inappropriate or inconsistent parental limitsetting (e.g., allowing children to sleep in the parents' bed after night wakings but not at sleep onset). In line with this, previous research has found that children with anxiety have less

consistency between weekend and weekday sleep schedules, compared with healthy controls (Hudson, Gradisar, Gamble, Schniering, & Rebelo, 2009), and that inconsistent sleep schedules are associated with bedtime resistance (Blader, Koplewicz, Abikoff, & Foley, 1997).

The ICSD-2 (AASM, 2005) described additional insomnia subtypes, which are distinguished based on daytime and night-time symptoms, as well as the proposed aetiology of the sleep complaints. These subtypes included psychophysiological insomnia (characterized by conditioned associations between the bedroom environment and arousal/alertness), paradoxical insomnia (characterized by subjective reports of severe insomnia that are not matched by objective sleep measures nor by daytime dysfunction), idiopathic insomnia (characterized by longstanding insomnia complaints beginning during infancy or early childhood with no identifiable trigger), inadequate sleep hygiene (characterized by the presence of daily activities that interfere with the maintenance of good sleep), and insomnia due to a mental or medical disorder (AASM, 2005). Most of these were uncommon in childhood (AASM, 2005), although both psychophysiological insomnia and idiopathic insomnia could occur during the school years (Vriend & Corkum, 2011). With the advent of the ICSD-3 (AASM, 2014), the distinction between these different subtypes of insomnia was removed because there was little evidence to support these distinct phenotypes and their utility in clinical practice was questionable (Zucconi & Ferri, 2014).

1.6.2 Delayed Sleep-Wake Phase Disorder

As described in section 1.4.1, significant changes to Process C and Process S at the onset of puberty commonly result in a pattern of delayed sleep timing, whereby adolescents are unable to fall asleep until very late at night and have difficulty waking in time for morning school commitments (Carskadon, Vieira, & Acebo, 1993; Crowley, Acebo, & Carskadon, 2007). In order to compensate for insufficient sleep during the school week, adolescents also tend to sleep-in late on weekend days (Carskadon & Acebo, 2002; Laberge et al., 2001), which may reduce sleep pressure on subsequent nights and/or cause further delay of the circadian sleep rhythm, producing an ongoing cycle (Taylor, Wright, & Lack, 2008). For some adolescents, this pattern is persistent

and associated with impaired daytime functioning (e.g., daytime sleepiness, depressed mood) resulting in a diagnosis of Delayed Sleep-Wake Phase Disorder (DSWPD; AASM, 2014). While DSWPD may occur during the middle childhood years, the prevalence of DSWPD increases markedly across the adolescent years, and is related to pubertal development (Carskadon, Vieira, & Acebo, 1993).

1.6.3 Parasomnias

A third group of sleep problems that are common in middle childhood are broadly classified as parasomnias, which are undesirable physical experiences that occur during sleep or during transitions from wakefulness to sleep and vice versa, and can include complex movements, behaviours, emotions, and cognitions (AASM, 2014). The most common parasomnias experienced during middle childhood are disorders of arousal (e.g., sleepwalking, sleep terrors), which are thought to occur as a result of incomplete arousal from deep sleep, and sleep enuresis (bedwetting), which is thought to be related to difficulty arousing from deep sleep despite the need to urinate (AASM, 2014). All of these parasomnias occur most often during childhood and tend to resolve spontaneously by early adolescence, consistent with the decrease in deep sleep that occurs during this developmental period (AASM, 2014); as such, they usually do not require treatment in their own right (Kotagal, 2009). However, disorders of arousal may be precipitated by sleep deprivation (Galbiati, Rinaldi, Giora, Ferini-Strambi, & Marelli, 2015; Kotagal, 2009; Owens & Mohan, 2016; Simon & Byars, 2016), which may be relevant when considering a child who presents with symptoms of insomnia and is chronically sleep-deprived.

1.7 Measurement of Sleep and Insomnia

Accurate assessment of childhood sleep disturbance is important for both research and clinical care, and the multi-dimensional nature of sleep has resulted in the development of a variety of methods of measurement (Lewandowski, Toliver-Sokol, & Palermo, 2011; Matricciani, 2013). The most common methods used in middle childhood include questionnaires, sleep diaries, wrist

actigraphy, and polysomnography (Figure 1.5). Not surprisingly, there are advantages and disadvantages to each approach, which will be addressed in turn.

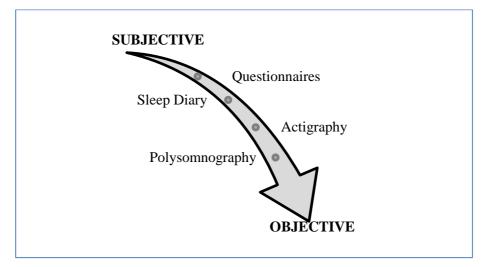


Figure 1.5. Methods of measurement for sleep and insomnia symptoms fall on a continuum. Note: Image developed by author.

Questionnaire measures of sleep usually collect information about typical sleep patterns, habits, or behaviours retrospectively from the child or parent, by asking them to report on typical patterns during a specified time period (Lewandowski et al., 2011). They provide the most subjective information about the child's sleep and are the least intrusive and time-consuming for families to complete; however, the nature of retrospective reporting means that they may be subject to biases in recall or reporting, and they do not allow for examination of night-to-night variability in sleep. There are a large number of questionnaires that have been developed in recent years; however, in many cases there has been little scrutiny of their psychometric properties (Spruyt & Gozal, 2011). The questionnaires used most commonly in middle childhood are the Children's Sleep Habits Questionnaire (parent-report; Owens, Spirito, & McGuinn, 2000) and the Sleep Disturbance Scale for Children (parent-report; Bruni et al., 1996), which both cover a range of sleep behaviours and have been independently verified as having a "well-established" evidence base (Lewandowski et al., 2011). Of the well-validated questionnaires currently available, there is unfortunately limited information about their sensitivity to longitudinal changes in sleep (e.g., as a result of maturation or treatment) and there are currently no questionnaires available to specifically assess insomnia symptoms in childhood (Lewandowski et al., 2011). Thus, they are not ideal for use as diagnostic tools or primary outcome measures in treatment studies for insomnia in childhood. However, questionnaire measures may provide useful information about subjective elements such as daytime consequences of poor sleep, with scales such as the Pediatric Daytime Sleepiness Scale (PDSS; Drake et al., 2003) being widely-used and well-validated in middle childhood (Lewandowski et al., 2011). According to AASM recommendations, such measures of daytime functioning are essential in monitoring outcomes for patients with insomnia (Edinger et al., 2015; Morgenthaler et al., 2015).

As previously mentioned, there is a high prevalence of night-time fears and anxiety in middle childhood, and this is understandably linked with sleep disturbance in this population. Anxiety is usually measured using standardised questionnaires, such as the Spence Children's Anxiety Scale (SCAS; Spence, 1998) or the Screen for Child Anxiety-Related Emotional Disorders (Birmaher et al., 1997), which have both parent-report and child self-report versions available. Previous research with similar populations has found that self-report questionnaires are more sensitive to treatment effects than parent-report questionnaires (Clementi & Alfano, 2014; Muris, Verweij, & Meesters; 2003; Paine & Gradisar, 2011; Stewart & Gordon, 2014). This is consistent with the classification of anxiety as an internalising problem, which may not be observable to parents (Muris et al., 2003). Thus, children should be considered the primary informant for their own fear and anxiety symptoms (Stallings & March, 1995).

Compared to sleep questionnaires, daily sleep diaries provide more detailed information about sleep (Buysse, Ancoli-Israel, Edinger, Lichstein, & Morin, 2006), including variables such as bedtime, lights-out time, sleep onset latency, time spent awake during the night, and wake-up time (Matricciani, 2013). This results in a more accurate measurement of total sleep time, compared to a single question about sleep duration (Matricciani, 2013). In middle childhood, sleep diaries may be completed by the child alone, parent alone, or the child and parent together (Matricciani, 2013). Trends in the literature suggest that researchers have used parent-report measures for children younger than 9 yrs and self-report for children older than 9 yrs (Matricciani, 2013). However, there is no empirical evidence to support this cut-off (Matricciani, 2013), and recent AASM recommendations suggest that self-report may be appropriate for children from 7 yrs (Edinger et al., 2015). Families are generally instructed to record information about the previous night's sleep immediately upon waking in the morning, thus minimising biases related to errors in recall and also allowing for observation of night-to-night variability in sleep patterns (Buysse et al., 2006; Matricciani, 2013). Sleep diaries are commonly used in research and clinical settings as a reliable and valid measure of an adult's subjective experience of insomnia (Buysse et al., 2006; Morin, 2003), and have been successfully used in previous studies with children (e.g., Gruber, Somerville, Bergmame, Fontil, & Paquin, 2016; Hudson, Rapee, et al., 2009; Paine & Gradisar, 2011; Papaconstantinou, Hodnett, & Stremler, 2018; Schlarb, Velten-Schurian, Poets, & Hautzinger, 2011; Schlarb, Bihlmaier, Velten-Schurian, Poets, & Hautzinger, 2013). According to AASM recommendations, sleep diaries are essential in monitoring sleep quality outcomes for patients with insomnia (Edinger et al., 2015; Morgenthaler et al., 2015).

Wrist actigraphy monitors are worn like a small wristwatch and provide an objective estimate of sleep-wake patterns based on gross motor movement (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). Movement data are collected continuously and, when downloaded, provide automatic scoring of wake vs sleep using a validated algorithm, which is reliable when used over at least 5 nights (at least for adolescents; Acebo et al., 1999). Actigraphy has become an increasingly popular method for estimating sleep parameters over the past 20 years because it provides a more objective measure than sleep diaries or questionnaires, is non-invasive, and can be used for extended periods of time in the home environment (Meltzer et al., 2012). The American Academy of Sleep Medicine recommends actigraphy "for delineating sleep patterns, and to document treatment responses in normal infants and children (in whom traditional sleep monitoring by polysomnography can be difficulty to perform and/or interpret)" (Morgenthaler et al., 2007, p.525).

In paediatric populations (i.e., 0-18yrs), research has consistently demonstrated that actigraphy provides accurate measurement of sleep, but not wake after sleep onset (Meltzer et al., 2012). While actigraphy is more objective than sleep diaries and questionnaires, there is also still a small degree of subjectivity in scoring and interpretation of actigraphy data, as most devices require sleep diary records of bedtime or lights-out time to calculate sleep onset latency.

PSG is the most objective method for measuring sleep and is often considered the "gold standard", especially for adults (Morin, 2003). PSG typically involves the use of electrodes to measure brain activity, eye movement, and muscle tone, which can be used together to identify sleep versus wakefulness and also to distinguish between different stages of sleep (e.g., stages 1-4, REM). Overnight sleep monitoring using PSG may be uncomfortable for the individual and is usually restricted to a single night of measurement for practical and financial reasons (i.e., it is expensive to pay a sleep technician for PSG setup and scoring), which may be problematic due to the so-called "first night effect" (i.e., the tendency for sleep to be worse than usual on the first night of monitoring) and the inability to examine night-to-night variability in sleep, which is particularly common for individuals with insomnia (Littner et al., 2003). For all of these reasons, PSG is not indicated for the routine evaluation of insomnia (Littner et al., 2003).

In summary, a comprehensive assessment of sleep and insomnia in middle childhood should include a multi-method approach (Buysse et al., 2006), including wrist actigraphy (an objective measure of sleep), daily sleep diaries (a subjective measure of sleep), and questionnaires (a subjective measure of daytime consequences of poor sleep). While both parent-report and child self-report questionnaires are available, previous research with similar populations has found that child self-report questionnaires are more sensitive to treatment effects than parent-report questionnaires (Clementi & Alfano, 2014; Paine & Gradisar, 2011). Therefore, only child self-report questionnaires will be used in the current thesis. However, parents can be a useful resource for information about a child's sleep (e.g., sleep history, parasomnias, etc.) and, as previously

mentioned, a diagnosis of insomnia also requires a comprehensive clinical interview, including detailed information about medical, psychiatric, and specific sleep history (Littner et al., 2003).

1.8 Consequences of Sleep Problems in Middle Childhood

As previously mentioned, poor sleep in middle childhood has been associated with negative outcomes in many domains, including poorer school performance (for a review, see Dewald et al., 2010), difficulties with attention, impulse control, behaviour regulation, and cognitive functioning (for a review, see Beebe, 2011), emotional difficulties (e.g., anxiety, depression; Gregory & Sadeh, 2012), and increased risk of overweight and obesity (for a review, see Fatima et al., 2015). Furthermore, daytime consequences of poor sleep are an essential diagnostic criteria for Chronic Insomnia Disorder (AASM, 2014) and may include symptoms such as daytime fatigue, sleepiness, difficulties with attention/concentration/memory, irritability, behavioural problems (e.g., hyperactivity, impulsivity, aggression), and impaired social/family/academic performance.

Sleep problems during middle childhood have wide-ranging implications, not only for the individual child, but for their whole family. Childhood sleep disorders have been associated with lower quality of maternal sleep (Meltzer & Mindell, 2007), maternal daytime sleepiness (Boergers, Hart, Owens, Streisand, & Spirito, 2007), lower maternal mood (Meltzer & Mindell, 2007), poorer parental mental health (Quack, Hiscock, & Wake, 2012; Quach, Mesnah, & Hiscock, 2016), parental marital conflict (Kelly & El-Sheikh, 2011), and higher levels of parenting stress (Byars, Yeomans-Maldonado, & Noll, 2011; Meltzer & Mindell, 2007). Interestingly, parental daytime functioning has been found to be a significant factor in help-seeking for insomnia in middle childhood (Newton, 2017). Using retrospective report, parents revealed that the impact of child sleep problems on their own daytime functioning was the most commonly reported non-child factor that motivated or would motivate them to seek treatment for their child's sleep problem (Newton, 2017). Furthermore, the likelihood of seeking help for child sleep problems increased with increases

in parental scores on the Depression Anxiety Stress Scales (DASS-21 [Lovibond & Lovibond, 1995]; Newton, 2017).

1.9 Interventions for Sleep Problems in Middle Childhood

Medication is not recommended as a first-line treatment for insomnia in childhood and should only be used in conjunction with cognitive-behavioural interventions and for a short period of time (Arboledas et al., 2017; Nunes & Bruni, 2015). Consistent with this ethos, recent research suggests that parents prefer behavioural interventions over homeopathic or pharmacological treatments (Goodday, Corkum, & Smith, 2014). However, middle childhood insomnia has been relatively under-studied, compared to other age groups (e.g., young children, 0-5yrs). A recent systematic review and meta-analysis of behavioural interventions for paediatric insomnia (0-18 yrs) found only 2 controlled trials for typically-developing primary school-aged children (age range not specified), and yet 12 for typically-developing young children (approx. 0-5 yrs; Meltzer & Mindell, 2014). In clinical practice, management of childhood insomnia seems to be largely based on research and practice guidelines for adults (Tikotzky & Sadeh, 2010; Vriend & Corkum, 2011), recommended treatments for comorbid conditions (e.g., anxiety; Tikotzky & Sadeh, 2010), or case studies and "clinical judgment" (Vriend & Corkum, 2011). While there are a number of clinical texts available (e.g., Durand, 2008; Meltzer & Crabtree, 2015), these also seem to justify behavioural sleep interventions in middle childhood using treatment outcome research conducted with other age groups (e.g., young children).

Cognitive-behaviour therapy for insomnia (CBT-i) is considered the "treatment of choice" for adult insomnia (Okajima, Komada, & Inoue, 2011; Riemann & Perlis, 2009) and there is strong evidence for a variety of cognitive and/or behavioural strategies used individually or in combination (e.g., Harvey et al., 2014; Miller et al., 2014; Morgenthaler, Kramer, et al., 2006). Two recent papers describing clients presenting to a paediatric outpatient sleep clinic suggest that behavioural interventions are commonly used in middle childhood (mean age 8 years; Meltzer, Moore, &

Mindell, 2008; Williamson, Patrick, Rubens, Moore, & Mindell, 2016). However, despite their common use in clinical practice, it is currently unknown whether CBT-i strategies maintain their effectiveness when used with children. A contemporary and comprehensive review of the literature in this area is presented in **Chapter 2** of the current thesis, including a description of each of the techniques that have been used in middle childhood.

As previously discussed, parents have a critical influence on sleep in middle childhood (e.g., Buxton et al., 2015; Meijer et al., 2001; Pyper et al., 2017), and thus it is essential that they are involved in treatment. Parents are time poor due to the many and varied demands involved in juggling work, family and social responsibilities. Furthermore, parents rarely discuss sleep problems with primary health care professionals (e.g., general practitioners [GPs]; Blunden et al., 2004) suggesting that sleep problems are seen as having a lower priority (compared to other health/medical problems). Therefore, there is value in developing brief interventions for sleep problems in middle childhood. Interestingly, there has also been a shift towards brief interventions for adult insomnia and adolescent sleep disturbance, and treatment outcome research suggests that they are effective (e.g., Bartel, Huang, Maddock, Williamson, & Gradisar, 2018; Buysse et al., 2011; Edinger & Sampson, 2003; Falloon, Elley, Fernando, Lee, & Arroll, 2015; Miller et al., 2014).

1.10 Common Comorbidities

In middle childhood, insomnia commonly co-occurs with other conditions such as developmental disorders (e.g., Autism Spectrum Disorder [ASD]; intellectual disability), behavioural disorders (e.g., Attention Deficit Hyperactivity Disorder [ADHD]) and emotional disorders (e.g., depression, anxiety). In fact, up to 85% of children with developmental disorders (Spruyt & Curfs, 2015) and up to 90% of children with anxiety disorders (Alfano, Ginsburg, & Kingery, 2007; Alfano, Pina, Zerr, & Villalta, 2010) also experience clinically-significant sleep problems.

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1.10.1 Sleep in Developmental and Behavioural Disorders

There has been a lot of research investigating sleep in developmental and behavioural disorders (e.g., ASD; ADHD; intellectual disability), and evidence suggests that cognitive-behavioural interventions for sleep problems are effective for children with these comorbid disorders (for a review, see Spruyt & Curfs, 2015). However, the validity of standard self-report measures of sleep and daytime functioning in these populations is unknown and may not be consistent with their psychometric properties for typically-developing children due to impairments in cognitive functioning (Finlay & Lyons, 2001). Furthermore, children with ASD may be unable to tolerate the use of objective measures of sleep (e.g., actigraphy) due to sensory sensitivities (Hodge, Parnell, Hoffman, & Sweeney, 2012). Therefore, sleep in children with developmental or behavioural disorders will not be a focus of the current thesis.

1.10.2 Sleep and Depression

While there is a high comorbidity between sleep disturbance and depression in adolescent samples (Bennett, Ambrosini, Kudes, Metz, & Rabinovich, 2005; Chorney, Detweiler, Morris, & Kuhn, 2008; Lovato & Gradisar, 2014; Lovato, Short, Micic, Hiller, & Gradisar, 2017; Roberts & Duong, 2013), this relationship is less clear in middle childhood (Chorney et al., 2008). One study suggested that up to 73% of Hungarian children (7-14 yrs) with a diagnosed depressive episode also reported disturbed sleep (Liu et al., 2007). However, it has also been consistently reported that the association between depression and sleep disorders becomes stronger as children move into adolescence (Gregory & O'Connor, 2002; Knowles & MacLean, 1990; Roberts, Roberts & Chen, 2002), and results were not reported separately for younger and older participants in the Hungarian sample. Furthermore, in an Australian sample of children (7-13 yrs) with insomnia, reports of depressive symptoms were low and there was no significant change in depressive symptoms during insomnia treatment (Paine and Gradisar, 2011). Therefore, depression will not be a focus of the present thesis.

Chapter 1: Introduction

1.10.3 Sleep and Anxiety

There is a high comorbidity between childhood sleep difficulties and anxiety symptoms, and this relationship is complex and bidirectional in nature (for reviews, see Gregory & Sadeh, 2012; Leahy & Gradisar, 2012a; McMakin & Alfano, 2015). Evidence from recent longitudinal studies suggests that sleep problems during childhood predict later anxiety symptoms (Gregory et al., 2005; Kelly & El-Sheikh, 2014; Sadeh, Tikotzky, & Kahn, 2014), and that late childhood (10-13yrs) may be a sensitive developmental period for this association (Kelly & El-Sheikh, 2014). However, anxiety is also a predictor of later sleep problems (albeit a weaker association; Kelly & El-Sheikh, 2014; McMakin & Alfano, 2015). One recent study reported that approximately one third of school-aged children presenting for treatment of insomnia also report clinically-significant anxiety (Van Dyk, Becker, & Byars, in press), while studies of children with anxiety disorders have reported prevalence of clinically-significant sleep problems of up to 90% (Alfano et al., 2007; Alfano et al., 2010). Treatment studies suggest that interventions for anxiety in childhood may also improve sleep problems (Caporino et al., 2017; Clementi, Alfano, Holly, & Pina, 2016; Donovan, Spence, & March, 2017; Peterman et al., 2016; Storch et al., 2008) and interventions for sleep problems may also improve anxiety symptoms (Paine & Gradisar, 2011), likely as a result of shared mechanisms.

According to Dahl and Harvey (2007), key psychological factors play a role in the association between sleep problems and anxiety. Children with anxiety experience physiological arousal and hypervigilance, as they shift their attention towards potential threats in their environment (Dudeney, Sharpe, & Hunt, 2015). However, sleep onset requires "switching off" awareness and responsiveness to the environment (Carskadon & Dement, 2011); thus sleep is inhibited (physiologically) under conditions of perceived threat (Dahl & Harvey, 2007). Thus, in a simply physiological sense, hypervigilance during the pre-sleep period inhibits sleep because sleep and arousal are opponent processes (see Figure 1.6; Dahl, 1996; Dahl & Harvey, 2007). Interestingly, while hypervigilance to potential threats is a common feature of anxiety disorders (APA, 2013), a recent study found hypervigilance to potential threats was associated with sleep

problem presence and severity, regardless of anxiety presence or severity (Ricketts et al., 2018). Other recent research has examined the roles of emotion, motivation, and self-regulation in relation to vigilance, although full discussion of these factors is beyond the scope of the present thesis. **Chapter 3** of the current thesis includes a review of two open trials that test elements of this theory

and inform the main study of this thesis.

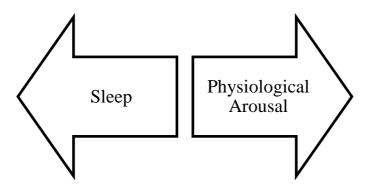


Figure 1.6. Children with insomnia experience physiological arousal at bedtime, which interferes with sleep onset because sleep and arousal are opponent processes. Note: Image developed by author.

Worry is a common phenomenon in middle childhood (Muris, Meesters, Merckelbach, Sermon, & Zwakhalen, 1998), and is defined as "an anticipatory cognitive process involving repetitive, primarily verbal thoughts related to possible threatening outcomes and their potential consequences" (Vasey & Daleiden, 1994, p.186). While there are strong links between worry and anxiety (Weems, Silverman, & La Greca, 2000), and worry is a prominent feature of several paediatric anxiety disorders (e.g., Generalized Anxiety Disorder, Separation Anxiety Disorder; APA, 2013), they also represent distinct constructs (Weems et al., 2000), as worry refers to a cognitive process and anxiety refers to an affective state. Worry may be particularly important in understanding the relationship between sleep and anxiety, as the time in bed prior to sleep onset provides an opportunity for worry to occur (McMakin & Alfano, 2015).

Another prominent feature of anxiety disorders is behavioural avoidance (APA, 2013). Socalled "safety behaviours" can emerge as a means of avoiding external cues (e.g., situations) or internal stimuli (e.g., thoughts) that have previously been associated with anxious affect (Helbig-Lang & Peterman, 2010). Cognitive-behavioural models suggest that safety behaviours play an important role in the maintenance of anxiety (Helbig-Lang & Peterman, 2010). In particular, they help us to understand why anxiety may persist despite the repeated experience that feared consequences do not occur (Helbig-Lang & Peterman, 2010; Lovibond, Mitchell, Minard, Brady, & Menzies, 2009). For example, children with insomnia who experience anxiety at bedtime may seek reassurance via parental presence while attempting to fall asleep (Alfano & Mellman, 2010; Paine & Gradisar, 2011), and never learn that feared consequences (e.g., personal harm) do not occur in the absence of parental presence. Alternatively, these children may resist or refuse to go to bed (in an attempt to avoid anxious affect previously associated with the bed or bedroom or the act of attempting to fall asleep) and thus develop further bedtime problems.

Considering the high degree of overlap between sleep problems and anxiety symptoms in middle childhood, the field needs to ensure that treatments for insomnia during this developmental period are able to target these cognitive-behavioural processes in order to break the vicious cycles maintaining insomnia for these children. Furthermore, more research is needed to understand the mechanisms linking sleep and anxiety in this population, which will ultimately provide further guidance for treatment recommendations. These aims will be addressed in the current thesis.

1.11 Outline of Thesis

Before designing and evaluating a new intervention for any physical or mental health condition, it is first necessary to understand what has been done before. Therefore, **Chapter 2** begins the current thesis with a narrative literature review summarising relevant research relating to the treatment of insomnia in middle childhood. This provides valuable background information that led to the development of the interventions described in subsequent chapters.

Chapter 3 describes unpublished findings from two open trials that provided preliminary evidence of the feasibility of brief interventions (i.e., bedtime restriction therapy, sleep restriction

therapy) for the treatment of insomnia in middle childhood, and also provided insight into the relationship between sleep and anxiety in this population, guiding the development of the randomised controlled trial (RCT) to follow. Whilst these pilot studies showed promising results, replication using a controlled design is important.

Chapter 4 describes an RCT to evaluate two brief single-component interventions (i.e., sleep restriction therapy, bedtime restriction therapy) for insomnia in middle childhood, including their efficacy for both sleep and anxiety symptoms, compared to an active control group who received bedtime regularisation. The intention of each treatment arm was to (1) reduce TIB (bedtime restriction therapy), (2) reduce both TIB and TST (sleep restriction therapy), or (3) not manipulate TIB (control), in order to test different mechanisms linking improvements in sleep and anxiety. Specifically, participants receiving bedtime restriction therapy were expected to experience decreases in worry as a result of reduced wakefulness in bed, while participants receiving sleep restriction therapy were expected to experience increased evening sleepiness as a result of reduced TST (and thus increased homeostatic sleep pressure), which may reduce anxiety as sleepiness and arousal are opponent processes (Dahl & Harvey, 2007). As well as providing full details of the design, implementation, and results of the RCT, this chapter also further discusses these potential mechanisms.

Chapter 5 explores possible contraindications for restriction therapies in middle childhood, as data on cognitive performance, school attention, and parasomnias were collected during the RCT to assess for such contraindications.

The thesis concludes with a final Discussion section (**Chapter 6**), which discusses possible mechanisms linking sleep and anxiety in middle childhood, clinical implications from the treatments tested, and directions for future research.

1.11.1 Summary of Thesis Objectives

Until recently, there has been very little research conducted into treatments for sleep problems in typically-developing children during the middle childhood years. The present thesis aims to address this gap in the literature, with the following key objectives:

- 1. Review evidence for cognitive and/or behavioural interventions for sleep problems in middle childhood.
- 2. Compare the efficacy of sleep restriction therapy, bedtime restriction therapy, and bedtime regularisation (control) for insomnia in middle childhood.
- 3. Examine effects of sleep restriction therapy, bedtime restriction therapy, and bedtime regularisation on anxiety symptoms in middle childhood.
- 4. Examine possible contraindications to treatment.
- 5. Conduct a preliminary investigation of possible process variables involved in the relationship between insomnia and anxiety in middle childhood.

1.11.2 Note to the Reader

The main body of this thesis includes three manuscripts that are being prepared for publication (i.e., Chapter 2, Chapter 4, Chapter 5). The format of these manuscripts necessitates some duplication of information, although this has been avoided wherever possible by referring to the same information presented in a previous chapter. To improve readability, references have been collated at the end of the thesis.

Chapter 2: Cognitive-Behavioural Interventions for Sleep Problems in Middle Childhood: A Review of Current Evidence

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Author Contributions

NC led review conceptualisation, literature search, results interpretation, and manuscript preparation. MG contributed to review conceptualisation and manuscript preparation.

Cain, N. & Gradisar, M. (in preparation). Cognitive-behavioural interventions for sleep problems in middle childhood: A review of current evidence.

2.1 Abstract

Sleep problems are common in middle childhood. However, until recently, there has been very little research conducted into treatments for sleep problems in typically-developing children during the middle childhood years. This chapter provides a review of existing research evaluating cognitive and/or behavioural treatment strategies for insomnia and other sleep problems in middle childhood, including a summary of individual strategies used and their combined effects on sleep outcome variables. In particular, articles included in this review (1) contain participants aged approximately 6-12 years, (2) include at least one cognitive or behavioural strategy aimed at improving sleep, and (3) measure at least one standardised sleep outcome variable (e.g., sleep diary, actigraphy, sleep questionnaire). Overall, the reviewed studies provide general support for the use of cognitive and/or behavioural sleep interventions in middle childhood. However, the heterogeneity of the treatment programs means that little can be inferred about the efficacy of individual treatment components. Implications and directions for future research are discussed.

2.2 Introduction

2.2.1 Sleep Problems in Middle Childhood

Sleep duration has been decreasing consistently during the past century, and data from a recent meta-analysis suggest that children have been obtaining about 30 mins less sleep than they need (Matricciani, Olds, Blunden, Rigney, & Williams, 2012). This is potentially concerning, as experimental research suggests that a reduction in sleep duration of as little as 30 mins per night can result in negative outcomes for school-aged children (Sadeh, Gruber, & Raviv, 2003). While the prevalence of sleep problems in infancy and early childhood is generally well-recognised by parents, health professionals, and organisations (e.g., National Sleep Foundation, https://sleepfoundation.org; Sleep Health Foundation, https://www.sleephealthfoundation.org.au; Raising Children Network, https://raisingchildren.net.au), there is less recognition that sleep problems can persist into middle childhood for some individuals, or that new sleep problems can emerge as children progress through different developmental stages (McDowall et al., 2017; Mindell, Moline, Zendell, Brown, & Fry, 1994; Mindell & Owens, 2003; Owens, 2001; Schreck & Richdale, 2011). A recent survey of Australian children at entry to school found that almost 40% of parents reported that their child had a "sleep problem", with the most common problems being difficulty falling asleep, difficulty sleeping alone, nocturnal wakefulness, and morning tiredness (Quach et al., 2012). As discussed in Chapter 1, children whose sleep difficulties are persistent and associated with daytime impairments (e.g., fatigue, mood disturbance, behavioural problems) may meet criteria for a diagnosis of insomnia (American Academy of Sleep Medicine [AASM], 2014).

2.2.2 Treatment of Insomnia in Middle Childhood

Over a decade ago, the AASM released evidence-based practice parameters for the treatment and management of insomnia for adults (Morgenthaler, Kramer, et al., 2006) and young children (i.e., under 5 years old; Morgenthaler, Owens, et al., 2006). Interestingly, however, there are no AASM practice parameters for the middle childhood years (i.e., 6-12 yrs), reflecting the relative absence of empirical research on treatment techniques during this developmental period.

