Flinders University

Supporting Literacy Skills for Children with Developmental Disabilities

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ABSTRACT

Learning to read is a fundamental human right and is associated with positive academic, employment, social and mental health outcomes. Yet, children with developmental disabilities are often underestimated in terms of their capacity to develop reading and writing skills and are more likely to experience poorer literacy outcomes than their typically developing peers. This is, at least in part, due to the quality of literacy instruction they receive. Comprehensive literacy instruction, incorporating five key skills, is widely regarded as evidence-based for typically developing children, though is relatively underexplored for children with developmental disabilities. This research aims to extend the current research base on comprehensive literacy instruction for children with developmental disabilities, with a focus on three prevalent disabilities: cerebral palsy, Down syndrome, and autism. This research includes a systematic review and three empirical studies involving delivery and evaluation of comprehensive literacy instruction. All empirical studies involved use of the ABRACADABRA literacy web application and shared book reading (SBR) methods.

The systematic review explored literacy instruction for children with cerebral palsy, with a focus on methods known to be evidence-based for typically developing children. This review revealed that no studies have explored comprehensive literacy instruction for children with cerebral palsy. Study 1 was a case study involving one child with cerebral palsy who participated in intensive literacy instruction using the ABRACADABRA program, delivered via telepractice, and supplemented by parent-led SBR. This child with cerebral palsy made modest gains in their letter-sound correspondence and decoding skills following participation in the literacy program. Study 2 was a pilot study involving six children with Down syndrome using the ABRACADABRA program, delivered via telepractice, and supplemented by parent-led SBR. Children with Down syndrome made gains in their word- and passage-level reading accuracy skills and functional reading comprehension skills. Study 3 was a quasi-experimental study involving 59 children with autism. This study was significantly disrupted by the COVID-19 pandemic and stay-at-home orders issued midway through instruction, leaving a final sample of 47 participants. This study initially involved small-group literacy instruction delivered face-to-face, though halfway through changed to one-to-one instruction delivered via telepractice because of the pandemic induced limitations. Children in

this study received either ABRACADABRA instruction only, or ABRACADABRA plus clinician-led SBR. Children who participated in instruction made statistically significant gains in their nonword reading skills, relative to a control group. There were no other statistically significant results; however, effect sizes for all other reading outcomes were similar to previous ABRACADABRA research with children with autism (medium effects for word reading accuracy and reading comprehension and large effect sizes for passage reading accuracy).

This research contributes original empirical evidence concerning comprehensive literacy instruction delivered to groups of children with developmental disabilities. Additionally, this research contributes new evidence to show that remote delivery of high-quality literacy instruction is feasible and can be effective for some children with developmental disabilities. This finding has the potential to increase the accessibility of some services. It is hoped that research such as this can help guide policies and practices that improve literacy outcomes for children with developmental disabilities.

DECLARATION

I certify that this thesis:

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

2. and the research within will not be submitted for any other future degree or diploma without the permission of Flinders University; and

3. 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.

Signed...... ....

Date......24/03/2024..

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Thank you to my family (and there are a lot of you!). To my parents and grandparents who all raised me and instilled me with the resilience, perspective, and determination to get through these last few years. To my siblings, Katie, Andrew, Laura, and Abbey, for always being there to lean on, and my niblings, Olivia, Cora, William, Charlotte, Florence, and Ruby, for always making me smile. Thank you to my friends who have tolerated me talking about this research for the last few years, have offered their support, and are still here! To my friend Ash who convinced me it was even possible that I could do this – I am so appreciative of your guidance and encouragement. And to my chosen family, Hollie, Tom, Shanthi, Paul, Jess and Tom, thank you for your unwavering emotional and practical support – I would not be at this point without it. I consider myself extremely fortunate to be surrounded by such generous people. Finally, I would like to thank my furry research assistants, Judy and Gertie, for keeping me company on the long days and nights of writing and for raising office morale. I look forward to having more time for walks and adventures!

DEDICATION

This thesis is dedicated to baby Mae, who never got to show the world how brilliant she could be.

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LIST OF ACRONYMS AND ABBREVIATIONS

AAC	Augmentative and alternative communication
ABRA	ABRACADABRA
ADD	Attention-deficit disorder
ADHD	Attention-deficit/hyperactivity disorder
APA	American Psychological Association
CAI	Computer-assisted instruction
CAS	Childhood apraxia of speech
COVID-19	Coronavirus Disease 2019
CP	Cerebral palsy
CSLP	Centre for the Study of Learning and Performance
CVC	Consonant vowel consonant
DIER	Direct and Indirect Effects Model of Reading
MMAT	Mixed Methods Appraisal Tool
NAP	Nonoverlap of All Pairs
NDIS	National Disability Insurance Scheme
NICHD	National Institute of Child Health and Human Development
NRA	Nonverbal reading approach
NRP	National Reading Panel
NSW	New South Wales
SBR	Shared book reading
SCED	Single-case experimental design
SES	Socioeconomic status
STM	Short-term memory
SVR	Simple View of Reading
UDL	Universal Design for Learning
UK	United Kingdom

ACRONYMS FOR STANDARDISED ASSESSMENTS

CC-2	Castles and Coltheart Test - Second Edition
CELF-4	Clinical Evaluation of Language Fundamentals – Fourth Edition
CTOPP-2	Comprehensive Test of Phonological Processing – Second Edition
NARA-3	Neale Analysis of Reading Ability – Third Edition
PPVT-5	Peabody Picture Vocabulary Test – Fifth Edition
TERC	Test of Everyday Reading Comprehension
TONI-4	Test of Nonverbal Intelligence – Fourth Edition
VABS-2	Vineland Adaptive Ability Scales – Second edition
WJ-IV	Woodcock-Johnson Tests of Achievement – Fourth Edition
WRAT-4	Wide Range Achievement Test – Fourth Edition
WRAT-5	Wide Range Achievement Test – Fifth Edition
YARC	York Assessment of Reading for Comprehension

LIST OF PUBLICATIONS AND PRESENTATIONS

Publications

Murphy, A., Bailey, B., & Arciuli, J. (2023). Exploring the effects of literacy instruction for children with cerebral palsy: A systematic review. *Language, Speech, and Hearing Services in Schools,* 54(1), 299-321. https://doi.org/10.1044/2022_LSHSS-22-00014

Murphy, A., Bailey, B., & Arciuli, J. (2023). ABRACADABRA literacy instruction for children with Down syndrome via telepractice during COVID-19: A pilot study. *British Journal of Educational Psychology*, 93(1), 333-352. https://doi.org/10.1111/bjep.12558

Murphy, A., Bailey, B., Savage, R., Parrila R., & Arciuli, J. (2024). An Effectiveness Trial of ABRACADABRA Literacy Instruction for Children with Autism during the COVID-19 Pandemic. [under review]

Presentations

Murphy, A., Bailey, B., & Arciuli, J. (2021, September 8-9). *Telepractice delivery of ABRACADABRA literacy instruction for children with Down syndrome: A preliminary study* [Oral presentation]. Psychology of Education Section Annual Conference, Online conference, United Kingdom.

Murphy, A., Bailey, B., & Arciuli, J. (2022, November 17-19). *Feasibility of literacy instruction for children with Down syndrome via telepractice during COVID-19* [Poster presentation]. American Speech-Language-Hearing Association (ASHA) Convention, New Orleans, LA, United States.

Murphy, A., Bailey, B., & Arciuli, J. (2023, July 19-22). *A systematic review of literacy instruction for children with cerebral palsy* [Poster presentation]. Society for the Scientific Study of Reading (SSSR) Conference, Port Douglas, QLD, Australia.

PREFACE

Chapters: This thesis comprises six chapters which are a combination of published and draft manuscripts formatted as journal articles, as well as unique chapters. Chapters 2 and 4 have been published in peer-reviewed international journals. Chapter 5 reports the main study outcomes of a project funded by the Australian Research Council (ARC). This project grant was awarded to Arciuli, Parrila and Savage and that manuscript is currently under review at a journal. Word document versions of published papers are included in this thesis, rather than copy edited versions. Minor amendments have been made to these chapters to ensure consistency of style and terminology throughout the thesis.

Study Abstracts: Abstracts are included at the beginning of all chapters formatted as journal articles (Chapter 2 to Chapter 5).

Acronyms: Acronyms used throughout this thesis are listed on page x and xi. Acronyms are re-introduced in each chapter the first time they appear to assist with ease of reading.

Tables and Figures: Tables and figures are embedded within each chapter as they are mentioned and are numbered according to the chapter. Table and figure numbers for published manuscripts have been amended to be consistent with the table number formatting throughout this thesis.

Footnotes: Where footnotes are used, these appear at the bottom of the page.

References: All references are included at the end of this thesis, rather than at the end of each chapter, to reduce repetition and assist with the thesis flow.

CHAPTER 1 INTRODUCTION

1.1 Brief Introduction to the Current Research Landscape

"Learning to read is not a privilege, but a basic and essential human right" (Ontario Human Rights Commission, 2022, p. 2). Yet, historically, many children with developmental disabilities have been underestimated in terms of their capacity to develop reading skills and some have been provided with literacy programs based on pseudoscientific theories about how these children learn to read (Accardo & Finnegan, 2019; Cologon, 2013; Cossu et al., 1993; Griffiths et al., 2016; Uccheddu et al., 2019). Reading instruction for children with developmental disabilities has often focused on developing emergent literacy skills (i.e., skills that precede literacy development, such as engagement with books and print awareness) or 'functional' literacy (e.g., sight word recognition of key words), rather than conventional literacy instruction (i.e., instruction targeted at developing reading and writing skills that adhere to the accepted norms in terms of structure, content, and application; (King, Rodgers, et al., 2022; Koppenhaver, 2000; Ruppar et al., 2011). Increasingly, research is demonstrating that the science of reading (the large body of interdisciplinary scientific research about reading processes, development, and instruction) applies to all children, including those with developmental disabilities (Allor, Mathes, Roberts, Cheatham, et al., 2010; Arciuli & Bailey, 2021; Browder et al., 2009). These children may require additional supports or modifications to ensure that instruction is accessible; however, there is now a general understanding that the same basic scientific principles of how children learn to read apply also to children with disabilities (Barton-Hulsey et al., 2021; Koppenhaver et al., 2007; Machalicek et al., 2010; Yorke et al., 2021). Despite recognition that children with developmental disabilities require access to the same evidence-based instruction as their peers, these children are underrepresented in scientific research on reading. Reduced understanding of inclusive evidence-informed literacy instruction practices contributes, at least in part, to the poorer literacy outcomes experienced by children with developmental disabilities (Accardo & Finnegan, 2019; Cologon, 2013).

1.2 Thesis Summary

This thesis explores some current literacy instruction practices for children with developmental disabilities, as well as investigating some new literacy instruction methods and models of service delivery. The methods of literacy instruction explored in this research are consistent with instruction methods known to be evidence-based for typically developing children. There is a focus on three prevalent developmental disabilities: cerebral palsy (CP), Down syndrome, and autism. Specifically, this thesis includes a published systematic review exploring the effects of literacy instruction for children with CP, as well as three empirical studies utilising the ABRACADABRA literacy web application and shared book reading (SBR) methods. The empirical studies include: (a) a case study involving one child with CP; (b) a published pilot study involving six children with Down syndrome; and (c) a quasi-experimental study involving 59 children with autism. This research was conducted during the global COVID-19 pandemic and as such instruction was primarily delivered via telepractice. The overall aim of this research is to provide a clearer understanding of effective reading instruction methods for children with developmental disabilities to help guide policies and practices that improve literacy outcomes for these children.

1.3 What is Literacy?

Literacy is a form of communication and includes both the ability to decode words and derive meaning from text (reading), as well as the ability to encode words and convey messages through print (writing). While literacy includes both reading and writing, reading is the primary focus of this thesis. The definition of reading provided by the Programme for International Student Assessment (PISA) applies throughout this thesis: “Reading literacy is understanding, using, reflecting on and engaging with written texts, in order to achieve one’s goals, develop one’s knowledge and potential, and participate in society” (OECD, 2013, p. 61). There is a reciprocal relationship between oral language and literacy skills, whereby oral language competency facilitates reading development and access to written language, which in turn increases exposure to more complex vocabulary and grammatical forms, further facilitating language growth (Nation, 2019; Snow, 2016). However, by comparison with oral language, written forms of language are more recent in human history and require explicit instruction to be acquired (Castles et al., 2018; Snow, 2020).

1.3.1 Importance of Literacy

The value of literacy in supporting independence and securing positive long-term outcomes cannot be underestimated. Literacy plays a pivotal role in furthering cognitive development and supporting participation with the education curriculum, as well as assisting with future employment opportunities and social engagement. Early success in literacy is associated with more positive academic achievement in later schooling (Singh, 2013), whilst low levels of literacy contribute to poorer academic, employment, and mental health outcomes (Francis et al., 2019; Hendren et al., 2018; Morrisroe, 2014). For individuals with complex communication needs, literacy can provide a means for independent communication and social participation and can facilitate self-advocacy and self-care (Caron & Light, 2016; Kitson et al., 2021; Koppenhaver & Williams, 2010). Literacy is not only a means for participation, independence, choice, and control, but can also be a source of enjoyment through reading for pleasure (Robinson et al., 2019; Tovli, 2014).

1.3.2 How Reading is Conceptualised in this Thesis

The science of reading is grounded in theoretical frameworks, such as the Simple View of Reading (SVR; Gough & Tunmer, 1986). Throughout this thesis, reading is conceptualised in line with the SVR which asserts that skilled reading is the product of two broad but distinct skills: word recognition and oral language comprehension. Skilled reading refers to the ability to accurately and efficiently extract meaning from text (Gough & Tunmer, 1986; Scarborough, 2001). The SVR is comprehensively explored in the next section (Section 1.4.1). Reading instruction incorporating five individual components (phonemic awareness, phonics, reading fluency, vocabulary, and reading comprehension) is viewed throughout this thesis as effective and evidence-informed and is further described in Section 1.5 (National Institute of Child Health and Human Development [NICHD], 2000).

1.4 Theoretical Models of Reading

Learning to read is a highly complex skill requiring the coordination of many cognitive and linguistic processes in order to become proficient. As stated by Perfetti and Stafura (2014), “there is no theory of reading, because reading has too many components for a single theory” (p. 1). However, many broad-scope theories and models exist that can help to explain the cognitive capacities involved in reading, as well as how this skill develops and where difficulties may arise. The SVR (Gough & Tunmer, 1986) is a highly influential model that has informed many other theories and frameworks and is the primary model that informs the conceptualisation of reading in this thesis. A selection of theories and frameworks based on the SVR that inform the instructional approaches used to teach reading in this thesis are outlined below.