Indeed, a recent systematic review and meta-analysis of behavioural interventions for paediatric insomnia (0-18 yrs) found only 2 controlled studies for typically-developing primary school-aged children (Meltzer & Mindell, 2014).

Management of childhood insomnia in clinical practice seems to be largely based on research and practice guidelines for adults (Tikotzky & Sadeh, 2010; Vriend & Corkum, 2011), recommended treatments for comorbid conditions (e.g., anxiety; Tikotzky & Sadeh, 2010), or case studies and "clinical judgment" (Vriend & Corkum, 2011). Cognitive behaviour therapy for insomnia (CBT-i) is considered the "treatment of choice" for adult insomnia (Okajima et al., 2011; Riemann & Perlis, 2009) and there is strong evidence for the use of a variety of cognitive and/or behavioural strategies used individually or in combination (e.g., Harvey et al., 2014; Miller et al., 2014; Morgenthaler, Kramer, et al., 2006). Two recent papers describing clients presenting to a paediatric outpatient sleep clinic suggest that behavioural interventions are commonly used in middle childhood (mean age 8 years; Meltzer et al., 2008; Williamson et al., 2016). Despite their common use in clinical practice, it is currently unknown whether CBT-i strategies maintain their effectiveness when used in middle childhood.

In their recent review of behavioural interventions for paediatric insomnia, Meltzer and Mindell (2014) summarised results relating to four key outcome variables (i.e., sleep onset latency [SOL], time spent awake after sleep onset [WASO], number of night wakings, and sleep efficiency [SE; percentage of time in bed actually asleep]). Positive results were reported for SOL (Paine & Gradisar, 2011), SE (Paine & Gradisar, 2011), and night wakings (Paine & Gradisar, 2011; Quach, Hiscock, Ukoumunne, & Wake, 2011). While these results are promising, and Meltzer and Mindell (2014) conclude that behavioural sleep interventions should be used in middle childhood, the results of their review provide no guidelines regarding what strategies should be used and how they should be implemented. It must also be acknowledged that these conclusions are based on only 2 controlled studies, and cognitive components of CBT-i were not covered in this review.

The current chapter will provide a review of existing research evaluating treatment strategies for insomnia and other sleep problems in middle childhood, including a summary of individual strategies used and their combined effects on sleep outcome variables. The chapter will conclude with a summary of implications and directions for future research.

2.3 Literature Search and Inclusion Criteria

To identify papers for this review, a literature search was conducted using the PsychInfo and PubMed databases and Google Scholar with a variety of search terms (e.g., [sleep OR insomnia] AND [intervention OR therapy OR treatment] AND [child OR paediatric]). Articles were also identified from the reference lists of these papers, and from the reference lists of relevant systematic reviews, meta-analyses, and book chapters. Included articles described original research studies that contained participants aged approximately 6-12 years (or partially covered this range) and were published in English. For studies that included a wider age range than this, the methodology was examined and the study was included provided the focus was on children attending primary school (also known as elementary school). Studies that focused on adolescents were excluded, due to pubertal changes in the accumulation of homeostatic sleep pressure and circadian factors (i.e, delayed sleep timing), which may mean that behavioural sleep strategies work differently for adolescents than they do in middle childhood. Papers were included if (1) treatment contained at least one cognitive or behavioural strategy aimed at improving sleep, and (2) measurements comprised at least one standardised sleep outcome variable (e.g., sleep diary, actigraphy, sleep questionnaire)¹. Studies of children with comorbid medical conditions (e.g., asthma, eczema) were included, and participants did not need to have a formal insomnia diagnosis (i.e., studies could include children with general 'sleep problems' or short sleep duration). However, studies of children with neurodevelopmental disorders (e.g., Autism Spectrum Disorder [ASD], Attention

¹ Papers were excluded when the focus was on treatment for night-time fears and the only outcome measures were fear questionnaires (e.g., Friedman & Ollendick, 1989; Muris et al., 2003; Stewart & Gordon, 2014) as these tend to measure emotion rather than sleep.

Deficit Hyperactivity Disorder [ADHD]) were excluded, due to several recent comprehensive reviews within this area (e.g., Rigney et al., in press; Spruyt & Curfs, 2015), the desire to include cognitive elements of treatment (which may operate differently in children with developmental disabilities), and to align with inclusion criteria for the randomised controlled trial (RCT) described in Chapter 4. Published abstracts, unpublished theses and dissertations were also excluded.

2.4 Summary of Findings

Table 2.1 provides descriptive information about the 14 studies included for review. Nine studies were identified that involved typically-developing children (Fehr, Russ, Ievers-Landis, 2016; Gruber et al., 2016; Hart, Hawley, & Wing, 2016; Paine & Gradisar, 2011; Quach et al., 2011; Schlarb et al., 2011; Schlarb et al., 2018; Tamura & Tanaka, 2014; Tamura & Tanaka, 2016). Two studies involved children hospitalised for comorbid medical conditions (Meltzer & Booster, 2016; Papaconstantinou et al., 2018). Three studies evaluated treatment programs for children with anxiety, which either included transdiagnostic components of CBT-i (e.g., exposure, relaxation, cognitive restructuring; Clementi et al., 2016; Donovan et al., 2017) or included additional sessions that focused on sleep (Clementi & Alfano, 2014). Further information about the design and results of each study is presented in Table 2.1.

Table 2.1

Descriptive information about studies included for review

(2014) N=4 Disorder Clementi et al. Open trial 7-16yrs Anxiety diso		Participants	Diagnosis	Treatment type	Standardised outcome measures	Main results Improvements in sleep, worry, and anxiety symptoms.		
		•	Generalised Anxiety Disorder	Manualised treatment program (2 sessions on sleep & 12 sessions on anxiety)	Sleep questionnaire (parent & self-report) Anxiety questionnaire (parent & self-report) Worry questionnaire (self-report)			
		Anxiety disorder or sub-clinical anxiety symptoms	Manualised treatment program (12 sessions, anxiety focus)	Sleep questionnaire (parent report) Anxiety questionnaires (self-report)	Improvements in sleep, anxiety, and pre-sleep arousal. Greater improvement in sleep-related problems for intervention group compared to control. Improvements were maintained at 6-month follow-up.			
Donovan et al. (2017)	(M=9.5yrs)deliveredN=63intervention withonline therapist		intervention with online therapist support (16 sessions,	Sleep questionnaire (parent report) Anxiety questionnaire (parent & self-report)				
Fehr et al. (2016)	Case report	4-6yrs (M=5.25yrs) N=4	Disordered sleep	Manualised intervention (3 child sessions & 1 parent session)	Sleep questionnaire (parent report) Fear questionnaires (parent & self-report)	Improvements in sleep, anxiety, bedtime resistance, total sleep problems, and general fears.		

Authors Study type I		Participants	Diagnosis	Treatment type	Standardised outcome measures	Main results		
Gruber et al. (2016)	Non- randomised controlled trial	7-11yrs (M=8.5yrs) N=71	No sleep disorder	Manualised intervention (6 sessions) delivered in school classroom	Sleep diary Wrist actigraphy Health questionnaire (parent report) School grades	Improvements in sleep (SOL, TST, SE) and school grades for intervention group relative to control.		
Hart et al. (2016)	RCT	8-11yrs (M=9.2yrs) N=12	Short sleep duration			Increase in actigraph sleep period by 40mins for intervention group, with a corresponding decrease of 16mins for control group. No change in SOL or WASO.		
Meltzer & Booster (2016)	Non- randomised controlled trial	M=7yrs N=93	No sleep disorder	Parent group education session	Child sleep questionnaire (parent report) Parent sleep questionnaires	Improvements in child and parent sleep occurred for both groups.		
Paine & Gradisar (2011)	RCT	7-13yrs (M=9.3yrs) N=42	Behavioural Insomnia of Childhood	Manualised treatment program (6 sessions)	Sleep diary Wrist actigraphy Anxiety & mood questionnaires (parent & self-report) Daytime sleepiness questionnaire (self- report)	Improvements in sleep (SOL, WASO, SE) and anxiety symptoms for intervention group relative to control. Improvements were maintained at 6-month follow-up.		

Authors Study type		Participants	Diagnosis	Treatment type	Standardised outcome measures	Main results		
Papaconstantinou et al. (2018)	RCT	4-10yrs (M=6.5yrs) N=48	No sleep disorder Hospitalised for acute or chronic illness or surgery	Manualised intervention (1 session)	Wrist actigraphy Sleep & anxiety questionnaires (parent report) Pain scale (self-report)	Improvements in sleep (TST, WASO) for intervention group relative to control. No differences in anxiety.		
Quach et al. (2011)	RCT	5-6yrs (M=5.7yrs) N=108	Parent-reported moderate/severe sleep problem	Strategies selected based on individual sleep problem (2-3 sessions)	Sleep, psychosocial health, & behaviour questionnaires (parent report)	Sleep problems improved in both groups, but improved quicker in the intervention group. Improvements were observed at 3-, 6- and 12-month follow-ups.		
Schlarb et al. (2011)	RCT	5-10yrs (M=7.8yrs) N=38	Insomnia	Manualised group treatment program (6 sessions)	Sleep diary Sleep questionnaires (parent report)	Improvements in sleep and sleep-related anxiety for intervention group, although control group also showed some improvements.		
Schlarb et al. (2018)	RCT	5-10yrs (M=8.1yrs) N=112	Chronic Insomnia Disorder	Manualised group treatment program (6 sessions)	Sleep diary Wrist actigraphy Sleep & daytime sleepiness questionnaires (parent & self-report)	Greater improvements in sleep for intervention group than control group, although control group improved on some variables. Improvements were maintained at 12- month follow-up.		

Authors	Study type	Participants	Diagnosis	Treatment type	Standardised outcome measures	Main results
Tamura & Tanaka (2014)	Cluster RCT	Elementary school grade 4-6 N=148	No sleep disorder (66% self-reported "poor sleep")	Sleep education session (delivered in school classroom) with self-selected strategies	Sleep diary	Improvements in sleep parameters for intervention group relative to control group.
Tamura & Tanaka (2016)	Cluster RCT	12-13yrs N=243	No sleep disorder	Sleep education session (delivered in school classroom) with self-selected strategies	Sleep diary	Improvements in sleep parameters for intervention group relative to control group.

Note: RCT = Randomised Controlled Trial.

2.4.1 Overall Effectiveness of Cognitive and Behavioural Strategies to Improve Sleep in Middle Childhood

Most of the studies reviewed here used questionnaire measures of sleep. The most commonly used questionnaire was the parent-report Children's Sleep Habits Questionnaire (Owens et al., 2000). Seven studies reported results for this measure (Clementi & Alfano, 2014; Clementi et al., 2016; Fehr et al., 2016; Papaconstantinou et al., 2018; Quach et al., 2011; Schlarb et al., 2011; Schlarb et al., 2018) and four of these observed significant improvements compared to a control group (Papaconstantinou et al., 2018; Quach et al., 2011; Schlarb et al., 2018). Improvements were reported for total sleep problems (Papaconstantinou et al., 2018; Quach et al., 2011; Schlarb et al., 2011; Schlarb et al., 2018), bedtime resistance (Quach et al., 2011; Schlarb et al., 2011), sleep anxiety (Schlarb et al., 2011), sleep onset delay (Quach et al., 2011; Schlarb et al., 2018), and sleep duration (Quach et al., 2011). In addition, Clementi and colleagues (2016) reported improvement in total sleep problems, bedtime resistance, and sleep anxiety, although they did not have a control group, thus the effect of historical changes unrelated to treatment, the maturation of participants, or unspecified treatment effects (e.g., therapeutic alliance) cannot be ruled out (Dimitrov & Rumrill, 2003; Shirk & Karver, 2003).

Fewer studies reported changes in actual sleep parameters from wrist actigraphy (5 studies: Gruber et al., 2016; Hart et al., 2016; Paine & Gradisar, 2011; Papaconstantinou et al., 2018; Schlarb et al., 2018) or sleep diaries (5 studies: Clementi & Alfano, 2014; Paine & Gradisar, 2011; Schlarb et al., 2018; Tamura & Tanaka, 2014; Tamura & Tanaka, 2016). Improvements were reported for total sleep time (TST) or sleep period duration (Gruber et al., 2016; Hart et al., 2016; Tamura & Tanaka, 2014; Tamura & Tanaka, 2016), SOL (Gruber et al., 2016; Paine & Gradisar, 2011; Schlarb et al., 2018; Tamura & Tanaka, 2016), number or duration of night wakings (Paine & Gradisar, 2011; Schlarb et al., 2018), and SE (Gruber et al., 2016; Paine & Gradisar, 2011; Schlarb et al., 2018). However, not all studies reported consistent results, with 4 studies finding no significant change in TST (Clementi & Alfano, 2014; Paine & Gradisar, 2011; Papaconstantinou et

al., 2018; Schlarb et al., 2018) despite reporting significant changes in other aspects of sleep. Furthermore, the clinical significance of these changes must be considered, as the magnitude of change in some variables was small (e.g., SOL improvement range 2-30mins; SE improvement range 2-7%; TST improvement range 15-40mins).

Follow-up data were only collected in 5 studies (Clementi & Alfano, 2014; Donovan et al., 2017; Paine & Gradisar, 2011; Quach et al., 2011; Schlarb et al., 2018) and the duration of the follow-up periods varied considerably. It is positive to note that all of these studies reported that improvements were maintained (or improved further) up to 3-month (Clementi & Alfano, 2014; Quach et al., 2011; Schlarb et al., 2018), 6-month (Donovan et al., 2017; Paine & Gradisar, 2011; Quach et al., 2011; Schlarb et al., 2018), and 12-month follow-ups (Quach et al., 2011, Schlarb et al., 2018). Interestingly, the multiple follow-up design implemented by Quach and colleagues (2011) allowed closer inspection of sequential changes in their sleep variables. In particular, they observed lower bedtime resistance at 3- and 6-months, reduced bedtime delay at 6- and 12-months, and longer sleep duration by 12-months post-treatment. This latter finding is particularly noteworthy, as not all studies report immediate improvements in TST following sleep interventions, and this work suggests that it may take up to 12-months for this to occur.

Overall, preliminary evidence from the reviewed studies suggests that CBT-i techniques may be effective in the treatment of insomnia and general 'sleep problems' in middle childhood. This is also consistent with the recommendation of Meltzer and Mindell (2014), despite the small number of studies in this age group included in their review. In the studies reviewed here, improvements were observed across a number of different measures of sleep (i.e., sleep diary, wrist actigraphy, questionnaires) and anxiety (although those studies measuring anxiety also included therapeutic components directly focused on anxiety). The results of the RCTs reviewed here also reinforce the importance of including a control group in sleep intervention research, because some of these control groups also showed improvement in sleep over the course of the study (Meltzer & Booster, 2016; Quach et al., 2011; Schlarb et al., 2011; Schlarb et al., 2018).

2.4.2 Individual Components of Treatment

A variety of cognitive and behavioural strategies have been used to improve sleep in middle childhood, and a summary of the strategies used in the reviewed studies is presented in Table 2.2. Almost all studies evaluated multi-component interventions, which either provided manualised treatment (i.e., all participants received the same psycho-education and implemented the same treatment strategies; 11 studies, 79%) or selected from a variety of techniques depending on the specific sleep problems experienced by the individual child (3 studies, 21%).

Across all of the reviewed studies, sleep hygiene education was the most common component of treatment (covered in 86% of reviewed studies). Sleep hygiene education typically includes education about health practices (e.g., exercise, diet, substances) and environmental factors (e.g., light, noise, temperature) that are believed to improve sleep (Hauri, 1991; Meltzer & Mindell, 2004). Pre-bedtime activities may also be considered as part of sleep hygiene education, although for the purposes of this review they have been considered separately (as bedtime routines may be particularly important for children). Eight studies (57%) included discussion about pre-bedtime routine or activities (either general education about appropriate pre-bedtime activities for children, or implementation of a structured pre-bedtime routine). Consideration of general sleep hygiene and pre-bedtime activities are important in order to ensure that the child is not engaging in any physically, emotionally, or psychologically stimulating activities prior to bedtime, which may interfere with the withdrawal of the sympathetic nervous system (SNS) and prevent sleep onset from occurring naturally. Furthermore, having a structured bedtime routine may help the child psychologically prepare for bedtime, as classical conditioning principles (Pavlov, 1906, 1927) may allow the individual steps in the bedtime routine (e.g., putting on pyjamas, getting into bed, etc.) to become conditioned stimuli for sleepiness (Stepanski & Wyatt, 2003).

	Cognitive and/or Behavioural Sleep Strategies											
Authors	Education about normal sleep	Sleep hygiene	Regular sleep schedule	Pre-bedtime activities / bedtime routine	Graded exposure	Graduated extinction / Checking	Bedtime fading / Sleep restriction	Sleep extension	Stimulus control instructions	Rewards	Relaxation	Cognitive therapy
Clementi & Alfano (2014)		Х		Х	X	х	X					
Clementi et al. (2016)					X					X	X	
Donovan et al. (2017)											х	X
Fehr et al. (2016)	Х	х		Х		Х						х
Gruber et al. (2016)		X		Х								
Hart et al. (2016)		х	х	Х				х		Х		
Meltzer & Booster (2016)	х	X	X	Х		х				х		
Paine & Gradisar (2011)	Х	X			X		х				х	X
Papaconstantinou et al. (2018)	Х	X	X	Х							х	
Quach et al. (2011)	Х	Х				х	X			X	х	
Schlarb et al. (2011)		Х	X	Х					х		х	X
Schlarb et al. (2018)	х	X	X	Х		х	X		х		х	X
Tamura & Tanaka (2014)		X										
Tamura & Tanaka (2016)		X										

Table 2.2

Relaxation strategies (e.g., controlled breathing, progressive muscle relaxation) were used in 50% of reviewed studies. These techniques aim to reduce physiological arousal at bedtime and encourage withdrawal of the SNS, allowing sleep onset to occur. Relaxation strategies have been brought into sleep interventions from the anxiety treatment literature (Alfano & Mellman, 2010).

Somewhat surprisingly, only six studies (43%) included education about aspects of normal sleep (e.g., how much sleep is needed for children, information about sleep stages and sleep cycles). This is surprising considering that parental knowledge about sleep is generally poor (McDowall et al., 2017; Schreck & Richdale, 2011) and knowledge is an important component in models of behaviour change (Prochaska & DiClemente, 1983).

Five studies (36%) included a regular sleep schedule, which involves setting a regular "lights out" time and wake-up time to be applied consistently 7 days a week, without increasing or decreasing the child's time in bed. This is based on the idea that regular bedtimes and wake times help to regulate the natural circadian rhythm for alertness and sleepiness across the 24 hour day, and evidence that sleep schedule regularity has been associated with better sleep quality (Buxton et al., 2015) and longer sleep duration (El-Sheikh et al., 2013; Erath & El-Sheikh, 2015; Kjeldsen et al., 2014; Suratt et al., 2007) in middle childhood.

Graduated extinction or "checking" was also included in 5 of the reviewed studies (36%). Graduated extinction involves gradual removal of parental presence at bedtime, using principles of operant conditioning (Skinner, 1963). After saying goodnight to their child, the parent ignores the child's protests (e.g., crying, calling out) and instead returns to the child's room after a predetermined length of time and provides brief, minimally stimulating contact and reassurance, without removing the child from bed. The parent gradually increases duration between return visits (therefore, this intervention is sometimes referred to as "checking") until the child falls asleep without the parent present. Most of the existing evidence for graduated extinction comes from research with infants and young children (0-5yrs, Morgenthaler, Owens, et al., 2006). Cognitive therapy was also included in 36% of reviewed studies. This involves identifying and challenging unhelpful thoughts and/or beliefs, and has been brought into paediatric sleep interventions from the anxiety treatment literature (Alfano & Mellman, 2010). Consistent with this, cognitive models of adult insomnia (e.g., Harvey, 2002) suggest that certain patterns of cognitive activity (e.g., selective attention toward and monitoring of insomnia symptoms, distorted perception of sleep and daytime consequences, dysfunctional beliefs about sleep) play a key role in the maintenance of insomnia in adults. Cognitive therapy is considered one of the critical components of CBT-i for adults (Harvey et al., 2014), and research suggests that children as young as 8 years old catastrophise about poor sleep and its consequences (Gregory et al., 2010). Thus, it is logical that cognitive therapy may be included in treatment for children who have reached this level of cognitive maturity.

Positive reinforcement (i.e. rewards) for compliance with desired sleep-related behaviours were also included in 5 studies (36%). This is often used in combination with other treatment strategies and is based on the theory of operant conditioning (Skinner, 1963). Operant conditioning theory predicts that behaviours are controlled by their consequences, such that positive reinforcement is likely to increase the frequency of a particular behaviour, and negative consequences (e.g., punishment or removal of reinforcement) are likely to decrease the frequency of the behaviour (Skinner, 1963). In the field of paediatric sleep, parental presence, attention, and reactions may be unknowingly providing positive reinforcement for undesirable sleep-related behaviours. Treatment strategies that use positive reinforcement may involve redirecting parental attention or implementing other rewards (e.g., toys, treats, special outings) for desired sleep-related behaviours (e.g., staying in own bed all night).

Bedtime fading was used in 4 studies (29%). This is analogous to sleep restriction therapy in adults (Spielman, Saskin, & Thorpy, 1987; Miller et al., 2014) and involves temporarily reducing the child's time in bed to more closely match their typical average sleep duration, usually by delaying their bedtime (Cooney, Short, & Gradisar, 2018; Piazza & Fisher, 1991). This allows the

child to build up greater homeostatic sleep pressure (Borbély, 1982), which aids sleep onset and improves sleep consolidation (Cooney et al, 2018). Furthermore, via principles of classical conditioning (Pavlov, 1906, 1927), the repeated experience of going to bed when sleepy strengthens the association between bed and sleep and, over time, the bed itself comes to elicit feelings of sleepiness. As SOL and WASO improve, the child's bedtime is gradually shifted earlier to extend their time in bed.

Graded exposure was used in 3 studies (21%). Graded exposure procedures are a common treatment component for anxiety (Alfano & Mellman, 2010; Rapee, Schniering, & Hudson, 2009), and in the reviewed studies could involve offering the child a stepped approach to changing their problematic bedtime behaviour. This is used for children who require parental presence to fall asleep due to anxiety about separating from parents at bedtime, and involves drawing up an "exposure hierarchy" whereby the parent's presence is gradually withdrawn, and thus the child is exposed to situations that are increasingly more anxiety-provoking, at the same time learning that their feared consequences do not occur (Rapee et al., 2009). Graded exposure may be coupled with positive reinforcement when each step is achieved, as a reward for the desired behaviour (i.e., sleeping more independently).

Stimulus control therapy was only used in 14% of studies, and involves setting up conditions by which the bed is only used for sleep and, if rapid sleep onset is not achieved, the child is instructed to get out of bed and only return to bed when feeling sleepy, repeating this process until rapid sleep onset is achieved (Bootzin, 1972). Stimulus control therapy is based on the idea that insomnia is caused by conditioned wakefulness in bed, and that by removing the individual from the bed until they are excessively sleepy (due to increasing homeostatic sleep pressure), this conditioned response will be extinguished (Bootzin, 1972). This is most commonly applied with adults (Chesson et al., 1999; Morgenthaler, Kramer, et al., 2006), although a similar intervention known as "response cost" is used with preschool children (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006). Response cost involves removing the child from their bed if they do not fall asleep

within a prescribed time, and then re-attempting sleep after a set period (e.g., 15-30min; Mindell et al., 2006). These interventions have been used infrequently in typically-developing children during the middle childhood years because they may reinforce the child's attempts to leave the bedroom and thus reward the child for not sleeping (Meltzer, 2010; Ortiz & McCormick, 2007).

Finally, sleep extension was only included in one study (7%). Sleep extension involves temporarily extending the child's time in bed, under the assumption that daytime tiredness and other associated daytime consequences have arisen as a result of insufficient sleep opportunity. This is not usually relevant for children with a diagnosed insomnia disorder, because the diagnosis of insomnia can only be applied if the individual experiences sleep difficulties despite adequate opportunity for sleep (AASM, 2014). However, it was included in this review as it may be an intervention worth considering for children with general sleep difficulties who have short sleep duration.

It is evident in Table 2.2 that most studies have evaluated multi-component interventions, thereby making it impossible to determine the relative contribution of each individual therapeutic technique. This leaves us with the question: *How do we determine which sleep strategy is appropriate for a particular child or sleep problem?* Luckily, the theoretical background for each of these strategies does provide some clues. However, for adults and young children, treatment decisions are guided by AASM practice parameters (Morgenthaler, Kramer, et al., 2006; Morgenthaler, Owens, et al., 2006), which have been built on a strong base of research evidence. This is clearly an important direction for future research in middle childhood. Interestingly, though, some researchers have speculated about which components of treatment resulted in the greatest effect for their child participants. Paine and Gradisar (2011) suggested that bedtime fading may have been a key component resulting in improvements in reduced nocturnal wakefulness for their child participants. Tamura and Tanaka (2014) reported that 3 key sleep hygiene behaviours were responsible for the improvements in sleep for their participants: getting up at approximately the same time every day, avoiding after-school naps, and reducing pre-bedtime television use. While

not a specific CBT-i component, Tamura and Tanaka (2016) also speculated that allowing children to self-select the specific behavioural targets for treatment was critical in the effectiveness of their sleep hygiene intervention.

2.5 Conclusions and Directions for Future Research

Research evaluating treatments for sleep problems in middle childhood is a relatively new area of focus. A thorough review of the existing literature found only 14 relevant studies, of which 9 reported on clinical RCTs, 2 reported on non-randomised controlled trials, and 3 were open trials or case reports. All of these studies were published within the past 7 years.

There were no studies identified in the current review that compared the relative effectiveness of different components of intervention for sleep problems in middle childhood. Instead, most of the reviewed studies either used manualised multi-component treatment programs (e.g., Paine & Gradisar, 2011; Schlarb et al., 2011; Schlarb et al., 2018), or allowed parents, therapists, or children to self-select appropriate strategies from a list of possible options (e.g., Quach et al., 2011; Tamura & Tanaka, 2014; Tamura & Tanaka, 2016). This means that the field lacks information about the broad effectiveness of these individual components of treatment.

Only one intervention, covered in 2 published studies (Tamura & Tanaka, 2014; Tamura & Tanaka, 2016), reported on treatment outcomes for what might be considered a single-component intervention, which involved education about sleep hygiene. This is particularly interesting because these broad recommendations are often considered a "first-line treatment" for children with sleep problems, and may be disseminated by primary health care professionals (e.g., general practitioners [GPs]) with very little training in sleep. However, in routine practice, these health professionals are unlikely to have time to provide the same level of detail (i.e., 45-60min appointment) and thus it is unclear whether these results would be replicated in a GP clinic setting. Furthermore, Tamura and Tanaka (2014, 2016) implemented their sleep hygiene intervention in schools with a general population sample of children without sleep disorders. Thus, while their results show promise for

sleep hygiene education as a general sleep improvement technique for children without sleep disorders, this does not provide evidence for its use in the treatment of insomnia.

In broad terms, future research evaluating treatments for sleep problems in middle childhood should work towards developing precise guidelines for the implementation of specific sleep strategies, in line with those available for adults (Morgenthaler, Kramer, et al., 2006) and young children (Morgenthaler, Owens, et al., 2006). This will involve conducting clinical RCTs that evaluate the use of individual cognitive and/or behavioural strategies (for example, those outlined in Table 2.2) in this population. Further research exploring the mechanisms of action of each of these strategies in this population may also be beneficial, as this may provide additional guidance about selection of appropriate treatments for individual children with sleep problems.

Chapter 3: Can Brief Sleep Interventions "Dampen" Anxiety in Middle Childhood?

3.1 Introduction

3.1.1 Sleep and Anxiety in Middle Childhood

There is robust evidence for the high comorbidity between childhood sleep difficulties and anxiety (for reviews, see Gregory & Sadeh, 2012; Leahy & Gradisar, 2012a; McMakin & Alfano, 2015). As previously mentioned in Chapter 1, psychological factors play a key role in this association (Dahl, 1996; Dahl & Harvey, 2007). Pre-sleep arousal experienced by children with anxiety disorders includes cognitive arousal (e.g., worries) and somatic arousal (e.g., restlessness, physical symptoms; Parker, Van Lenten, & Pina, 2017). Children with anxiety are hypervigilant to potential threats in their environment, and hypervigilance during the pre-sleep period inhibits sleep, because sleep and arousal are opponent processes (Dahl, 1996; Dahl & Harvey, 2007). For adults with insomnia, sleep and its associated stimuli (e.g., bed, bedroom, clock) can be seen as "threats" in and of themselves. The individual attending to these stimuli then needs to exert effort to aid sleep onset, which interferes with the automaticity of sleep (Espie, Broomfield, MacMahon, Macphee, & Taylor, 2006). While a cognitive-behavioural model of insomnia is as yet untested in middle childhood (Byars & Simon, 2014), children as young as 8 years old have been reported to catastrophise about poor sleep and its consequences (Gregory et al., 2010) - a common feature of adult insomnia.

During middle childhood, children experience a peak in night-time fears and worries (Muris et al., 2001). In order to prevent associated increases in nocturnal arousal, children with insomnia often rely on safety behaviours (e.g., sleeping with a parent), which unfortunately only serve to maintain the child's night-time fears (Paine & Gradisar, 2011, Muris, et al., 2001; Peterman, Carper, & Kendall, 2014). In fact, persistent reluctance or refusal to go to sleep without parental presence at night is a symptom of Separation Anxiety Disorder (American Psychiatric Association [APA], 2013), which is the most common anxiety disorder for children under 12 years (Cartwright-Hatton, McNichol, & Doubleday, 2006). It is common for anxious children to have sleep schedules that are inappropriate (e.g., putting children to bed before they are sleepy) or inconsistent (e.g.,

varying bedtime each night; Hudson, Gradisar, et al., 2009). This may also contribute to insomnia symptoms, as children sent to bed too early are unlikely to experience enough homeostatic sleep pressure in order to initiate sleep quickly. Thus, via increased time spent awake in bed, these children are likely to associate bedtime with alertness (Borbély, 1982; Paine & Gradisar, 2011).

According to Dahl and Harvey (2007), if sleepiness is an opposing process to the alerting effects of anxiety, then increased sleepiness may act to "dampen" anxiety symptoms (Dahl & Harvey, 2007). Thus, the implementation of a consistent and later bedtime routine, which allows children to build up homeostatic sleep pressure and go to bed when sleepy, may act to "dampen" bedtime arousal and reduce the opportunity for maladaptive conditioning to occur (Paine & Gradisar, 2011). Consistent with this, Kyle, Morgan, Spiegelhalder, and Espie (2011) reported that adults described "dampened mentation", both at sleep onset and during night wakings, while undergoing sleep restriction therapy (which aims to treat insomnia by reducing time in bed [TIB]; Spielman et al., 1987; Miller et al., 2014). Oualitative data revealed that these adults began to "crave" sleep, which in turn decreased their anxiety and worry when initiating sleep, increased their confidence in their ability to sleep, reduced their application of "effort" when attempting to fall asleep, and allowed sleep to return to an automatic process (Kyle et al., 2011). Furthermore, Vallieres and colleagues (2013) found their adult participants had lower salivary cortisol levels during sleep restriction therapy (compared to baseline), suggesting a decrease in physiological arousal during treatment. In children, a similar technique that is congruent with overcoming these maladaptive processes is bedtime fading (Piazza & Fisher, 1991), or as described here, bedtime *restriction therapy.*

3.1.2 Previous Research

Previous studies linking restricted sleep duration with impairments in mood and emotion regulation (e.g., Gruber, Cassoff, Frenette, Wiebe, & Carrier, 2012; Vriend, et al., 2013; Vriend, Davidson, Rusak, & Corkum, 2015) suggest that sleep interventions which restrict sleep opportunity (e.g., bedtime restriction therapy) may be contraindicated for children with pre-existing

anxiety symptoms. However, a randomised controlled trial (RCT) evaluating a multi-component cognitive-behaviour therapy (CBT) program that incorporated elements of CBT for adult insomnia (e.g., bedtime restriction therapy²) and CBT for child anxiety (e.g., cognitive restructuring, graded exposure) found that school-aged children (7-13 years) showed significant improvements in sleep and anxiety symptoms, compared to a wait-list control group, and continued to show improvements in anxiety at the 1-month follow-up (Paine & Gradisar, 2011). These improvements in anxiety occurred with less sessions (i.e., six) compared to standard CBT for childhood anxiety (i.e., 8-17 sessions; Rapee et al., 2009). While this research provides preliminary support for restricting bedtimes and observing reductions in anxiety (Paine & Gradisar, 2011), given the multi-component treatment design, the unique contribution of restricting TIB on this dampening process cannot be adequately determined outside the potential influence of skill transference from the anxiety treatment components. Thus, Paine and Gradisar (2011) recommended future dismantling studies, and speculated that bedtime restriction therapy may be a key intensive treatment component.

3.2 Open Trial: Bedtime Restriction Therapy

Building upon the work of Paine and Gradisar (2011), Leahy and Gradisar (2012b) were interested in whether, in the absence of specific anxiety-focused treatment strategies (i.e., cognitive and exposure therapies), bedtime restriction therapy alone would result in improvements in anxiety. Specifically, their open trial examined the feasibility of bedtime restriction therapy for school-aged children with insomnia, and also provided insight into the relationship between sleep and anxiety in this population.

Participants were 12 children aged 5 to 13 years who met criteria for a diagnosis of Behavioural Insomnia of Childhood (American Association of Sleep Medicine [AASM], 2005), sampled from January to November 2011. Treatment consisted of 2 sessions of bedtime restriction

² Paine and Gradisar (2011) referred to this component of treatment as *bedtime fading*. However, for consistency across the remainder of this thesis, the term *bedtime restriction therapy* will be used.

therapy over a period of 2 weeks. During the first treatment session, families were advised to temporarily delay the child's 'lights out time' (i.e., the time at which they attempted sleep) to their average sleep onset time (i.e., the time they usually fell asleep), as indicated by their pre-treatment sleep diary. Children also adhered to a fixed scheduled wake time across the week (including weekends), and were advised to abstain from napping. Families were provided with relevant sleep education regarding individual differences in sleep need, sleep architecture (i.e., stages of sleep), and homeostatic sleep pressure (Borbély, 1982) as a treatment rationale for bedtime restriction therapy. No information about the possible dampening effects of increased sleep homeostatic pressure on anxiety was provided. After one week, prescribed TIB was increased by advancing lights-out time by 15 mins if the family reported significant daytime consequences (e.g., daytime sleepiness, attention or concentration problems, behavioural issues), or the child was falling asleep quickly with minimal wake after sleep onset (WASO). If there were no significant daytime consequences, prescribed TIB remained unchanged. Children completed a daily sleep diary (i.e., measuring sleep onset latency [SOL], WASO, sleep efficiency [SE], total sleep time [TST]) continuously during this period, and the Spence Children's Anxiety Scale - Self-Report (SCAS; Spence, 1998) and the Pediatric Daytime Sleepiness Scale (PDSS; Drake et al., 2003) at the end of each week.

Linear mixed models analyses revealed improvements in both sleep and anxiety from pre- to post-treatment (see Appendix B.1). Child participants complied with the protocol, as demonstrated by a significant decrease in TIB (reducing by an average of 37 mins during the first week of treatment). As predicted, children demonstrated significant improvements in wakefulness in bed (i.e., SOL, WASO, and a trend towards significance for SE) and a significant reduction in anxiety (i.e., SCAS total score, but not SCAS separation anxiety subscale or diary-reported 'fear of sleeping alone'). There was no significant change in TST or daytime sleepiness, which is not surprising as bedtime restriction therapy only aimed to reduce wakefulness in bed but not restrict actual sleep time.

The authors concluded that the reduction in wakefulness in bed experienced during bedtime restriction therapy may hold the key to understanding some of the observed improvements in anxiety (Leahy & Gradisar, 2012b). They postulated that, through the process of classical conditioning (Pavlov, 1906, 1927), children's time spent awake in bed prior to treatment may have been associated with a heightened state of anxiety (i.e., worry, vigilance, physiological arousal), which may have been further reinforced by avoidant coping mechanisms (e.g., bedtime resistance, parental presence). Thus, the reduction in wakefulness experienced in bed following treatment may have allowed extinction of this conditioned response (i.e., anxiety).

3.3 Open Trial: Sleep Restriction Therapy

Following the success of Leahy and Gradisar (2012b), a second open trial (Watherston & Gradisar, 2013) examined the feasibility of an intervention that used a stronger manipulation of sleepiness (i.e., via further restricting TIB) to more adequately assess whether sleep and arousal may be opponent processes. As this treatment aimed to not only restrict TIB but also TST, it is described as *sleep restriction therapy*.

Participants were 12 children aged 5 to 12 years with Behavioural Insomnia of Childhood (AASM, 2005), sampled from March 2012 to July 2013, and treatment consisted of 2 sessions of sleep restriction therapy over a period of 2 weeks. Watherston and Gradisar's (2013) treatment protocol for sleep restriction therapy was almost identical to Leahy and Gradisar's (2012b) bedtime restriction therapy, with the exception of how the child's bedtime schedule was calculated. During the first week of treatment, children's TIB was reduced to 30 min less than their pre-treatment TST. Following the first week of sleep restriction, prescribed TIB was increased by advancing lights-out time by 15 mins if the family reported significant daytime consequences (e.g., daytime sleepiness, attention or concentration problems, behavioural issues), or the child was falling asleep quickly with minimal WASO. If there were no significant daytime consequences, prescribed TIB remained unchanged for the second week of treatment. Children completed a daily sleep diary (i.e.,

measuring SOL, WASO, SE, TST) continuously during this period, and the SCAS – Self-Report (Spence, 1998). The inclusion of a measure of worry (i.e., the Worry Scale for Children [WSC; Muris et al., 1998]) and evening sleepiness (via daily ratings provided on the sleep diary) also allowed preliminary examination of these potential process variables. Specifically, significant change in children's worry during treatment might indicate that reduction in anxiety was related to reduced nocturnal wakefulness and thus reduced opportunity for worry while awake in bed (as proposed by Dahl & Harvey, 2007). Alternatively, significant change in evening sleepiness might indicate that reduction in anxiety was related to 'dampened' arousal at bedtime (also proposed by Dahl & Harvey, 2007).

Again, analysis of sleep diary data revealed that child participants complied with treatment, as evidenced by significant decreases in both TIB (reducing by an average of 79 mins during the first week of treatment) and TST (reducing by an average of 43 mins during the first week of treatment; see Appendix B.2). As predicted, children demonstrated significant improvements in wakefulness in bed (i.e., SOL, SE, but not WASO) and anxiety (i.e., SCAS total score, diary-reported 'fear of sleeping alone', and a trend towards significance for SCAS separation anxiety subscale). For process variables, significant improvements were observed for children's worry (i.e., WSC total score) and, in particular, worry about personal harm³. As predicted, evening sleepiness also significantly increased, suggesting that the intervention was effective at increasing homeostatic sleep pressure.

3.4 Discussion

These novel and brief open trials examined the feasibility of bedtime restriction therapy and sleep restriction therapy for the treatment of insomnia in middle childhood. Despite the counterintuitive nature of therapies that reduce time in bed, not only for adults (Kyle et al., 2011)

³ Paine and Gradisar (2011) noted that this was a specific worry that children volunteered during cognitive therapy, and excessive worry about personal harm is also a symptom of Separation Anxiety Disorder (APA, 2013).

but also children (Hill, 2011), participants in both studies complied with the protocol, and experienced improvements in both wakefulness in bed (e.g., SOL) and anxiety (SCAS total score) over the 2-week treatment period. The concordance of findings between these 2 studies is noteworthy.

Consistent with its aims, bedtime restriction therapy did not result in significant changes in total sleep time; rather, it served to decrease wakefulness in bed (i.e., SOL and WASO) without actually restricting sleep. In contrast, sleep restriction therapy resulted in a small but significant decrease in total sleep time (26min) and an increase in evening sleepiness, suggesting that the manipulation was successful in increasing homeostatic sleep pressure and subsequently reducing SOL. Together with the observed improvement in evening ratings of fear of sleeping alone, and reductions in worry and total anxiety, these results are consistent with the theory that an increase in evening sleepiness resulted in "dampened" anxiety for these children. Of course, the relationships between these process variables need further investigation in future studies, which is an aim of the current thesis.

In school-aged children, inconsistent sleep schedules have been associated with a variety of consequences such as behavioural difficulties and impaired cognitive functioning (Biggs, Lushington, van den Heuvel, Martin, & Kennedy, 2011; Kelly, Kelly, & Sacker, 2013a; Pesonen et al., 2009), and a longitudinal study found that changes from irregular to regular bedtimes resulted in improvements in behaviour (Kelly, Kelly, & Sacker, 2013b). It is possible that some of the effects of bedtime restriction and sleep restriction therapies observed in the present studies are due to a regular, albeit restricted, sleep schedule. Consistent with this, another study of children aged 10-18 years found that a sleep intervention combining regular sleep schedules and sleep hygiene education resulted in significant improvements in sleep and daytime sleepiness (Tan, Healey, Gray, & Galland, 2012). Therefore, future RCTs should compare these restriction therapies with a "control" condition of bedtime regularisation (without restricting TIB).

Adults undergoing sleep restriction therapy have been reported to experience a number of daytime "side effects" during treatment, including fatigue/exhaustion, extreme sleepiness, reduced motivation/energy, headache/migraine, changes in appetite, irritability, and low mood (Kyle et al., 2011). Furthermore, research examining the consequences of experimental sleep restriction in children has suggested that a 30 min reduction in sleep duration may be associated with impairments in cognitive performance and increased evening sleepiness, although interestingly no change in morning alertness (Sadeh et al., 2003). Future research evaluating sleep restriction therapies in middle childhood should also include measures of daytime consequences of shortened sleep duration. This will be another aim of the present thesis, which will be explored in Chapter 5.

3.4.1 Strengths

The two open trials reported here were conducted in a real-world clinical setting using an intervention that was delivered in only 2 treatment sessions. This not only suggests good external validity, but is also consistent with recent trends in the insomnia literature, which have favoured reducing treatment duration in order to improve dissemination in primary care settings (e.g., Buysse et al., 2011). For individuals with anxiety, who may be seeking treatment for both an anxiety disorder and sleep problem, this may provide an opportunity for one of these brief restriction therapies to be applied as an adjunct to traditional CBT for anxiety. Finally, the replication of effects between these two open trials is noteworthy.

3.4.2 Limitations

These preliminary results must be interpreted with caution due to several limitations. First, an obvious limitation of both of these studies is in the absence of a control group. A lack of control group can introduce confounding factors such as the effect of historical changes unrelated to treatment, the maturation of participants, or unspecified treatment effects (e.g., therapeutic alliance; Dimitrov & Rumrill, 2003; Shirk & Karver, 2003). In these open trials it is unlikely that 2 weeks was enough to observe maturation effects. This is particularly the case in a clinical setting, where

sleep problems are generally chronic before help is sought (Newton, 2017). Furthermore, while no random allocation to different treatments was possible (these interventions were evaluated in two separate open trials), the treatment was provided to all children who entered the clinic (i.e., they were not specifically selected for research participation) and baseline characteristics were comparable for both groups. It may be possible that improvements in sleep and anxiety did not occur because of the treatment, rather they occurred because children were seeking help from a health professional (as has been observed in previous studies of children with anxiety disorders; e.g., Hudson, Rapee, et al., 2009) and thus expected to experience improvements in one or both of these variables. In order to reduce demand effects on self-report measures there was no explicit mention of anxiety at all during either of the treatment programs; however, it is possible that children still expected that their anxiety *should* be decreasing and thus under-reported their symptoms after treatment.

Second, as these were pilot intervention studies, the sample sizes were small. This may limit the generalisability of results and increase the chance of a false-positive result or over-estimation of the size of an effect (Hackshaw, 2008). Preliminary studies of small sample sizes are advantageous because they allow a new research hypothesis to be tested without consuming too many resources when there is no real effect; however, any observed associations must be replicated in larger confirmatory studies (Hackshaw, 2008). As discussed earlier, larger RCTs are required to further examine the effects of bedtime restriction and sleep restriction therapies.

Finally, treatment compliance was not formally assessed or validated with objective measures (i.e., wrist actigraphy) and thus the possibility that families did not adequately follow the bedtime restriction or sleep restriction therapy protocol, as instructed during treatment, cannot be excluded. That said, based on parental feedback during treatment sessions, and significant decreases in TIB following treatment (as seen in the weekly sleep diaries), it appears that children and parents followed treatment recommendations in both studies. Nonetheless, it is advisable that future studies

include an objective measure of sleep (i.e., wrist actigraphy) to confirm compliance with the treatment protocol.

3.4.3 Conclusions

It is promising to note that a brief sleep intervention (i.e., bedtime restriction therapy or sleep restriction therapy) delivered in only 2 treatment sessions, has the potential to produce meaningful improvements in both sleep (e.g., SOL) and anxiety (e.g., SCAS total score), consistent with the improvements observed by Paine and Gradisar (2011) for their 6-session multi-component treatment program. Larger RCTs, which compare these brief interventions with a control group, will provide further insight into both the effectiveness of these interventions and the mechanisms linking sleep and anxiety in middle childhood. As well as being valuable stand-alone interventions, sleep restriction therapies may also be potentially useful adjuncts to traditional CBT for anxiety in school-aged children. However, before such implementation can occur, it is important to identify potential benefits alongside any contraindications of treatment. These issues are addressed in Chapter 4 and Chapter 5 of the current thesis.

Chapter 4: A Randomised Controlled Trial of Sleep Restriction Therapy as a Transdiagnostic Intervention for Insomnia and Anxiety in Middle Childhood

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NC contributed to study conceptualisation and design, and led recruitment, data collection, data analysis, results interpretation, and manuscript preparation. CR and KB assisted with recruitment and data collection. HW and JR assisted with data collection. MG contributed to study conceptualisation, design, recruitment, data collection, results interpretation, and manuscript preparation. All authors approved the final manuscript.

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4.1 Abstract

In recent years, there has been an increasing focus on transdiagnostic interventions, which aim to address common underlying processes across different forms of psychopathology. During the middle childhood years, there is considerable overlap between sleep problems and anxiety, and preliminary research suggests that multi-component cognitive-behavioural sleep interventions may also improve anxiety symptoms. The present study aimed to further investigate this effect using two brief interventions (sleep restriction therapy, bedtime restriction therapy), compared to an active control group (bedtime regularisation). 61 children (6-14yrs; 54% female) with Chronic Insomnia Disorder participated in a randomised controlled trial. Both restriction groups experienced reductions in total sleep time and associated increases in evening sleepiness during the 2-week treatment, as well as improvements in sleep parameters (i.e., sleep onset latency, sleep efficiency). However, contrary to expectations, all three groups reported similar improvements in anxiety and worry. Further research is needed to draw conclusions about the potential mechanisms underlying the relationship between sleep and anxiety in middle childhood.

4.2 Introduction

In recent years, there has been an increasing focus on transdiagnostic processes and interventions across clinical psychology, as a move away from the categorical approach that has been used for many years (Harvey, Watkins, Mansell, & Shafran, 2004). The categorical diagnostic system has been criticised due to high rates of comorbidity between psychiatric disorders, substantial overlap in symptoms between disorders, and evidence of shared risk factors and maintaining processes between disorders (Harvey, 2009; Harvey et al., 2004; Mansell, Harvey, Watkins, & Shafran, 2008; Meidlinger & Hope, 2017). This focus on distinct diagnostic categories has resulted in the independent development and evaluation of diagnosis-specific treatment manuals, many of which contain common elements, although the categorical focus means that research findings have not been generalised to other disorders (Harvey, 2009; Harvey et al., 2004; Mansell et al., 2008). It has been proposed that transdiagnostic treatments, which focus on addressing common underlying processes across different forms of psychopathology, may have greater clinical utility (Harvey, 2009; Harvey et al., 2004; Mansell et al., 2008; Meidlinger & Hope, 2017).

As disordered sleep has causal links with many forms of psychopathology, it has been suggested that sleep disturbance may itself be a transdiagnostic process (Harvey, Murray, Chandler, & Soehner, 2011). This is supported by evidence from research in genetics and neurobiology (Harvey et al., 2011) and is consistent with recent changes in both the Diagnostic and Statistical Manual for Mental Disorders – 5th Edition (DSM-5; American Psychiatric Association [APA], 2013) and the International Classification of Sleep Disorders – 3rd Edition (ICSD-3; American Academy of Sleep Medicine [AASM], 2014). These manuals have both seen a shift away from identifying distinct insomnia subtypes (e.g., psychophysiological insomnia, paradoxical insomnia, insufficient sleep hygiene) and a shift towards a single diagnostic category to encompass all clinically significant insomnia symptoms. Furthermore, these manuals now allow an individual to have a diagnosis of insomnia alongside other medical or psychological conditions, without needing

to specify one of these as the "primary" condition, which reflects the building evidence that cognitive behaviour therapy for insomnia (CBT-i) is effective in treating insomnia with a range of comorbid conditions (e.g., chronic pain: Jungquist et al., 2010; breast cancer: Savard, Simard, Ivers, & Morin, 2005; review including mixed diagnoses: Smith, Huang, & Manber, 2005).

As described in Chapter 2, there has been relatively little research evaluating interventions for sleep problems in middle childhood (6-12 yrs), compared to other populations (e.g., infants, adults). In middle childhood, the most common sleep difficulties are associated with anxiety or night-time fears, including fear of the dark, worries about personal security or environmental threats, or separation anxiety (Muris, Merckelbach, Gadet, & Moulaert, 2000, Gregory et al., 2010). To address this gap in the literature, Paine and Gradisar (2011) developed a 6-session treatment program that combined age-appropriate components of CBT-i (i.e., 2 sessions focused on sleep education and bedtime restriction therapy⁴) and cognitive behaviour therapy (CBT) for anxiety (i.e., 2 sessions of cognitive restructuring, followed by 2 sessions of graded exposure). Compared to a waitlist control group, their child participants (7-13 years) showed significant improvements in sleep (i.e., sleep onset latency [SOL], wake after sleep onset [WASO], sleep efficiency [SE]) and anxiety symptoms post-treatment, and continued to show improvements in anxiety one month after completion of the intervention (Paine & Gradisar, 2011).

Paine and Gradisar (2011) speculated that bedtime restriction therapy may have been the critical treatment component that resulted in the observed improvements for their participants, and they recommended future dismantling studies to understand specific mechanisms involved. Subsequently, as described in Chapter 3, a small open trial was conducted to examine whether bedtime restriction therapy alone would result in similar improvements in sleep and anxiety, in the absence of specific anxiety-focused treatment strategies (i.e., cognitive and exposure therapies; Leahy & Gradisar, 2012b). Twelve children (5-13 years) participated in a 2-week bedtime

⁴ Paine and Gradisar (2011) referred to this component of treatment as *bedtime fading*. However, for consistency across this thesis, the term *bedtime restriction therapy* will be used.

restriction therapy intervention, which involved temporarily delaying the child's 'lights out time' (i.e., the time at which they attempted sleep) to match their average sleep onset time (i.e., the time they usually fell asleep), as indicated by a pre-treatment sleep diary. Children also adhered to a fixed scheduled wake-up time across the week (including weekends) and families were provided with relevant education regarding sleep architecture, individual differences in sleep need, and homeostatic sleep pressure (Borbély, 1982) as a treatment rationale for bedtime restriction therapy. Consistent with the results of Paine and Gradisar (2011), these school-aged children experienced improvements in both sleep (SOL, WASO) and anxiety, which is impressive considering the brevity of this intervention and the fact that it did not contain any specific anxiety-focused treatment components.

So how does bedtime restriction therapy alone result in improvements in anxiety, without explicitly addressing anxiety during treatment? According to Dahl and Harvey (2007), sleep and arousal (i.e., anxiety) are opponent processes. Thus, sleep-onset difficulties among children with anxiety occur due to hyperarousal during the pre-sleep period, which directly inhibits sleep onset. Conversely, it is plausible that increased sleepiness during the pre-sleep period would result in reductions in arousal (i.e., anxiety). Thus, the implementation of a consistent and later bedtime routine during the bedtime restriction therapy intervention, which allowed children to go to bed when sleepy, may have resulted in "dampened" anxiety (see Figure 4.1). Furthermore, it is also common for children with insomnia to worry while awake in bed (McMakin & Alfano, 2015). Thus, the reduced wakefulness in bed (i.e., SOL, WASO) experienced during bedtime restriction therapy may have also resulted in less opportunity for worry, consequently reducing overall anxiety. However, the lack of measurement of evening sleepiness and worry, as well as the limitations of the study design (e.g., small sample size, lack of control group) mean that these potential mechanisms could not be confirmed.

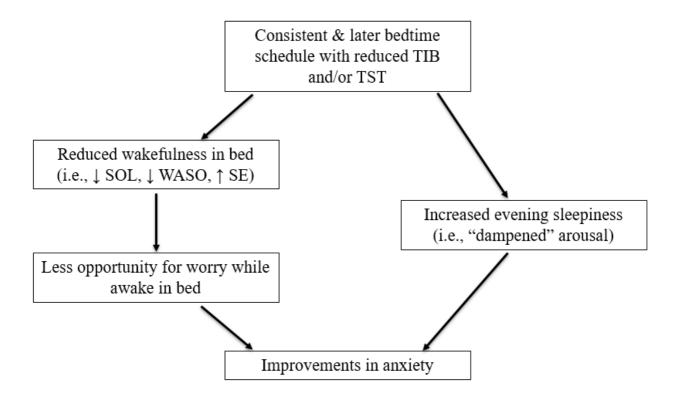


Figure 4.1. Potential mechanisms linking the sleep intervention to improvements in anxiety. Note: TIB = time in bed; TST = total sleep time; SOL = sleep onset latency; WASO = wake after sleep onset; SE = sleep efficiency. Image developed by author.

A second open trial was designed to further investigate these potential mechanisms (Watherston & Gradisar, 2013). Twelve children (5-12 years) participated in a 2-week intervention designed to induce a greater manipulation of evening sleepiness. Sleep restriction therapy involved temporarily delaying the child's 'lights out time' so that their scheduled time in bed was reduced to 30 min less than their pre-treatment total sleep time, while they adhered to a fixed scheduled wakeup time across the week. Families also received relevant sleep education as described previously. Again, children experienced improvements in both sleep (SOL, SE) and anxiety. As expected, evening sleepiness also significantly increased during treatment, although the precise relationship between sleep, evening sleepiness, and anxiety could still not be confirmed due to limitations of the study design.

The first limitation of these two open trials is the absence of a control group, which introduces confounding factors such as unspecified treatment effects (e.g., therapeutic alliance;

Dimitrov & Rumrill, 2003; Shirk & Karver, 2003). Thus it is impossible to conclude that the observed improvements occurred as a direct result of the interventions. Secondly, small sample sizes may also limit the generalisability of results and increase the chance of a false-positive result or over-estimation of effect size (Hackshaw, 2008). Finally, treatment compliance was not formally assessed using an objective measure of sleep (i.e., wrist actigraphy) and thus the possibility that families did not adequately follow the bedtime restriction or sleep restriction therapy protocol, as instructed during treatment, cannot be excluded.

In an attempt to overcome these limitations, the current chapter describes a randomised controlled trial (RCT) that was designed to directly compare and evaluate sleep restriction therapy and bedtime restriction therapy, and compare them to an active control group, using both subjective and objective measures of sleep. This design also allows for further exploration of the mechanisms linking improvements in sleep and anxiety in this population. There were 3 main research questions:

- 1. Do sleep restriction and bedtime restriction therapies improve sleep?
- 2. Do sleep restriction and bedtime restriction therapies 'dampen' anxiety? Are there potential mechanisms explaining the relationship between sleep and anxiety?
- 3. Do these brief (2 week) interventions provide *sustained* benefits to sleep and anxiety?

4.3 Method

4.3.1 Participants

Participants were 61 children aged 6-14 years⁵ (M = 9.1yrs \pm 2.1, 54% female, SES SEIFA score⁶=7.2 \pm 2.5) who were recruited from April 2014 to April 2017. At pre-treatment, 91% of parents reported that their child experienced difficulty falling asleep, 47% reported difficulty

⁵ Only 1 participant was 14 years old and 4 participants were 13 years old. They were included in this study as they met all other inclusion criteria, their sleep difficulties were identical to those experienced by the younger participants in this study, and they showed no signs of a pubertal delay in circadian rhythm timing.

⁶ The SEIFA index of socioeconomic advantage and disadvantage ranges from low (1) to high (10) socioeconomic status (Australian Bureau of Statistics, 2011).

staying asleep, and 72% reported that their child required parental presence to fall asleep or return to sleep after waking. All participants met criteria for Chronic Insomnia Disorder (AASM, 2014) and reported symptoms of anxiety (Spence Children's Anxiety Scale total score: $M = 31.8 \pm 16.5$). Inclusion criteria were (1) presence of clinically significant symptoms of insomnia, consistent with a diagnosis of Chronic Insomnia Disorder, and (2) presence of anxiety symptoms. Children were excluded if their sleep difficulties were better explained by another sleep disorder (e.g., a Circadian Rhythm Sleep Disorder), if they had a previously diagnosed behaviour disorder (e.g., Attention Deficit Hyperactivity Disorder) or a developmental delay (e.g., intellectual disability, Autism Spectrum Disorder), which may have resulted in difficulties complying with study requirements (i.e., questionnaires, sleep diaries). Other psychiatric diagnoses (e.g., depression) were not specific exclusion criteria; however, no participants reported any current or previous psychiatric diagnoses (except anxiety, as described earlier). This study was approved by the Southern Adelaide Clinical Human Research Ethics Committee and registered with the Australian New Zealand Clinical Trials Registry (ACTRN12614000109606). Parents provided written informed consent for their child's participation. A participant flow diagram is presented in Figure 4.2.

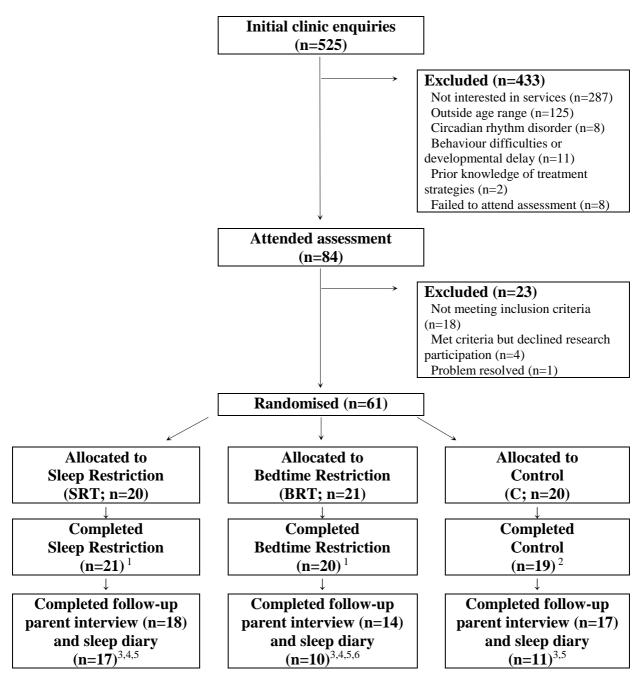


Figure 4.2. Participant flow diagram.

Note: ¹Allocated to bedtime restriction but received sleep restriction due to therapist error (n=1). ²Did not complete treatment due to parental health issues (n=1). ³Lost contact with family (SRT: n=2, BRT n=4, C: n=2). ⁴Withdrew from follow-up for further treatment (SRT: n=1, BRT: n=1). ⁵Failed to complete sleep diary (SRT: n=1, BRT: n=2, C: n=6). ⁶Withdrew from follow-up for overseas travel (n=1).

4.3.2 Design

The study employed a 3 (treatment: sleep restriction, bedtime restriction, control) x 4 (time: pre-treatment, mid-treatment, post-treatment, 4-week follow-up) mixed-model design.

4.3.3 Materials & Measures

7-day sleep diary (see Appendix C.1). In collaboration with their parents, children completed a 7-day sleep diary to record their daily sleep patterns (e.g., bed time, lights-out time, wake-up time, SOL, WASO). Sleep diaries are commonly used in insomnia treatment studies as a reliable and valid measure of an individual's subjective experience of insomnia (Buysse et al., 2006; Morin, 2003) and have been successfully used in previous studies with children diagnosed with insomnia (Paine & Gradisar, 2011). Based on the information entered, additional variables were calculated for total sleep time (TST), time in bed (TIB; i.e., the total duration from lights-out time until out-of-bed time), and SE (calculated as TST/TIB x 100). Children also provided daily ratings of their sleepiness just prior to bedtime using a 4-point scale (1 = not at all; 4 = extremely).

Self-report questionnaires (see Appendix C.2). At all time points, children completed the Spence Children's Anxiety Scale (SCAS; Spence, 1998), and the Worry Scale for Children (WSC; Muris et al., 1998). For younger children, parents were instructed that they could help their child to read and understand the items, but that the response recorded must be the child's own response.

The SCAS is a 44-item self-report scale containing 38 anxiety symptom items (e.g., *I feel afraid*) and 6 positive filler items (to reduce negative response bias; e.g., *I am good at sports*). Ratings are provided on a 4-point scale (0 = never; 3 = always) to indicate the frequency of anxiety symptoms experienced in the past week, and anxiety items are summed to produce a total score from 0 to 114, with higher scores indicating greater anxiety. The SCAS also yields 6 domain scores according to anxiety subtypes. For the present study, only the separation anxiety subscale was calculated, as Separation Anxiety Disorder (APA, 2013) is typically associated with bedtime sleep difficulties (i.e., separating from parents; Alfano et al., 2007) and was the subscale showing the greatest decrease in response to treatment in previous studies with this population (Paine &

Gradisar, 2011). In the current sample, internal validity for the SCAS was excellent (Chronbach's alpha = 0.91).