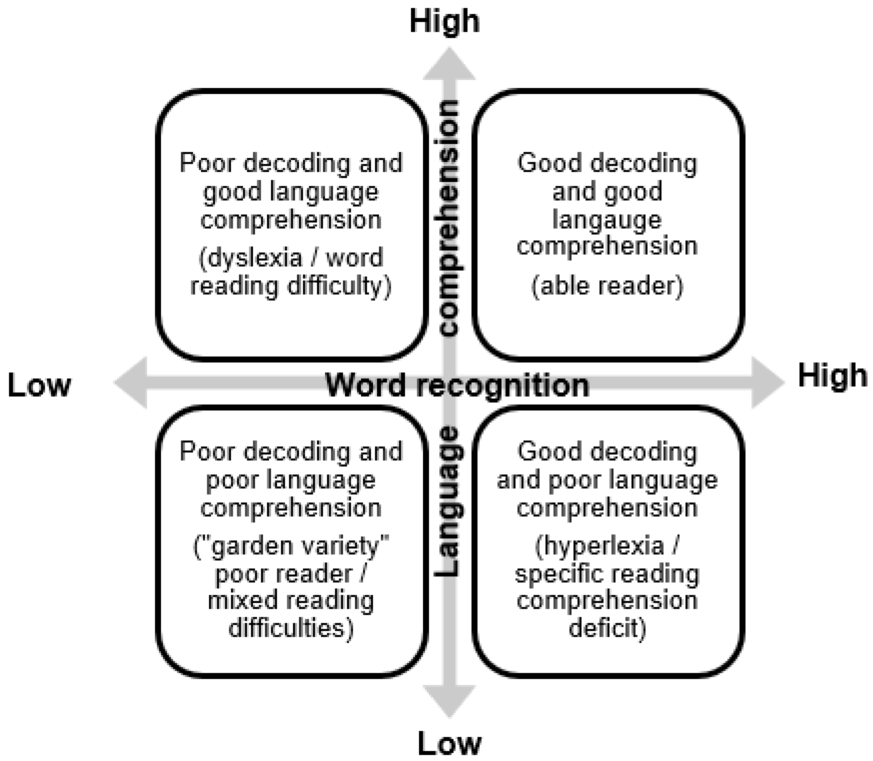
1.4.1 The Simple View of Reading

The SVR is an empirically supported conceptual model for skilled reading (Gough & Tunmer, 1986; Hoover & Gough, 1990; Hoover & Tunmer, 2018). According to the SVR, reading comprehension is the goal of reading and refers to the ability to obtain literal and inferential meaning from printed text. The model posits that reading comprehension is the product of two separate but interacting components: word recognition and oral language comprehension. Word recognition refers to the ability to recognise printed words fluently and accurately (including both phonological decoding and sight-based word identification). Language comprehension refers to the ability to understand literal and inferential meaning from spoken oral language. Both skills are necessary and equally important for successful reading comprehension and deficits in either, or both, of these domains results in reading difficulties.

The SVR has a robust evidence base supporting its utility and has been widely used throughout reading research. The framework has also had an impact on educational policy and clinical guidelines (e.g., Department for Education, 2023; Rose, 2006; Speech Pathology Australia, 2021). Studies consistently support that the components of the SVR are accurate in capturing variance in children’s reading comprehension skills. For example, Hoover and Tunmer (2018) summarised three empirical studies involving hundreds of students from early to late primary grades, which all demonstrated that differences in reading comprehension could be accounted for by word recognition and oral language comprehension skills. Support for the SVR has been shown across alphabetic and non-alphabetic languages (e.g., Lonigan et al., 2018; Peng et al., 2020), as well as typically developing children and children with developmental disabilities (e.g., Asbell et al., 2010; Hoover & Gough, 1990; Nation, 2019; Roch et al., 2021; Sorenson Duncan et al., 2021). Application of the SVR to children with developmental disabilities is further explored in Section 1.6.5 of this chapter.

The SVR provides a theoretical framework for explaining variation in reading comprehension at all stages of learning to read and can be used to inform reading assessment, instruction, and intervention. According to the SVR, difficulties with reading comprehension may be a result of: (a) impaired word recognition; (b) impaired oral language comprehension skills; or (c) both. These distinct reading profiles are displayed in Figure 1.1. Understanding where a child falls on the continuum of reading profiles shown in Figure 1.1 provides insight into why the child is having difficulty with reading and what the focus of instruction or intervention should be. In the original description of the SVR, Gough and Tunmer (1986) described these three varieties of reading difficulties as: hyperlexia, dyslexia, or ‘garden variety’ reading disability. Terminology for these reading profiles has changed over time, though research continues to support these discrete reading patterns (e.g., Catts et al., 2003; Nation, 2019).

Figure 1.1 *Reading Profile Subgroups Based on the Simple View of Reading (Gough & Tunmer, 1986)*



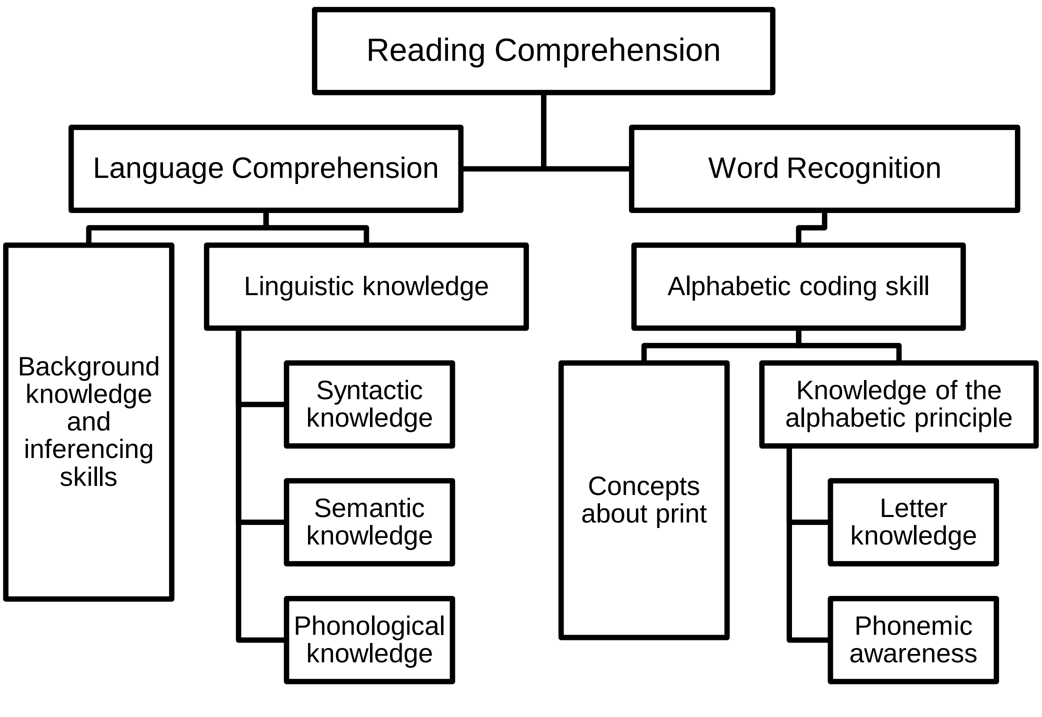
The SVR model acknowledges that both word recognition and language comprehension are highly complex skills made up of many contributing components, but that together, these two broad components comprise the big picture of skilled reading. Over time, the relative contributions of word recognition and language comprehension to skilled reading change. Initially, reading comprehension is constrained by a child's decoding and word recognition skills; however, as a child's decoding skills increase and word recognition becomes more fluent, oral language comprehension skills become more important for reading comprehension (e.g., Lonigan et al., 2018). Whilst changes in relative contributions of skills can be seen through use of the model, the SVR is a static model of reading and does not explain how reading develops over time.

Some have been critical of the SVR, highlighting the model's limitations. For example, Cervetti et al. (2020) discussed that the SVR does not clearly define what skills belong within each of the two broad SVR components, limiting the models utility for guiding reading instruction. In particular, Cervetti et al. (2020) noted that the model does not provide sufficient detail to explain variance in reading comprehension, and the skills required for successful comprehension, in the later school years. Duke and Cartwright (2021) argued that not all reading difficulties are caused by deficits in word recognition or oral language comprehension skills as theorised in the SVR, nor do each of these components influence reading separately (i.e., skills such as vocabulary, morphological awareness, and reading fluency are affected by both word recognition and oral language comprehension skills). For example, some children with age-appropriate word reading and listening comprehension skills still demonstrate reading comprehension difficulties (e.g., Catts et al., 2003; Ebert & Scott, 2016). Furthermore, some have suggested that the SVR does not account for the numerous additional components involved in reading which can influence reading comprehension, such as theory of mind (e.g., Larusso et al., 2016; Tong et al., 2020) and active self-regulation (Duke & Cartwright, 2021), including executive functions (e.g., Taboada Barber, Cartwright, et al., 2020), motivation and engagement (e.g., Taboada Barber, Lutz Klauda, et al., 2020), and strategy use or metacognitive skills (e.g., Ahmed et al., 2016).

1.4.2 Cognitive Foundations Framework

The Cognitive Foundations Framework is a developmental model based on the SVR that outlines the cognitive capacities involved in learning to read (Tunmer & Hoover, 2019). The model describes the subcomponents involved within each of the two SVR components (word recognition and oral language comprehension) and specifies the relationship between these various subcomponents (see Figure 1.2). The framework supports a developmental progression of skills and outlines that more advanced skills cannot be gained if other foundational skills are not mastered. For example, phonological decoding cannot be fully achieved until letter knowledge and phonemic awareness are developed. However, a child does not need to completely master all lower-level skills before higher-level skills can start to develop. Once a lower-level skill is developed to a certain point, there is a bi-directional relationship where this skill continues to develop simultaneously, in a reciprocal manner, with the skills immediately above and below it. For example, children's linguistic knowledge (phonological, semantic, and syntactic knowledge) continues to develop through engaging with more advanced texts which in turn exposes children to more complex language structures. As such, the skills outlined in the Cognitive Foundations Framework all rely on each other and cannot be taught in isolation. The framework can be used to guide assessment and to inform individualised instruction to remediate reading difficulties. This Framework includes only the cognitive skills that directly contribute to reading comprehension and does not include other factors that indirectly affect reading, such as motivation, self-efficacy, or quality of literacy instruction (Tunmer & Hoover, 2019).

Figure 1.2 *The Cognitive Foundations Framework (adapted from Tunmer & Hoover, 2019, p. 76)*

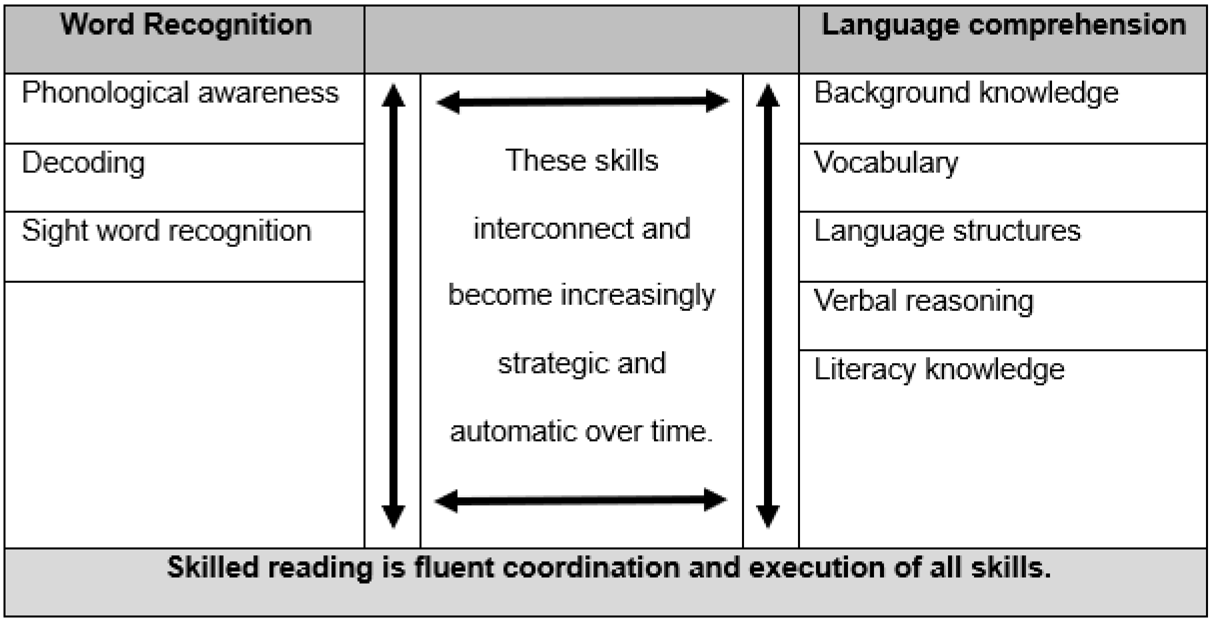


1.4.3 Scarborough's Rope

The 'Reading Rope' by Scarborough (2001) is another model that builds upon the SVR to explain the many sub-skills and cognitive processes that contribute to word recognition and oral language comprehension. The model uses the metaphor of a rope to explain how these skills are interwoven and developed over the course of becoming a skilled reader. The skills are interrelated and develop simultaneously, and integration of skills becomes increasingly automatic and strategic over time. The sub-skills involved in each strand of the 'Reading Rope' are listed in Figure 1.3. This model highlights the ubiquitous continuum of skill development which provides important considerations for supporting the literacy skills of children with developmental disabilities. According to this model, there are no 'prerequisite' skills for learning literacy, but rather children are continuously developing these skills throughout all speaking, listening, reading, and writing activities.

Although the 'Reading Rope' model unpacks the processes involved in both word recognition and language comprehension in more detail than the SVR, some have argued that the model still does not accurately represent all constructs involved in skilled reading. For example, Duke and Cartwright (2021) argued that the model does not reflect recent science of reading research which has demonstrated the role of factors such as theory of mind, engagement and motivation, morphological awareness, and executive function skills on reading. In addition, the 'Reading Rope' does not indicate that any skills across word recognition and language comprehension are overlapping or shared, only that the skills are intertwined in later reading development. For instance, 'vocabulary' is listed only in language comprehension, though is known to influence word recognition skills (Mitchell & Brady, 2013).