The WSC is a 15-item self-report questionnaire assessing the intensity of specific worries common in children (e.g., *How much do you worry about school? How much do you worry about future events?*) and contains items that align with common worries experienced by insomniacs when attempting sleep. Each item is rated on a 5-point scale (0 = none; 4 = very, *very much*) and items are summed to produce a total score from 0 to 60, with higher scores indicating more intense worry symptoms. The WSC possesses excellent reliability and validity (Muris et al., 1998) and had good internal consistency in the current sample (Chronbach's alpha = 0.86).

Parent-report questionnaire (see Appendix C.3). At baseline only, parents completed the 7item anxiety subscale of the Depression Anxiety Stress Scales (DASS-21; Lovibond & Lovibond, 1995) as an indication of their own anxiety symptoms (e.g., *I felt scared without any good reason*). Responses were given using a 4-point scale (0 = *never*; 3 = *almost always*) and were summed to produce a total score from 0 to 21. This was included in the present study because parental anxiety is a key risk factor for child anxiety (Cobham, 1997), and research suggests that treatment outcomes following CBT for anxiety may differ for children whose parents also report high anxiety (Cobham, Dadds, & Spence, 1998). Thus, it was important to identify any group differences in parental anxiety in the present study.

Clinical sleep history interview (see Appendix C.4). This semi-structured interview was developed to diagnose paediatric sleep disorders as per the International Classification of Sleep Disorders – 2nd Edition (ICSD-2; AASM, 2005), and updated consistent with changes to relevant diagnostic categories in the ICSD-3 (AASM, 2014). Relevant to Chronic Insomnia Disorder, this interview elicited information relating to Criterion A (specific sleep difficulties), Criterion B (daytime consequences of poor sleep), Criterion C (adequate opportunity and circumstances for sleep), Criterion D (symptom frequency), Criterion E (symptom duration), and Criterion F (differential diagnosis).

Wrist actigraphy. Wrist actigraphy monitors are worn like a small wristwatch and provide an objective estimate of sleep-wake patterns based on gross motor movement (Meltzer et al., 2012). Movement data are collected continuously and, when downloaded, provide automatic scoring of wake vs sleep using a validated algorithm, which is reliable when used over at least 5 nights (Acebo et al., 1999). For the present study, children wore actigraphy (MicroMini Motionlogger; Ambulatory Monitoring, Inc., Ardsley, NY) on their non-dominant wrist for 3 continuous weeks (i.e., pre-treatment, mid-treatment, post-treatment) and these data were used to confirm that children complied with treatment instructions. Data were collected using zero-crossing mode, in 1-min epochs, and scored using a validated algorithm (AMI-Sadeh, Action 4, Ambulatory Monitoring Inc, Ardsley, NY). Sleep diary records were used to identify lights-out time and out-of-bed time, sleep onset and offset were defined according to predetermined criteria (i.e., the "3/5 minute rule", described in Meltzer & Westin, 2011). Data files were de-identified and scored by 2 research assistants who were blind to the participant's treatment condition, and a random sample of files were double-scored by the first author (NC) to ensure accuracy.

Treatment. All families attended 2 treatment sessions with their psychologist, each held 1 week apart and lasting approximately 50 mins. All treatment sessions were conducted on an individual basis and were attended by the child and at least one parent (for full treatment manuals see Appendix C.5).

Sleep restriction therapy involved educating children and parents about children's sleep needs and the function of sleep homeostatic pressure. Specifically, they were informed about how temporarily restricting sleep would build sleep pressure and consolidate their child's sleep. Families were instructed to implement a sleep schedule in which the child would reduce their TIB to 30 mins less than their pre-treatment average TST, using consistent bedtimes and rise times as specified by the psychologist, tailored to the child/family (see Figure 4.3).

Participants in the bedtime restriction therapy group were educated that restricting time in bed awake would result in a more consolidated sleep for their child. Families were instructed to

implement a sleep schedule in which the child would reduce their TIB to match their pre-treatment average TST (Figure 4.3).

Participants allocated to the control condition were educated about the importance of a regular sleep schedule for children, to allow the circadian rhythm to regulate sleep. These families were advised to implement a sleep schedule which involved maintaining a consistent bedtime and rise time across the week, but which matched the child's pre-treatment average TIB (i.e., no change to the amount of TST or time awake in bed; Figure 4.3). This was chosen for the "control" condition as it was not intended to manipulate wakefulness in bed (i.e., SOL, WASO) nor TST, and may be considered the usual care provided by primary health care professionals (e.g., general practitioners [GPs]) and is often recommended as the first point of intervention by sleep specialists (e.g., Durand, 2008; Meltzer & Crabtree, 2015).

Participants in all three groups also received information about appropriate pre-bedtime activities for children, with specific advice to avoid stimulating activities before bed.

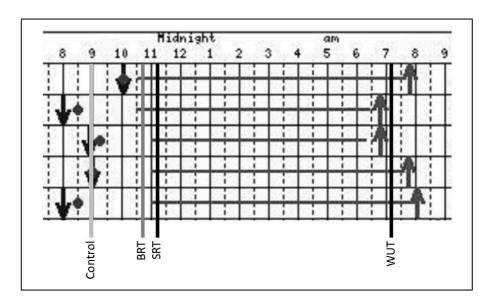


Figure 4.3. Example sleep diary showing proposed scheduled bedtimes for Sleep Restriction Therapy (SRT), Bedtime Restriction Therapy (BRT), and Bedtime Regularisation (Control) assuming a consistent wake-up time of 7.15am (WUT).

Note: Image developed by author.

In the sleep restriction and bedtime restriction conditions, prescribed TIB was increased by 15 mins for the second week of treatment if the family reported significant daytime consequences (e.g., daytime sleepiness, attention or concentration problems, behavioural issues), or the child was falling asleep quickly with minimal WASO. If there were no significant daytime consequences, prescribed TIB remained unchanged. Participants in the control condition were advised to continue with the same regular sleep schedule during the second week of treatment.

At the end of the second week of treatment, parents of all participants were given written instructions about how to manage their child's sleep schedule during the 4-week follow-up period (see Appendix C.6). Parents of children in the sleep restriction and bedtime restriction conditions were advised to continue to increase their scheduled TIB in 15-min increments each week if the child experienced significant daytime consequences (e.g., daytime sleepiness, attention or concentration problems, behavioural issues), or was falling asleep quickly with minimal WASO. For children in the control condition, parents were advised to continue with the same regular sleep schedule during the follow-up period.

Brief Follow-up Interview. At the completion of the 4-week follow-up period, parents were contacted by telephone and asked to provide information about the child's current sleep pattern (i.e., *Does your child currently have difficulty falling asleep? Does your child currently have difficulty staying asleep? Does your child require special conditions in order to fall asleep or return to sleep?*), whether the child still had a "sleep problem", and whether the child required any further treatment for their sleep difficulties. Yes/No responses were recorded for all items, and parents were given the opportunity to provide further details about any special conditions required for sleep onset, in particular to identify whether the child required parental presence. A copy of the interview can be found in Appendix C.7.

4.3.4 Procedure

Participants were recruited via usual intake processes at the Child & Adolescent Sleep Clinic at Flinders University, including referrals from GPs and advertisements in school newsletters and Facebook. An initial screening call or email confirmed the child was aged 6-14 years and obtained brief information about their primary sleep problem. Parents obtained a referral from a GP for their child, and paid a once-off fee of AUD\$200. Families completed a 7-day sleep diary and web-based questionnaires prior to attending a 1-hr assessment interview with either a psychologist, clinical psychologist, or provisional psychologist with specialist training in assessment and treatment of sleep problems (mean 4.7yrs experience in behavioural sleep medicine). After the assessment, research eligibility was confirmed in a group consensus meeting. Participants were then randomised to 1 of 3 treatment groups using random permuted blocks with random block size. The randomisation sequence was generated by the first author (NC) and was concealed from other members of the research team until after a decision about research eligibility was reached at the consensus meeting.

Families completed 7-day sleep diaries and children completed self-report questionnaires weekly during treatment, and children wore a wrist actigraphy monitor (MicroMini Motionlogger, Ambulatory Monitoring Inc., Ardsley, NY) for 3 continuous weeks. Manualised treatment was provided by the psychologist in 2 weekly 50-min sessions, and all sessions were attended by the child and at least one parent. At post-treatment, families were provided with brief written information about how to continue to manage their child's sleep. Four weeks later, families returned a final 7-day sleep diary, children completed self-report questionnaires, parents completed a brief follow-up interview, and families were offered further treatment for their child's sleep problem if necessary (e.g., cognitive restructuring, exposure therapy). The timing of all measures is presented in Table 4.1.

Table 4.1

Timing of measures

	Pre	Mid	Post	4wk FU
Clinical sleep history interview	•			
Sleep diary	•	•	•	•
Actigraphy	•	•	•	
DASS-21 (Anxiety scale)	•			
SCAS	•	•	•	•
WSC	•	•	•	•
Brief follow-up interview				•

Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), 4wk FU = 4-week follow-up, DASS-21 = Depression Anxiety Stress Scales, SCAS = Spence Children's Anxiety Scale, WSC = Worry Scale for Children.

4.4 Results

4.4.1 Statistical Analyses

All quantitative data analyses were conducted using the Statistical Package for the Social Sciences (SPSS v.22, SPSS Inc., Chicago, USA) and an alpha level set to 0.05. Sleep variables from actigraphy and sleep diaries (i.e., TST, TIB, SOL, WASO, SE) were averaged across the week because previous studies of children with insomnia (Paine and Gradisar, 2011) and anxiety (Hudson, Gradisar, et al., 2009) have found no difference between school-day and weekend sleep patterns in middle childhood. An average was calculated if data was available for >3 nights (out of a possible 7). One-way ANOVAs were used to investigate baseline differences between groups. Linear mixed model (LMM) analyses were used to analyse the 3 (group) x 4 (time) mixed model design for sleep diary, anxiety, worry, and sleepiness variables, and the 3 (group) x 3 (time) mixed model design for actigraphy sleep variables, and post-hoc analyses were conducted to address each of the research questions. LMM analyses reduce the potential impact of missing data (~37% from

pre-treatment to follow-up) by imputing missing values using expectation maximisation, based on a regression line for each individual, which is preferable to other statistical techniques for repeated measures (e.g., ANOVA; Landau & Everitt, 2004). For sleep variables expressed as units of time, effect sizes are presented as the magnitude of change in minutes. For other variables, Cohen's *d* was calculated as $d=M_1-M_2/(SD_{pooled})$ to establish the magnitude of within- and between-subjects differences. Non-parametric McNemar's Tests were used to examine changes in parental perception of child sleep difficulties from pre-treatment to 4-week follow-up, and Spearman's Correlations were used to examine relationships between changes in key variables (i.e., sleep, anxiety, worry, and sleepiness). Conservative *a priori* calculations predicting medium effects against a control group, using a power of 0.90 and inter-correlations between measures of r=0.51, and factoring a low attrition rate of 8%, it was determined that a target sample size of 60 (i.e., 20 per group) would be sufficient to detect significance (Faul, Erdfelder, Lang, & Buchner, 2007).

4.4.2 Baseline Sample Characteristics

There was no significant difference between groups in age, F(2,58)=.97, p=.39, gender distribution, $\chi^2(2, N=61)=3.24$, p=.20, socioeconomic status, F(2,54)=.89, p=.42, child anxiety, F(2,57)=1.80, p=.17, or parent anxiety, F(2,56)=1.17, p=.32. There was a significant group difference in sleep diary SOL, F(2,58)=3.34, p=.04; however, controlling for this in subsequent analyses did not change the overall pattern of results. There was no significant difference between groups for any of the other sleep diary or actigraphy sleep variables (all p>.05). Across the whole sample, 90% of children lived in a dual-parent household and 87% had one or more siblings. Descriptive statistics for demographic and sleep variables are presented in Table 4.2.

		Treatment Group			
		Sleep Restriction (N=21)	Bedtime Restriction (N=20)	Control (N=20)	Whole Sample (N=61)
Age (M \pm SD, in yrs)		9.5 ± 2.4	9.1 ± 2.0	8.6 ± 2.0	9.2 ± 2.1
Gender (% Female)		43%	50%	70%	54%
Socioeconomic Status ¹ (M \pm SD)		6.9 ± 2.3	7.8 ± 2.0	6.8 ± 3.0	7.2 ± 2.5
Parent Anxiety (M \pm SD)		2.2 ± 2.6	2.9 ± 3.5	3.8 ± 3.9	2.9 ± 3.4
Family Structure	Dual-parent household (%)	86%	100%	85%	90%
	\geq 1 sibling (%)	76%	94%	90%	87%
	Oldest child (%)	80%	53%	70%	68%
Sleep Diary	SOL ($M \pm SD$, mins)	37.4 ± 20.4	57.2 ± 38.6	37.0 ± 22.9	43.8 ± 29.5
	WASO (M ± SD, mins)	12.1 ± 20.0	11.5 ± 23.6	10.7 ± 14.7	11.4 ± 19.4
	SE (M \pm SD, %)	89.7 ± 6.8	86.6 ± 8.0	90.4 ± 5.8	88.9 ± 7.0
	TST (M \pm SD, hrs)	9.4 ± 0.8	9.2 ± 1.0	9.5 ± 0.8	9.4 ± 0.9
	TIB $(M \pm SD, hrs)$	10.5 ± 0.5	10.6 ± 0.9	10.6 ± 0.8	10.5 ± 0.7
Actigraphy	SOL (M \pm SD, mins)	36.0 ± 22.0	43.6 ± 37.3	33.5 ± 26.2	37.6 ± 28.7
	WASO (M \pm SD, mins)	49.9 ± 26.9	51.6 ± 22.3	50.1 ± 14.2	50.5 ± 21.6
	SE (M \pm SD, %)	85.6 ± 7.3	85.4 ± 6.6	87.1 ± 4.3	86.1 ± 6.2
	TST ($M \pm SD$, hrs)	8.9 ± 0.8	9.0 ± 0.6	9.2 ± 0.5	9.1 ± 0.6

Table 4.2

Pre-treatment descriptive statistics for demographic and sleep variables

Note: ¹ The SEIFA index of socioeconomic advantage and disadvantage ranges from low (1) to high (10) socioeconomic status (Australian Bureau of Statistics, 2013).

4.4.3 Treatment Compliance and Manipulation Check

Significant interactions were observed for both TIB, F(6,152)=6.98, $p<.001^7$ (Figure 4.4A), and sleep diary TST, F(6,149)=6.00, p<.001 (Figure 4.4B). Post-hoc testing confirmed that, consistent with treatment instructions, the sleep restriction and bedtime restriction groups both experienced reduced TIB (sleep restriction: F(3,150)=30.4, p<.001, 83mins; bedtime restriction: F(3,153)=19.8, p<.001, 71mins) and TST (sleep restriction: F(3,148)=16.3, p<.001, 52mins; bedtime restriction: F(3,150)=4.41, p=.005, 25mins), while the control group experienced no significant changes (TIB: F(3,153)=1.72, p=.17, 14mins; TST: F(3,150)=0.93, p=.43, 8mins). Actigraphy data similarly showed reduced TST during the first treatment week for both sleep restriction, F(2,79)=13.6, p<.001, 49mins, and bedtime restriction, F(2,76)=6.4, p=.003, 27mins, with no significant change for the control group, F(2,79)=1.7, p=.20, 19mins, but this interaction failed to reach statistical significance, F(4,78)=1.84, p=.13 (Figure 4.4C).

⁷ Based upon statistical advice, LMM analyses were conducted using data from all 4 time points. However, analyses relating to treatment compliance and the first 2 research questions (i.e., Do sleep restriction and bedtime restriction therapies improve sleep?; Do sleep restriction and bedtime restriction therapies 'dampen' anxiety?) will focus on post-hoc comparisons for the first 3 time points only.

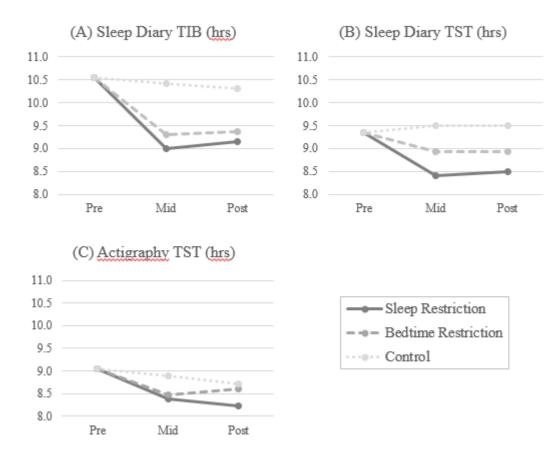


Figure 4.4. Estimated marginal means for sleep diary time in bed (hrs), sleep diary total sleep time (hrs), and actigraphy total sleep time (hrs) for each of the three treatment groups.

Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), TIB = time in bed; TST = total sleep time. Values have been equated at pre-treatment to assist in interpretation of change.

4.4.4 Do Sleep Restriction and Bedtime Restriction Therapies Improve Sleep?

Significant interactions occurred for SOL for both sleep diary, F(6,150)=5.51, p<.001(Figure 4.5A), and actigraphy, F(4,82)=3.72, p=.008 (Figure 4.5B). Post-hoc testing confirmed that the sleep restriction and bedtime restriction groups both experienced reduced SOL during the 2 treatment weeks (sleep restriction: F(3,149)=13.5, p<.001, 21mins; bedtime restriction: F(3,151)=27.2, p<.001, 35mins), with no significant change for the control group, F(3,151)=0.80, p=.50, 5mins. There was also a significant interaction for SE (diary: F(6,151)=2.22, p=.045 [Figure 4.5E]; but not actigraphy: F(4,80)=1.84, p=.13 [Figure 4.5F]), and post-hoc testing confirmed that significant increases in diary SE were observed for sleep restriction, F(3,149)=8.38, p<.001, d=0.84, and bedtime restriction, F(3,151)=16.0, p<.001, d=1.21, but not the control group, F(3,151)=1.74, p=.16, d=0.58. There were no significant interactions for WASO (diary: F(6,146)=0.38, p=.89 [Figure 4.5C]; actigraphy: F(4,81)=0.69, p=.60 [Figure 4.5D]).

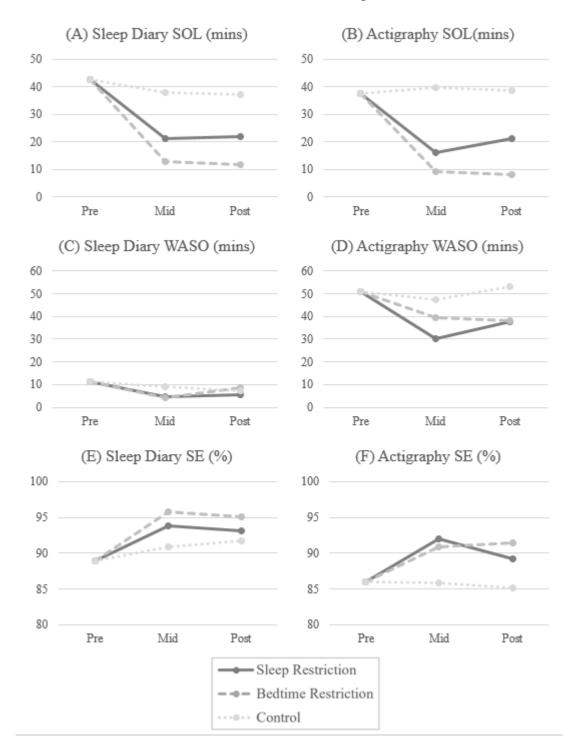


Figure 4.5. Estimated marginal means for sleep diary and actigraphy measures of sleep for each of the three treatment groups.

Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), SOL = sleep onset latency, WASO = wake after sleep onset, SE = sleep efficiency. Values have been equated at pre-treatment to assist in interpretation of change.

4.4.5 Do Sleep Restriction and Bedtime Restriction Therapies 'Dampen' Anxiety?

There was no significant interaction for SCAS total score, F(6,135)=0.64, p=.70 (Figure 4.6A), although an interaction for the separation anxiety subscale approached statistical significance, F(6,135)=2.08, p=.059 (Figure 4.6B). Contrary to expectations, anxiety improved similarly for participants in all 3 groups from pre- to post-treatment, as evidenced by significant main effects for time for both SCAS total score, F(3,135)=16.5, p<.001, d=0.31, and separation anxiety subscale scores, F(3,135)=8.35, p=.001, d=0.30.

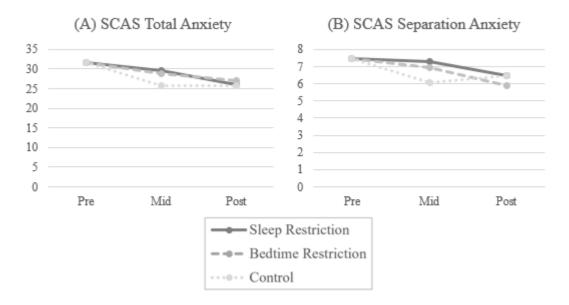
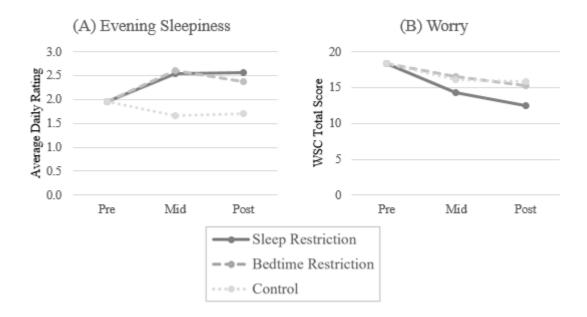


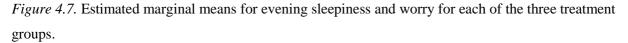
Figure 4.6. Estimated marginal means for anxiety variables for each of the three treatment groups. Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), SCAS = Spence Children's Anxiety Scale. Values have been equated at pre-treatment to assist in interpretation of change.

4.4.6 Exploratory Testing of Potential Mechanisms Explaining the Relationship between Sleep and Anxiety

Although there were no interactions for anxiety outcome variables, it was nevertheless important to explore changes – or lack of changes – in the process variables expected to explain any links between sleep and anxiety. First, it was proposed that sleep restriction and bedtime restriction therapies could reduce anxiety due to an increase in evening sleepiness. As expected, there was a significant interaction effect for evening sleepiness, F(6,146)=5.15, p<.001 (Figure 4.7A). Post-hoc testing confirmed that there was a significant increase in evening sleepiness for sleep restriction, F(3,145)=6.44, p<.001, d=0.80, and bedtime restriction, F(3,147)=6.39, p<.001, d=0.55, with no significant change for the control group, F(3,147)=1.94, p=.13, d=0.34.

Second, it was proposed that a reduction in the time spent awake in bed during sleep restriction and bedtime restriction therapies would in turn reduce time spent worrying. However, there was no significant interaction for worry, F(6,133)=0.57, p=.78 (Figure 4.7B), but there was unexpected improvement in worry for all three treatment groups, F(3,133)=6.83, p<.001, d=0.33. Post-hoc testing also revealed greater reduction in worry for the sleep restriction group, F(3,133)=5.42, p=.002, d=0.51, compared to the other two groups (bedtime restriction: F(3,134)=1.12, p=.35, d=0.27; control: F(3,133)=1.61, p=.19, d=0.22.





Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), WSC = Worry Scale for Children. Values have been equated at pre-treatment to assist in interpretation of change.

As per the model proposed earlier (Figure 4.1), change scores were calculated by subtracting the pre-treatment value from the post-treatment value for key variables, and relationships between these change scores were examined with Spearman's correlations. As there were no group differences in the reductions observed in anxiety (SCAS total score), the correlations were calculated for the whole sample (Figure 4.8). Using all 3 groups also reduces the chance of not detecting significant correlations due to a restricted range of scores for the variables of interest.

Moderate significant correlations were found between a change in TIB and both a change in SE and a change in evening sleepiness. There was also a significant moderate relationship between a change in worry and a change in anxiety (SCAS total score). However, there were no significant correlations between a change in SE and changes in worry or anxiety (SCAS total score). There was also no relationship between a change in evening sleepiness and a change in anxiety (SCAS total score).

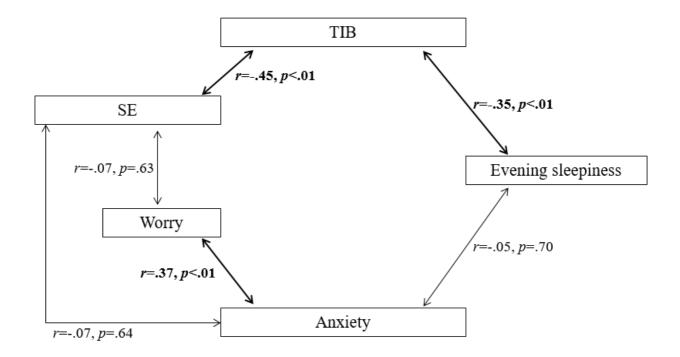


Figure 4.8. Spearman's correlations between changes in key variables from pre- to post-treatment. Note: TIB = time in bed; SE = sleep efficiency; Worry = Worry Scale for Children total score; Anxiety = Spence Children's Anxiety Scale total score. Image developed by author.

4.4.7 Do Sleep Restriction and Bedtime Restriction Therapies Provide Sustained Benefits to Sleep and Anxiety?

Post-hoc testing revealed that, consistent with treatment instructions, participants in the sleep restriction group experienced significant increases in both TIB (32mins, p=.006) and sleep diary TST (38mins, p<.001) from post-treatment to 4-week follow-up, and the bedtime restriction group experienced a significant increase in sleep diary TST (33mins, p=.01) while the control group experienced no significant changes in TIB or TST from post-treatment to 4-week follow-up (p>.05; see Figure 4.9). There were no significant changes in SOL, WASO, or SE for any group from post-treatment to 4-week follow-up (all p>.05; Figure 4.9). Post-hoc testing for anxiety variables also revealed no significant changes in SCAS total score and SCAS separation anxiety for any group from post-treatment to 4-week follow-up (all p>.05; Figure 4.10).

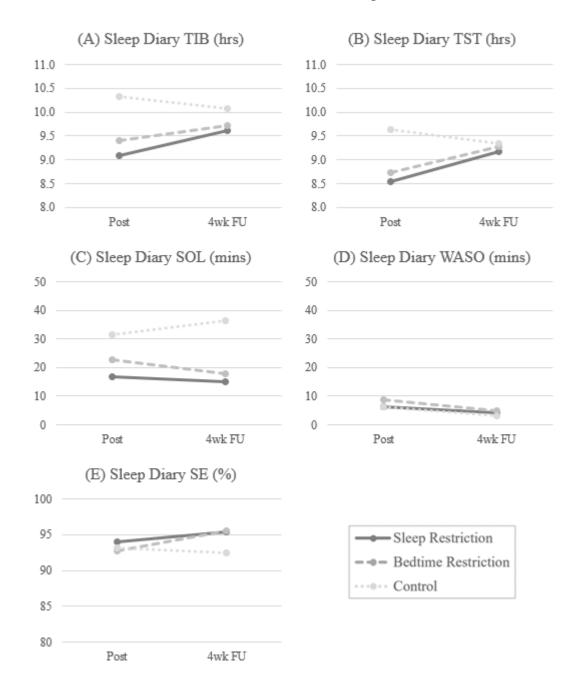


Figure 4.9. Estimated marginal means for sleep variables for each of the three treatment groups. Note: Post = Post-treatment (end of second treatment week), 4wk FU = 4-week follow-up; TIB = time in bed; TST = total sleep time; SOL = sleep onset latency, WASO = wake after sleep onset, SE = sleep efficiency.

Chapter 4: A Randomised Controlled Trial

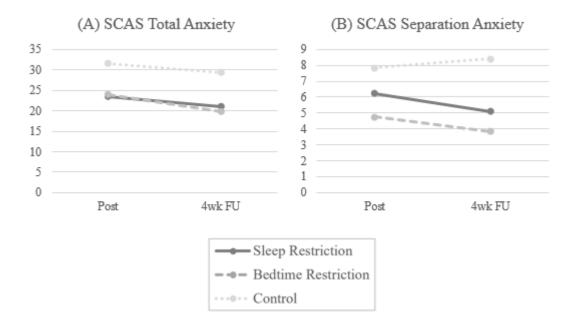


Figure 4.10. Estimated marginal means for anxiety variables for each of the three treatment groups. Note: Post = Post-treatment (end of second treatment week), 4wk FU = 4-week follow-up; SCAS = Spence Children's Anxiety Scale.

Changes in parental perception of child sleep difficulties from pre-treatment to 4-week follow-up were also examined. Using non-parametric McNemar's Tests, and considering the sample as a whole, there was a statistically significant reduction in the percentage of parents reporting that their child (A) had difficulty falling asleep, p<.001, (B) had difficulty staying asleep, p<.001, (C) required parental presence to fall asleep or return to sleep, p=.02, and (D) had a "sleep problem", p<.001. Results for each of the treatment groups separately are presented in Figure 4.11. All groups saw improvement in at least one of these items and, as expected, sleep restriction therapy was associated with the largest improvements. After completion of the 4-wk follow-up period, parents were also asked whether their child required any further treatment at the Child & Adolescent Sleep Clinic. Despite 30% fewer parents in the sleep restriction group requesting further treatment (compared to the control group), Pearson's Chi-Square analysis revealed no significant difference between groups, $\chi^2(2, N=48)=3.00$, p=.22 (% seeking further treatment after follow-up: sleep restriction 29%; bedtime restriction 43%; control 59%).

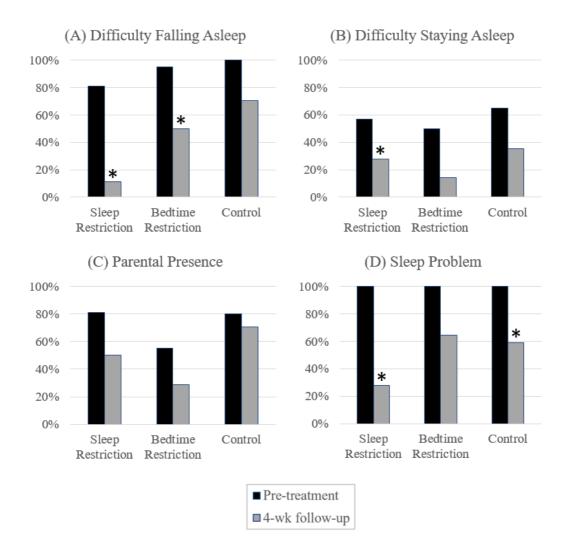


Figure 4.11. Percentage of parents reporting that their child (A) had difficulty falling asleep, (B) had difficulty staying asleep, (C) required parental presence to fall asleep or return to sleep, and (D) had a "sleep problem".

Note: * indicates statistically significant change from pre-treatment to follow-up.

4.5 Discussion

This RCT evaluated the efficacy of two brief interventions (i.e., sleep restriction therapy, bedtime restriction therapy) for school-aged children with Chronic Insomnia Disorder, compared to an active control group (i.e., bedtime regularisation). The intention of each treatment arm was to (1) reduce TIB (bedtime restriction therapy), (2) reduce both TIB and TST (sleep restriction therapy), or (3) not manipulate TIB (control), in order to explore different mechanisms linking improvements in sleep and anxiety (Dahl & Harvey, 2007).

Participants complied with treatment instructions by reducing TIB, as expected, during the 2 treatment weeks, with no significant change for the control group. TST also decreased significantly following sleep restriction therapy by 58mins (as expected), and bedtime restriction therapy resulted in a significant decrease in TST (slightly unexpected) although of a smaller magnitude (26mins). Both sleep restriction and bedtime restriction therapy resulted in significant improvements in SOL and SE during treatment, and these improvements were maintained up to the 4-week follow-up, with no significant changes in sleep experienced for the control group. The magnitude of improvement in SOL experienced by children in the sleep restriction and bedtime restriction groups (~20-30mins per night) can be considered clinically meaningful, and is similar to that observed in previous studies using multi-component cognitive-behavioural therapy (e.g., Paine & Gradisar, 2011)⁸. These results add to the growing body of evidence supporting the use of cognitive-behavioural treatments for insomnia in typically-developing school-aged children (e.g., Paine & Gradisar, 2011; Quach et al., 2011; Schlarb et al., 2016) and provide preliminary support for sleep restriction and bedtime restriction therapies as "stand alone" brief interventions in this population.

Pilot data from the two open trials described in Chapter 3 (Leahy & Gradisar, 2012b; Watherston & Gradisar, 2013), together with theory considering sleep and arousal to be opponent processes (Dahl, 1996; Dahl & Harvey, 2007), suggest the three treatment groups in the present study would experience different changes in anxiety. In particular, it was expected that changes in sleep experienced by the sleep restriction and bedtime restriction groups would result in improvements in anxiety, with no change in anxiety expected for the control group. However, contrary to expectations, anxiety improved similarly for participants in all three groups from pre- to post-treatment, and this improvement was maintained up to the 4-week follow-up. Compared to Paine and Gradisar (2011) the magnitude of improvement in SCAS total scores was smaller in the

⁸ Few previous treatment studies have measured SOL and the magnitude of improvement observed in the present study is greater than those reported by others (e.g., 5min, Schlarb et al., 2016).

present study (i.e., 18 points for Paine & Gradisar [2011] vs 8 points in the present study), which may suggest that the additional anxiety-focused treatment components did confer additional benefits for anxiety over-and-above any transdiagnostic effect induced by bedtime restriction therapy.

Similar to studies using sleep restriction therapy with adults (e.g., Kyle et al., 2011; Miller et al., 2014; Vallieres et al., 2013), both sleep restriction and bedtime restriction therapies were associated with an increase in evening sleepiness during treatment, suggesting that these interventions both resulted in an increase in sleep homeostatic pressure. This is also consistent with Watherston and Gradisar's (2013) open trial using the same sleep restriction therapy protocol, and is understandable considering that both groups experienced a decrease in TST during the 2 weeks of treatment. While previous research suggests that behavioural disinhibition (e.g., hyperactivity) is common among children with short sleep duration (Paavonen et al., 2009), it is worth noting that we did not observe hyperactivity in any participants who received sleep restriction or bedtime restriction therapies, and that this is consistent with previous research on experimental sleep restriction in school-aged children (for a review, see Lundahl, Kidwell, Van Dyk, & Nelson, 2015).

Based on Dahl and Harvey (2007), it was further hypothesised that a reduction in anxiety following restriction of TIB may be related to reduced opportunity for worry while awake in bed. However, contrary to expectations, but consistent with results pertaining to anxiety, worry improved similarly for participants in all three groups from pre- to post-treatment. Using the same measure of worry, Watherston and Gradisar (2013) also reported significant improvement following sleep restriction therapy, although again the magnitude of improvement was smaller in the present study (i.e., 11 points for Watherston & Gradisar [2013] vs 3-6 points in the present study).

Four-weeks after completion of treatment, parental perceptions about the child's current sleep pattern were collected via email or telephone interview. Considering that attrition is common with follow-up data (Dumville, Togerson, & Hewitt, 2006), response rates of 70-86% for each group (80% overall) can be considered adequate. Interestingly, parents in all three groups reported

improvements in their child's sleep since commencing treatment. Parents reported significant reductions in difficulty falling asleep (for both sleep restriction and bedtime restriction groups), difficulty staying asleep (sleep restriction group), and perception of their child's sleep as a "problem" (sleep restriction and control groups). Similar magnitudes of improvement were also reported for the bedtime restriction group (e.g., for reduced difficulty staying asleep, and perception of sleep as a "problem") but these failed to reach statistical significance, probably as a result of a slightly lower response rate for this group. It is interesting to note that these reports do not completely align with the improvements observed using sleep diary and actigraphy measures of sleep (i.e., for those in the control group), and suggest that parental perceptions of children's sleep difficulties are multi-faceted.

As previously mentioned, the control group in this study implemented a regular sleep schedule and saw little change in sleep parameters during treatment. These results alone suggest that, for school-aged children with Chronic Insomnia Disorder, sleep regularisation may be insufficient to produce significant improvements in sleep. This is interesting because it is often recommended as the first point of intervention by sleep specialists (e.g., Durand, 2008; Meltzer & Crabtree, 2015) and may be considered the usual care provided by primary health care professionals (e.g., GPs). Further research with a larger sample is required to confirm this finding.

Interestingly, approximately 40% of parents who implemented the bedtime regularisation (i.e., control) intervention reported that their child no longer had a "sleep problem" 4 weeks after the completion of treatment, and approximately 30% reported that their child no-longer had difficulty falling asleep. Although it is possible that this reflects a demand effect, it is also possible that participating in the treatment program had changed parents' attitudes towards their child's sleep and thus they no longer viewed their child's sleep as a "problem", despite little change in actual sleep parameters (SOL, WASO, TST). Again, this finding requires future replication.

The control group also reported unexpected improvements in total anxiety and worry, despite no change in TST or evening sleepiness. This may suggest that simply having contact with a

therapist or knowing that they were seeking treatment for their sleep problem helped to reduce selfreported anxiety symptoms (as has been reported in previous studies of children with anxiety disorders; Hudson, Rapee, et al., 2009). The results for separation anxiety may provide some insight here, as this interaction approached significance and suggested that separation anxiety continued to improve for the sleep restriction and bedtime restriction groups up to the 4-week follow-up, but the control group's score returned to pre-treatment level (when they were no longer having contact with their sleep therapist). As there is a strong relationship between parent anxiety and child anxiety (Craske & Waters, 2005; Ginsburg, Silverman, & Kurtines, 1995; Ginsburg & Schlossberg, 2002), it is also possible that implementing a structured sleep plan under the supervision of a sleep therapist reduced parents' anxiety about their child's sleep and, consequently, child anxiety was also reduced. However, this cannot be confirmed because parent anxiety was not measured at posttreatment.

Unfortunately, exploratory correlations between changes in sleep, evening sleepiness, worry, and anxiety failed to confirm the predicted relationships in the present sample. Thus, further research is required to investigate the potential mechanisms linking improvements in sleep and anxiety following sleep restriction therapy in middle childhood.

4.5.1 Strengths, Limitations, and Recommendations for Future Research

Strengths of this study include the use of actigraphy as an objective measure of sleep, and the presence of an active control group who received a treatment commonly recommended and disseminated in primary care (i.e., bedtime regularisation; Durand, 2008; Meltzer & Crabtree, 2015). This study was also conducted in a real-world clinical setting, and thus has good external validity. While many treatment studies are limited by attrition, it is worth noting that 98% of participants (60 out of 61) completed the 2-week treatment program and 95% (58 out of 61) provided sleep diary and questionnaire data at post-treatment. Replication studies using these treatments are needed to confirm the high uptake of these therapies, although these preliminary

results suggest that sleep restriction, bedtime restriction, and bedtime regularisation may all be wellreceived by parents of school-aged children with sleep problems.

This study used a broad age range (6-14 yrs) so as to maximise sample size during a limited recruitment period. Pubertal status was not explicitly measured, although the average onset of puberty in western populations is around 10-12 yrs (Kall & Cavanaugh, 2010; Mensah et al., 2013; Warren & Yu, 2015) so it is likely that some of the participants in our sample had entered the early stages of puberty. As previously mentioned, the onset of puberty is often associated with delayed sleep timing (i.e., evening chronotype). Thus, it is possible that the later bedtimes implemented during sleep restriction and bedtime restriction therapies were effective in reducing SOL due to alignment with a delayed circadian rhythm in our older participants. However, this is unlikely as we attempted to reduce the impact of circadian influences on treatment outcome by specifically excluding individuals with DSWPD.

As with previous studies of multi-component cognitive-behavioural sleep interventions (e.g., Paine & Gradisar, 2011), our population comprised predominantly mid- to high-SES families, which may limit generalisability of our findings to lower-SES groups. Similarly, none of the participants in the present study reported current or previous mood disorders (e.g., depression) or other psychiatric symptoms (e.g., suicidality, psychosis), and thus these findings may not generalise to children with more complex psychiatric presentations. Although this study was powered to detect differences between treatment groups, further dividing the groups into responders/non-responders would reduce statistical power by 50%. Therefore, it is recommended that characteristics of responders/non-responders to treatment be examined in a future study with a larger sample size.

The present study used a short follow-up period of only 4 weeks, so as to not prevent children who required further treatment from accessing it. However, this means that conclusions cannot be drawn about the longer-term effectiveness of these brief interventions. Future studies should therefore continue to monitor participants for longer follow-up periods (e.g., 6 months) after the completion of treatment. Furthermore, future studies are needed to further examine the relationship between improvements in sleep and anxiety, and the potential mechanisms linking them.

4.5.2 Conclusions

The present study suggests that two brief sleep interventions (i.e., sleep restriction therapy or bedtime restriction therapy) delivered over 2 weeks, have the potential to produce meaningful improvements in sleep (i.e., SOL, SE) for school-aged children with Chronic Insomnia Disorder, compared to an active control group (who received bedtime regularisation). Furthermore, all three groups experienced improvement in anxiety and worry during treatment, and showed improvements in parental perception of child sleep problems. Future studies are needed to replicate these findings, to investigate whether these improvements are maintained in the longer-term, and to further investigate the potential mechanisms underlying the relationship between sleep and anxiety in middle childhood.

Chapter 5:

Cognitive Performance, Classroom Attention, and Parasomnias: Contraindications for Sleep Restriction Therapy in School-Aged Children with Chronic Insomnia Disorder?

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Author Contributions

NC contributed to study conceptualisation and design, and led recruitment, data collection, data analysis, results interpretation, and manuscript preparation. CR and KB assisted with recruitment and data collection. AP, HW, and JR assisted with data collection. MG contributed to study conceptualisation, design, recruitment, data collection, results interpretation, and manuscript preparation. All authors approved the final manuscript.

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5.1 Abstract

Sleep restriction therapy is effective in the treatment of adult insomnia, and recent studies suggest that similar techniques may also be used for school-aged children. However, clinicians may be understandably cautious about restricting sleep opportunity during the school years, due to the unknown impact on cognitive performance, school attention, or parasomnias. 61 participants (6-14yrs; 54% female) with Chronic Insomnia Disorder were randomly allocated to one of 3 groups (sleep restriction therapy, bedtime restriction therapy, bedtime regularisation [control]). Both restriction therapies improved sleep onset latency (p<0.01). However, there were no significant interactions for cognitive performance and classroom attention measures, nor any difference in parasomnias between groups (all p>.05). Therefore, clinicians may not need to be overly concerned about these potential contraindications.

5.2 Introduction

There is a good evidence base for the use of sleep restriction therapy in the treatment of adult insomnia (Miller et al., 2014). This behavioural sleep intervention, which involves temporarily reducing the individual's time in bed (TIB) to more closely match their baseline average total sleep time (TST), aims to improve sleep by increasing homeostatic sleep pressure and extinguish the learned conditioned alertness that the individual experiences in bed. Consistent with the expected increase in sleep pressure, adults undergoing sleep restriction therapy experience a temporary increase in daytime sleepiness (Kyle et al., 2011; Miller et al., 2014; Vallieres et al., 2013), followed by improvements in sleep onset latency (SOL; i.e., time taken to fall asleep; Miller et al., 2014; Vallieres et al., 2013; Whittall, Pillion, & Gradisar, 2018) and wake after sleep onset (WASO; i.e., time spent awake during the night; Miller et al., 2014; Whittall et al., 2014; Vallieres et al., 2013) and a subjective experience of improved sleep quality and attitude towards sleep (e.g., via the novel sensation of "craving" sleep; Kyle et al., 2011).

With school-aged children, similar techniques (also known as "faded bedtime" or "bedtime fading") are generally used and recommended, despite a lack of strong empirical evidence. Several recent studies suggest that these techniques may be effective as part of multicomponent treatment programs for insomnia in middle childhood (Corkum et al., 2016; Paine & Gradisar, 2011; Quach et al., 2011; Schlarb et al., 2016). However, clinicians may remain understandably cautious about restricting sleep opportunity in this critical period of development and thus these techniques may be under-utilised. In fact, some clinical texts specifically advise against using sleep restriction therapy "at times … when academic functioning is a concern" (Meltzer & Crabtree, 2015, pp.149-150). Some clinical texts also advise against using sleep restriction therapy for individuals with comorbid sleep disorders which may be exacerbated by sleep restriction (e.g., parasomnias; Meltzer & Crabtree, 2015), although others simply acknowledge that an increase in frequency of parasomnias may occur during sleep restriction, and that parents should "be patient … [because] these should

abate when the bedtime is moved earlier" (Durand, 2008, p.63). Each of these potential contraindications will be addressed in turn.

5.2.1 Cognitive Performance

Firstly, it is plausible that sleep restriction therapies may be contraindicated for children due to potential consequences on cognitive functioning during this critical developmental period. In good-sleeping children, one night of experimental sleep restriction (i.e., reducing sleep opportunity to 4-5 hours) does not appear to negatively impact basic cognitive functions such as sustained attention (Fallone, Acebo, Arnedt, Seifer, & Carskadon, 2001), memory (Randazzo et al., 1998) or processing speed (Carskadon et al., 1981); however, more complex cognitive functions such as abstract reasoning and verbal creativity do appear to be affected (Randazzo et al., 1998). Studies involving milder sleep restriction (i.e., reducing sleep opportunity by 1 hour relative to baseline for 3-4 consecutive nights) have found deficits in simple reaction time (Sadeh, Gruber, & Raviv, 2003), attention (Vriend et al., 2013), and short-term memory (Vriend et al., 2013), although significant differences were only detected when comparing to a sleep extension condition rather than a "normal sleep" condition. In these studies, experimental sleep restriction resulted in an increase in subjective sleepiness (Sadeh et al., 2003; Vriend et al., 2013) as well as improvements in SOL and objective measures of sleep quality (i.e., sleep percent, WASO; Sadeh et al., 2003) which may be similar to changes observed during sleep restriction therapy. Thus, while the sleep restriction in these experimental studies was brief (relative to sleep restriction therapy) and performed on goodsleeping children, it may be anticipated that 14 days of sleep restriction therapy would also result in increased subjective daytime sleepiness and impaired cognitive performance for school-aged children.

5.2.2 Classroom Attention

Studies using parent or teacher questionnaire ratings as indicators of basic cognitive processes, such as attention, have also found impairments following 3-5 nights of experimental

sleep restriction (Fallone et al., 2005; Gruber et al., 2012; Vriend et al., 2013). Vriend and colleagues (2013) found impairments in attention after sleep restriction (compared to sleep extension) using a parent-report questionnaire. Using a similar teacher questionnaire, improvements in attention were observed after experimental sleep extension (compared to baseline; Gruber et al., 2012) and impairments in attention were observed after experimental sleep restriction (Fallone et al., 2005; Gruber et al., 2012). Thus, it is anticipated that 14 nights of sleep restriction therapy would also result in impairments in teacher-reported attention.

5.2.3 Parasomnias

Sleep deprivation is often understood to be a trigger for parasomnias, and individuals who experience parasomnias classified as disorders of arousal (e.g., sleep terrors, sleepwalking; American Academy of Sleep Medicine [AASM], 2014), which occur during slow wave sleep, are particularly advised to avoid sleep deprivation (Galbiati et al., 2015; Kotagal, 2009; Owens & Mohan, 2016; Simon & Byars, 2016) and irregular sleep schedules (Owens & Mohan, 2016). It is generally understood that sleep restriction can increase time spent in slow wave sleep, and thus increase the occurrence of parasomnia episodes (Owens & Mohan, 2016). This is supported by results of experimental studies with adults (Joncas, Zadra, Paquet, & Montplaisir, 2002; Pilon, Montplaisir, & Zadra, 2008; Zadra, Pilon, & Montplaisir, 2008); although these studies involved a full night of sleep deprivation, which may be more extreme than the mild restriction of sleep opportunity that occurs during sleep restriction therapy. A case report study of sleep restriction therapy for two 4-year-old girls, with sleep difficulties and comorbid developmental disorders, observed the occurrence of sleepwalking and sleep terrors in one of their participants after treatment (Durand & Christodulu, 2004), although the complexity of these cases and the prolonged observation period (> 6 months) make it difficult to determine whether these events were a direct result of the sleep restriction intervention. To our knowledge, there have been no systematic evaluations of the increase of parasomnias during sleep restriction therapies for school-aged children with insomnia, which leaves an important gap in the literature.

5.2.4 Study Aims

The current study investigated changes in subjective daytime sleepiness, cognitive performance, classroom attention, and the occurrence of parasomnias in school-aged children undergoing 2 weeks of restriction therapies, as part of a larger randomised controlled trial (RCT; see Chapter 4). Guidelines vary for the implementation of sleep restriction therapy among school-aged children: some recommend reducing TIB during treatment to match baseline TST (e.g., Roane & Taylor, 2013), while others recommend longer (eg., Meltzer & Crabtree, 2015) or even shorter (e.g., Durand, 2008) prescribed TIB. The current study compared two "doses" of sleep restriction therapy, which we have labelled "bedtime restriction therapy" (reducing TIB to match baseline TST) and "sleep restriction therapy" (reducing TIB to 30 mins less than baseline TST), against an active control group.

5.3 Method

5.3.1 Participants

Participant details were presented in Chapter 4 and, therefore, will not be repeated here.

5.3.2 Design

The study employed a 3 (treatment: sleep restriction, bedtime restriction, control) x 3 (time: pre-treatment, mid-treatment, post-treatment) mixed-model design.

5.3.3 Materials and Measures

Descriptions of the 7-day sleep diary (Appendix C.1), clinical sleep history interview

(Appendix C.4) and wrist actigraphy measures were presented in Chapter 4 and, therefore, will not be repeated here.

Cognitive testing. Cognitive functioning was measured with the Neuropsychological Evaluation System (NES), which comprises 6 tasks completed on a desktop computer. The NES has been successfully used with school-aged children (e.g., Arcia, Ornstein, & Otto, 1991; Sadeh et al.,

2002; Sadeh et al., 2003) and is a good predictor of classroom attention and performance (Arcia et al., 1991). A research assistant blind to treatment allocation administered these tasks.

1. Finger tapping test: The child used their index finger to tap a button on the keyboard as many times as possible within a 10-sec interval. Five consecutive trials were completed with each hand. The task measured vigilance and motor reaction with a total score calculated as the overall mean number of taps (Groth-Marnat, 2000).

2. *Simple reaction-time test*: The child pressed a key as quickly as possible when a large red circle appeared on the screen. The circle was presented at random time intervals (range: 2000ms-8000ms). In the first set of trials, the circle was consistently presented in the centre of the screen and on the second set of trials the circle was presented in random locations. The task measured sustained visual attention, response inhibition and reaction time with a total score obtained for mean response latency.

3. Continuous performance test: Seven different animal silhouettes were presented on the computer screen one by one. The child was instructed to respond only when a specific animal was presented (the target) and to avoid pressing the key whenever a different animal was presented (distractors). This task measured sustained visual attention, response inhibition and reaction time with scores being obtained for (1) mean reaction time, (2) percentage of omission errors (i.e., not responding to target stimulus), and (3) percentage of commission errors (i.e., responding to non-target stimulus).

4. Symbol-digit substitution: Nine symbol-digit pairs were presented at the top of the screen. The child was given a grid of mixed order symbols and asked to pair as many symbols with their corresponding number as quickly as possible within a 2-min time limit. The symbol-digit substitution task measured visual memory, visual scanning and visual-motor speed, with a total score calculated by subtracting the error count from the number of correct responses.

5. *Visual digit span test*: Sequences of digits were presented on the computer screen and the participant was asked to repeat the sequence by typing on the computer keyboard either in the same

order (i.e., forwards) or in reverse order (i.e., backwards). Increasingly longer spans were presented. This task measured visual short-term memory with scores being obtained for longest span length forwards and backwards.

6. Serial digit learning test: A sequence of 9 single digits was presented to the child in succession and they were asked to recall as many numbers in the correct order as possible. The same digits were presented over 8 trials. The task measured learning and visual short-term memory, with a total error score calculated by summing the number of errors across the 8 trials.

Teacher-report questionnaire (see Appendix D.1). The School Situations Questionnaire – Revised (SSQ-R, DuPaul & Barkley, 1992) contains 8 items that assess whether the child has difficulty paying attention or concentrating in a variety of situations (e.g., "during individual work"). The teacher initially provides a yes or no response for each item, followed by a severity rating (1=*mild*, 9=*severe*) for all items endorsed for the child. Two final scores were obtained: (1) The total problems score was the number of items on the questionnaire to which a 'yes' response was provided; and (2) The mean severity score was the mean of scores provided (1-9) for all questions answered with a 'yes'.

Daytime sleepiness (see Appendix D.2). The Paediatric Daytime Sleepiness Scale (PDSS; Drake et al., 2003) is an 8-item self-report scale of daytime sleepiness (e.g., *How often do you fall asleep or feel drowsy in class?*). Ratings are provided on a 5-point scale (0 = never; 4 = always). One item is reverse scored (*Are you usually alert during the day?*) and responses are summed to produce a total score from 0 to 32. For younger children, parents were instructed that they could help their child to read and understand the items, but that the response recorded must be the child's own response. In the current sample, internal validity was acceptable (Chronbach's alpha = 0.78).

Treatment. All families attended 2 treatment sessions with their psychologist, each held 1 week apart. Descriptions of the treatment protocols were presented in Chapter 4, and full treatment manuals can be found in Appendix C.5.

At each treatment session, the psychologist noted whether the child had experienced any parasomnias during the past week (and if so, how many). If necessary, the parents' concerns were discussed and any modifications to the treatment plan were negotiated (e.g., increase in TIB) based on recommended clinical care for parasomnias (e.g., Galbiati et al., 2015; Kotagal, 2009; Owens & Mohan, 2016; Simon & Byars, 2016).

5.3.4 Procedure

Details of participant recruitment, screening, assessment, and randomisation were presented in Chapter 4. In addition to the procedures described in Chapter 4, children completed the PDSS and the computerised NES on 3 occasions (pre-treatment, mid-treatment, post-treatment) and the child's usual classroom teacher completed the SSQ-R at pre-treatment and post-treatment. The timing of all measures relevant to the current chapter is presented in Table 5.1.

Table 5.1

Timing of measures

	Pre	Mid	Post
Clinical sleep history interview	•		
Sleep diary	•	•	•
Actigraphy	•	•	•
PDSS	•	•	•
NES	•	•	•
SSQ-R	•		•

Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), PDSS = Pediatric Daytime Sleepiness Scale, NES = Neuropsychological Evaluation System, SSQ-R = School Situations Questionnaire, Revised.

5.3.5 Statistical Analyses

All quantitative data analyses were conducted using the Statistical Package for the Social Sciences (SPSS v.22, SPSS Inc., Chicago, USA) and an alpha level of 0.05. One-way ANOVAs were used to investigate baseline differences between groups. Linear mixed model (LMM) analyses were used to analyse the 3 (group) x 3 (time) mixed model design for sleep diary, actigraphy and cognitive performance variables, and the 3 (group) x 2 (time) design for teacher-rated attention. For sleep variables expressed as units of time, effect sizes are presented as the magnitude of change in minutes. For other variables, Cohen's *d* was calculated as $d=M_1-M_2/(SD_{pooled})$ to establish the magnitude of within- and between-subjects differences. Non-parametric Chi-square analyses were used to compare differences in reported history of parasomnias and occurrence of parasomnias during treatment.

5.4 Results

5.4.1 Baseline Sample Characteristics

There was no significant difference between groups in age, F(2,58)=.97, p=.39, gender distribution, $\chi^2(2, N=61)=3.24$, p=.20, or socioeconomic status, F(2,54)=.89, p=.42. Across the whole sample, 90% of children lived in a dual-parent household and 87% had one or more siblings. Descriptive statistics for demographic and sleep variables are presented in Table 5.2. There were also no pre-treatment differences in cognitive performance, teacher ratings of attention, or history of parasomnias (all p>.05).

Table 5.2

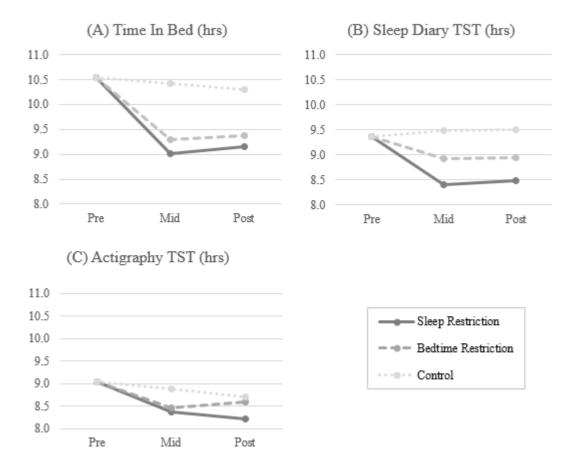
Pre-treatment descriptive statistics for demographic and sleep variables

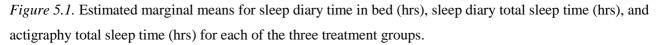
		Treatment Group			
		Sleep Restriction (N=21)	Bedtime Restriction (N=20)	Control (N=20)	Whole Sample (N=61)
Age (M ± SI	D, in yrs)	9.5 ± 2.4	9.1 ± 2.0	8.6 ± 2.0	9.2 ± 2.1
Gender (% F	Female)	43%	50%	70%	54%
Socioeconor	nic Status ¹ ($M \pm SD$)	6.9 ± 2.3	7.8 ± 2.0	6.8 ± 3.0	7.2 ± 2.5
Family Structure	Dual-parent household (%)	86%	100%	85%	90%
	\geq 1 sibling (%)	76%	94%	90%	87%
	Oldest child (%)	80%	53%	70%	68%
Sleep Diary	SOL (M \pm SD, in mins)	37.4 ± 20.4	57.2 ± 38.6	37.0 ± 22.9	43.8 ± 29.5
	WASO (M \pm SD, in mins)	12.1 ± 20.0	11.5 ± 23.6	10.7 ± 14.7	11.4 ± 19.4
	SE (M \pm SD, %)	89.7 ± 6.8	86.6 ± 8.0	90.4 ± 5.8	88.9 ± 7.0
	TST (M \pm SD, in hrs)	9.4 ± 0.8	9.2 ± 1.0	9.5 ± 0.8	9.4 ± 0.9
	TIB (M \pm SD, in hrs)	10.5 ± 0.5	10.6 ± 0.9	10.6 ± 0.8	10.5 ± 0.7
Actigraphy	SOL (M \pm SD, in mins)	36.0 ± 22.0	43.6 ± 37.3	33.5 ± 26.2	37.6 ± 28.7
	WASO (M \pm SD, in mins)	49.9 ± 26.9	51.6 ± 22.3	50.1 ± 14.2	50.5 ± 21.6
	SE (M \pm SD, %)	85.6 ± 7.3	85.4 ± 6.6	87.1 ± 4.3	86.1 ± 6.2
	TST ($M \pm SD$, in hrs)	8.9 ± 0.8	9.0 ± 0.6	9.2 ± 0.5	9.1 ± 0.6

Note: ¹ The SEIFA index of socioeconomic advantage and disadvantage ranges from low (1) to high (10) socioeconomic status (Australian Bureau of Statistics, 2011).

5.4.2 Treatment Compliance and Manipulation Check

There were significant interactions for both TIB, F(4,114)=8.83, p<.001, and sleep diary TST, F(4,113)=6.35, p<.001. This suggests that, consistent with treatment instructions, the sleep restriction and bedtime restriction groups both experienced reduced TIB and TST relative to the control group (Figure 5.1). Actigraphy data similarly showed reduced TST during the first treatment week for both sleep restriction and bedtime restriction groups compared to the control group, but this interaction failed to reach statistical significance, F(4,78)=1.84, p=.13.





Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), TST = total sleep time. Values have been equated at pre-treatment to assist in interpretation of change.

5.4.3 Daytime Sleepiness

There was a significant interaction for daytime sleepiness, F(4,106)=2.96, p=.023. However, contrary to expectations, there was no change for the sleep restriction and bedtime restriction groups, while the control group reported a decrease in daytime sleepiness during treatment (Figure 5.2).

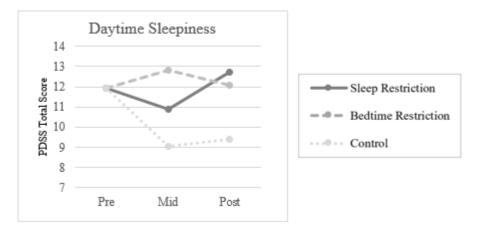


Figure 5.2. Estimated marginal means for subjective sleepiness for each of the three treatment groups. Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week). Values have been equated at pre-treatment to assist in interpretation of change.

5.4.4 Cognitive Performance and Classroom Attention

There were no significant interactions for any of the cognitive performance variables as measured by the NES (all p>.05, Table 5.3). However, significant main effects for time were observed for simple reaction time, F(2,47)=11.48, p<.001, d=0.47, symbol-digit substitution, F(2,60)=40.52, p<.001, d=0.82, and the serial digit learning test, F(2,91)=18.32, p<.001, d=0.60. For symbol-digit substitution and the serial digit learning test performance improved over time, while there was a slowing of responses on the simple reaction time task for participants in all three treatment groups.