Figure 1.3 *Scarborough's Reading Rope (based on Scarborough, 2001, p. 98)*

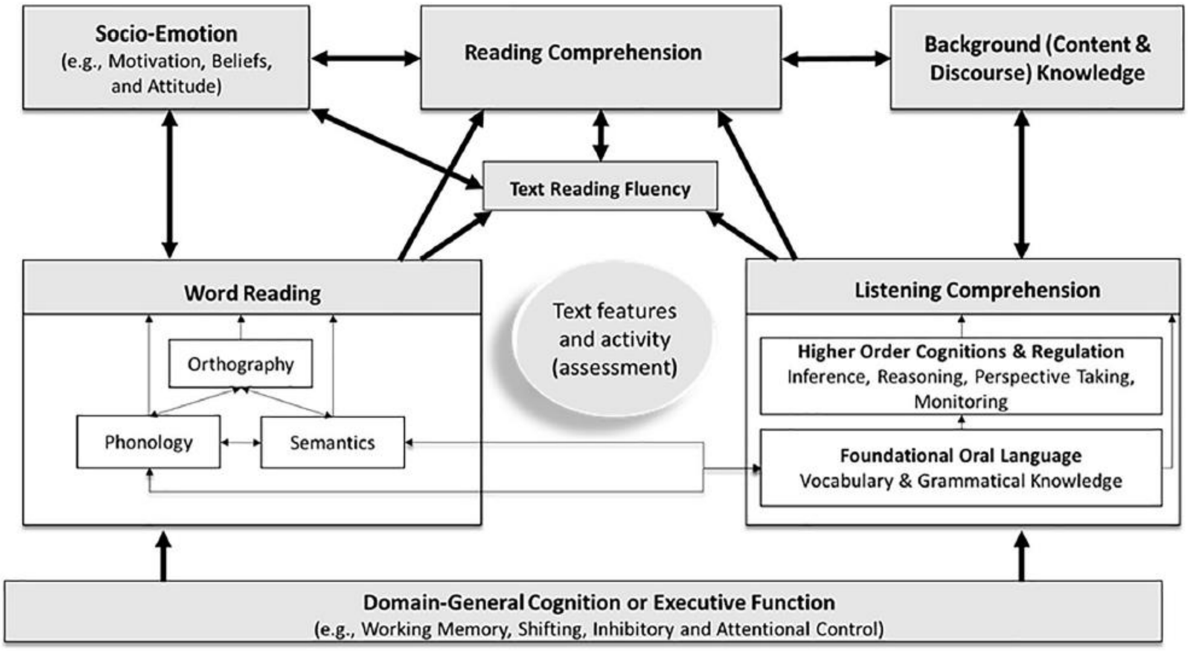


1.4.4 Direct and Indirect Effects Model of Reading

The direct and indirect effects model of reading (DIER) is a more comprehensive model for understanding the component skills involved in skilled reading (see Figure 1.4). This model integrates the broad skills identified in the SVR with additional language, cognitive, and socio-emotional skills that are necessary for reading comprehension (Kim, 2017, 2020b). In comparison with the aforementioned models, the DIER model specifies the relationship between the component skills more explicitly. This includes a hierarchical relationship between component skills, where some skills contribute directly to reading comprehension (e.g., word reading and listening comprehension), whilst others have an indirect effect (e.g., attention and working memory; Kim, 2020b). In this model, reading fluency mediates the effects of word recognition and listening comprehension on reading comprehension. Word recognition is comprised of component skills, including orthography, phonology, and semantics. Listening comprehension comprises foundational language skills (i.e., vocabulary and morphosyntax) which support the use of higher order language skills (e.g., inferencing, theory of mind, comprehension monitoring). These component skills all indirectly contribute to reading comprehension via word reading and listening comprehension abilities. Cognitive functions (e.g., attentional control and working memory) are foundational to both word reading and listening comprehension skills and thus also have an indirect effect on reading comprehension. The skills have a hierarchical effect on reading comprehension, for example, cognitive functions predict foundational language skills, which predict higher order language skills, which then predict listening comprehension, and in turn, reading comprehension.

The DIER model also specifies a dynamic and interactive relationship between component skills, whereby skills develop through interaction with each other and environments, and use of skills is influenced by factors such as the task and text complexity. For example, texts with more complex orthography, language, and cognitive demands will require use of different component skills than simpler texts. Component skills develop through interaction with each other, where increased experience reading results in more expert development of skills. As with the SVR, the DIER model recognises that the role of various component skills in reading comprehension changes as an individuals' reading proficiency develops. The DIER model also includes socio-emotional factors, such as motivation, anxiety, and self-efficacy, as well as background knowledge, as having an indirect effect on reading comprehension. Such socio-emotional factors contribute to the interactive relationship between skills. For example, children who are motivated to read are likely to read more often, facilitating greater reading skill and leading to further motivation to read. Components of the DIER model are based on the extensive research literature which demonstrates the role of the included skills in reading comprehension. The validity of the model has been shown through empirical studies involving typically developing children only (e.g., Kim, 2017, 2020a, 2020b). While the DIER model thoroughly describes the processes involved in skilled reading, the model is more limited in its practical use for informing reading assessment, instruction, and intervention for struggling readers (Duke & Cartwright, 2021).

Figure 1.4 *Direct and Indirect Effects Model of Reading (Kim, 2020b, p. 470)*



Note. Figure included with permission: Kim, Y.-S. G. (2020b). Toward integrative reading science: The Direct and Indirect Effects model of Reading. *Journal of Learning Disabilities*, 53(6), 469-491. https://doi.org/10.1177/0022219420908239.

1.5 Evidence-Based Literacy Instruction

The theoretical models outlined above address the many components involved in reading; however, these theories do not describe the pedagogical approaches that should be used to teach children the complex skill of reading. For this we look to large-scale national enquiries on literacy instruction from the United States (NICHD, 2000), the United Kingdom (Rose, 2006), and Australia (Rowe, 2005). These enquiries have provided strong scientific consensus for five key skills that are essential for early reading instruction: phonemic awareness, phonics, reading fluency, vocabulary, and reading comprehension (see Table 1.1). Instruction incorporating all five of these skills (the Big Five) is known as comprehensive reading instruction and is widely regarded as the gold standard for early literacy instruction for any beginning reader (e.g., Castles et al., 2018; Snow & Juel, 2005).

Table 1.1 *Big Five Components of Comprehensive Literacy Instruction*

Component	Description	Examples
Phonemic Awareness	The ability to identify and manipulate individual sounds in spoken words.	○ Segmenting words into individual sounds. ○ Blending sounds to create a word. ○ Deleting sounds in words to make a new word.
Phonics	Understanding letter-sound correspondences to read and spell words.	○ Systematic synthetic phonics instruction. ○ Teaching grapheme-phoneme correspondences, including decoding and encoding.
Reading Fluency	Reading accurately with appropriate speed and expression.	○ Repeated readings of a text with guidance and feedback. ○ Frequent reading practice.
Vocabulary	Understanding the meaning of words and how words are used in different contexts.	○ Explicit pre-teaching of vocabulary prior to reading. ○ Word analysis (e.g., word roots or affixes). ○ Inferring word meanings from texts.
Reading Comprehension	Understanding and making meaning from texts, including using specific strategies to support understanding.	○ Explicit comprehension strategies e.g., comprehension monitoring, summarising, using graphic organisers. ○ Cooperative learning.

The United States National Reading Panel (NRP; NICHD, 2000) was the first of the national enquiries to explore reading instruction and employed separate subcommittees to examine the available research for each of the Big Five components. Each of these five subcommittees made use of different inclusion criteria when conducting their search of the available research. As a result, children with disabilities were inconsistently included in the research studies contributing to the NRPs recommendations. Across the subgroups, children with cognitive disabilities were included in phonemic awareness instruction and children with learning disabilities or autism were included in some reading fluency studies. Yet, children with neurological, behavioural, or emotional disorders were excluded from research on phonics instruction. The comprehension subgroups (vocabulary and text comprehension) stated that:

> The Panel did not focus on special populations such as children whose first language is not English and children with learning disabilities. It did not review the research evidence concerning special populations and thus cannot say that its conclusions are relevant to them. (NICHD, 2000, p. 4-2)

As such, the recommendations of the NRP and the national enquiries that followed were largely based on research involving typically developing children, and children with developmental disabilities or special educational needs were not explicitly included in these recommendations or the supporting research.

Many have advocated for children with developmental disabilities to have access to this same comprehensive literacy instruction, noting that these children can make progress with reading when provided with instruction incorporating the Big Five (e.g., Arciuli & Bailey, 2021; Browder et al., 2009). However, research on comprehensive literacy instruction for children with disabilities is severely lacking in comparison to research involving children who are typically developing (e.g., Allor, Mathes, Roberts, Cheatham, et al., 2010; Bailey & Arciuli, 2020; Browder et al., 2006; Stauter et al., 2017). Comprehensive literacy instruction for children with developmental disabilities is explored further in the sections below.

1.6 Developmental Disabilities

Developmental disabilities are a group of conditions resulting from impairments in cognition, language, behaviour, or physical abilities during the developmental period (Zablotsky et al., 2019). These conditions are permanent and lifelong and affect approximately 13-17% of children in the United States, with this number increasing significantly over the last 15 years (Boulet et al., 2009; Zablotsky et al., 2019). In Australia, approximately 8% of children have a disability (Australian Institute of Health and Welfare, 2022). In 2023, this included almost 100,000 children under seven years of age accessing the National Disability Insurance Scheme (NDIS), with 11% of all boys and 5% of all girls between 5-7 years of age accessing the scheme (National Disability Insurance Agency, 2023).

This thesis focuses on three prevalent developmental disabilities: cerebral palsy (CP), Down syndrome, and autism. CP is the most common physical disability in childhood, occurring in approximately two per 1,000 live births in Australia (Australian Cerebral Palsy Register Group, 2016; Sellier et al., 2020). Down syndrome is the most common genetic cause of cognitive impairment, affecting one in 1,158 live births in Australia (de Graaf et al., 2022). Autism is the most common developmental disability amongst children in Australia and affects approximately 1% of the population worldwide (Australian Bureau of Statistics, 2018; Baxter et al., 2015). Children with these developmental disabilities may experience cognitive, language, behavioural, and physical impairments that can adversely impact literacy development; however, they do not necessarily prevent acquisition of literacy skills. Children with CP, Down syndrome, or autism are more likely to experience poorer literacy and academic outcomes than their typically developing peers (Nation et al., 2006; van Bysterveldt & Gillon, 2014; Wotherspoon et al., 2023). The sections below outline: (a) each developmental disability in more detail, including defining characteristics and how these may impact literacy skill development; (b) the literacy profiles of children with each developmental disability; and (c) literacy instruction methods have been used with each group.

1.6.1 Cerebral Palsy

CP is a heterogeneous condition that affects muscle coordination and control, caused by a disturbance to the developing brain, that is frequently accompanied by associated impairments, including impairments in cognition, communication, and sensation (Rosenbaum et al., 2007). This well accepted definition of CP includes activity limitations and co-occurring impairments as core to the diagnosis of CP, highlighting the widespread impact these secondary impairments can have on an individual. Co-occurring impairments may influence participation as well as educational, vocational, and social outcomes (Bourke-Taylor et al., 2018; Koppenhaver et al., 1991; Raghavendra et al., 2012; Wotherspoon et al., 2023).

Motor impairments, which are a core feature of CP, and can directly and indirectly impact children’s literacy development. For instance, motor disturbances may impact the muscles involved in coordinating eye movements and the muscles used for speech (Lampe et al., 2014; Mei, Reilly, et al., 2020), both of which can adversely impact learning to read. Motor impairments can also result in activity limitations which can affect children's early literacy experiences and access to books, writing materials, and other alphabetic materials. Communication difficulties can further impact literacy development and are particularly complex for children with CP as they may be on the background of motor impairments, cognitive-linguistic differences, or social influences. When considering communication for children with CP, it is important to differentiate between speech, receptive language, expressive language, functional communication, vision and hearing, and cognition (Geytenbeek, 2016). Approximately 80% of children with CP have speech difficulties (Mei, Reilly, et al., 2020), 60% of children have a receptive and/or expressive language impairment (Mei et al., 2016; Vaillant et al., 2023), 55-70% have impaired functional communication (Himmelmann et al., 2013; Kristoffersson et al., 2020), more than 34% experience impairments in vision (Heydarian et al., 2022), 40% have a hearing impairment (Weir et al., 2018), and approximately 50% have an intellectual impairment (Novak et al., 2012; Wotherspoon et al., 2023). Approximately 25% of children with CP do not use speech to communicate and may use augmentative and alternative communication (AAC) methods (Novak et al., 2012). Literacy and AAC is discussed further in Section 1.6.6.3 of this Chapter. Communication difficulties are most prevalent in children with more severe motor and cognitive impairments (Mei, Fern, et al., 2020; Voorman et al., 2010). In addition, children with CP are also more likely to experience impairments with executive function, working memory, and attention (Critten et al., 2023; Micheletti et al., 2023; Sakash et al., 2018). As noted in the theoretical models outlined earlier, such language and cognitive functions play a foundational role in skilled reading.

1.6.1.1 What are the Literacy Skills of Children with CP? As a group, children with CP experience poorer literacy outcomes than their typically developing peers. Whilst some individuals achieve advanced literacy skills, many do not reach their full literacy potential for a range of reasons. Several studies have found that even when a child's intellectual abilities are within the average range, children with CP are still more likely to experience reading and learning difficulties than their peers (Critten et al., 2019, 2023; Micheletti et al., 2023; Wotherspoon et al., 2023). For example, Wotherspoon et al. (2023) explored the cognitive and academic performance of 93 children with CP from 8 to 12 years of age. Word reading and spelling scores on standardised measures were significantly below average for all children with CP. This remained consistent even when only children with intellectual abilities within the average range were included in the analysis. Micheletti et al. (2023) found that in a sample of children with CP aged 7 to 16 years who communicated using speech, had more mild motor impairments, and had intelligence within the average range, 45% presented with reading disorders. Similarly, Critten et al. (2019) found that in a sample of 15 children with CP aged from 6 to 11 years whose language skills were within the average range, only five children presented with reading and spelling skills that were age-appropriate. Critten et al. (2019) and Micheletti et al. (2023) included only children who used speech to communicate and Wotherspoon et al. (2023) included primarily children who communicated using speech.

Studies including nonspeaking children have found an even more concerning disparity in reading skills for children with CP (Dahlgren Sandberg, 2006). For example, Larsson et al. (2009) explored the literacy skills of 28 nonspeaking children with CP who had severe speech and physical impairments (aged 5-13 years) and 28 typically developing children (aged 4-10 years), matched for receptive vocabulary. Overall, the children with CP in this study scored lower than the typically developing children on all reading and spelling measures. However, there were no significant differences between the children with CP and the typically developing children on phonemic awareness tasks. In this study, Larsson et al. (2009) found that phonemic awareness was the strongest predictor of reading and spelling abilities for both children with CP and typically developing children.