Table 5.3

Estimated marginal means and standard errors for cognitive performance variables

		Cognitive Performance Scores ($M \pm SE$)			
		Sleep Restriction	Bedtime Restriction	Control	
Finger Tapping	Pre	48.0 ± 1.97	49.5 ± 2.08	49.5 ± 2.14	
(overall mean number of taps)	Mid	49.0 ± 1.97	49.1 ± 2.08	48.7 ± 2.14	
	Post	49.4 ± 1.98	49.3 ± 2.09	49.1 ± 2.15	
Simple Reaction Time	Pre	434 ± 36.6	425 ± 38.0	416 ± 37.6	
(ms)*	Mid	478 ± 36.6	466 ± 37.6	505 ± 38.0	
	Post	496 ± 37.3	461 ± 38.4	538 ± 38.0	
Symbol-Digit	Pre	26.8 ± 3.54	25.4 ± 3.74	26.0 ± 3.74	
Substitution (total score)*	Mid	34.2 ± 3.56	36.8 ± 3.74	30.0 ± 3.77	
	Post	37.6 ± 3.59	41.0 ± 3.80	37.4 ± 3.77	
Continuous	Pre	600 ± 15.9	578 ± 16.8	593 ± 16.8	
Performance Test (reaction time)	Mid	587 ± 15.9	568 ± 16.8	610 ± 16.8	
	Post	596 ± 16.3	563 ± 17.4	580 ± 17.1	
Continuous	Pre	3.29 ± 1.86	4.89 ± 1.97	5.60 ± 1.97	
Performance Test (omission errors %)	Mid	4.59 ± 1.86	5.16 ± 1.97	5.64 ± 1.97	
	Post	2.92 ± 1.91	5.91 ± 2.04	9.13 ± 2.00	
Continuous	Pre	1.12 ± 0.50	1.12 ± 0.53	1.83 ± 0.53	
Performance Test (commission errors %)	Mid	1.53 ± 0.50	1.24 ± 0.53	2.16 ± 0.53	
	Post	1.22 ± 0.52	1.68 ± 0.56	1.63 ± 0.54	
Digit Span Forwards	Pre	5.32 ± 0.24	5.65 ± 0.25	5.24 ± 0.25	
	Mid	5.42 ± 0.24	5.70 ± 0.26	5.28 ± 0.26	
	Post	5.65 ± 0.25	5.79 ± 0.27	5.44 ± 0.26	
Digit Span Backwards	Pre	4.16 ± 0.27	4.29 ± 0.29	4.47 ± 0.29	
	Mid	4.26 ± 0.27	4.32 ± 0.29	4.01 ± 0.29	
	Post	4.42 ± 0.28	4.59 ± 0.30	4.12 ± 0.29	

		Cognitive 1	Cognitive Performance Scores (M \pm SE)			
		Sleep Restriction	Bedtime Restriction	Control		
Serial Digit Learning Test (error score)*	Pre	35.4 ± 4.15	36.4 ± 4.52	39.3 ± 4.52		
	Mid	32.5 ± 4.15	36.0 ± 4.52	32.4 ± 4.52		
	Post	26.3 ± 4.23	22.8 ± 4.63	29.6 ± 4.57		

Note: Pre = Pre-treatment, Mid = Mid-treatment (end of first treatment week), Post = Post-treatment (end of second treatment week), * = significant main effect for time (p < .05).

There were no significant main effects and no significant interactions for frequency and severity of attention problems, as rated by teachers using the School Situations Questionnaire (all p>.05; Table 5.4). However, the teachers' response rate was poor, with only 38 out of 61 questionnaires completed at pre-treatment (62%) and only 23 completed at post-treatment (37%). Therefore, these results must be interpreted with caution.

Table 5.4

Estimated marginal means and standard errors for teacher ratings on the School Situations Questionnaire (all p>.05)

		School Situations Questionnaire Scores (M \pm SE)			
		Sleep Restriction	Bedtime Restriction	Control	
Number of problems	Pre	2.56 ± 0.66	3.33 ± 0.76	2.40 ± 0.83	
	Post	2.68 ± 0.70	2.84 ± 0.81	2.40 ± 0.89	
Severity of problems	Pre	2.49 ± 0.58	2.61 ± 0.68	1.50 ± 0.74	
	Post	2.71 ± 0.65	2.52 ± 0.76	1.58 ± 0.82	

Note: Pre = Pre-treatment, Post = Post-treatment (end of second treatment week).

5.4.5 Parasomnias

At pre-treatment, 27 out of 61 participants (44%) reported a history of parasomnias (e.g., sleep terrors, sleepwalking) and these were evenly split across the 3 treatment groups (Table 5.5). Of these, 21 out of 27 (78%) had no re-occurrence of parasomnias during treatment; the remaining 6 children did experience parasomnias during treatment. For 5 of these children this was infrequent, and parents' were not concerned, so treatment continued without modification. For 1 child (bedtime restriction group) the frequency of parasomnias increased and the treatment plan was modified (i.e., prescribed TIB was increased by 45mins and education was provided about scheduled awakenings as the recommended treatment for sleep terrors; Galbiati et al., 2015; Kotagal, 2009; Owens & Mohan, 2016; Simon & Byars, 2016). None of the children without a history of parasomnias experienced them for the first time during treatment, and Chi-square analyses revealed no significant difference between treatment groups in reported history of parasomnias, $\chi^2(2, N=61)=.004$, *p=.*99.

			History of Parasomnias	
		-	No	Yes
Parasomnias during	No	Sleep Restriction	13	6
treatment		Bedtime Restriction	11	7
		Control	10	8
	Yes	Sleep Restriction	0	2
		Bedtime Restriction	0	2
		Control	0	2

Table 5.5

Number of parents	reporting of	occurrence of p	parasomnias b	y treatment group

5.5 Discussion

This RCT considered possible "contraindications" for sleep restriction and bedtime restriction therapies for school-aged children with Chronic Insomnia Disorder, with a particular focus on cognitive performance, classroom attention, and the occurrence of parasomnias. This study extends previous work by other authors in that it examined the effects of sleep restriction on cognitive functioning and school attention (1) in a clinical sample of children with Chronic Insomnia Disorder, (2) during a longer period of sleep restriction (i.e., 14 days), and (3) observed changes in the occurrence of parasomnias during therapeutic sleep restriction.

Compliance was good during the 2 treatment weeks, with TIB decreasing as expected for participants in each of the 2 treatment groups (i.e., sleep restriction therapy and bedtime restriction therapy), but surprisingly no significant increase in daytime sleepiness. Similarly, there were no significant interactions for any of the cognitive performance measures, nor for teacher ratings of attention. This suggests that clinicians may not need to be overly concerned about possible impacts on cognitive functioning for school-aged children experiencing sleep problems who undergo therapies that restrict TIB (and even sleep). These results are inconsistent with previous studies that have monitored cognitive performance during experimental sleep restriction (Sadeh et al., 2003; Vriend et al., 2013) despite a similar magnitude of reduction in TST (~30-45 mins per night on average) and a similar increase in evening sleepiness. Considering that these previous studies (Sadeh et al., 2003; Vriend et al., 2013) only restricted sleep for 3-4 consecutive nights, we predicted that 14 nights of sleep restriction in our sample of school-aged children would also result in impaired cognitive performance. However, previous studies (Sadeh et al., 2003; Vriend et al., 2013) only found significant differences when comparing sleep restriction to a sleep extension condition (rather than a "no change" group) and none of our treatment groups experienced an increase in TST during the 2 weeks of treatment, thus we cannot make this comparison with our data. It is also important to note that these previous studies were done using children without sleep disorders, and it is possible that the children in the present study were already experiencing

difficulties with attention and other aspects of cognitive functioning as a result of their longstanding sleep difficulties. Future studies should therefore compare cognitive performance during sleep restriction therapy with a control group of age-matched good-sleeping children.

While there were no significant interactions for any of the cognitive performance variables, participants in all 3 groups experienced improved performance on the symbol-digit substitution and serial digit learning tasks, which may indicate a practice effect for these tasks. There was also a slowing of responses on the simple reaction time tasks for participants in all 3 groups. These findings are consistent with previous research (Sadeh et al., 2003).

Only one participant (out of 61) experienced an increase in frequency of parasomnias (i.e., sleep terrors) which required modification to his bedtime restriction therapy plan (i.e., increasing TIB by 45 mins). For the rest of the sample (i.e., 98% of participants), there was no increase in the occurrence of parasomnias during the 2 weeks of treatment – even for children whose parents reported a history of these behaviours. It is widely recognised that these disorders of arousal are most prevalent during early childhood and tend to resolve spontaneously by early adolescence (AASM, 2014; Laberge, Tremblay, Vitaro, & Montplaisir, 2000). For example, two separate large community surveys found very high incidence of parasomnias during early childhood (almost 90%; Petit, Touchette, Tremblay, Boivin, & Montplaisir, 2007), with much lower rates described for children aged 11-13yrs (Laberge et al., 2000). However, these studies also point to different developmental courses for individual parasomnias, with prevalence of sleep terrors peaking early and decreasing significantly after 4yrs of age, but sleepwalking continuing to increase until at least 6yrs (Petit et al., 2007) and declining by 11-13yrs (Laberge et al., 2000). Furthermore, parasomnias (especially sleep terrors) have been associated with higher anxiety among children (Laberge et al., 2000; Petit et al., 2007) and are understood to be precipitated by sleep deprivation (Galbiati et al., 2015; Kotagal, 2009; Owens & Mohan, 2016; Simon & Byars, 2016), which may be relevant when considering children with insomnia (i.e., chronic sleep-deprivation) and anxiety symptoms. Thus, in the current sample, the risk of recurrence of parasomnias during treatment may have been low due

to a combination of (1) the overall declining incidence of parasomnias during the school years, (2) the observed improvements in anxiety during treatment (Chapter 4) may have reduced risk of future sleep terrors, and (3) the observed improvements in sleep quality during treatment (Chapter 4) may have counteracted any potential risk related to short-term restriction of sleep opportunity. This is important because it means that school-aged children (6-14yrs) experiencing both insomnia and parasomnias may not need to miss out on potentially beneficial treatment for their insomnia. To our knowledge, this is the first systematic evaluation of the occurrence of parasomnias during sleep restriction therapies for school-aged children with insomnia.

5.5.1 Strengths and Limitations

Strengths of this study include the use of actigraphy as an objective measure of sleep, and the presence of an active control group who received a treatment commonly recommended and disseminated in primary care (i.e., bedtime regularisation; Durand, 2008; Meltzer & Crabtree, 2015). While many treatment studies are limited by attrition, it is worth noting that 98% of participants (60 out of 61) completed the 2-week treatment program and 95% (58 out of 61) provided sleep diary and questionnaire data at post-treatment. Thus, replication studies using these treatments are needed to confirm the high uptake of these therapies, as well as evaluating any ongoing sleep benefits in the longer-term.

As with previous studies (i.e., Paine & Gradisar, 2011), our population comprised predominantly mid- to high-SES families, which may limit generalisability of our findings to lower-SES groups. Similarly, none of the participants in the present study reported current or previous mood disorders (e.g., depression, bipolar disorder) or other psychiatric symptoms (e.g., suicidality, psychosis), and thus these findings may not generalise to children with more complex psychiatric presentations. In fact, it is possible that symptoms of depression, suicidality, and/or psychosis may be exacerbated during sleep restriction for children with a history of such symptoms, as previous research has found links between sleep problems, depression, and suicidality in adolescents (Liu &

Buysse, 2006; Sarchiapone et al., 2014; Wong & Brower, 2012). This potential contraindication may be an important consideration for future research.

5.5.2 Conclusions

The present study suggests that clinicians may not need to be overly concerned about the impacts of sleep restriction and bedtime restriction therapies on daytime sleepiness, cognitive performance, or classroom attention, nor do they need to completely avoid using these techniques with school-aged children (6-14yrs) who have a history of parasomnias. Further studies are needed to confirm these positive findings, which may encourage the scientific and clinical communities to implement these brief sleep treatments.

Chapter 6: General Discussion

Chapter 6: Discussion

6.1 Summary of Main Findings

This thesis adds to the growing body of evidence supporting the use of cognitivebehavioural interventions for sleep problems in middle childhood. In particular, this research suggests that sleep restriction therapies⁹ can produce meaningful improvements in sleep parameters (i.e., sleep onset latency [SOL], sleep efficiency [SE]) and parental perception of child sleep problems (Chapter 4). Improvements in anxiety and worry also occurred; however, these improvements could not be attributed solely to the sleep restriction treatments as the control group reported similar improvements over the same time period (Chapter 4). As a consequence, it is unsurprising that correlations between sleep, anxiety, worry, and sleepiness provided little insight regarding potential mechanisms linking changes in sleep and anxiety, suggesting that other factors (e.g., changes in parent anxiety) may also contribute to this relationship.

Implementation of any new treatment requires careful consideration of both the potential benefits and potential risks associated with its use. An examination of changes in daytime sleepiness, cognitive performance, school attention, and incidence of parasomnias during our randomised controlled trial (RCT) suggested that clinicians may not need to be concerned about the impacts of sleep restriction therapies on daytime sleepiness, cognitive performance or classroom attention, nor do they need to completely avoid using these techniques with school-aged children (6-14yrs) who have a history of parasomnias (Chapter 5). This means that children experiencing both insomnia and parasomnias (e.g., sleep terrors) may not need to miss out on potentially beneficial treatment for their insomnia.

⁹ As results were similar for both sleep restriction therapy and bedtime restriction therapy, they will be collectively referred to as "sleep restriction therapies".

6.2 How Thesis Findings Extend Current Knowledge about Sleep Interventions in Middle Childhood

6.2.1 Replication of Past Research

Replication of research findings is essential for building knowledge (Moonesinghe, Khoury, & Janssens, 2007; Open Science Collaboration, 2015; Schmidt, 2009). According to the Open Science Collaboration (2015), "[understanding] is achieved through multiple, diverse investigations that provide converging support for a theoretical interpretation and rule out alternative explanations" (p.6). This implies that replication does not only refer to repetition of identical methodology (sometimes referred to as *direct replication*; Schmidt, 2009) but also allows introduction of differences in sampling, measurement, and experimental procedures, which actually increase the potential confirmatory power of the replication study (Schmidt, 2009).

In a broad sense, the findings presented in the current thesis are consistent with results of previous research suggesting that cognitive-behavioural sleep interventions may be effective in middle childhood. The literature review presented in Chapter 2 found three previous RCTs aimed directly at improving sleep for children with insomnia (i.e., Paine & Gradisar, 2011; Schlarb et al., 2011; Schlarb et al., 2016), with a further six RCTs aimed at improving sleep for children without a specific sleep disorder diagnosis (i.e., Donovan et al., 2016; Hart et al., 2016; Papaconstantinou et al., 2016; Quach et al., 2011; Tamura & Tanaka, 2014; Tamura & Tanaka, 2016), and five other studies (e.g., case reports; open trials; non-randomised controlled trials, Clementi & Alfano, 2014; Clementi et al., 2016; Fehr et al., 2016; Gruber et al., 2016; Meltzer & Booster, 2016). It is promising to note that all of the RCTs reported positive results for their intervention group relative to their control group, and that this is consistent with the findings presented in Chapter 4 of the current thesis.

For studies that reported changes in actual sleep parameters, consistent improvements have been observed for SOL (Chapter 4; Gruber et al., 2016; Paine & Gradisar, 2011; Schlarb et al., 2018; Tamura & Tanaka, 2016) and SE (Chapter 4; Gruber et al., 2016; Paine & Gradisar, 2011; Schlarb et al., 2018). Improvements in number or duration of night wakings have been reported in previous studies (Paine & Gradisar, 2011; Schlarb et al., 2018) but were not found in the present RCT (Chapter 4). Similarly, improvements in total sleep time (TST) or sleep period duration were reported by some (Gruber et al., 2016; Hart et al., 2016; Tamura & Tanaka, 2014; Tamura & Tanaka, 2016) but not all studies (Chapter 4; Clementi & Alfano, 2014; Paine & Gradisar, 2011; Papaconstantinou et al., 2018; Schlarb et al., 2018). Considering the clinical significance of these improvements, the magnitude of change observed in the present RCT was similar to or greater than the magnitude of change reported in previous studies (e.g., SOL improvement 20-30mins [previous studies 2-30mins]; SE improvement 4-6% [previous studies 2-7%]).

6.2.2 Extension of Knowledge

As described in Chapters 1 and 2, interventions for middle childhood insomnia have been relatively under-studied, compared to other age groups (e.g., young children, 0-5yrs). Instead, management of insomnia in middle childhood has been largely based on research and practice guidelines for other age groups (e.g., young children, adults). Therefore, it is important to compare the findings of the present research to the results of similar research conducted in early childhood (0-5yrs) and adulthood.

6.2.2.1 Sleep Restriction Therapies in Early Childhood (0-5yrs) vs Middle Childhood (6-14yrs).

Previous studies evaluating sleep restriction therapies (e.g., bedtime fading) as stand-alone interventions for typically-developing young children (0-5yrs) have reported improvements in both SOL and wake after sleep onset [WASO] following brief (1-2 week) interventions (Cooney et al., 2018; Gradisar et al., 2016). The improvements in SOL are consistent with those observed in the present study (Chapter 4). While we did not observe significant improvement in WASO, this is likely because WASO was already low prior to treatment for our sample (Chapter 4).

6.2.2.2 Sleep Restriction Therapies in Adulthood vs Middle Childhood.

Adults undergoing sleep restriction therapy for the treatment of insomnia typically report increased daytime sleepiness during treatment (Kyle et al., 2011; Kyle et al, 2014; Vallieres et al., 2013), as well as by improvements in SOL (Kyle et al, 2014; Miller et al., 2014; Vallieres et al., 2013; Whittall et al., 2018), WASO (Kyle et al, 2014; Miller et al., 2014; Whittall et al., 2018), and SE (Kyle et al, 2014; Miller et al., 2014; Vallieres et al., 2013), and a subjective experience of improved sleep quality and attitude towards sleep (Kyle et al., 2011). The child participants in our RCT who undertook sleep restriction therapies similarly experienced improvements in SOL and SE, and parents reported subjective improvements in sleep (e.g., less "difficulty falling asleep", less "difficulty staying asleep", and less likelihood of describing their child's sleep as a "problem"; Chapter 4). There was no significant improvement in WASO, although this is likely because WASO was already low prior to treatment (Chapter 4). No increase in self-reported daytime sleepiness was observed for children who implemented sleep restriction therapies in the present study (Chapter 5), although there was an increase in *evening* sleepiness, consistent with the build-up of sleep pressure expected with a consistently later bedtime over consecutive nights during treatment (Chapter 4). It may be relevant that not all adult studies have found a significant increase in daytime sleepiness during treatment with a smaller magnitude of sleep restriction (i.e., total sleep time [TST] reduced by ~30 mins; Whittall et al., 2018), in contrast to other observations that TST reduction >1 hour during sleep restriction therapy does produce significant increases in daytime sleepiness (Kyle et al., 2014).

Sleep misperception is common among adults with insomnia (Carskadon, Dement, Mitler, Guilleminault, Zarcone, & Spiegel, 1976; Edinger & Krystal, 2003; Manconi et al., 2010). This is expressed as a tendency to underestimate sleep duration, with a discrepancy between objective and subjective sleep duration that may be as large as 50-80 mins for adults engaging in treatment for insomnia (Lund, Rybarczyk, Perrin, Leszczyszyn, & Stepanski, 2013; Morin, Colecchi, Stone, Sood, & Brink, 1999; Omvik, Pallesen, Havik, Kvale, & Nordhus, 2006). Considering that

polysomnography is not indicated for the routine assessment of insomnia (Littner et al., 2003), sleep restriction therapy protocol dictates that prescribed time-in-bed [TIB] during treatment is determined relative to baseline subjective TST (from a 7-day sleep diary; Glovinsky & Spielman, 1991; Spielman et al., 1987; Miller et al., 2014). Interestingly, though, this means that adults undergoing sleep restriction therapy may experience an acute reduction of objective TST, which in one study was found to be as large as 90 mins on average during the first week of treatment (Kyle et al., 2014). Sleep misperception is likely to operate differently in children, where sleep diaries are commonly completed by parents, or collaboratively by the parent and child together. In fact, research suggests that parents of children with insomnia may overestimate sleep duration by >90mins (compared to actigraphy; Dayyat, Spruyt, Molfese, & Gozal, 2011). While polysomnography was not available in the current study, there were only small discrepancies between subjective (sleep diary) and objective (wrist actigraphy) measures of TST (Chapter 4). Therefore, the reduction in objective TST during the acute phase of treatment for our child participants is likely to be much smaller than that reported in previous studies with adults. This may have important clinical implications for the implementation of sleep restriction therapies in middle childhood. That is, individual differences in sleep perception and/or accuracy of sleep diary reporting may mean that there is little difference between participants who schedule TIB to match TST (i.e., bedtime restriction therapy), and those who reduce it by a further 30mins (i.e., sleep restriction therapy). This is consistent with the findings of our RCT.

6.2.2.3 Sleep Restriction Therapies in Middle Childhood.

There has been a clear sequence of research progression, from Paine and Gradisar's (2011) original RCT, to the open trials described in Chapter 3 (Leahy & Gradisar, 2012b; Watherston & Gradisar, 2013), to the RCT presented in Chapter 4. A clear strength of this research progression is the use of comparable treatment manuals and research protocols across all of these studies, allowing direct comparison of results. For example, identical treatment manuals were used for sleep

restriction therapy and bedtime restriction therapy in the RCT and the two open trials (Chapter 3), and there was also consistency between sleep diaries and questionnaires (i.e., Spence Children's Anxiety Scale [SCAS; Spence, 1998], Pediatric Daytime Sleepiness Scale [PDSS; Drake et al., 2003]) used across all studies. This is advantageous, because the literature reviewed in Chapter 2 suggests that there has been a lot of variability in the treatment strategies and measures used in previous treatment studies for middle childhood sleep problems, making it more difficult to compare results across these studies.

Table 6.1 contains data that allows a direct comparison of changes in sleep variables reported by Paine and Gradisar (2011), Leahy and Gradisar (2012b), Watherston and Gradisar (2013) and the present RCT (Chapter 4). While identical treatment manuals were used for sleep restriction therapy and bedtime restriction therapy in the RCT (Chapter 4) and the two open trials (Chapter 3), the treatment protocol used by Paine and Gradisar (2011) was multi-component in nature and included additional information about sleep hygiene, cognitive restructuring, and graded exposure, which was not presented in the other studies. Thus, their treatment was longer (i.e., 6 sessions vs 2 sessions) and their post-treatment and follow-up data should be interpreted with this in mind.

As seen in Table 6.1, there are notable similarities in primary sleep outcome variables (i.e., SOL, SE) between single-component sleep restriction therapy (Chapter 3; Chapter 4), singlecomponent bedtime restriction therapy (Chapter 3; Chapter 4), and multi-component CBT-i (Paine & Gradisar, 2011). In addition, all of these 3 interventions look to be superior to a waiting list control group (Paine & Gradisar, 2011) and active control group (i.e., bedtime regularisation; Chapter 4). There was consistent improvement in SOL following bedtime restriction therapy, sleep restriction therapy, and multi-component CBT-i, with a magnitude of approx. 20-30mins. Similarly, there was also consistent improvement in SE following bedtime restriction therapy, sleep restriction therapy, and multi-component CBT-i, with a magnitude of approx. 4-6%.

Interestingly, in the present RCT (Chapter 4) both sleep restriction therapy and bedtime restriction therapy resulted in continued improvement in SOL and SE up to the 4-week follow-up, while Paine and Gradisar's (2011) participants reported no further improvement between posttreatment and follow-up (see Table 6.1). This may be explained by the shorter treatment duration in the present RCT, as participants were expected to continue to adjust their TIB following prescribed guidelines during the follow-up period. There were also differences between Paine and Gradisar's (2011) waitlist control group and the active control group in the present RCT (Chapter 4). In particular, there were small decreases (i.e., improvements) in SOL and WASO for the active control group, with small increases in the same variables for the waitlist control group. This suggests that bedtime regularisation may be better than "doing nothing". However, it is also possible that this reflects non-specific treatment effects (e.g., therapist attention), as the active control group reverted to baseline levels by the 4-week follow-up (when they were no longer having contact with their sleep therapist). A future RCT could include both an active control group (i.e., bedtime regularisation) and a waitlist control group to further delineate these effects. For example, it is possible that while the active control group did not experience statistically significant change compared to their own baseline (and compared to the performance of the sleep restriction and bedtime restriction groups), but perhaps their outcomes may be superior to a waitlist control group whose sleep may actually worsen over time.

Table 6.1

Comparison of changes in sleep variables observed by Paine and Gradisar (2011), Leahy and Gradisar
(2012b), Watherston and Gradisar (2013), and the present RCT

		Pre	Post	4wk FU	Pre-Post Change
SOL (mins)					
Paine & Gradisar (2011)	CBT-i	41.90^	11.19	13.98	-30.71
	WL control	41.90^	50.21	47.05	8.31
Leahy & Gradisar (2012b)	BRT	36.29	18.47	-	-17.82
Watherston & Gradisar (2013)	SRT	42.34	17.80	-	-24.54
Present RCT (2018)	SRT	42.76^	22.02	20.31	-20.74
	BRT	42.76^	11.60	6.63	-31.16
	Control	42.76^	37.32	42.18	-5.44
WASO (mins)					
Paine & Gradisar (2011)	CBT-i	14.02^	0.03	1.06	-13.99
	WL control	14.02^	18.54	20.86	4.52
Leahy & Gradisar (2012b)	BRT	22.21	7.95	-	-14.26
Watherston & Gradisar (2013)	SRT	14.67	3.21	-	-11.46
Present RCT (2018)	SRT	11.42^	5.80	3.69	-5.62
	BRT	11.42^	8.53	4.99	-2.89
	Control	11.42^	7.14	3.86	-4.28

		Pre	Post	4wk FU	Pre-Post Change
SE (%)					8
Paine & Gradisar (2011)	CBT-i	85.88^	92.39	91.91	6.51
	WL control	85.88^	88.10	86.80	2.22
Leahy & Gradisar (2012b)	BRT	89.78	93.96	-	4.18
Watherston & Gradisar (2013)	SRT	89.90	95.70	-	5.80
Present RCT (2018)	SRT	88.93^	93.16	94.54	4.23
	BRT	88.93^	95.09	97.80	6.16
	Control	88.93^	91.71	90.88	2.78
TST (hrs)					
Paine & Gradisar (2011)	CBT-i	9.35^	9.02	9.50	-0.33
	WL control	9.35^	9.36	9.64	0.01
Leahy & Gradisar (2012b)	BRT	9.29	9.19	-	-0.10
Watherston & Gradisar (2013)	SRT	9.37	8.93	-	-0.44
Present RCT (2018)	SRT	9.36^	8.49	9.12	-0.87
	BRT	9.36^	8.94	9.49	-0.42
	Control	9.36^	9.49	9.20	0.13

Note: Pre = pre-treatment; Post = post-treatment; 4wk FU = 4-week (or 1-month) follow-up; CBT-i = multi-component cognitive behavioural therapy for insomnia (including bedtime restriction therapy); WL = waitlist; BRT = bedtime restriction therapy; SRT = sleep restriction therapy; RCT = randomised controlled trial; SOL = sleep onset latency; WASO = wake after sleep onset; SE = sleep efficiency; TST = total sleep time. [^]For Paine & Gradisar (2011) (2 groups) and the present RCT (3 groups), raw mean values have been adjusted to equate groups on pre-treatment data and observe group changes over time.

While there are clear strengths of this research progression, it is not without its limitations. Replication by independent researchers is important to reduce the potential impact of experimenter bias and increase confidence that the findings of a particular study reflect knowledge that is independent of the circumstances in which it was gathered (e.g., time, place; Schmidt, 2009). While the use of the same measures of sleep and anxiety across these studies improves our ability to compare results, this could also increase the risk of measurement bias (i.e., if these measures do not accurately capture the intended constructs). Furthermore, drawing participant samples from the same clinic may increase the risk of selection bias (i.e., the study sample not being representative of the target population). Therefore, replication of these findings by independent researchers is an important direction for future research.

6.2.2.4 Do Sleep Restriction Therapies 'Dampen' Anxiety in Middle Childhood?

Table 6.2 contains data that allows a direct comparison of changes in SCAS total anxiety scores reported by Paine and Gradisar (2011), Leahy and Gradisar (2012b), Watherston and Gradisar (2013) and the present RCT (Chapter 4). Taken together, these results suggest that sleep restriction therapies are associated with decreases in anxiety (both total anxiety and separation anxiety). However, the control group in the present study also reported decreases in total and separation anxiety (Chapter 4) and thus this cannot be attributed solely to processes associated with sleep restriction therapies (e.g., sleepiness and anxiety being opponent processes).

Interestingly, previous research that has observed changes in SCAS (self-report) total scores following 10-weeks of cognitive behaviour therapy (CBT) for anxiety has also reported improvement in SCAS scores for a waitlist control group (March, Spence, & Donovan, 2009) or an active control group (Hudson, Rapee, et al., 2009). So why did these control groups improve, while there was no improvement in anxiety reported for Paine and Gradisar's (2011) waitlist control group? And why did some control groups improve more than others?

Paine & Gradisar (2011) reported no change in SCAS total anxiety for their waitlist control group, yet the active control group in the present RCT (Chapter 4) experienced a small but significant improvement in the same measure. This may suggest that simply having contact with a therapist or knowing that they were seeking treatment for their sleep problem helped to reduce self-reported anxiety symptoms (consistent with Hudson, Rapee, et al., 2009). As there is a strong

relationship between parent anxiety and child anxiety (Craske & Waters, 2005; Ginsburg et al., 1995; Ginsburg & Schlossberg, 2002), it is also possible that implementing a structured sleep plan under the supervision of a sleep therapist reduced parents' anxiety about their child's sleep and, consequently, child anxiety was also reduced. However, this cannot be confirmed because changes in parent anxiety during treatment have not been reported in any of these studies. This may be an interesting direction for future research.

Considering the two studies with waitlist control groups in Table 6.2, March and colleagues (2009) reported improvements in SCAS anxiety, while Paine & Gradisar (2011) found no significant changes. It is possible that the nature of the measures employed in these studies may explain the difference in findings. While both of these studies used the SCAS as a measure of anxiety symptoms, March and colleagues (2009) employed a comprehensive anxiety-focused assessment interview (the Anxiety Disorders Interview Schedule [ADIS]; Silverman & Albano, 1996), while Paine and Gradisar (2011) employed a comprehensive sleep-focused assessment interview (the Clinical Sleep History Interview; see Appendix C.4). Perhaps the act of reflecting on their anxiety symptoms during the assessment interview resulted in some improvement in child participants' perception of their anxiety, and this was reflected in "post-treatment" SCAS scores. Similarly, the two studies with active control groups revealed that the group completing an anxietyfocused assessment interview (the ADIS; Silverman & Albano, 1996) reported greater improvement in anxiety during "treatment" (Hudson, Rapee, et al., 2009) compared to those completing a sleepfocused assessment interview (the Clinical Sleep History Interview; Chapter 4). For these latter two studies, it is also noteworthy that a greater magnitude of improvement in anxiety was reported with a longer active control intervention (i.e., 10wks [Hudson, Rapee, et al., 2009] vs 2wks [Chapter 4]). This suggests that a longer duration of therapist contact may produce greater non-specific treatment effects.

Table 6.2

Comparison of changes in Spence Children's Anxiety Scale total scores reported in previous studies and the present RCT

Authors		Pre	Post	Pre-Post Change	Treatment Duration
Paine & Gradisar (2011)	CBT-i	35.54^	25.36	-10.18	8wks
	WL control	35.54^	34.73	-0.81	8wks
Leahy & Gradisar (2012b)	BRT	35.33	22.33	-13.00	2wks
Watherston & Gradisar (2013)	SRT	35.82	24.82	-11.00	2wks
Present RCT (2018)	SRT	29.10	23.00	-5.65	2wks
	BRT	28.70	23.94	-4.76	2wks
	Active control	37.45	31.52	-5.93	2wks
Hudson, Rapee, et al. (2009)	CBT-A	35.55	23.43	-12.12	10wks
	Active control	34.48	21.81	-12.67	10wks
March et al. (2009)	CBT-A	40.00	27.36	-12.64	10wks
	WL control	38.56	29.72	-8.84	10wks

Note: Pre = pre-treatment; Post = post-treatment; CBT-i = multi-component cognitive behavioural therapy for insomnia (including bedtime restriction therapy); CBT-A = multi-component cognitive behavioural therapy for anxiety; WL = waitlist; BRT = bedtime restriction therapy; SRT = sleep restriction therapy, RCT = randomised controlled trial. [^] For Paine & Gradisar (2 groups), raw mean values were adjusted to equate groups on pre-treatment data and observe group changes over time.

6.2.2.5 Potential Contraindications for Sleep Restriction Therapies in Middle Childhood.

The present thesis provides empirical evidence relating to potential contraindications for sleep restriction therapies in middle childhood (Chapter 5). This research found no significant changes in daytime sleepiness, various cognitive performance abilities, classroom attention, or occurrence of parasomnias (disorders of arousal) during treatment. These results are inconsistent with previous studies that have observed significant changes in cognitive performance during

experimental sleep restriction (Sadeh et al., 2003; Vriend et al., 2013) despite a similar magnitude of reduction in TST (~30-45 mins per night on average).

One potential explanation for this discrepancy is that the participants in previous studies were children without sleep disorders, whereas the participants in the present study were children with Chronic Insomnia Disorder. It is possible that children with sleep disorders (i.e., like the participants in the present study) were already experiencing cognitive performance impairments as a result of their long-standing sleep difficulties, and thus the effect of short-term (i.e., 2 weeks) restriction of sleep opportunity during treatment did not confer any additional impairment to their performance. These differential effects of short-term therapeutic sleep restriction on cognitive performance between good sleepers and children with insomnia should be investigated in future research.

6.3 Theoretical Implications of Thesis Findings

6.3.1 Aetiology and Maintenance of Chronic Insomnia Disorder in Middle Childhood

According to Carr (2006), the development of any psychological disorder in childhood is influenced by a combination of risk factors (predisposing children to developing psychological difficuties), precipitating factors (triggering the onset of problems), maintaining factors (perpetuating the problems once they have developed), and protective factors (having implications for prognosis and treatment outcome). Bringing together findings of previous research (described in Chapters 1 and 2), along with insight gained from two recent open trials (Chapter 3) and an RCT (Chapters 4 and 5), the present thesis suggests that a complex array of biological, environmental, cognitive, and behavioural factors may contribute to the aetiology and maintenance of Chronic Insomnia Disorder in middle childhood. This information is collated in Figure 6.1.

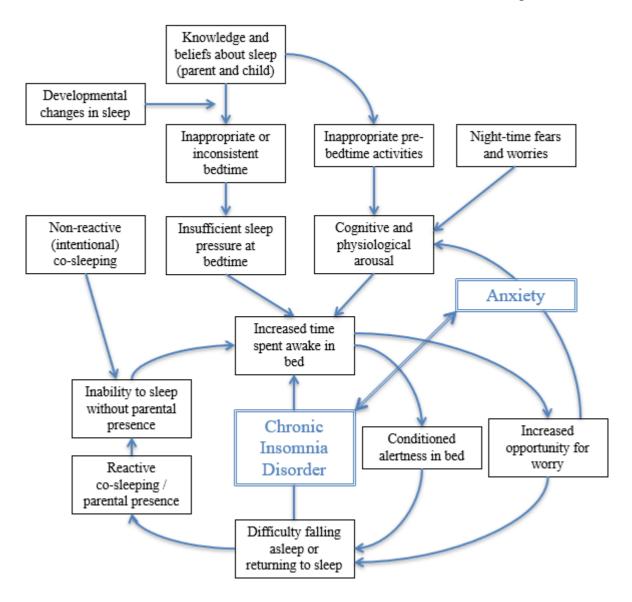


Figure 6.1 A cognitive-behavioural model for the aetiology and maintenance of Chronic Insomnia Disorder in middle childhood, including proposed links with anxiety. Image developed by author.

As outlined in Figure 6.1, parent and child knowledge and beliefs about sleep are critical in determining sleep behaviour (McDowall et al., 2017; Owens & Jones, 2011; Owens et al., 2011), but parental knowledge about child sleep is generally poor (McDowall et al., 2017). Along with developmental changes that result in a gradual decreasing need for sleep during the middle childhood years (Price et al., 2014), this may result in inappropriate or inconsistent bedtimes for children, which may result in increased time spent awake in bed due to insufficient opportunity to build up sleep pressure during the waking hours. Alternatively (or simultaneously), inappropriate

pre-bedtime activities and/or night-time fears and worries (which are common in middle childhood; Gordon et al., 2007; Muris et al., 2001) may result in increased cognitive and physiological arousal prior to bedtime, which also result in increased time spent awake in bed as sleep and arousal are opponent processes (Dahl, 1996; Dahl & Harvey, 2007).

Over time, repeated experiences of prolonged wakefulness in bed may result in the development of an association between the bed and alertness (rather than the bed and sleep) via principles of classical conditioning (Pavlov, 1906, 1927). In addition, increased time spent awake in bed provides an opportunity for worry while awake in bed (Dahl & Harvey, 2007; McMakin & Alfano, 2015), which further compounds the child's difficulty falling or staying asleep and may also provide a key link between sleep problems and anxiety symptoms.

Parent-child co-sleeping (i.e., sharing a bed or bedroom) can be either reactive (i.e., occurring as a means of coping with a child's sleep difficulties) or non-reactive (i.e., intentional, occurring for personal or cultural reasons unrelated to sleep problems; Madansky & Edelbrock, 1990). Either way, principles of classical conditioning (Pavlov, 1906, 1927) suggest that the repeated experience of co-sleeping (for either part or whole night) may result in an inability to fall asleep or return to sleep without an important stimulus present – the parent(s). As children grow older, parents may be less willing to engage in co-sleeping (e.g., due to parental beliefs about child independence) and children may themselves wish to be able to fall asleep without parental presence (e.g., to attend school camps and sleepovers), but they may find that the removal of parental presence while attempting to fall asleep results in increased time spent awake in bed, and thus contributes to the cycle of Chronic Insomnia Disorder.

While it is acknowledged that correlation does not infer causation, the findings presented in the present thesis support the proposed model insofar as associations between some of these factors¹⁰ were found in the current sample of school-aged children. Prior to commencement of

¹⁰ Not all of these factors were measured, so as to not overburden research participants.

treatment, all participants reported either difficulty falling asleep or difficulty staying asleep (or both), and all reported excessive time spent awake in bed (usually prior to sleep onset, as WASO was generally low in the present sample; Chapter 4). At baseline, 72% of parents reported that their child required parental presence to either fall asleep or return to sleep after waking at baseline (Chapter 4). While the distinction between reactive and non-reactive co-sleeping was not made in the assessment interview (see Appendix C.4), it is the clinical impression of the therapists involved that reactive co-sleeping was more prevalent in this sample. Similarly, knowledge and beliefs about sleep were not formally assessed; however, anecdotal reports from parents of participants reflected deficits in knowledge relevant to their child's sleep (e.g., *"I thought he needed more sleep than he does."*) Additional research is clearly needed to further investigate the proposed relationships between these variables.

The data presented in Chapter 4 provides conflicting information regarding the relationship between worry and time spent awake in bed. While there was no significant correlation between changes in wakefulness in bed (i.e., SE) and changes in worry from pre- to post-treatment, and all three groups reported unexpected improvement in worry, there was nonetheless a greater reduction in worry reported by the sleep restriction group compared to the other two groups (i.e., bedtime restriction therapy, bedtime regularisation). Thus, this component of the proposed model also requires further investigation.

As seen in Figure 6.1, it is proposed that excessive time spent awake in bed is central to the aetiology and maintenance of Chronic Insomnia Disorder in middle childhood. Considering that cognitive-behavioural interventions aim to help individuals to modify key factors involved in the maintenance of their difficulties (Carr, 2006), it follows that sleep restriction therapy (which aims to reduce TIB and thus time spent awake in bed) may be an appropriate treatment for Chronic Insomnia Disorder in middle childhood. However, no previously-published studies have evaluated sleep restriction therapy as a single-component intervention in this population.

6.3.2 Potential Mechanisms for Improvements in Sleep and Anxiety following Sleep Restriction Therapy

Based on the model of aetiology and maintenance presented in Figure 6.1, a graphical representation of potential mechanisms involved in changes to sleep and anxiety following sleep restriction therapy is presented in Figure 6.2.

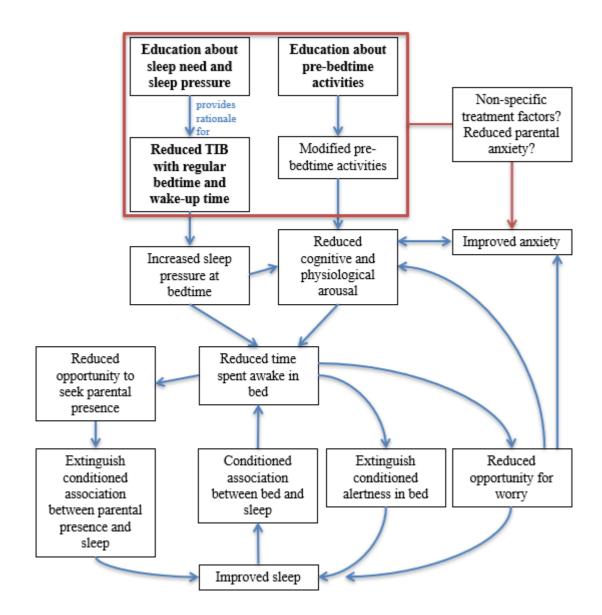


Figure 6.2 Potential mechanisms involved in improvements to sleep and anxiety following sleep restriction therapy.

Image developed by author.

Chapter 6: Discussion

6.3.2.1 How Does Sleep Restriction Therapy Improve Sleep?

The protocol for sleep restriction therapy and bedtime restriction therapy used in the present study involved providing education about sleep need and sleep pressure as rationale for reducing TIB during treatment (consistent with sleep restriction therapy protocol for adults: Vallieres et al., 2013; Whittall et al., 2018; see treatment manuals for the present RCT in Appendix C.5). A discussion of pre-bedtime activities was also included, to help the family learn to manage increased time available in the evening before the child's new scheduled bedtime, and to ensure that they stay awake until the scheduled bedtime (Glovinsky & Spielman, 1991). These components of treatment aimed to directly target factors involved in the aetiology and maintenance of Chronic Insomnia Disorder in middle childhood (i.e., poor knowledge about sleep, inappropriate or inconsistent bedtime, inappropriate pre-bedtime activities), as outlined in Figure 6.1.

According to Borbély (1982), the homeostatic sleep drive causes a gradual build-up of sleep pressure during wakefulness, which is slowly dissipated during sleep. Sleep loss results in an increase of sleep pressure the following day, which continues to build during subsequent days if insufficient sleep opportunity is provided on subsequent nights (Borbély, 1982). Therefore, reduction of TIB (and consequently TST) during sleep restriction therapy allows the individual to build up greater homeostatic sleep pressure during the acute phase of treatment (Spielman et al., 1987). This increase in sleep pressure can be measured subjectively via an increased feeling of sleepiness prior to bedtime, and objectively via decreased sleep onset latency (Taylor et al., 2005), both of which have been reported by adults following sleep restriction therapy (Kyle et al., 2011; Kyle et al., 2014; Miller et al., 2014; Vallieres et al., 2013). The results of the RCT are consistent with this theory, because there was a significant increase in evening sleepiness and a significant decrease in SOL for the two restriction groups during the acute phase of treatment, with no change for the control group (who had no significant change in TIB during treatment; Chapter 4).

In the present RCT, sleep restriction therapies resulted in significant improvements in wakefulness in bed (i.e., \downarrow SOL, \uparrow SE) and parental perception of the child's difficulty falling asleep

or staying asleep (Chapter 4). Interestingly, though, there was less change reported in children's desire for parental presence, and this was the most common reason for seeking further treatment after the 4-week follow-up period. It was hypothesised that increasing evening sleepiness (i.e., sleep pressure) during treatment would 'dampen' pre-sleep anxiety and therefore reduce children's fear of sleeping alone. In cases where some anxiety remained, increasing evening sleepiness would result in shorter SOL and therefore parental presence would be required less prior to sleep onset. Furthermore, improved sleep consolidation (i.e., reduced WASO) during treatment should result in less opportunity to seek out parental presence during the night. Over time, we expected that these changes would allow extinction of the conditioned association between parental presence and sleep. However, this prediction was not supported, and over 60% of children still required parental presence to fall asleep or return to sleep following sleep restriction therapy - even though other aspects of their sleep had improved. While Paine and Gradisar (2011) did not report changes in the prevalence of these behaviours, they did report significant improvement in sleep onset associations (e.g., parental presence), suggesting that multi-component treatment (including cognitive therapy and graded exposure to directly target fear of sleeping alone) may be required for children who require parental presence to fall asleep or return to sleep.

6.3.2.2 How Could Sleep Restriction Therapy Contribute to Improvements in Anxiety?

The theoretical link between sleep restriction therapy and improvements in anxiety is the idea that sleep and arousal are opponent processes (Dahl, 1996; Dahl & Harvey, 2007). This suggests that increased evening sleepiness (i.e., sleep pressure) during sleep restriction therapy may act to "dampen" pre-sleep arousal (Dahl & Harvey, 2007). Reduced time spent awake in bed also results in reduced opportunity for worry to occur (McMakin & Alfano, 2015), and together these improvements in cognitive arousal (e.g., worry) and physiological arousal may result in overall improvements in anxiety.

Our results were unable to confirm this prediction because the control group also experienced unexpected improvements in both anxiety and worry (Chapter 4). This suggests that non-specific treatment effects (e.g., therapist attention) may also influence anxiety in children with Chronic Insomnia Disorder. This possibility is consistent with the lack of improvement observed by Paine and Gradisar (2011) for their waiting list control group, who did not have contact with a therapist. Worth noting though is that treatment studies of children with anxiety disorders have found improvements for both active control groups and waiting list control groups over a 10-week treatment period (Hudson, Rapee, et al., 2009; March et al., 2009). Furthermore, there are strong links between parent anxiety and child anxiety (Craske & Waters, 2005; Ginsburg et al., 1995; Ginsburg & Schlossberg, 2002); therefore, implementing a structured sleep plan under the supervision of a therapist may have reduced control parents' anxiety about their child's sleep and, consequently, control child anxiety was also reduced. However, this cannot be confirmed because parent anxiety was not measured at post-treatment in the present study.

6.4 Clinical Implications of Thesis Findings

The findings of the present thesis have important clinical implications for the treatment of insomnia in middle childhood. In broad terms, the literature review presented in Chapter 2 supports the use of cognitive-behavioural interventions for children experiencing insomnia and other sleep problems (e.g., short sleep duration without a diagnosed sleep disorder), including children with other comorbid psychological or medical conditions (e.g., anxiety, asthma, eczema). More specifically, the results of the present thesis' RCT (Chapter 4) provide insight for clinicians attempting to choose between three brief behavioural interventions for school-aged children with insomnia, namely sleep restriction therapy, bedtime restriction therapy, and bedtime regularisation.

The results of the RCT suggest that clinicians can be more confident in using both sleep restriction therapy and bedtime restriction therapy to improve sleep for children with Chronic Insomnia Disorder (Chapter 4). While clinicians should always be mindful of comorbid conditions for the individual child, our results further suggest that these interventions may not exacerbate parasomnias (disorders of arousal). Clinicians may also reassure families that these techniques have not been associated with increases in *daytime* sleepiness or deficits in cognitive performance or classroom attention, despite the common experience of increased *evening* sleepiness during treatment. Considering that there was little difference in changes in objective TST during treatment for the sleep restriction and bedtime restriction groups in the RCT, this suggests that the two different sets of instructions (i.e., reduce TST to match baseline TIB, or reduce TST to 30mins less than baseline TIB) may be used interchangeably by clinicians.

The control group in the present RCT implemented bedtime regularisation, which is commonly recommended in primary health settings, or as a first step in specialist cognitivebehavioural sleep interventions (Durand, 2008; Meltzer & Crabtree, 2015). Recent research interest in the ill effects associated with sleep schedule variability (e.g., Biggs et al., 2011; Fuligni & Hardway, 2006; Kelly et al., 2013a; Kelly et al., 2013b; Lemola, Ledermann, & Friedmann, 2013; Manber, Bootzin, Acebo, & Carskadon, 1996; Pesonen et al., 2009) suggests that bedtime regularisation would be an intuitive technique to use. The findings of the present thesis suggest that bedtime regularisation was associated with small improvements in anxiety and daytime sleepiness, as well as improvements in parental perception of the child's sleep as a "problem"; however, bedtime regularisation alone was insufficient to result in clinically significant change in actual sleep parameters. It may be beneficial to educate primary health care providers (e.g., general practitioners) and specialist clinicians (e.g., sleep physicians; psychologists) that other brief interventions (i.e., sleep restriction therapies) are available and may be more effective than bedtime regularisation. However, in cases where families may be resistant to restricting their child's time in bed, bedtime regularisation may be a useful first step in treatment, as our results suggest that this may result in improvements in both child and parent perception of sleep (even in the absence of improvements in actual sleep parameters).

Chapter 6: Discussion

6.5 Limitations of Research

The RCT described in Chapter 4 used a short follow-up period of only 4 weeks, so as to not prevent children who required further treatment from accessing it. However, this means that conclusions cannot be drawn about the long-term efficacy of these brief interventions. Future studies should include a longer follow-up period (e.g., 6 months) to determine the long-term effects of sleep restriction therapies in middle childhood. Furthermore, as with previous studies (i.e., Paine & Gradisar, 2011), the implementation of our treatment study within an existing private clinic setting resulted in a participant sample of predominantly mid- to high-SES families without comorbid mood disorders (e.g., depression) or other psychiatric symptoms (e.g., suicidality, psychosis). This may limit the generalisability of our findings to lower-SES groups and children with more complex psychiatric presentations.

Measures of anxiety and worry used in the present study did not distinguish between anxiety experienced at bedtime and anxiety experienced throughout the day. In light of the finding that daytime sleepiness did not change during sleep restriction therapies (Chapter 5), but evening sleepiness did significantly increase (Chapter 4), it is plausible that the dampening effect of sleepiness on arousal occurs primarily during the pre-sleep period. Therefore, future research using sleep restriction therapy in middle childhood should examine specific changes in *pre-sleep* arousal. For example, a child version of the adult Pre-Sleep Arousal Scale (Nicassio, Mendlowitz, Fussell, & Petras, 1985) has been used successfully in previous studies of children with sleep difficulties (Gregory et al., 2008) and anxiety (Alfano et al., 2010; Clementi et al., 2016). The Pre-Sleep Arousal Scale measures both cognitive and physiological arousal, allowing future research to differentiate between these two components of arousal and their potential relationships with evening sleepiness.

The findings of the RCT described in Chapter 4 suggest that changes in parent anxiety during treatment may have contributed to the observed changes in child anxiety for all 3 treatment groups. In particular, it is plausible that implementing a structured sleep plan under the supervision

of a trained therapist reduced parental anxiety about their child's sleep. However, this cannot be confirmed because parent anxiety was not measured at post-treatment. Future studies may benefit from measuring changes in parent anxiety following sleep restriction therapy, to further explore this effect.

A final limitation of the RCT research design is the failure to include a waitlist control group. This study was designed with an active control group (i.e., bedtime regularisation) in order to control for non-specific treatment effects (e.g., therapist attention). However, all 3 groups reported improvements in anxiety and (to a lesser extent) parental perceptions of sleep, which may have reflected a demand effect. In order to reduce demand effects on self-report measures of anxiety and worry there was no explicit mention of anxiety at all during any of the treatment programs. It is possible, though, that children still expected that their anxiety *should* be decreasing and thus they under-reported their symptoms after treatment. Similarly, it is possible that parents expected that their child's sleep should be improving and this was reflected in their responses to the follow-up interview, despite little change in actual sleep parameters. Future studies should consider including a waitlist control group, although this must be weighed against the reduced statistical power (or larger participant sample) associated with the inclusion of an additional group.

6.6 Directions for Future Research

6.6.1 Replication

As mentioned earlier, recent trends in the field of psychology suggest that the best way to build knowledge is with "multiple, diverse investigations that provide converging support for a theoretical interpretation and rule out alternative explanations" (Open Science Collaboration, 2015, p.6). Therefore, replication of research findings is essential (Moonesinghe et al., 2007; Open Science Collaboration, 2015; Schmidt, 2009) as this increases our confidence that the findings of a particular study reflect knowledge that is independent of the circumstances in which it was gathered (e.g., time, place; Schmidt, 2009). To the authors' knowledge, the current thesis provides the first systematic evaluation of sleep restriction therapies for middle childhood insomnia; thus, future replication studies will be required to confirm the effects described.

In addition to future replication studies, the results of the present thesis suggest a number of potential avenues for future research investigating sleep restriction therapy in middle childhood. Particular areas of interest are: (1) investigating developmental changes in the effectiveness of sleep restriction therapy, (2) whether sleep restriction therapy may be a useful adjunct to CBT for anxiety, and (3) how rapid eye movement (REM) sleep fragmentation may contribute to the link between sleep and anxiety. Each of these areas is described further below.

6.6.2 Developmental Changes in Sleep Restriction Therapy

Considerable developmental changes occur during the middle childhood years, including domains such as physical growth, cognitive development, social skills, and independence (Eccles, 1999). In the field of sleep medicine, the middle childhood years have traditionally been viewed as a cohesive group, nestled between early childhood (0-5yrs) and adolescence (the onset of puberty to the onset of adulthood). Recent studies suggest that there are sufficient differences in sleep patterns and processes within the middle childhood years that they should not be considered a homogenous group (Campbell, Burright, Kraus, Grimm, & Feinberg, 2017; Gruber, Somerville, Wells, Keskinel, & Santisteban, 2018). Splitting the middle childhood years into three discrete groups (i.e., 6-7yrs, 8-9yrs, 10-11yrs), Gruber and colleagues (2018) found a progressive delay in sleep onset time, shortening of sleep duration, and increasing discrepancy between weekday and weekend wake-up time with increasing age. Also using cross-sectional data, Campbell and colleagues (2017) found that objectively-measured daytime sleepiness (measured in the afternoon) increased with age (10-14yrs) and, while increasing TIB decreased daytime sleepiness, this effect was greater for the younger children within this age group (i.e., younger children showed greater benefit from sleep extension). However, previous research grouping participants by developmental stage (rather than chronological age) has found that pubertally-mature adolescents (approx. 14yrs) accumulate sleep pressure at a significantly slower rate, compared to pre-pubertal children (approx. 11yrs; Jenni,

Achermann, & Carskadon, 2005; Taylor et al., 2005). These changes in sleep patterns and processes during the middle childhood years may be relevant to the mechanisms involved in sleep restriction therapy.

Future research should examine the effectiveness of sleep restriction therapy for children at different ages within the middle childhood years (as per Gruber et al., 2018) or at different stages of pubertal development (i.e., using guidelines developed by Tanner, 1962). This study should also include an objective measure of evening sleepiness (i.e., multiple sleep latency test; Carskadon, 1986) before and after sleep restriction therapy to allow (1) objective examination of changes in sleep pressure accumulation during treatment, and (2) whether this changes with age or developmental stage. This research would contribute to our knowledge of the effectiveness of sleep restriction therapy across the middle childhood years.

6.6.3 Sleep Restriction Therapy as an Adjunct to CBT for Anxiety

As described in Chapter 1, clinically-significant sleep difficulties occur in up to 90% of children with anxiety disorders (Alfano, Ginsburg, & Kingery, 2007; Alfano et al., 2010). Multicomponent CBT is the treatment-of-choice for anxiety in middle childhood (In-Albon & Schneider, 2007; Reynolds, Wilson, Austin, & Hooper, 2012) and recent studies suggest that CBT for anxiety may also improve sleep (Caporino et al., 2017; Clementi et al, 2016; Donovan et al., 2017; Peterman et al., 2016; Storch et al., 2008). These treatment programs usually involve 8-17 sessions (Rapee et al., 2009), and include components such as psychoeducation, relaxation training, cognitive restructuring, and graded exposure (Rapee et al., 2009). Considering the observed improvements for both sleep and anxiety reported in the present thesis following brief (2 session) sleep restriction or bedtime restriction therapy for children with anxiety *symptoms*, it would be interesting to see whether these results hold for children with a diagnosed anxiety disorder. And, if so, whether sleep restriction therapy may be a useful adjunct to CBT for anxiety.

With a similar hypothesis, Clementi and Alfano (2014) conducted a pilot study (N=4; 7-12 yrs) that included two sessions of CBT for insomnia (e.g., sleep hygiene education, regular sleep

scheduling, improving pre-bedtime activities, and/or graduated extinction for children requiring parental presence to fall asleep) as part of a larger treatment program for children with generalised anxiety disorder (total 14 sessions). Improvements were seen in child-reported (but not parentreported) sleep and worry, and no child met diagnostic criteria for generalised anxiety disorder at 3month follow-up. Of course, the observed improvements in anxiety are not unexpected considering that 12 of the 14 sessions were directly focused on anxiety. While the sample size was small, Clementi and Alfano (2014) concluded that "the brief format of the sleep intervention used (2 sessions) was inadequate to produce lasting changes in sleep" and "the sleep difficulties experienced by some children with [generalised anxiety disorder] may require more intensive intervention" (p221). However, Clementi and Alfano (2014) did not use sleep restriction or bedtime restriction therapy; instead, components of their intervention were strikingly similar to the control group in our present study (i.e., regular sleep scheduling and improving pre-bedtime activities) who experienced no significant improvements in sleep parameters (Chapter 4).

A future RCT should examine whether sleep restriction therapy improves outcomes (relative to treatment-as-usual) as an adjunct to CBT for anxiety in middle childhood. This would involve comparing three groups: (1) 2-week waiting period + treatment-as-usual (an established program of CBT for anxiety; e.g., *Coping Koala*, Barrett, Dadds, & Rapee, 1996; *Coping Cat*, Kendall, 1990; *Cool Kids*, Lyneham, Abbott, Wignall, & Rapee, 2003; *BRAVE*, Spence, Holmes, March, & Lipp, 2006), (2) sleep restriction therapy + treatment-as-usual, and (3) bedtime regularisation + treatment-as-usual. This third group is important because the control group in the present study reported improvements in anxiety, despite relatively no change in sleep parameters (Chapter 4), and would allow comparison with the similar study by Clementi and Alfano (2014). This future study would provide more information about whether sleep restriction therapy truly is a useful transdiagnostic intervention for sleep problems and anxiety in middle childhood.

6.6.4 Does REM Sleep Fragmentation Contribute to the Link Between Sleep and Anxiety?

For adults with insomnia, research suggests that shorter REM sleep duration and greater REM sleep fragmentation (i.e., micro- and macro-arousals during REM sleep) contribute to the subjective perception of nocturnal wakefulness and, as a result, to the poor subjective sleep quality reported by people experiencing insomnia (Feige et al., 2008). Research investigating the relationship between sleep and emotion has found that REM sleep strengthens conditioned fear responses and improves discrimination between threatening and non-threatening stimuli after a period of sleep, as well as facilitating the subsequent extinction of these conditioned fear responses (Goldstein & Walker, 2014; Menz et al., 2013). In contrast, sleep deprivation impairs these processes, resulting in impairments to the generation and maintenance of fear responses in dangerous situations, and the inhibition of fear responses in the presence of safety cues (Goldstein & Walker, 2014).

According to Riemann and colleagues (2012), improvements in subjective sleep quality following cognitive-behavioural sleep interventions may be associated with reductions in microand macro-arousals from REM sleep following treatment, although this is on the verge of being empirically tested. Similarly, it is possible that reduced REM sleep fragmentation following treatment may improve discrimination between stimuli associated with threat vs safety, and facilitate the extinction of conditioned fear responses, thus resulting in improvements in anxiety.

It is not yet known whether REM sleep fragmentation also occurs in middle childhood insomnia, and if this changes following cognitive-behavioural treatment. Future research using sleep restriction therapy in middle childhood should use polysomnography as an objective measure of sleep (before and after treatment), in addition to subjective measures of sleep and anxiety. This would allow for examination of (1) the presence of REM sleep fragmentation in middle childhood insomnia, (2) changes in REM sleep fragmentation during sleep restriction therapy, and (3) potential relationships between changes in subjective sleep, REM sleep fragmentation, and anxiety.

Chapter 6: Discussion

6.7 Conclusions

Sleep restriction therapies are efficacious, brief interventions that have the potential to improve the sleep of school-aged children with Chronic Insomnia Disorder, without notable negative effects on daytime sleepiness, cognitive performance, classroom attention, or parasomnias. Sleep restriction therapies may also improve anxiety (even without directly addressing anxiety during treatment), although further research is needed to elucidate the mechanisms involved.

Bedtime regularisation does not appear to be sufficient to improve sleep in school-aged children with Chronic Insomnia Disorder, although it may improve parental perception of child sleep difficulties and confer benefits for daytime sleepiness and anxiety. Therefore, it may be a useful first step for families who are resistant to restricting their child's sleep opportunity via reducing their time in bed.

Overall, the findings of the present thesis support the use of cognitive-behavioural interventions for Chronic Insomnia Disorder in typically-developing school-aged children.

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Appendices

Appendix A: Diagnostic Criteria

A.1 Diagnostic criteria for Chronic Insomnia Disorder according to the International Classification of Sleep Disorders (3rd Edition; AASM, 2014)

- A. The patient reports, or the patient's parent or caregiver observes, one or more of the following:
 - 1. Difficulty initiating sleep.
 - 2. Difficulty maintaining sleep.
 - 3. Waking up earlier than desired.
 - 4. Resistance to going to bed on appropriate schedule.
 - 5. Difficulty sleeping without parent or caregiver intervention.
- B. The patient reports, or the patient's parent or caregiver observes, one or more of the following related to the nighttime sleep difficulty:
 - 1. Fatigue/malaise.
 - 2. Attention, concentration, or memory impairment.
 - 3. Impaired social, family, occupational, or academic performance.
 - 4. Mood disturbance/irritability.
 - 5. Daytime sleepiness.
 - 6. Behavioural problems (e.g., hyperactivity, impulsivity, aggression).
 - 7. Reduced motivation/energy/initiative.
 - 8. Proneness for errors/accidents.
 - 9. Concerns about or dissatisfaction with sleep.
- C. The reported sleep/wake complaints cannot be explained purely by inadequate opportunity (i.e., enough time is allotted for sleep) or inadequate circumstances (i.e., the environment is safe, dark, quiet, and comfortable) for sleep.
- D. The sleep disturbance and associated daytime symptoms occur at least three times per week.
- E. The sleep disturbance and associated daytime symptoms have been present for at least three months.
- F. The sleep/wake difficulty is not better explained by another sleep disorder.