Others have identified different predictors of literacy for children with CP when compared to typically developing children. For example, Peeters, Verhoeven, de Moor and van Balkom (2009) conducted a longitudinal study of 52 children with CP and 65 children without disabilities and found that speech production was the primary predictor of word reading ability in children with CP, whilst phonological awareness and phonological short-term memory were the primary predictors for typically developing children. Asbell et al. (2010) found that in a cohort of 41 children with CP and 74 typically developing children, phonemic awareness, receptive vocabulary, and general reasoning skills predicted reading comprehension skills in both the children with CP and typically developing children. However, the impact of phonemic awareness skills on reading comprehension was moderated by age in the typically developing, but not the CP, group suggesting that children with CP may rely on phonemic processing skills for a longer period in their reading development. In this study, fine and gross motor abilities had no statistically significant impact on reading comprehension skills, highlighting that physical ability should not be equated with capacity to develop reading skills.

1.6.1.2 Literacy Instruction for Children with CP. Few studies have explored literacy instruction methods specifically for children with CP. Studies investigating literacy instruction for this group have primarily utilised single-case study designs, many of poor quality, and none have considered the effects of comprehensive literacy instruction. No previously published studies have attempted to synthesise any aspects of literacy instruction specifically for children with CP. Chapter 2 provides a systematic review of studies in this area.

1.6.2 Down Syndrome

Down syndrome, also known as trisomy 21, is a genetic disorder caused by an extra copy of chromosome 21 (Bull, 2020). The presence of this extra chromosome results in intellectual and developmental disabilities and causes certain physical characteristics and other health related issues (Fidler, 2005; Kazemi et al., 2016). Children with Down syndrome typically exhibit relative strengths in receptive language, visual short-term memory, visuospatial processing, and social communication, and relative weaknesses with expressive language, phonological processing skills, and verbal short-term memory (Fidler, 2005; King, Lemons, et al., 2022; Lim et al., 2014; Næss et al., 2011; Versaci et al., 2021). Such difficulties are known to adversely impact literacy development. There is also a high prevalence of sensory impairments, including hearing (over 80%) and vision (55%), as well as motor impairments (Bull, 2020; Grieco et al., 2015). Speech intelligibility can be variable amongst individuals with Down syndrome and many have complex communication needs (Kent & Vorperian, 2013; Versaci et al., 2021; Wild et al., 2018).

Children with Down syndrome typically demonstrate a distinctive behavioural phenotype, including intellectual impairment and difficulties with cognitive functions, such as attention and working memory, as well as increased non-compliant behaviours (Fidler, 2005; Grieco et al., 2015). A behavioural phenotype is a set of cognitive, linguistic, motor, and social behaviours that are more likely to be present in individuals with a certain genetic condition (O'Brien, 2000). However, applicability of a behavioural phenotype to any individual will vary greatly.

1.6.2.1 What are the Literacy Skills of Children with Down Syndrome? Many children with Down syndrome develop at least some reading skills; however, on average, the level of literacy attained is poorer than their typically developing peers. This has been demonstrated across many cross-sectional and longitudinal studies. For example, van Bysterveldt and Gillon (2014) investigated the reading profiles of 77 children with Down syndrome aged between 5 and 14 years. The majority of children in this study could read one or more words in isolation, with phoneme awareness and letter sound knowledge strongly linked to reading performance. Only 25 of these children had the skills to participate in passage-level reading assessment and performed better on passage reading accuracy compared with passage reading comprehension. A longitudinal study by Byrne et al. (2002) followed 24 children with Down syndrome over two years and found that children made steady progress with reading accuracy, but limited progress with reading comprehension over this time. Whilst the children’s word reading accuracy improved over time, it remained significantly below average for their age. In their study, Byrne et al. (2002) found that the language skills of children with Down syndrome were lower than their equivalent reading age, suggesting that the children’s reading skills were more advanced than their language skills. Næss et al. (2012) conducted a meta-analysis of nonword decoding skills in children with Down syndrome, compared with typically developing children. In this analysis, the children with Down syndrome had weaker nonword decoding skills than the typically developing children matched for word recognition skills; however, this difference was not statistically significant. In this meta-analysis, the children with Down syndrome demonstrated poorer vocabulary and phonological awareness skills than the typically developing children, and differences in vocabulary skills, but not phonological awareness skills, predicted nonword decoding skills.

Throughout the literature, children with Down syndrome present with a reading profile of poorer comprehension skills compared with their reading accuracy. However, in line with the SVR framework, reading comprehension can still be predicted by listening comprehension and word recognition skills in this group (Laws et al., 2016). Although reading comprehension skills are likely to be poorer amongst children with Down syndrome, many children do comprehend what they read (Cologon et al., 2011; Groen et al., 2006). However, accurately measuring reading comprehension for children with Down syndrome can be difficult given that many reading comprehension assessments rely on verbal responses and may be measuring expressive oral language, rather than reading comprehension skills (Cologon, 2013).

1.6.2.2 Literacy Instruction for Children with Down Syndrome. In the past, some believed that children with Down syndrome did not benefit from explicit phonics instruction and recommended teaching using a ‘whole word’ approach, whereby children were taught to recognise words by sight. This line of reasoning was based largely on research by Cossu et al. (1993) that investigated the single word reading and phonological awareness skills of 10 Italian children with Down syndrome and 10 younger typically developing children matched for reading ability. This study found that both groups of children were able to read real and nonwords at a similar level, despite the children with Down syndrome performing significantly more poorly on phonemic awareness tasks. Cossu et al. (1993) asserted that the children with Down syndrome’s “gross failure on phonological awareness tasks, has not prevented them from acquiring reading” (p. 134). This study concluded that as some children with Down syndrome can develop advanced reading skills in the absence of phonological awareness skills, reading instruction should focus on reading skills and not phonological awareness. A 'whole word' approach to instruction was further supported by arguments about weaknesses in phonological processing and strengths in visuo-spatial processing for children with Down syndrome (Byrne et al., 2002; Fidler, 2005; Kernan & Sabsay, 1996; Næss et al., 2011). More recent research has challenged these views. There is now general consensus that, while phonological awareness skills may be weaker in children with Down syndrome, phonemic awareness and phonics still play an essential role in reading development and instruction for children with Down syndrome (e.g., Allor, Mathes, Roberts, Jones, et al., 2010; Baylis & Snowling, 2012; Cologon, 2013; Cupples & Iacono, 2002; Lemons & Fuchs, 2010; Lim et al., 2019; Næss, 2016).

Research involving children with Down syndrome has typically focused on instruction targeting one or a few reading sub-skills (Snowling et al., 2008). No published studies have explored literacy instruction incorporating the Big Five for children with Down syndrome, even though this is known to be the gold standard for developing foundational reading skills in typically developing children. Studies exploring individual components of the NRP Big Five in reading instruction for children with Down syndrome have shown that these instruction methods can be effective. For example, studies exploring phonemic awareness and phonics instruction for children with Down syndrome have reported gains in letter-sound knowledge and word reading skills (e.g., Baylis & Snowling, 2012; Burgoyne et al., 2013; Cupples & Iacono, 2002; Goetz et al., 2008; Lemons et al., 2018; Lemons et al., 2012; Lim et al., 2018; Lim et al., 2019), as have studies exploring combined phonics and vocabulary instruction (Burgoyne et al., 2012), and phonics and reading comprehension instruction (Cologon et al., 2011). Most reading studies involving children with Down syndrome have targeted word reading skills, with very few studies explicitly exploring reading comprehension instruction. Some studies exploring literacy instruction for children with intellectual disabilities have included children with Down syndrome and provide results that may be beneficial when considering reading instruction for this group. For example, a systematic review by Joseph et al. (2023) indicated that explicitly teaching reading comprehension strategies to individuals with mild to moderate intellectual disabilities is effective in achieving reading comprehension gains for these children.

Several studies targeting reading sub-skills for children with Down syndrome have explored interventions specifically designed for the behavioural phenotype of these children (King, Lemons, et al., 2022; Lemons et al., 2018; Lemons et al., 2017; Lemons et al., 2015). Within intervention tasks, these studies have reduced demands on expressive language, working memory, and auditory processing skills, and capitalised on strengths in visual short-term memory, visuo-spatial processing, receptive language, and social skills. As a result of interventions designed for the specific phenotype of children with Down syndrome, children across these studies have made modest gains in phonological awareness skills and word reading on measures closely aligned to instruction targets (King, Lemons, et al., 2022; Lemons et al., 2018; Lemons et al., 2017; Lemons et al., 2015). Other studies have explored the use of computer-assisted instruction (CAI) as a mode of instruction delivery to cater to the specific profile of children with Down syndrome. These studies have demonstrated that CAI is effective in improving word reading and vocabulary skills for these children (Felix et al., 2017; Næss et al., 2022; Nakeva von Mentzer et al., 2021).

1.6.3 Autism

Terminology preferences have received increasing attention in autism research (Bottema-Beutel et al., 2021; Bury et al., 2023; Kenny et al., 2016). The term ‘Autism Spectrum Disorder’, used in the Diagnostic and Statistical Manual of Mental Disorders – Fifth Edition (DSM-5; American Psychiatric Association [APA], 2022), is generally rejected by the autism community as overly medicalised, deficit-based, and stigmatising and, as such, is not used throughout this thesis. Terms such as ‘high-‘ or ‘low-functioning’ are also widely rejected by members of the autism community as stigmatising and not accurately reflecting personal experiences and again are not used in this work (Bottema-Beutel et al., 2021; Kenny et al., 2016). While there are differing views on person-first and identify-first language in relation to autism, the decision here is to use both terms in an attempt to respect the varied preferences of the autistic and autism community (Bottema-Beutel et al., 2021; Bury et al., 2023; Kenny et al., 2016; Lei et al., 2021; Monk et al., 2022; Shakespeare, 2018).

Autism is a neurodevelopmental difference that is diagnosed based on an individual meeting characteristics related to specific social and behavioural criteria, including: (a) differences in social communication and social interaction, such as difficulties with social-emotional reciprocity, nonverbal communication, and relationships; and (b) the presence of restricted and repetitive patterns of behaviours or interests, such as repetitive motor movements, intense interests, and unusual reactiveness to sensory input (APA, 2022). These characteristics present early in development and significantly impact an individual's daily functioning. There is a gender bias in receiving an autism diagnosis, with males far more likely to receive a diagnosis than females (three males to every one female; Loomes et al., 2017). This ratio has started to shift in recent years with greater awareness of how autism presents in females (Russell et al., 2022). The presentation of autism is highly heterogeneous and is frequently accompanied by co-occurring difficulties, such as behavioural disorders, intellectual impairment, and language difficulties (APA, 2022; Anderson et al., 2007). Approximately 25-30% of autistic children are nonspeaking or minimally verbal (Anderson et al., 2007; Rose et al., 2016) and mental health conditions such as anxiety, depression, and attention-deficit/hyperactivity disorder (ADHD) are common (Lai et al., 2019). Together, all these factors place children with autism at a greater likelihood of experiencing literacy difficulties.

1.6.3.1 What are the Literacy Skills of Children with Autism? Autistic children demonstrate great variability in their reading skills, with some children demonstrating very poor reading abilities and others demonstrating above average skills (Brown et al., 2013; Liu et al., 2023; McIntyre, Solari, Grimm, et al., 2017). Research has demonstrated that the variance in autistic children's reading abilities can be accounted for by both word reading and oral language comprehension skills, supporting the legitimacy of the SVR with this group (Jones et al., 2009; Liu et al., 2023; McIntyre, Solari, Grimm, et al., 2017; Nation et al., 2006; Norbury & Nation, 2011; Ricketts et al., 2013; Sorenson Duncan et al., 2021). Early research on autism and literacy often focused on ‘hyperlexia’ or ‘precocious reading’ to the exclusion of other aspects of reading (e.g., Cardoso-Martins & Da Silva, 2010; Grigorenko et al., 2003; Newman et al., 2007; Sparks, 2004). The definition of hyperlexia is inconsistent throughout the literature, though typically refers to advanced word reading and decoding skills relative to text comprehension skills (Grigorenko et al., 2003; Nation et al., 2006; Sparks, 2004). There is now greater recognition of variability in autistic children’s word reading and comprehension skills and hyperlexia receives less attention in the autism and literacy research literature than it once did.

McIntyre, Solari, Grimm, et al. (2017) identified four profiles of reading ability in children with autism, including children who: (a) are average readers and score within the average range on reading accuracy, oral language, and reading comprehension measures; (b) are poor comprehenders but whose decoding, phonological processing, and vocabulary skills are within the average range; (c) are poor readers globally; or (d) are very poor readers globally. Children with poor or very poor reading skills globally (i.e., below or extremely below average on word reading accuracy, oral language, and reading comprehension measures) comprised almost 50% of the sample in the McIntyre, Solari, Grimm, et al. (2017) study, with average readers only accounting for approximately one third of the group. Solari et al. (2019) identified similar reading profiles in their longitudinal study of autistic children, though found that these profiles shifted over time. Some children from the ‘poor comprehender’ and ‘poor reader’ groups in this study moved into the ‘average reader’ group over a period of 30 months. This indicates that children’s reading profiles are not fixed and may change if provided with appropriate instruction. Despite this movement, approximately 68% of the sample in Solari et al. (2019) had reading comprehension difficulties and 50% of the sample had difficulties with word reading accuracy at the final assessment timepoint. Liu et al. (2023) identified similar reading profiles in a sample of Chinese speaking children with autism, though identified one additional profile: children with above average reading and language skills. A reading profile consistent with dyslexia (language comprehension skills within the average range but poor word reading accuracy skills; Catts et al., 2003) has not been noted in any studies profiling the reading abilities of children with autism (Liu et al., 2023; Lucas & Norbury, 2014; McIntyre, Solari, Grimm, et al., 2017; Norbury & Nation, 2011).

Autistic children who demonstrate good word reading abilities, but poor reading comprehension have received a lot of attention throughout the extant literature (Brown et al., 2013; Nation et al., 2006; Wei et al., 2015). Across studies, 37-68% of samples of autistic children have demonstrated reading comprehension difficulties (Brown et al., 2013; Jones et al., 2009; McIntyre, Solari, Grimm, et al., 2017; Nation et al., 2006; Norbury & Nation, 2011; Ricketts et al., 2013). Research indicates that reading comprehension difficulties are related to the social communication, cognitive, and behavioural characteristics of autism and are negatively associated with these traits (i.e., children who have more difficulty with social communication, lower cognitive skills and more complex behavioural needs are more likely to have poorer reading comprehension skills; Jones et al., 2009; McIntyre, Solari, Grimm, et al., 2017; Norbury & Nation, 2011; Ricketts et al., 2013; Solari et al., 2019). For example, McIntyre, Solari, Grimm, et al. (2017) found that lower levels of autism characteristics, as measured by the Autism Diagnostic Observation Schedule (ADOS-2; Lord et al., 2012), were associated with higher reading comprehension scores. Given that reading is a method of written communication, it makes sense that this skill would be impacted by social communication abilities. Many of the same aspects of social cognition that place children with autism at greater likelihood for higher-level language difficulties also impact reading comprehension skills. These include cognitive processes associated with verbal reasoning, inferencing, theory of mind, and central coherence (Davidson, 2021; Happé & Frith, 2006; McIntyre et al., 2018; McIntyre, Solari, Gonzales, et al., 2017; Randi et al., 2010; Ricketts et al., 2013). Jones et al. (2009) identified that many autistic children present with poorer reading skills than would be expected based on their intellectual abilities and poor reading comprehension was the most prevalent academic attainment discrepancy amongst children with autism. This suggests that current literacy instruction practices may not be sufficient to support children with autism to reach their potential with reading comprehension.

Studies investigating the reading profiles of children with autism have largely included only children who use verbal speech to communicate and those without intellectual impairment (e.g., McIntyre, Solari, Gonzales, et al., 2017; McIntyre, Solari, Grimm, et al., 2017; Roch et al., 2021; Solari et al., 2019). This is likely due to many reading assessments requiring verbal responses. There is a distinct lack of research on the reading skills of children with autism who do not speak, even though approximately 25-30% of autistic children are nonspeaking or minimally verbal and use AAC methods (Anderson et al., 2007; Rose et al., 2016). Some studies have explored literacy instruction methods for autistic children with limited verbal communication (e.g., Benedek-Wood et al., 2016; Browder et al., 2012; Coleman-Martin et al., 2005; Leytham et al., 2015) and there is growing awareness of the need to focus on supporting these children.

1.6.3.2 Literacy Instruction for Children with Autism. In comparison with research on literacy instruction for other groups of children with developmental disabilities, research on literacy instruction methods for children with autism has explored a wider range of instructional methods and reading skill outcomes. Even still, methods of comprehensive literacy instruction are relatively underexplored with autistic children. Bailey and Arciuli (2020) conducted a systematic review of studies published between 2009 and 2017 that investigated literacy instruction methods for children with autism that were consistent with the NRP recommendations. They identified only three studies exploring comprehensive reading instruction (i.e., studies that incorporated the Big Five). This systematic review was a follow-up study to a review by Whalon et al. (2009) who investigated studies published prior to 2009. Only two studies exploring multicomponent instruction (targeting both reading accuracy and reading comprehension skills) were identified in the Whalon et al. (2009) review. Across the studies identified in these reviews and additional recent empirical studies, research has shown that autistic children can make gains when provided with instruction that is evidence-based for typically developing readers (e.g., Arciuli & Bailey, 2019; Bailey et al., 2017; Grindle et al., 2013; Kamps et al., 1994). .

Much of the research on literacy instruction for children with autism has focused on developing children's reading comprehension skills, and several reviews have specifically examined methods of reading comprehension instruction for this group (e.g., Conner, Allor, et al., 2022; El Zein et al., 2014; Finnegan & Mazin, 2016; Randi et al., 2010; Senokossoff, 2016; Singh et al., 2021; Tárraga-Mínguez et al., 2020). These studies have found that practices consistent with evidence-based reading comprehension instruction for children with typical development are also effective for autistic children. Some of these practices include, visually cued instruction (e.g., graphic organisers), metacognitive strategy instruction (e.g., question generation, anaphoric cueing, comprehension monitoring, predicting, summarising), and cooperative learning. Whalon (2018) described important instructional components designed specifically for autistic children that have been used throughout the literature 'before', 'during', and 'after' reading a text within reading comprehension instruction (e.g., scripts to generate questions, prompting hierarchies, task completion checklists, visual supports). According to Solis et al. (2016) it is these additional behavioural strategies and supports that improve reading comprehension performance over simply reading comprehension instruction alone for children with autism. Some studies have incorporated additional elements into instruction that are specifically designed to support the needs of autistic children, including integrating special interests into texts (Marshall & Myers, 2021; Solis et al., 2016; Solis et al., 2021), supporting social-communication skills during reading instruction (Kamps et al., 1994; Reutebuch et al., 2015; Whalon & Hanline, 2008), utilising technology in instruction (Khowaja et al., 2020), and incorporating visual scaffolds and supports (Solis et al., 2016; Stringfield et al., 2011; Whalon et al., 2015).

1.6.4 Dual Diagnoses

Although CP, Down syndrome, and autism are explored separately in the coming chapters, these diagnoses are not mutually exclusive as neurodevelopmental disorders often co-occur in individuals (APA, 2022; Lingiardi & McWilliams, 2017). For example, ADHD co-occurs in approximately 28% individuals with autism (Simonoff et al., 2008), 19% to 35% individuals with CP (Craig et al., 2019), and up to 34% of individuals with Down syndrome (Oxelgren et al., 2017). This section, however, focuses on the co-occurrence of the developmental disabilities that are central to this thesis (i.e., CP, Down syndrome, and autism). Until recently, co-occurrence of developmental disabilities, such as Down syndrome and autism, had received little attention in the literature and consequently clinical practice. Recognition that individuals may present with more than one developmental disability is slowly gaining attention, in part due to greater awareness and more accurate assessment and identification of individuals with more than one disability. Research suggests that approximately 5% to 37% of children with Down syndrome meet diagnostic criteria for autism (DiGuiseppi et al., 2010; Moss et al., 2012; Warner et al., 2014). A similar prevalence is seen amongst children with CP, with approximately 3% to 30% of children with CP also having an autism diagnosis (Craig et al., 2019; Påhlman et al., 2020). These rates are far higher than estimates of autism in the general population (approximately 1%; Australian Bureau of Statistics, 2018; Baxter et al., 2015). There is no published work on the co-occurrence of Down syndrome and CP, though anecdotally this does occur.

Dual diagnosis of autism with other developmental disabilities typically occurs much later than diagnosis of autism in isolation (i.e., children typically receive an isolated autism diagnosis between 3 to 5 years of age, yet the mean age of a dual CP and autism diagnosis is 6 to 7 years and a dual Down syndrome and autism diagnosis is 14 years; Leader, Hogan, et al., 2022; Leader, Mooney, et al., 2022; Rasmussen et al., 2001; Reilly, 2009; Smile et al., 2013). Due to the physical characteristics associated with CP and Down syndrome, children typically receive these initial diagnoses at a much earlier age (Bull, 2020; Te Velde et al., 2021). Reasons for delayed dual diagnosis of autism and Down syndrome or CP are varied, though typically autism characteristics may be confused with overlapping behavioural features of the original disability diagnosis (Smile et al., 2013; Versaci et al., 2021). In addition, gold standard diagnostic methods for autism are often not suitable for individuals with vision, hearing, or motor impairments which can further delay dual diagnoses. Delay in accurate diagnosis consequently impedes access to targeted early interventions, which can negatively impact development and longer-term outcomes.

Accurate identification of dual diagnoses is essential as the features and pattern of impairments and support needs seen in individuals with dual diagnoses is distinct from what is observed in the developmental disabilities in isolation. For example, children with co-occurring Down syndrome and autism present with atypical autism characteristics compared to those with an isolated autism diagnosis (Warner et al., 2014). These children tend to have less severe social-communication impairments, potentially due to relative strengths in social skills associated with the Down syndrome phenotype (Godfrey et al., 2019; Hamner et al., 2020; Warner et al., 2014). However, children with a dual autism and Down syndrome diagnosis are more likely to present with emotional and behavioural challenges than children with a Down syndrome only diagnosis (Warner et al., 2014). Children with a dual autism and Down syndrome diagnosis also tend to have lower verbal and nonverbal cognitive abilities than children with either diagnosis in isolation, including poorer language skills, and are less likely to communicate using speech (Hamner et al., 2020; Versaci et al., 2021; Ward & Sanoudaki, 2021; Warner et al., 2014). Similar atypical autism diagnostic characteristics can be seen in children with co-occurring CP and autism. For example, these children are likely to present with higher adaptive behaviour abilities than children with autism alone, though present with similar social-communication skills to children with autism only (Leader, Mooney, et al., 2022). Children with dual CP and autism diagnoses are more likely to present with communication difficulties, cognitive impairment, and aggressive behaviours than children with a CP only diagnosis (Smile et al., 2013).

In summary, greater awareness of co-occurring developmental disabilities is critical as earlier identification results in access to targeted interventions and better outcomes. To our knowledge, no studies have explored the literacy skills of children with developmental disability dual diagnoses or literacy instruction for these individuals. This is an important area for future research.

1.6.5 The Simple View of Reading and Children with CP, Down syndrome, or Autism

The SVR has been well documented to explain variation in reading comprehension skills amongst typically developing children (e.g., Hoover & Tunmer, 2018; Nation, 2019). Across the literature, the validity of the SVR has also been shown for children with CP, Down syndrome, or autism (e.g., Asbell et al., 2010; Laws et al., 2016; Norbury & Nation, 2011; Roch et al., 2021; Roch & Levorato, 2009). However, for some groups, additional factors may contribute to reading comprehension in addition to the SVR components (i.e., word recognition and oral language comprehension). For autistic children, social skills and theory of mind have been found to contribute to reading comprehension above and beyond what is predicted by the components of the SVR (Davidson, 2021; Dore et al., 2018; McIntyre et al., 2018; Norbury & Nation, 2011; Ricketts et al., 2013; Tong et al., 2020). Such factors have not been found to impact reading comprehension for children with Down syndrome or CP. For children with CP, a study by Asbell et al. (2010) indicated that verbal expressive language may independently contribute to reading comprehension, though this is mediated by phonemic awareness.

Despite the differences and heterogeneity in reading profiles across each of the groups, studies involving children with CP, Down syndrome, or autism suggest that variability in reading comprehension is largely explained by differences with oral language comprehension, rather than word reading skills. For example, Roch et al. (2021) examined the facets of the SVR amongst three groups: typically developing children, children with Down syndrome, and children with autism. Results of this study indicated that outcomes on reading accuracy and listening comprehension measures predicted reading comprehension for typically developing children, whereas only the listening comprehension measures predicted reading comprehension for children with Down syndrome or autism. Furthermore, the children with Down syndrome or autism demonstrated poorer reading comprehension skills than would be anticipated based on their vocabulary and word reading skills. This is consistent with a 'poor comprehender' profile in the SVR. Across the literature, children with any of the focus developmental disabilities of this thesis are more likely to fall within the 'poor comprehender' (good word recognition and poor comprehension skills) profile (Dorman, 1987; McIntyre, Solari, Grimm, et al., 2017; Nash & Heath, 2011; Roch et al., 2021; Roch & Levorato, 2009; Solari et al., 2019; Tong et al., 2020). The component skills that contribute to language comprehension (e.g. vocabulary, language structures, verbal reasoning) are the most important predictors of reading comprehension for children with CP, Down syndrome, or autism (Asbell et al., 2010; Dorman, 1987; Lucas & Norbury, 2014; Norbury & Nation, 2011; Roch et al., 2021; Roch & Levorato, 2009).

1.6.6 Additional Factors Impacting Literacy for Children with CP, Down Syndrome, or Autism

Whilst children with CP, Down syndrome, or autism all have unique profiles and support needs, there are many important commonalities that can impact literacy development. For instance, children with any of these developmental disabilities are likely to have associated complex medical issues which can mean that these children may have more school absences (due to extended hospitalisations or frequent specialist appointments) leading to missed learning opportunities. Additional factors are outlined in the sections below.

1.6.6.1 Home Literacy Environments. The home literacy environment typically encompasses literacy materials in the home, parents’ attitudes towards literacy, and the frequency and nature of literacy activities (Biggs et al., 2023). It is well recognised that the home literacy environment influences early literacy development for children. Many studies have suggested that children with developmental disabilities have access to similar literacy materials and opportunities in the home environment as typically developing children (e.g., Biggs et al., 2023; Dynia et al., 2014; Justice et al., 2016; Peeters, Verhoeven, van Balkom, et al., 2009; Ricci, 2011). However, these same studies indicated that some characteristics of shared reading in the home environment may differ for children with developmental disabilities, such as shorter shared reading duration (Lucas & Norbury, 2018), fewer books per reading session (Ricci, 2011), and fewer word orienting activities during book reading (Peeters, Verhoeven, van Balkom, et al., 2009). Differences in home literacy environments are not only observed between typically developing children and children with disabilities, but also between children with different developmental disability diagnoses. For example, Westerveld and van Bysterveldt (2017) compared the home literacy environments of preschool-aged children with Down syndrome or autism. This study found that while both groups had access to similar literacy resources in the home, parents of children with Down syndrome read significantly more frequently with their child than parents of children with autism did. Parents of children with Down syndrome in this study also reported higher child interest in reading, though parents of autistic children pointed out signs and words in the environment to their child more frequently.

For children with developmental disabilities, child characteristics appear to influence the nature of literacy activities in the home more so than for typically developing children. Peeters, Verhoeven, van Balkom, et al. (2009) found that children with CP with lower speech intelligibility had less active participation in word-level activities (such as naming pictures or letters in words) and children with lower fine motor skills were less actively engaged in story orientation activities with their parents. These distinctions did not exist amongst the typically developing sample in this study. Studies involving children with Down syndrome have similarly found differences in home literacy environments based on child characteristics (e.g., Ricci, 2011), as have studies of children with autism. In a study involving school-aged children with autism and their typically developing peers, Lucas and Norbury (2018) found that parents of children with autism and co-occurring language disorders spent more time on shared reading activities and discussing reading than did parents of children with autism who had age-appropriate language skills. Across the literature, parents of children with CP, Down syndrome, or autism viewed literacy as important and attempted to provide literacy rich home environments; however, the frequency and nature of literacy activities varied and was impacted by child and disability characteristics (Barton-Hulsey et al., 2022; Biggs et al., 2023; Dynia et al., 2014; Peeters, Verhoeven, van Balkom, et al., 2009; Trenholm & Mirenda, 2006).

1.6.6.2 Sleep, Pain, and Fatigue. Sleep plays an important role in maintaining health and wellbeing and is essential for learning and memory (El-Sheikh & Sadeh, 2015). Children with CP, Down syndrome, or autism are all more likely to experience sleep disorders than typically developing children. Approximately 23% of children with CP experience pathological sleep disorders (Novak et al., 2012), up to 65% of children with Down syndrome have sleep problems (Hoffmire et al., 2014), and approximately 45% of children with autism have sleep problems significant enough to disrupt family life (Hirata et al., 2016; Maskey et al., 2013). Sleep disturbances amongst these children are typically the result of co-occurring medical, physiological, or behavioural conditions. Such disturbances can negatively impact daily functioning and are associated with increased behavioural problems in children with developmental disabilities (Chawla et al., 2020; Hirata et al., 2016). Amongst typically developing children, poor sleep is associated with poorer academic and cognitive functioning (Blunden et al., 2005; Bourke et al., 2011) and several studies suggest a link between sleep problems and poorer language abilities in children (Bonuck et al., 2021). In studies involving adults, sleep deprivation has been shown to have adverse effects across all cognitive domains and particularly impact performance on tasks that require high levels of attention, working memory, and other executive functions, such as reading (Fostick et al., 2014; Lim & Dinges, 2010). There is currently limited research on the impact of sleep disorders and disturbances on reading and learning for children with developmental disabilities (Chawla et al., 2020). Given the significant impact of sleep on cognitive and behavioural functioning, the high incidence of sleep problems amongst children with disabilities may contribute to their poorer literacy outcomes.