A.2 Diagnostic criteria for Behavioural Insomnia of Childhood according to the International Classification of Sleep Disorders (2nd Edition; AASM, 2005)

- A. A child's symptoms meet the criteria for insomnia based upon reports of parents or other adult caregivers.
- B. The child shows a pattern consistent with either the sleep-onset association or limit-setting type of insomnia described below.
 - i. Sleep-onset association type includes each of the following:
 - 1. Falling asleep is an extended process that requires special conditions.
 - 2. Sleep-onset associations are highly problematic or demanding.
 - 3. In the absence of the associated conditions, sleep onset is significantly delayed or sleep is otherwise disrupted.
 - 4. Nighttime awakenings require caregiver intervention for the child to return to sleep.
 - ii. Limit-setting type includes each of the following:
 - 1. The individual has difficulty initiating or maintaining sleep.
 - 2. The individual stalls or refuses to go to bed at an appropriate time or refuses to return to bed following a nighttime awakening.
 - 3. The caregiver demonstrates insufficient or inappropriate limit setting to establish appropriate sleeping behaviour in the child.
- C. The sleep disturbance is not better explained by another sleep disorder, medical or neurological disorder, mental disorder, or medication use.

Appendix B: Supplementary Data for Chapter 3

Appendices

Table B.1

Descriptive statistics and effect sizes for sleep and anxiety measures from pre- to post-treatment (Leahy & Gradisar, 2012b)

	H	Pre	Ν	/lid	Pre-	Р	ost	Pre-
					Mid			Post
	М	SD	М	SD	d	М	SD	d
Sleep								
TIB (hrs)	10.36	1.18	9.73	1.02	.57	9.77	0.82	.59
SOL (min)	36.29	20.02	27.50	25.26	.39	18.47	14.18	1.04
WASO (min)	22.21	19.14	7.00	6.15	1.20	7.95	11.79	.92
SE (%)	89.78	5.17	92.97	4.90	63	93.96	5.67	77
TST (hrs)	9.29	1.09	9.04	1.03	.23	9.19	1.01	.10
Daytime Sleepin	ess							
PDSS	13.33	5.98	12.56	4.67	.14	12.89	4.57	.08
Anxiety								
SCAStotal	35.33	14.76	24.33	6.74	1.02	22.33	7.94	1.15
SAD	8.00	4.52	6.12	6.25	.35	5.50	2.88	.68
SDfear	3.05	0.73	2.46	0.81	.77	2.72	0.75	.45

Note: Cohen's d effect size magnitude: .20 = small, .50 = moderate, .80 = large; Pre = pre-treatment; Mid = mid-treatment; Post = post-treatment; SOL = sleep onset latency; WASO = wake after sleep onset; TST = total sleep time; TIB = time in bed; SE = sleep efficiency; PDSS = Pediatric Daytime Sleepiness Scale total score; SCAStotal = Spence Children's Anxiety Scale total score; SAD = SCAS separation anxiety subscale; SDfear = sleep diary, fear of sleeping alone.

Appendices

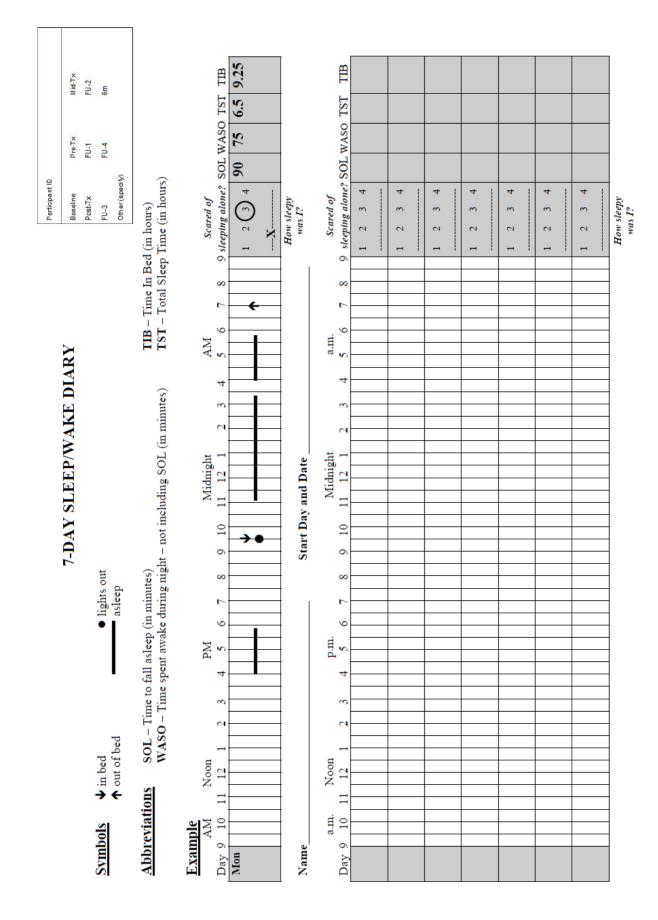
Table B.2

Descriptive statistics and effect sizes for sleep and anxiety measures from pre- to post-treatment (Watherston & Gradisar, 2013)

		Pre	N	lid	Pre-	Pe	ost	Pre-
					Mid			Post
	М	SD	М	SD	d	М	SD	d
Sleep								
TIB (hrs)	10.42	0.43	9.10	0.78	2.63	9.32	0.81	1.63
SOL (min	42.34	23.88	22.36	15.85	.87	17.80	15.22	1.25
WASO (min)	14.67	20.64	4.88	4.05	.49	3.21	5.16	.53
SE (%)	89.90	4.23	95.00	2.92	-1.25	95.70	4.17	-1.19
TST (hrs)	9.37	0.63	8.65	0.86	1.70	8.93	0.93	1.07
Anxiety								
SCAStotal	35.82	14.47	26.73	15.21	.69	24.82	18.41	.69
SAD	8.45	3.53	7.00	2.97	.40	6.73	4.13	.61
SDfear	2.91	0.95	2.13	0.84	.95	1.99	0.80	1.00
Process Variables								
WorryTotal	22.73	10.91	13.00	9.02	1.38	11.91	10.97	1.03
WorryQ10	2.40	1.78	2.10	1.60	.18	1.10	1.45	.85
SDsleepiness	2.02	0.79	3.12	0.58	-1.33	3.17	0.49	-1.49

Note: Cohen's d effect size magnitude: .20 = small, .50 = moderate, .80 = large; Pre = pre-treatment; Mid = mid-treatment; Post = post-treatment; SOL = sleep onset latency; WASO = wake after sleep onset; TST = total sleep time; TIB = time in bed; SE = sleep efficiency; SCAStotal = Spence Children's Anxiety Scale total score; SAD = SCAS separation anxiety subscale; WorryTotal = Worry Scale for Children total score; WorryQ10 = "How much did you worry about personal harm?"; SDfear = sleep diary, fear of sleeping alone; SDsleepiness = sleep diary, evening sleepiness.

Appendix C: Supplementary Materials for Chapter 4



C.1 7-Day Sleep Diary

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INSTRUCTIONS FOR THE 7-DAY SLEEP/WAKE DIARY

Just before going to bed at night:

- 1. Place a 'down arrow' (\checkmark) at the time you go to bed
- 2. Place a just before you turn out your light

When you get up in the morning:

- Mark the time you actually got out of bed with an 'up arrow' (\uparrow).
- Circle "O" a number for how you felt the night before for this question: "I feel scared of sleeping on my own":

1	2	3	4
Not at all	A little bit	Moderately	Extremely

• Place a mark " \mathbf{X} " on the dotted line to show how sleepy you felt just before bed:

Not at all	A little bit	Moderately	Extremely

- Enter how long (minutes) it took you to fall asleep after turning out the light in the SOL column.
- Enter how long (minutes) you felt you were awake during the night after initially falling asleep and before getting out of bed in the WASO column.
- Enter how long you slept (hours) in the TST column.

C.2 Self-Report Questionnaires

Spence Children's Anxiety Scale

Please put a circle around the word that shows how often each of these things happen to you. There are no right or wrong answers.

1.	I worry about things	Never	Sometimes	Often	Always
2.	I am scared of the dark	Never	Sometimes	Often	Always
3.	When I have a problem, I get a funny feeling in my stomach	Never	Sometimes	Often	Always
4.	I feel afraid	Never	Sometimes	Often	Always
5.	I would feel afraid of being on my own at home	Never	Sometimes	Often	Always
6.	I feel scared when I have to take a test	Never	Sometimes	Often	Always
7.	I feel afraid if I have to use public toilets or bathrooms	Never	Sometimes	Often	Always
8.	I worry about being away from my parents	Never	Sometimes	Often	Always
9.	I feel afraid that I will make a fool of myself in front of people	Never	Sometimes	Often	Always
10.	I worry that I will do badly at my school work	Never	Sometimes	Often	Always
11.	I am popular amongst other kids my own age	Never	Sometimes	Often	Always
12.	I worry that something awful will happen to someone in my family	Never	Sometimes	Often	Always
13.	I suddenly feel as if I can't breathe when there is no reason for this	Never	Sometimes	Often	Always
14.	I have to keep checking that I have done things right (like the switch is off, or the door is locked)	Never	Sometimes	Often	Always
15.	I feel scared if I have to sleep on my own	Never	Sometimes	Often	Always
16.	I have trouble going to school in the mornings because I feel nervous or afraid	Never	Sometimes	Often	Always
17.	I am good at sports	Never	Sometimes	Often	Always
18.	I am scared of dogs	Never	Sometimes	Often	Always
19.	I can't seem to get bad or silly thoughts out of my head	Never	Sometimes	Often	Always

20.	When I have a problem, my heart beats really fast	Never	Sometimes	Often	Always
21.	I suddenly start to tremble or shake when there is no reason for this	Never	Sometimes	Often	Always
22.	I worry that something bad will happen to me	Never	Sometimes	Often	Always
23.	I am scared of going to the doctors or dentists	Never	Sometimes	Often	Always
24.	When I have a problem, I feel shaky	Never	Sometimes	Often	Always
25.	I am scared of being in high places or lifts (elevators)	Never	Sometimes	Often	Always
26.	I am a good person	Never	Sometimes	Often	Always
27.	I have to think of special thoughts to stop bad things from happening (like numbers or words)	Never	Sometimes	Often	Always
28.	I feel scared if I have to travel in the car, or on a bus or a train	Never	Sometimes	Often	Always
29.	I worry what other people think of me	Never	Sometimes	Often	Always
30.	I am afraid of being in crowded places (like shopping centres, the movies, buses, busy playgrounds)	Never	Sometimes	Often	Always
31.	I feel happy	Never	Sometimes	Often	Always
32.	All of a sudden I feel really scared for no reason at all	Never	Sometimes	Often	Always
33.	I am scared of insects or spiders	Never	Sometimes	Often	Always
34.	I suddenly become dizzy or faint when there is no reason for this	Never	Sometimes	Often	Always
35.	I feel afraid if I have to talk in front of my class	Never	Sometimes	Often	Always
36.	My heart suddenly starts to beat too quickly for no reason	Never	Sometimes	Often	Always
37.	I worry that I will suddenly get a scared feeling when there is nothing to be afraid of	Never	Sometimes	Often	Always
38.	I like myself	Never	Sometimes	Often	Always
39.	I am afraid of being in small closed spaces, like tunnels or small rooms	Never	Sometimes	Often	Always
40.	I have to do some things over and over again (like washing my hands, cleaning or putting things in a certain order)	Never	Sometimes	Often	Always

or putting things in a certain order)

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41.	I get bothered by bad or silly thoughts or pictures in my mind	Never	Sometimes	Often	Always
42.	I have to do some things in just the right way to stop bad things happening	Never	Sometimes	Often	Always
43.	I am proud of my school work	Never	Sometimes	Often	Always
44.	I would feel scared if I had to stay away from home overnight	Never	Sometimes	Often	Always
45.	Is there something else that you are really afraid of?	YES	NO		
	Please write down what it is:				
	How often are you afraid of this thing?	Never	Sometimes	Often	Always

Worry Scale for Children

This form is about worrying. Worrying happens when you are scared about something and you think about it a lot.

	he answer that best tells <i>how much</i> you have worried about the item <u>in</u>		it			/ much
<u>the pa</u>	<u>st week.</u>	None	A Little Bit	Some	A Lot	Very, very much
1.	How much did you worry about <u>school</u> ?	0	0	0	0	0
2.	How much did you worry about your <u>performance</u> ?	0	0	0	0	0
3.	How much did you worry about your <u>classmates</u> ?	0	0	0	0	0
4.	How much did you worry about your <u>friends</u> ? (e.g., being left out/excluded by friends, betrayed by friends)	0	0	0	0	0
5.	How much did you worry about war?	0	0	0	0	0
6.	How much did you worry about <u>disasters</u> ? (e.g., earthquakes, bush fires, floods, tsunamis)	0	0	0	0	0
7.	How much did you worry about <u>money</u> ?	0	0	0	0	0
8.	How much did you worry about your <u>health</u> ?	0	0	0	0	0
9.	How much did you worry about <u>future events</u> ? (<i>e.g.</i> , <i>holidays</i> , <i>moving house/school</i> , <i>family & friends</i>)	0	0	0	0	0
10.	How much did you worry about <u>personal harm</u> ? (e.g., being kidnapped, house being robbed, accidents)	0	0	0	0	0
11.	How much did you worry about <u>little things</u> ?	0	0	0	0	0
12.	How much did you worry about your <u>appearance</u> ?	0	0	0	0	0
13.	How much did you worry about your <u>family</u> ? (e.g., parents arguing, siblings, step family, grandparents dying)	0	0	0	0	0
14.	How much did you worry about not being able to sleep?	0	0	0	0	0
15.	How much did you worry about other things not covered above?	0	0	0	0	0

C.3 Parent-Report Questionnaire

The following questions are to be completed by the child's parent or guardian. We would like you to answer these questions about yourself (<u>not</u> about your child).

Please read each statement and circle a number which indicates how much the statement applied to you *over the past week*. There are no right or wrong answers. Do not spend too much time on any statement.

The rating scale is as follows:

- 0 Did not apply to me at all (NEVER)
- 1 Applied to me to some degree, or some of the time (SOMETIMES)
- 2 Applied to me to a considerable degree, or a good part of the time (OFTEN)
- **3** Applied to me very much, or most of the time (ALMOST ALWAYS)
- 1. I was aware of dryness of my mouth

0	1	2	3
NEVER	SOMETIMES	OFTEN	ALMOST ALWAYS

2. I experienced breathing difficulty (e.g., excessively rapid breathing, breathlessness in the absence of physical exertion)

0	1	2	3
NEVER	SOMETIMES	OFTEN	ALMOST ALWAYS

3. I experienced trembling (e.g., in the hands)

0	1	2	3
NEVER	SOMETIMES	OFTEN	ALMOST ALWAYS

4. I was worried about situations in which I might panic and make a fool of myself

0	1	2	3
NEVER	SOMETIMES	OFTEN	ALMOST ALWAYS

5. I felt I was close to panic

7.

NEVER

0	1	2	3
NEVER	SOMETIMES	OFTEN	ALMOST ALWAYS

6. I was aware of the action of my heart in the absence of physical exertion (e.g., sense of heart rate increase, heart missing a beat)

0 NEVER	1 SOMETIMES	2 OFTEN	3 ALMOST ALWAYS
. I felt scared with	out any good reason		
0	1	2	3

OFTEN

SOMETIMES

ALMOST ALWAYS

C.4 Clinical Sleep History Interview

Assessme	nt Intervi	iew				
DATE:	DATE:		Pe	P&C (6-18)		
In attendance: In	In attendance: Interviewer		Parent: Mun	Parent: Mum / Dad / Neither / Siblings		
them answering and the parents t	questions. Howe o talk by themse	ver, **for teer lves for a cou	ıs** there will l ple of minutes.	day is going to involve both be an opportunity for the to Remind them that the ess they or another person to	een	
Age:		Grad	le:			
Fat	ther her lings					
How much assist	ince did <child's< td=""><td>name> need 1</td><td>with completing</td><td>the questionnaires?</td><td></td></child's<>	name> need 1	with completing	the questionnaires?		
0 No hel	500	2 ne help	3 lots of help	4 help with it all		
How much assist				•		
110w much assiste 0	ince ala ~crata s	2	vun ine sieep ai 3	<i>ury:</i> 4		
No he	p so	me help	lots of help	help with it all		
			UNCTIONING			
Do any of the fol	owing occur for	< child's nam		poor sleep? (tick if YES)		
Tiredness / fatigua Attention, concern Problems socialisi Poor school perfor Moody or irritable Sleepy during the Lack of energy / r Behavioural issue Accident prone Tension, headache Worries about slee Other health prob	ration, or memor ng mance (e.g. grad day notivation s (e.g., hyperactives, or stomach pro- ep ems	y problems es) vity, aggression oblems		Parent		
Are these difficul	ties present at le	east 3 days pei	r week? 🔲 YI	ES 🔲 NO		

SLEEP HISTORY	
Specify to parent/child that you are interested in the nature of the current problem How long has the current sleep problem been an issue? YearsMonthsWeeks	
Was there a trigger event that you associate with the onset of the problem? If yes, clarify trigger event	
If no, "Has the sleep problem come about gradually?"	
Has the sleep problem changed/worsened since you first noticed it?	
Now broaden sleep history Have there been other sleep issues in the past? (get info about type of symptoms, age of onset, duration, treatment,etc.)	
What was <child's name="">'s sleep like as an infant?</child's>	
***For teens** During the primary school years, did <child's name=""> require parental presence to sleep?</child's>	
Is < child's name's > sleep like: Mum's Dad's Dibling's DK	
Is there anyone in Mum's / Dad's family that has similar sleep? (in particular, give details about similar morningness/eveningness)	

CATACTRODUCTING INTERVIEW
CATASTROPHISING INTERVIEW For this next part, we really need to hear the answers from the child So you've said you have trouble < falling /staying asleep >, does it worry you? Always Sometimes Not really Not at all
Arways Sometimes not really inot at all Go to next section
What is it that worries you about nights when you have problems sleeping?
What is it that worries you about < previous worry >?
What is it that worries you about < previous worry >?
What is it that worries you about < previous worry >?
What is it that worries you about < previous worry >?
What is it that worries you about < previous worry >?
What is it that worries you about < previous worry >?
Number of catastrophising thoughts:
BEDTIME ROUTINE
What does < child's name > usually do in the last hour before going to bed?

< Child's name >, do you do anything close to bedtime that may that might wake you up a bit							
either mentally (like going on the	internet) , j	physically	(exercising)	, or ei	notiona	lly (watch	ing
disturbing TV or movie)?	_	_					
YES if YES, list:		NO					
			how often?	1	nights/w	k	
			how often?		nights/w		
			how often?		nights/w		
			how often?		nights/w		
			now onen.				
Does < child's name > resist or re	fuse to go	to bed at	night?				
YES NO							
If YES, please rate level of a	esistance:						
Mild Moderate	Severe 3						
Reasons given for resisting	-	not faolina	tired or clas		and of	dowb)	
Reasons given for resisting	, beu (eg, i	loi jeeiing	they or siee	py, sc	ureu oj i	шк)	
To fall asleep (initial onset or night ti	me awaken	ing) , does					
					-	lem is it to	
			-			Moderate	
To read / watch TV / listen to music	:		Yes	1	2	3	4
A drink/food	Ļ	= =	Yes	1			4
To be in other room	Ļ		Yes	1		3	4
Other (ie, Person, eg, Mum)	L	No 🗌	Yes	1	2	3	4
Any other object, or setting?							
If <u>YES</u> , does the child have difficul				e?			
NO YES	📃 descrit	be:					
	SLEE	P HYGI	ENE				
Does < child's name >:							
	No	Yes	5				
Read in bed							
Watch TV in bed	H	H					
	H	H					
Study in bed Talk on the phone (or text) in bed	H	H					
Taik on the phone (or text) in bed							
Is their:							
Pad comfortable							
Bed comfortable	H						
Bedroom dark at night	H						
Bedroom quiet at night							

Anything else they do in bed besides sleeping (eg, DS, PSP)?_____

*** Ask parent and teen if they feel comfortable for the parent to wait for a couple of minutes in waiting room so you can ask the teen some questions. Before asking questions on nicotine and alcohol, advise teen that you can keep this information confidential unless it is harmful to them. If they wish, they can tell their parents – but we cannot if there is no harm involved ***

How much of the following do you have (on average) each day?

Cola Drink	0	·	glasses per day	Last drink 🔲 a.m. 🔲 p.m.
Coffee/Tea			_ cups per day	Last drink 🔲 a.m. 🔲 p.m.
Chocolate			pieces per day	Last piece 🔲 a.m. 📃 p.m.
Energy drinks			_ cans per day	Last drink 🔲 a.m. 🔲 p.m.
*Alcohol			standard per day	Last drink 🔲 a.m. 🔲 p.m.
*Nicotine			_ cigarettes per day	Last one 🔲 a.m. 🔲 p.m.

*** Invite parent back in

At night, does < child's name > look to see what time it is? YES NO If <u>YES</u> , with what (eg, mobile phone, illuminated clock, etc.)? when?: (can tick more than one) trying to fall asleep at the start of the night when waking up during the night when waking up in the morning (ie, before getting out of bed)
when waking up in the morning (ie, before getting out of bed)
DELAYED CIRCADIAN PHASE?
Does < child's name > nap during the day?
If <u>YES</u> , how often?: 1 time per week 2-3 times per week 4-5 times per week 6-7 times per week
If <u>YES</u> , i) time of nap

ii) length of nap _____ minutes / hours
Does < the child > lack energy/motivation/is flat after school? YES NO

Does < the child > become alert after dinner? YES No	0
Does < the parent > need to repeatedly ask < child's name > to get out a mornings? YES NO If <u>YES</u> , how many times on an average morning?	of bed on <i>school</i> times
Does <child's name=""> hate bright light? NO YES Image: describe:</child's>	
Does <child's name=""> 'wake up by themselves' on School mornings YES NO describe:</child's>	
Is <child's name=""> late getting to school?</child's>	

NO YES describe:

Does <child's name=""> 'wake up by themselves' on Weekends YES NO describe:</child's>							
Think about the last time you were able to choose your own sleep timing (e.g., weekends or holidays, without any morning commitments) - What time did you naturally wake-up? - Do you think you slept better? - Do you think you slept longer?							
DIFFERENTIAL DIAGNOSIS							
Has < child's name > recently travelled interstate or overseas? YES NO VES If YES, describe below: Destination: No. of time zones travelled: How recent? mths wks days							
Has < child's name > been diagnosed with a MEDICAL condition?							
\longrightarrow If <u>YES</u> , list:							
diagnosed by? diagnosed by?							
diagnosed by?							
Did the sleep problem occur at the same time as <the disorder="" medical="">? NO ☐ ⇔ go to q13 YES ☐</the>							
Which would you consider worse, the sleep problem or <the disorder="" medical="">? Sleep Problem Medical Disorder</the>							
Has < child's name > been diagnosed with a PSYCHIATRIC condition? (eg, depression, anxiety, etc.)? YES NO S If YES, list:							
diagnosed by?							
diagnosed by? diagnosed by?							
Did the sleep problem occur around the same time as <the disorder="" psychiatric="">?</the>							
Does the sleep problem 'come and go' with changes in the severity of the <diagnosed disorder="" psychiatric="">?</diagnosed>							
Is < child's name > on any MEDICATION(please list)?							
dose when taken							
dose when taken dose when taken							

Does < child's name > experience restless legs (*ie*, feel need to move legs, feeling of insects crawling over legs)?

6-7 nights per week

LIES LINO	
If <u>YES</u> , how often?:	🔲 1 night per week
	2-3 nights per week
	4-5 nights per week

Does < child's name > have regular twitching legs (or arms) during sleep (e.g. is their bed in a mess in morning? YES NO If <u>YES</u>, how often?: 1 night per week 2-3 nights per week 4-5 nights per week 🔲 6-7 nights per week Does < child's name > snore in their sleep? YES DK 🔲 NO 🔲 □→ If YES, how often?: 1 night per week 2-3 nights per week 🔲 4-5 nights per week 6-7 nights per week Generally, how loud is the snoring? 🔲 light moderately loud 🔲 very loud Does < child's name > stop breathing for short periods (eg, 10-20 secs) during your sleep? YES 📃 DK 🔲 NO 🔲 If YES, how often?: 1 night per week 2-3 nights per week 4-5 nights per week

6-7 nights per week

*** For teens, ask if teen and parent comfortable with teen waiting outside for a couple of minutes, to allow parent to say some things ***

Parent comments

^{***} If interviewing a teen, invite them back in ***

Appendices

Child's Goa	ls:	1)
		2)
		3)
		4)
Parent's Go	als:	1)
		2)
		3)
		4)
Local GP	Name:	
	Practice:	
	Address:	
Scho	ol name:	
	her's name:	
Princ	ipal's name:	
Coun	sellor's name:	

Inform parent and child/adolescent of:

- clinic meeting to confirm they meet research criteria.
- what happens if they don't meet research criteria.
- we will call or email later today to confirm next appointment.

ICSD-3 Classification(s):

DSM-5 Classification(s):

C.5 Treatment Manuals

These documents have been removed due to copyright restrictions

C.6 Ongoing Sleep Management Handouts

Ongoing Management of my Child's Sleep Schedule (Restriction Groups)

Now that your child is starting to sleep better during their reduced time-in-bed, we need to determine their optimal sleep schedule.

It is best to keep a consistent rise time in the mornings, and stick to this 7 days a week (yes, even on weekends!).

If your child is falling asleep quickly, has minimal waking during the night, and/or reports feeling very sleepy at "lights out" time, then you can gradually extend their time-in-bed by bringing their "lights out" time 15 minutes earlier. Each time you adjust their "lights out" time, try to stick with that sleep schedule for a few days to monitor how their sleep changes. The easiest way to continue to monitor their sleep is with a sleep diary.

Managing disruptions to your child's routine

We understand that there may be occasions when a special event means you need to stay up late one night, or get up early one morning. If so, you can adjust your child's "lights out" time and rise time so that their time-in-bed remains the same. For example, if the time-in-bed should be 8.5 hours and the usual schedule is 10.30 pm - 7 am, but you need to stay up until 11.30 pm one night, then you can have a later rise time of 8 am the next morning. Then try to get back to the usual schedule the following night.

Monitoring your child's sleep

We would like you to continue to keep a sleep diary for the next few weeks. We would also like your child to complete questionnaires once per week on Tuesday or Wednesday. This is another way that we will continue to monitor your child's progress over the coming weeks.

Please log in to the online system at: https://socsci.flinders.edu.au/psyc/research/cain

First Name:
Last Name:
Password:

What happens next?

We will contact you by phone at the end of the follow-up period. You will then also receive some brief written information about your child's sleep and cognitive functioning (as measured by the computer tasks). If necessary, we will discuss options for further treatment at this time.

Please let us know if you plan to commence any new treatments (for sleep or other issues) over the next few weeks.

Ongoing Management of my Child's Sleep Schedule (Control Group)

Over the next few weeks, your child will continue to adjust to their new regular sleep schedule.

As a general "rule", it is best to keep a consistent "lights out" time and a consistent rise time in the mornings, and stick to this 7 days a week (yes, even on weekends!).

However, we understand that there may be occasions when a special event means you need to stay up late one night, or get up early one morning. If so, you can adjust your child's "lights out" time and rise time so that their time-in-bed remains the same. For example, if the time-in-bed should be 9.5 hours and the usual schedule is 9.30pm - 7am, but you need to stay up until 10.30pm one night, then you can have a later rise time of 8am the next morning. Then try to get back to the usual schedule the following night.

Also remember to keep your child's bedtime routine as consistent as possible, avoiding stimulating activities and instead engaging in relaxing activities under dim light.

Monitoring your child's sleep

We would like you to continue to keep a sleep diary for the next few weeks. We would also like your child to complete questionnaires once per week on Tuesday or Wednesday. This is another way that we will continue to monitor your child's progress over the coming weeks.

Please log in to the online system at: https://socsci.flinders.edu.au/psyc/research/cain

First Name:	•••
Last Name:	••
Password:	••••

What happens next?

We will contact you by phone at the end of the follow-up period. You will then also receive some brief written information about your child's sleep and cognitive functioning (as measured by the computer tasks). If necessary, we will discuss options for further treatment at this time.

Please let us know if you plan to commence any new treatments (for sleep or other issues) over the next few weeks.

C.7 Brief Follow-up Interview

Does your child have difficulty falling asleep?	YES	NO
Does your child have difficulty staying asleep?	YES	NO
Does your child require special conditions in order to fall asleep or return to sleep? <i>If YES, please describe:</i>	YES	NO
Do you think your child has a "sleep problem"?	YES	NO
Do you think your child needs further treatment here?	YES	NO

Please give reasons why (or why not) further treatment is requested

Appendix D: Supplementary Materials for Chapter 5

D.1 Teacher-Report Questionnaire

School Situations Questionnaire-Revised

Name of child:

Name of person completing this form:

Does this child have problems paying attention or concentrating in any of these situations? If so, indicate how severe these attentional difficulties are.

Situation	Yes (circle		Mild	Ι	f yes,	how	sever	e (ciro	cle or	ne)	Severe
During individual desk work	Yes	No	1	2	3	4	5	6	7	8	9
During small group activities	Yes	No	1	2	3	4	5	6	7	8	9
During free play time in class	Yes	No	1	2	3	4	5	6	7	8	9
During lectures to the class	Yes	No	1	2	3	4	5	6	7	8	9
On field trips	Yes	No	1	2	3	4	5	6	7	8	9
During special assemblies	Yes	No	1	2	3	4	5	6	7	8	9
During movies, filmstrips	Yes	No	1	2	3	4	5	6	7	8	9
During class discussions	Yes	No	1	2	3	4	5	6	7	8	9

Appendices

D.2 Daytime Sleepiness Scale

Please answer the following questions as honestly as you can, thinking about how you have been feeling in the <u>past week</u>. Circle one answer only.

1. How often do you fall asleep or feel drowsy in class?								
Always	Frequently	Sometimes	Sometimes Seldom					
2. How often do y	ou feel sleepy or	drowsy while	doing your ho	mework?				
Always	Frequently	Sometimes	Seldom	Never				
3. Are you usually	alert during the	day?						
Always	Frequently	Sometimes	Seldom	Never				
4. How often do y	ou feel tired and	grumpy during	the day?					
Always	Frequently	Sometimes	Seldom	Never				
5. How often do y	ou have trouble	getting out of b	ed in the morr	ning?				
Always	Frequently	Sometimes	Seldom	Never				
6. How often do y	ou fall back to sl	eep after being	woken in the	morning?				
Always	Frequently	Sometimes	Seldom	Never				
7. How often do y	ou need someon	e to wake you i	n the morning	?				
Always	Frequently	Sometimes	Seldom	Never				
8. How often do y	ou think that you	need more sle	ep?					
Always	Frequently	Sometimes	Seldom	Never				