Many children with CP or Down syndrome also experience pain and physical fatigue associated with their motor impairments (Bull, 2020; Novak et al., 2012). Only one study has explored the role of pain and fatigue on academic functioning for children with CP. In this study, Berrin et al. (2007) found a relationship between higher-levels of pain and fatigue and reduced school functioning, based on parent-report. This study did not explicitly explore the impact of pain and fatigue on literacy, nor have any studies involving children with Down syndrome. Recent studies involving children with hearing loss have explored the impact of fatigue on reading outcomes and have found that higher levels of self-reported fatigue are associated with lower scores on reading and spelling measures for this group (Camarata et al., 2018; Werfel & Hendricks, 2016). Potentially, fatigue is playing a similar role in literacy outcomes for children with other developmental disabilities, though this area requires further research.

1.6.6.3 Children who use Augmentative and Alternative Communication. As outlined in Sections 1.6.1 to 1.6.3, many children with CP, Down syndrome, or autism have complex communication needs (are entirely or mostly nonspeaking) and utilise AAC methods. AAC is any form of communication that is used to supplement verbal speech for individuals with speech and/or language difficulties and includes low-tech and high-tech options, such as signing, alphabet boards, picture symbols, and symbol or text-based speech generating devices. Children who use AAC often receive inadequate literacy instruction and as such experience poorer literacy outcomes, with up to 90% not acquiring basic functional literacy skills by adulthood (Dahlgren Sandberg et al., 2010; Foley & Wolter, 2010; Koppenhaver, 2000). These children are provided with fewer opportunities to participate In authentic literacy learning activities and often do not have easy access to their AAC to allow for full and active participation in tasks (Andzik et al., 2018; Sturm et al., 2006). Most AAC systems are based on communication at a whole word or sematic level, yet most early literacy instruction practices require children to verbally produce sounds and parts of words. These children require adapted and individualised literacy instruction that allows them to fully participate. Research on this topic is slowly gaining attention (e.g., see Cheng & Chavers, 2023). A recent systematic review exploring phonemic awareness and phonics-based instruction for children who use AAC reported positive effects for all interventions that were adapted to meet the individual needs of children (Yorke et al., 2021). Barton-Hulsey et al. (2021) reported that AAC systems can be successfully used to support early language and literacy skills in preschool classrooms for children with Down syndrome or autism. However, there is still a long way to go in supporting consistent and meaningful access to foundational literacy learning opportunities in the classroom for children who use AAC and further research is warranted. Literacy instruction for children who utilise AAC is outside the scope of this thesis, though is an important factor impacting literacy outcomes for many children with CP, Down syndrome, or autism.

1.6.6.4 Self-efficacy, Self-concept, and Mental Health.

Research involving typically developing children indicates that reading difficulties can negatively affect children's self-efficacy, self-concept, and mental health (Kargiotidis & Manolitsis, 2024; McArthur et al., 2020). Many studies have demonstrated a statistically significant relationship between reading difficulties and anxiety in children (see systematic review and meta-analysis by Francis et al., 2019). Longitudinal studies suggest that poor reading impacts mental and emotional health for children over time, and not vice versa (McArthur et al., 2022). Many children with developmental disabilities experience mental health conditions, including children and adolescents with CP, Down syndrome, or autism (Downs et al., 2018; Raffaele et al., 2022). For instance, up to 70% of autistic individuals have a co-occurring mental health diagnosis, such as depression or anxiety disorders, and approximately 40% have two or more co-occurring mental health conditions (APA, 2022). Children with disabilities are also more likely to have lower self-concept (McCoy et al., 2016).

Currently, few studies have explored the relationship between self-efficacy, self-concept, mental health, and literacy abilities for children with developmental disabilities. Levy et al. (2013) found that children with ADHD and at least one co-occurring mental health condition, such as depression or generalised anxiety disorder, were more likely to have reading difficulties than children with either ADHD or a mental health condition in isolation. Conversely, Eldblom et al. (2021) found no significant relationship between mental health conditions and word reading skills in adolescents with intellectual and severe developmental disabilities. Investigation of the relationship between self-efficacy, self-concept, mental health, and literacy abilities for children with developmental disabilities is outside the scope of research in this thesis. However, given the relationship between these factors in typically developing children, and the high number of children with disabilities experiencing mental health concerns, this should be considered in future research.

1.7 Social Barriers Impacting Literacy Outcomes

Psychosocial and environmental factors play a key role in literacy development and ultimately attainment of literacy skills for all individuals. For children with developmental disabilities, their experiences of literacy instruction, including the type and quality of instruction, as well as expectations of key stakeholders can be vastly different from those of their typically developing peers. For example, teachers and clinicians may not always utilise evidence-based reading methods when working with children with developmental disabilities and may have lower expectations for their level of literacy attainment (Accardo & Finnegan, 2019; Cologon, 2013; Peeters, Verhoeven, & de Moor, 2009; Ruppar et al., 2011). Poorer literacy outcomes for children with developmental disabilities can at least partially be attributed to these external factors. The sections below explore such barriers that can influence literacy outcomes for children with CP, Down syndrome, or autism.

1.7.1 Stakeholder Expectations

Parents and educators often have different expectations for children with developmental disabilities when compared to their typically developing peers (Shifrer, 2013). For example, Peeters, Verhoeven and de Moor (2009) explored teachers' literacy expectations for children with CP and typically developing children in their first year of schooling. Many teachers reported not knowing what to expect for the future reading and writing abilities of children with CP, whilst holding high expectations for typically developing children. In this study, only 35% of teachers expected that a child with CP would be able to read complex texts in the future and 24% expected their student with CP to read only single letters or words by the end of primary school. Similarly, Conner, Jones, et al. (2022) found that only 75% of teachers believed that their students with intellectual and developmental disabilities could acquire some level of reading ability. The setting in which teachers are based can impact literacy expectations, with teachers working within inclusive settings more likely to believe that children can benefit from literacy instruction than teachers in special education settings (Ruppar et al., 2011). Low expectations can act as a barrier to quality literacy instruction (Trenholm & Mirenda, 2006; Zascavage & Keefe, 2004) and may impact what and how educators decide to teach literacy (Cologon, 2013; Ruppar et al., 2011).

Parental expectations can impact long-term educational outcomes for children with disabilities (Doren et al., 2012; McCoy et al., 2016). A longitudinal study of Australian children with and without disabilities by O'Donnell et al. (2022) explored parental expectations and child school functioning across three timepoints (from 12 to 17 years of age). This study found that higher parental expectations for education at the first timepoint predicted greater academic achievement at the final timepoint for children with a disability, but not for typically developing children. Educational expectations for children with learning difficulties and speech impairments were lower than for children with physical disabilities in this study. Across studies, parental literacy expectations have been linked to children's communication and cognitive skills (e.g., Fleury & Lease, 2018; Peeters, Verhoeven, van Balkom, et al., 2009) and are also influenced by parental socioeconomic status (Bush et al., 2017; Doren et al., 2012; O'Donnell et al., 2022). Lower expectations of children with disabilities can impact the educational opportunities that these children are provided at home (Peeters, Verhoeven, de Moor, van Balkom, et al., 2009; Skibbe et al., 2022), as well as influencing development of autonomy and self-concept (Doren et al., 2012; McCoy et al., 2016), both of which are associated with educational outcomes.

1.7.2 Opportunity Barriers

Children with developmental disabilities often experience stigma regarding their capacity for learning, creating an 'opportunity gap' where they are provided with fewer quality learning opportunities than their typically developing peers (Shifrer, 2013; Wolter, 2016). Whilst children with disabilities spend more time in one-to-one and small group literacy instruction, this instruction is less frequent, involves limited engagement with their peers, is frequently disrupted, and often involves passive participation (Foley & Wolter, 2010; Koppenhaver & Yoder, 1993; Ruppar, 2014). Mike (1995) found that children with CP in a self-contained classroom had significantly less time allocated for literacy activities (30 minutes per day) than would be allocated for typically developing children. These findings, taken together with studies demonstrating that children with developmental disabilities can make reading progress when provided with robust instruction (e.g., Allor, Mathes, Roberts, Jones, et al., 2010; Bailey et al., 2017), suggest that poor reading outcomes for these children may be a result of limited learning opportunities, not limited capacity for learning.

Stigma and discrimination within the education setting, resulting in access to a less ambitious curriculum, can be a contributing factor to the poorer literacy outcomes experienced by children with disabilities (Shifrer, 2013; Zascavage & Keefe, 2004). These children are often excluded from high-quality literacy instruction and have reduced access to rich literacy experiences. Many have identified lack of inclusion in the general education curriculum as a barrier to evidence-based literacy instruction (e.g., Foley & Wolter, 2010; Zascavage & Keefe, 2004). In a survey of special education teachers by Ruppar et al. (2011), teachers reported preferring to teach literacy related to life-skills or 'functional literacy', rather than teaching foundational literacy skills (such as phonics) for children with disabilities. According to Zascavage and Keefe (2004):

The goal of functional literacy is to be able to understand and use printed material in daily activities of living. The educator assumes that students with disabilities will have no use for the traditional literacy curriculum. An attitude that marginalizes and devalues the potential of an individual with a disability. (p. 232)

Inclusive education is one factor impacting access to quality literacy instruction for children with developmental disabilities. The definition of inclusive education has shifted over time from an initial focus on location (i.e., children with disabilities learning in the same place as children without disabilities), to a focus on providing the same high-quality evidence-based instruction, consisting of appropriately challenging content, to children with disabilities (Shurr et al., 2023; Wehmeyer, 2006). There has been a significant increase in the number of studies published on inclusive education since 2017 (Shurr et al., 2023), potentially reflecting increased expectations for students with disabilities. Universal Design for Learning (UDL) is one method of facilitating inclusion and access to the curriculum for all children. UDL is an educational framework that emphasises designing flexible and inclusive instructional materials and methods that accommodate diverse learner needs, preferences, and abilities (Capp, 2017). These principles allow children with diverse needs to receive information and demonstrate their knowledge in multiple ways. Technology in instruction can be one feature of UDL that can promote access; however, research indicates that stigma around assistive technology and lack of adequate training for educators means that technology is not always available to those who need it most (Barton-Hulsey et al., 2022; Parette & Scherer, 2004; Zascavage & Keefe, 2004). For example, Barton-Hulsey et al. (2022) found that children with developmental disabilities with lower verbal speech skills were given less access to technology for literacy instruction than children with greater speech ability.

1.7.3 Educational Policies

Children with disabilities have long been ignored in policies surrounding literacy instruction. Educational legislation in Australia is clear that education providers must make reasonable adjustments to ensure that children with disabilities can access the curriculum and participate in educational activities on the same basis as their peers without a disability (based on the *Disability Standards for Education 2005*, a subordinate legislation to the *Disability Discrimination Act 1992*). Yet, influential reports relating directly to reading instruction have largely excluded these children. Children with disabilities received very little mention in the pivotal United States NRP report (NICHD, 2000), and similar reports from England (Rose, 2006) and Australia (Rowe, 2005) around this time. As previously described, the NRP report stated that they did not review research evidence including children with disabilities, and as such cannot conclude that the recommendations made by the panel are relevant to these children. The Rose report (2006) made more reference to children with special educational needs, and noted that these children would likely require "highly personalised interventions" (p. 46), though referred to other national policies already in place (e.g., the special educational needs code of practice; Department for Education and Skills, 2001) rather than providing specific guidance around supporting reading development for children with disabilities. The *National Enquiry into the Teaching of Literacy in Australia* (Rowe, 2005) did not make explicit mention of children with disabilities within their recommendations, other than to note the diversity of children within Australian schools, all of whom are expected to learn to read and write. It is not surprising then that many teachers report feeling unsure and underprepared for how to support children with disabilities to learn to read in the classroom (Accardo & Finnegan, 2019; Cologon, 2013; Conner, Jones, et al., 2022). In recent years, policies around teaching reading have become more inclusive (e.g., *The Reading Framework* by the UK Department for Education, 2023). These recent policy changes will be explored in Chapter 6 in light of the findings from the empirical studies in this thesis.

1.8 Literacy Instruction Methods

Literacy instruction can encompass a diverse range of methods and modalities, all designed to foster effective reading and writing skills. While it is generally agreed that evidence-based literacy instruction is comprehensive in incorporating five key skills (NICHD, 2000); how this instruction is delivered to children can take many forms. Instruction may be delivered at the whole class, small group, or individual level, and may be provided face-to-face or via remote service delivery. Use of technology in reading instruction has become increasingly prevalent and is commonplace in a contemporary classroom environment (McTigue & Uppstad, 2019). These factors are explored in the sections below.

1.8.1 Computer-Assisted Instruction

CAI is the use of computer technology to facilitate interactive and individualised learning experiences. Successful implementation of CAI is not based on the technology itself, but rather, how the technology aligns with pedagogical content and the quality of teaching that it enables. Effective CAI should foster motivation for reading by developing children's reading competence and autonomy (McTigue & Uppstad, 2019). A systematic review by Cheung and Slavin (2013) indicated small positive effects of CAI on the reading accuracy and reading comprehension skills of children with reading difficulties when compared with business-as-usual control groups. In this review, Cheung and Slavin (2013) found that CAI implemented within small-group instruction resulted in the largest effect sizes, though these findings were based on small-scale studies. How CAI for literacy is implemented can significantly impact results, with higher levels of adult-child interaction within CAI typically leading to more positive reading outcomes (Cheung & Slavin, 2013; McTigue et al., 2020; McTigue & Uppstad, 2019). This interaction may support transfer of skills from computer activities to authentic reading contexts.

Several studies have explored CAI within explicit literacy instruction for children with autism (e.g., Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022; Henderson-Faranda et al., 2022; Khowaja et al., 2020), Down syndrome (e.g., Næss et al., 2022; Nakeva von Mentzer et al., 2021), or CP (e.g., Coleman-Martin et al., 2005; Hetzroni & Schanin, 2002; Holyfield et al., 2019). These studies have indicated that technology in instruction can result in reading gains and that many children are able to generalise their skills to non-computerised contexts and maintain these skills post-instruction. Literacy-based CAI for children with developmental disabilities can increase academic engagement and decrease challenging behaviours during learning activities for some children (LeJeune & Lemons, 2021). Some have suggested that multimedia technology is particularly suited to children with developmental disabilities, as providing information via both auditory and visual channels can reduce the cognitive load involved in learning activities (Mayer, 2008; Mayer & Moreno, 2003). In addition, many computer-assisted multimedia literacy programs provide flexibility and personalised learning opportunities which can be adapted to meet a child's individual learning needs (Major et al., 2021). Overall, CAI has the potential to enhance the learning experiences and accessibility of educational materials for children with diverse needs (Capp, 2017; Dalton et al., 2011; Proctor et al., 2011).

1.8.1.1 ABRACADABRA. ABRACADABRA (hereafter referred to as ABRA; Centre for the Study of Learning and Performance [CSLP], 2019) is an interactive multimedia web-based program that is used throughout the empirical studies in this thesis. ABRA comprises modular game-based activities centred around age-appropriate texts for early readers and targets foundational literacy skills in key areas that reflect the NRP Big Five: alphabetics (phonemic awareness and phonics), reading fluency, reading comprehension (including vocabulary), and writing (spelling). ABRA is freely accessible online and has been used globally in both high- and low-resource environments (e.g., Lysenko et al., 2019; Vousden et al., 2022; Wolgemuth et al., 2011). The ABRA program was selected for this research based on several factors, including that: (a) it is evidence-based and has been well researched with typically developing children (Abrami et al., 2020); (b) it has been previously trialled with children with developmental disabilities (specifically children with autism; Arciuli & Bailey, 2019; Bailey et al., 2017); and (c) it is cost effective, making ABRA an equitable option for instruction and intervention (McNally et al., 2016).

Abrami et al. (2020) conducted a meta-analysis of the effects of ABRA on reading outcomes in studies published from 2008 to 2017. They identified 17 high-quality randomised controlled trial or quasi-experimental studies, involving 7,388 children from pre-kindergarten to Grade 3. Across the studies, children received between 3.5 to over 30 hours of instruction over 8 to 30 weeks, in a one-to-one, small group, or whole class setting. Included studies primarily used standardised reading outcome measures. Overall positive effect sizes were reported in the meta-analysis for phonemic awareness, phonics, reading fluency, vocabulary, reading comprehension, and listening comprehension following ABRA instruction. This analysis found that poorer performing readers and children from economically disadvantaged backgrounds made the largest reading gains following ABRA instruction. One-to-one instruction with Grade 2 students yielded the largest effects, highlighting the importance of a high-quality adult instructor in ABRA implementation. A recent large-scale efficacy trial of ABRA, funded by the Education Endowment Foundation (EEF), included 1,884 mainstream Year 1 students across 48 schools in England (McNally et al., 2016). This trial compared 20 weeks (four x 15-minute sessions per week) of small-group computer-based ABRA or paper-based ABRA (using the same activities and texts) delivered by trained teaching assistants with a business-as-usual control group. Children who received the computer- or paper-based ABRA intervention made 2- and 3-months' additional reading progress respectively when compared with the control group. This reading progress was maintained at one year post-intervention. Again, children from lower socioeconomic backgrounds and with poorer reading skills at pre-assessment made the greatest reading gains. Across this sample, 15% of students had special educational needs, though the impact of intervention for these children is not disaggregated from the larger sample. A follow-up large-scale effectiveness trial, also funded by the EEF, was recently completed

(Bell et al., 2022) and found contrasting results (children who received the paper-based ABRA made an additional 2-months' reading progress, compared with a control group, whilst children who participated in the computer-based ABRA made no additional progress in reading). This study is discussed further in the final chapter of this thesis.

ABRA has demonstrated positive effects on the reading skills of children with autism when delivered on a one-to-one basis (Bailey et al., 2017) and in small groups (Arciuli & Bailey, 2019), but not when delivered via telepractice in a recent study (Bailey et al., 2022). ABRA has not yet been trialled with any other group of children with developmental disabilities. Given the positive effects of ABRA when used with children with autism and other low progress readers (Abrami et al., 2020; Bailey et al., 2017; McNally et al., 2016), and the potential benefits of CAI and multimedia programs for children with developmental disabilities (e.g., LeJeune & Lemons, 2021; Mayer & Moreno, 2003), ABRA warrants further exploration with this group.

1.8.2 Shared Book Reading

Another method of literacy instruction central to this thesis is SBR. SBR is referred to by many names throughout the literature, including dialogic reading, interactive shared reading, or read-alouds, and can encompass a range of behaviours from naturalistic to more structured reading practices (Akemoglu et al., 2020; Justice et al., 2015; Noble et al., 2020; Noble et al., 2019; Pillinger & Vardy, 2022; Swanson et al., 2011; What Works Clearinghouse, 2010). Many definitions for SBR and related practices exist, though there is consensus that SBR typically involves some level of interaction between an adult and child engaged in reading a book together (National Early Literacy Panel [NELP], 2008; Noble et al., 2019). Whilst SBR can describe both a naturalistic activity between an adult and a child and also a mode of instruction or intervention (Biggs et al., 2023), the focus of this thesis is on SBR as an intervention mode.

Research indicates that quality SBR interventions can have positive effects on children's oral language, phonological awareness, and print awareness skills for both typically developing children and children with developmental disabilities (e.g., Boyle et al., 2019; National Early Literacy Panel [NELP], 2008; Pillinger & Vardy, 2022; Sim & Berthelsen, 2014; Swanson et al., 2011; Towson et al., 2021; What Works Clearinghouse, 2010). Much of the literature around SBR has focused on children's oral language outcomes. Yet, a recent meta-analysis of SBR interventions for typically developing children reported only small effects of SBR interventions on language outcomes (Noble et al., 2019). Noble et al. (2019) found that while SBR can have modest positive effects on oral language skills, the effects were non-significant in studies that utilised an active control group (i.e., control groups who engaged in alternative language-based activities). There is no agreement on the optimal dosage of SBR interventions across the literature; though, in a systematic review of interactive SBR interventions, the average dosage was 5 to 15 minutes per day for 6-weeks (Pillinger & Vardy, 2022).

SBR may take place within the home, preschool, school, or clinic environment. As such, SBR interactions may be parent-, teacher-, or clinician-led across contexts (National Early Literacy Panel [NELP], 2008; Roth & Baden, 2001). Much of the literature has focused on parent-led SBR and training parents to deliver SBR programs to their child in the home setting (e.g., Dodge-Chin et al., 2022; Sim & Berthelsen, 2014). These studies have generally reported high fidelity of parent SBR implementation, though mixed findings for child outcomes (e.g., Biggs et al., 2023; Noble et al., 2020; Pierson et al., 2021). Teacher- or clinician-led SBR involves reading with children either individually or in groups and may involve the adult reading the text aloud or shared reading of text between the adult and child (Justice & Pullen, 2003; Westerveld & Gillon, 2008; Yorke et al., 2018). Teacher-led SBR is an important part of classroom literacy instruction. In one study, teachers reported that students engaged in SBR to develop their word-level reading skills at least four times per week in first grade and two to three times per week in third grade (Sturm et al., 2006). Clinician-led SBR, when fastidiously implemented, can provide a dynamic intervention context that can be adapted to suit a child's communication, language, and literacy goals (Roth & Baden, 2001; Westerveld & Gillon, 2008; Yorke et al., 2018).

Research on SBR for children with developmental disabilities has primarily focused on preschool-aged children (e.g., Akemoglu et al., 2020; Fleury et al., 2014; Jeremic et al., 2023; Towson et al., 2021; Westerveld et al., 2021). Much less attention has been given to SBR methods for school-aged children (Biggs et al., 2023). Research has primarily focused on developing oral language and comprehension related skills, and very few studies have explored how SBR can be used to support print related skills for children with developmental disabilities. This is despite both meaning and print-based knowledge being essential for skilled reading (Gough & Tunmer, 1986). A recent scoping review of home-based SBR interventions for children with intellectual and developmental disabilities found that only 20% of studies aimed to increase parent's use of print-related strategies, while 80% focused on parent's use of language or meaning-based strategies (Biggs et al., 2023). Children with Down syndrome, autism, or CP accounted for the majority of children in this scoping review. More research is needed to understand how SBR can be best utilised with children with developmental disabilities to support both print and meaning-based skill development. In particular, how parents can be better supported to help their child to develop both of these important skills. SBR research and practices for children with CP, Down syndrome, or autism are described in Chapters 3, 4, and 5 of this thesis respectively.

1.8.3 Instruction Modalities

1.8.3.1 Individual and Small Group Instruction. Within the school environment, literacy instruction is typically delivered on a whole-class level. Where children require additional support, small group and individual instruction are provided. Many schools globally and within Australia use a three-tiered response to intervention (RTI) framework (or a Multi-tiered System of Supports [MTSS]) to ensure that timely and appropriate reading intervention is provided to children who require it (Fuchs & Fuchs, 2006; Gersten et al., 2009; Scott, 2023; Siegel, 2020). Tier-1 involves core reading instruction (evidence-based whole class teaching). Most children will develop their literacy skills at this level; however, for children with literacy difficulties Tier-1 instruction only is insufficient (Fuchs & Fuchs, 2006; Torgesen, 2002). In Tier-2, children who are identified as not responding to whole class teaching are provided with additional provisions to support them to catch-up to their peers (typically small-group-based intervention targeting specific skills). This small-group intervention increases practise opportunities and allows for more targeted instruction and specific feedback (Fien et al., 2014; Gersten et al., 2009; Torgesen, 2002). Tier-3 is for children who are not responding to Tier-2 interventions and require more intensive, explicit, and specialised supports, typically delivered at an individual level (Denton et al., 2013; Gersten et al., 2009; Torgesen, 2002). Within a clinical setting, literacy intervention is primarily delivered at the individual or small-group level, or may be provided in an interprofessional consultative or collaborative manner aligned with the three-tiered support framework (American Speech-Language-Hearing Association, n.d.-b; Speech Pathology Australia, 2021). Many children with developmental disabilities require Tier-2 and Tier-3 intervention supports to develop their literacy skills (Denton et al., 2013; Rose, 2006) and, as such, need access to evidence-based and efficacious interventions at the small-group and individual level.

1.8.3.2 Telepractice. Telepractice uses video conferencing and other telecommunication technologies to connect health and/or education professionals with individuals in real-time to deliver consultation, assessment, intervention, or education services (American Speech-Language-Hearing Association, n.d.-a). Different terms have been used throughout the literature to reference these services, including ‘telehealth’, ‘telemedicine’, or ‘telerehabilitation’; however, telepractice is used throughout this thesis in recognition that such services can be used outside of a health care setting (American Speech-Language-Hearing Association, n.d.-a). Clinical guidelines for telepractice exist (e.g., American Speech-Language-Hearing Association, n.d.-a; Royal College of Speech and Language Therapists, 2022; Speech Pathology Australia, 2023). Many of these guidelines were emerging at the time the studies in this thesis took place, though are now a standard part of clinical practice. These guidelines make clear that clinical services provided via telepractice must be equivalent in quality to services that would be provided face-to-face. Large scale studies indicate that speech and language interventions delivered via telepractice are equivalent to services delivered face-to-face for school-aged children

(Coufal et al., 2018; Musaji et al., 2021). Studies involving children with developmental disabilities have also reported that telepractice can be an efficient and cost-effective method of providing services to this group, though advocate that more research is needed (Bekteshi et al., 2022; Valentine et al., 2021).

Literacy instruction has traditionally been delivered face-to-face, though the efficacy of literacy instruction delivered via telepractice has received increasing attention in recent years. This is largely in response to the global COVID-19 pandemic which has brought this service delivery mode into mainstream use (Bolden & Grogan-Johnson, 2022; Hermes et al., 2021). A rapid review by Furlong et al. (2021) explored literacy assessment, instruction, or intervention delivered via telepractice from 2005 to 2020 and identified nine relevant studies, including two studies exploring literacy assessment via telepractice and seven investigating online literacy instruction or intervention. Included studies involved primarily typically developing children (aged 4-19 years), all of whom presented with literacy difficulties at baseline. One study included children with hearing loss and one included children with specific learning disorders; however, no other developmental disabilities were represented across the included studies. Of the seven instruction or intervention studies, two used non-randomised controlled designs, four utilised single-subject experimental designs, and one used a group pre-test/post-test design. Across these studies, sessions were delivered two to four times per week for 30-60 minutes, over 8 to 18 weeks. Six of the seven instruction studies followed programs that included set tasks, and time per task, for each session. Instruction across the included studies targeted reading and spelling skills, including phonological awareness, reading accuracy and fluency, vocabulary knowledge, and reading comprehension, with a variety of standardised assessments used to measure outcomes. Furlong et al. (2021) concluded that online literacy assessment returned similar results to assessments administered face-to-face, and that interventions via telepractice could be feasible and engaging, though more research is needed to draw conclusions regarding the efficacy of such services.

Since this rapid review by Furlong et al. (2021), new studies have emerged exploring literacy assessment and intervention via telepractice (e.g., Collins, 2021; Nelson Nickola & Plante, 2022), including some studies involving children with developmental disabilities (e.g., Bailey & Arciuli, 2022; Henry et al., 2023). For example, Conner, Henry, et al. (2022) explored the feasibility of delivering literacy assessments (oral language, reading, and writing) via telepractice to 13 school-aged autistic children. This study focused on behaviours with the potential to impact assessment outcomes, including parental involvement, child disengagement, and technology issues. The study concluded that while literacy assessments via telepractice for children with autism are feasible, clear guidelines on how best to conduct and adapt assessments via this mode for children with developmental disabilities are needed. Several studies have also explored parent coaching via telepractice to deliver SBR interventions to their child with a developmental disability (Akemoglu et al., 2021; Dodge-Chin et al., 2022; Pierson et al., 2021). These studies have all found that parents can effectively implement SBR programs following coaching via telepractice, though have reported mixed effects on child outcomes (Akemoglu et al., 2021; Dodge-Chin et al., 2022; Pierson et al., 2021). For example, Akemoglu et al. (2021) reported an increase in communicative acts from children following participation in the SBR program, while Dodge-Chin et al. (2022) reported no changes in communication for four out of five child participants. Studies exploring parent coaching in SBR programs via telepractice have all investigated communication and oral language outcomes for children and have not studied explicit literacy skills.

1.9 Significance of thesis

Given that literacy provides a foundation for educational success and contributes to broader quality of life outcomes, there is a pressing need to improve reading outcomes for children with developmental disabilities. The research presented throughout this thesis explores high-quality comprehensive literacy instruction for children with developmental disabilities, a key gap in the current evidence base. Instruction methods used throughout this research are consistent with evidence-based practice for typically developing children and have the potential to significantly increase reading outcomes for children with developmental disabilities. The findings from this thesis can help educators, clinicians, and families to make informed decisions around literacy interventions, potentially increasing the quality of instruction provided to children with disabilities. These findings can also be used to guide policy and practice around reading instruction and intervention for children with diverse needs. In addition, the service delivery modes explored in this thesis have the potential to make instruction more accessible to some children.

1.10 Aims of the Thesis

This research aims to address the broad research question: *Which literacy instruction methods are effective in supporting the literacy skills of children with developmental disabilities?* In order to address this question throughout this thesis, I, together with my supervisors, explore the effects of a freely available evidence-based multimedia literacy web application (ABRACADABRA), supplemented by SBR, on the reading skills of children with three prevalent developmental disabilities. We aim to provide a clearer understanding of effective reading instruction methods and models of service delivery for children with developmental disabilities in order to help guide policies and practice and improve literacy outcomes for these children. By exploring a freely available literacy program, we aim to provide more equitable access to evidence-based literacy instruction for children with disabilities.

1.11 Overview of this Thesis

Chapter 2 presents a systematic review of the literature on literacy instruction for children with CP: *Exploring the Effects of Literacy Instruction for Children with Cerebral Palsy: A Systematic Review*. The three chapters that follow are presented as empirical papers exploring the effects of the ABRACADABRA program on reading outcomes for children with developmental disabilities, including:

- Chapter 3: Empirical study 1 - *Computer-assisted Literacy Instruction via Telepractice for a Child with Cerebral Palsy: A Case Study*
- Chapter 4: Empirical study 2 - *ABRACADABRA Literacy Instruction for Children with Down Syndrome via Telepractice during COVID-19: A Pilot Study*

- Chapter 5: Empirical study 3 - *An Effectiveness Trial of ABRACADABRA Literacy Instruction for Children with Autism during the COVID-19 Pandemic*

Chapters 2 and 4 have been submitted and accepted for publication in the journals *Language, Speech, and Hearing Services in Schools* and the *British Journal of Educational Psychology* respectively. Chapter 5 has been submitted to a peer-reviewed journal and is currently under review. The same methodology was used in the empirical studies presented in Chapters 3 and 4. This PhD was completed during the COVID-19 pandemic and as such the impact of the global pandemic on literacy instruction delivery and outcomes is touched on within each of the experimental studies and is explored in detail in Chapter 6. The final chapter of this thesis, Chapter 6, consolidates and provides a discussion of the thesis findings, including the implications of these findings for educators, clinicians, and policy makers, and recommendations for future research.

CHAPTER 2 SYSTEMATIC REVIEW

Murphy, A., Bailey, B., & Arciuli, J. (2023). Exploring the effects of literacy instruction for children with cerebral palsy: A systematic review. *Language, Speech, and Hearing Services in Schools,* 54(1), 299-321. https://doi.org/10.1044/2022_LSHSS-22-00014

2.1 Abstract

Purpose: Some children with cerebral palsy (CP) have difficulty acquiring conventional reading and writing skills. This systematic review explores the different types of literacy instruction and their effects on the reading and writing skills of children with CP.

Method: Relevant studies published between 2000 and 2020 were identified using electronic databases and terms related to cerebral palsy and literacy. Data on participant characteristics, instruction characteristics, and instruction outcomes were extracted. A standardised measure of effect size was used to quantify reported treatment effects.

Results: The systematic search identified 2,970 potentially relevant studies, of which 24 met inclusion criteria. These studies included 66 children with cerebral palsy aged 5 to 18 years. One of the included studies utilised a group research design while the remaining used single-subject designs. Studies investigated literacy instruction methods designed to teach phonics, sight-word recognition, reading fluency, reading comprehension, spelling, or written expression skills, or multicomponent instruction (instruction methods encompassing three or more of these skills). Most instruction methods were associated with gains in reading and writing skills with medium to large effects; however, our analysis of methodological rigour suggest that these findings need to be interpreted with caution.

Conclusion: We propose that literacy instruction utilising evidence-based principles can be effective for children with CP, provided instruction is accessible and allows children to demonstrate and receive feedback on their skills; however, further research is greatly needed. Clinical implications and priorities for future research are discussed.

2.2 Introduction

Cerebral palsy (CP) is the most common physical disability of childhood and can impact children’s functional abilities in many ways (Rosenbaum et al., 2007; Sellier et al., 2020). Up to 80% of children with CP have communication or speech difficulties, with approximately one in four communicating nonverbally, one in two having an intellectual disability, and one in twenty-five having a severe hearing impairment (Mei, Reilly, et al., 2020; Novak et al., 2012). These cognitive and linguistic factors, along with children’s broader social context and learning environments can all influence literacy development. Literacy is essential for inclusion and access in society (National Commission on Writing, 2004). For individuals with complex communication needs, literacy provides access to independent and meaningful communication (Koppenhaver & Williams, 2010), supports friendships and social participation (Caron & Light, 2016), and facilitates self-advocacy, self-determination, and self-care (Kitson et al., 2021). Yet, literacy instruction practices are often not inclusive of children with severe speech and physical impairments, creating an ’opportunity gap’ where children are provided fewer opportunities to engage in quality literacy instruction than their typically developing peers (Wolter, 2016; Zascavage & Keefe, 2004). In addition, educators, clinicians, parents, and peers may underestimate the capacity of children with CP to acquire literacy skills (Peeters, Verhoeven, & de Moor, 2009). Given these factors, it is not surprising that many children with CP have difficulty achieving a level of literacy required to fully participate in daily life (Koppenhaver, 2000). This paper presents a systematic review on the effects of literacy instruction for children with CP.

2.2.1 Effective Literacy Instruction

Literacy includes both reading and writing. Reading is the ability to decode words and derive meaning from text and writing is the ability to encode and compose messages that express ideas (Department for Education, 2021a). According to the Simple View of Reading (SVR), skilled reading is the product of two distinct abilities, decoding skills and language comprehension (Gough & Tunmer, 1986). It is well known that a child’s reading and writing skills are influenced by many underlying abilities, including early receptive and expressive language skills (Snow, 2020). Moreover, as children's literacy skills develop, there is a reciprocal relationship between oral and written language, where development in one domain results in gains in the other (Adlof, 2019).

There is now a well-established body of research on effective reading instruction for typically developing children, including several large-scale reviews. One such review was conducted by the United States National Reading Panel (NRP; National Institute of Child Health and Human Development [NICHD], 2000) which found that effective reading instruction focuses on five key skills, known as the Big Five: (a) phonemic awareness (ability to hear and manipulate individual sounds in words); (b) phonics (knowledge of letter-sound correspondences and their use to read and spell words); (c) reading fluency (reading with speed, accuracy, and expression); (d) vocabulary (understanding of word meaning); and (e) reading comprehension (use of specific cognitive strategies to increase understanding of what is read). Comprehensive reading instruction involving integration of all five of these skills during explicit instruction is the current gold standard for typically developing children in the beginning years of conventional literacy development.

Research on effective writing instruction is less well established (National Commission on Writing, 2003). Broadly, writing instruction can be grouped into two approaches: (a) writing skills (e.g. handwriting, spelling, grammar); and (b) writing processes (e.g. planning, revising, self-evaluation; Cutler & Graham, 2008; Graham et al., 2002). Currently, the optimal quantity and combination of approaches in writing instruction is unclear based on meta-analyses of writing instruction for typically developing children (e.g. Graham et al., 2012).

Much of the research to date has included only typically developing children and has excluded children with developmental disabilities. There is mounting evidence that literacy instruction embodying the principles above is effective for all beginning readers, including those with developmental disabilities such as autism, Down syndrome, and hearing impairment (Bailey & Arciuli, 2020; Lim et al., 2019; Schirmer & McGough, 2005), but much less is known about children with physical disabilities and/or complex communication needs, such as those with CP.

2.2.2 Cerebral Palsy and Literacy

Many children with CP, including those who communicate verbally and nonverbally, experience some level of literacy difficulty (Beal et al., 2000; Critten et al., 2019). Co-occurring disabilities, such as communication and/or vision impairment, contribute to these group differences but do not fully account for the gap in children's reading and writing skills. Even when children's expressive communication, receptive language, social skills and intelligence are within the average range, some children with CP demonstrate lower literacy levels than their typically developing peers (Beal et al., 2000; Berninger & Gans, 1986; Critten et al., 2019).

Importantly, many children with CP who are delayed in development of emergent literacy skills fall further behind their typically developing peers over time (Dahlgren Sandberg, 2006). For example, Critten et al. (2019) found that for a group of children with CP (*n* =15) and age-appropriate language skills, only one third demonstrated age-appropriate reading and spelling skills. For those children with poorer literacy skills, difficulties were associated with lower phonological awareness and visual-spatial perception skills. Peeters, Verhoeven, de Moor and van Balkom (2009) conducted a longitudinal study involving 52 children with CP and 65 children without disabilities from 5 to 7 years of age. By the second year of schooling, children with CP were behind their peers on early reading measures. Speech production was found to be the most important predictor of reading success for these children, followed by phonological awareness and speech perception. Dahlgren Sandberg (2006) assessed the reading and spelling skills of six children with CP with average intelligence and significant speech and physical impairments at 6, 9, and 12 years of age. These children were not able to read or spell any whole words at 6 years of age despite demonstrating age-appropriate phonological awareness skills. Their reading and spelling skills increased significantly by 9 years of age, following their first three years of formal schooling. Children's word- and passage-level reading accuracy, as well as spelling skills, showed almost no improvement between 9 and 12 years of age.

Access to literacy learning opportunities likely contributes to the reading and writing difficulties experienced by some children with CP. Some children have fewer opportunities to engage with books and writing materials during their early years and have insufficient communication supports in place which can reduce the quality of interactions around early reading (Peeters, Verhoeven, van Balkom & de Moor, 2009). Similar barriers are encountered at school resulting in some children with CP spending less time on academic instruction than their typically developing peers (Jenks et al., 2007; Zascavage & Keefe, 2007). Lower literacy expectations from parents and educators can further limit children's access to quality literacy instruction, particularly for children with severe speech and physical impairments whose cognitive capacity may be underestimated (Peeters, Verhoeven, & de Moor, 2009; Stadskleiv, 2020). Additionally, many forms of 'typical' literacy instruction may be inaccessible to children with communication or physical impairments (e.g., tasks which require a verbal response or accessing a pencil or keyboard). Given that quality literacy instruction is essential for all beginning readers (Ontario Human Rights Commission, 2022), there is a need for high-quality evidence-based literacy instruction methods that accommodate all children, including those who communicate and participate in a variety of ways.

2.2.3 Previous Systematic Reviews

Relatively few systematic reviews have investigated literacy instruction for children with severe speech and physical impairments, although there has been some attention directed at individuals who use augmentative and alternative communication (AAC). Reviews have investigated different aspects of literacy instruction, including emergent literacy instruction and participation (Stauter et al., 2017) and use of assistive technology in literacy instruction (Stauter et al., 2019). One review by Machalicek et al. (2010) investigated the effects of literacy instruction for children with physical and developmental disabilities who use AAC with a focus on the NRP Big Five, identifying 18 relevant studies published between 1989 and 2009. Of the 41 participants in these studies, only five had a diagnosis of CP. Most studies implemented instruction aimed at improving multiple behaviours, including non-literacy outcomes such as communication and participation. Communication skills were the most common instruction target, followed by phonics skills (33%) and sight-word reading (22%). Systematic instruction was found to be the most effective instructional strategy (e.g., language scaffolding, least-to-most prompting), with improved literacy outcomes reported in most studies.

Several reviews have investigated literacy instruction for children with intellectual and developmental disabilities and/or multiple disabilities. These reviews have investigated both individual components of reading instruction (such as phonics activities; e.g. Hill, 2016), as well as reading instruction more broadly (e.g. Alquraini & Rao, 2020; Browder et al., 2006). The aforementioned studies report positive effects for systematic and explicit instruction, and all have called for more rigorous research in this area. The reviews outlined in this section provide some insight into literacy instruction methods for children with developmental disabilities, such as CP, and have the potential to help guide instructional practices for these children in the real world. See also the recent review by Arciuli and Bailey (2021) which is cited in the UK *Reading Framework* policy report as a key resource on literacy instruction methods for children with special educational needs (Department for Education, 2021a).

2.2.4 The Current Systematic Review

As far as we are aware, no previous review has investigated literacy instruction specifically for children with CP. Given that children with CP are more likely to experience literacy difficulties than their typically developing peers and are highly heterogeneous in both their abilities and support needs, a review on literacy instruction for children with CP is needed. We provide a systematic review and quality analysis for the research investigating the effects of literacy instruction for children with CP. Systematic review methodology was selected over other review procedures to ensure that all peer-reviewed research on this topic was systematically and explicitly identified and appraised (Moher et al., 2009). Our aim is to support clinicians', educators', and families' informed selection of literacy instruction methods which best support individuals with CP. We highlight gaps in the current research to guide future research directions and answer the following questions:

RQ1. What are the effects of literacy instruction designed to teach phonemic awareness, phonics, sight-word recognition, reading fluency, vocabulary, and/or reading comprehension on the reading abilities of school-aged children with CP?

RQ2. What are the effects of literacy instruction designed to teach spelling or written expression skills on the writing abilities of school-aged children with CP?

RQ3. What is the quality of these literacy instruction studies involving children with CP?

2.3 Method

This review was carried out using a protocol submitted to an international prospective register of systematic reviews (PROSPERO, http://www.crd.york.ac.uk/PROSPERO, registration number: CRD42020202330) and is reported according to the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) statement (Moher et al., 2009).

2.3.1 Inclusion Criteria

Studies which met the following criteria were included in the review: (a) included at least one school-aged participant (5-18 years) with a diagnosis of CP; (b) investigated the effects of literacy instruction with at least one dependent variable related to reading and/or writing outcomes (phonemic awareness, phonics, word recognition, vocabulary, reading fluency, reading comprehension, written expression, or spelling); (c) used a group or single-subject research design reporting baseline and treatment phase measures or pre- and post-outcome measures; (d) published in English; (e) published in a peer reviewed journal; and (f) published from 2000-2020 (this range was selected in order to capture the most up-to-date and relevant research). Studies were excluded if they did not directly report literacy outcomes. For example, studies investigating participation, turn-taking, or engagement in literacy activities (e.g. Browder et al., 2008) or access to literacy via assistive technology (e.g. Stasolla et al., 2019) were excluded if they did not report outcomes directly related to participants' reading and/or writing skills.

2.3.2 Search Procedure

A comprehensive search was conducted to identify all peer-reviewed journal articles on reading and/or writing outcomes for children with CP in response to literacy instruction. Potentially relevant studies were first identified through a search of the following electronic databases: PubMed, PsycINFO (Ovid), MEDLINE (Ovid), Cochrane Library data bases, ERIC (ProQuest), Education database (ProQuest). Search terms related to CP (Cerebral Palsy OR Multiple disabilit* OR Physical disabilit*) and literacy (Literacy OR Read* OR Spell* OR Writ* OR Word* OR Letter OR Phon* OR Alphabet* OR Vocabulary OR Fluency OR Comprehension). This search was limited to articles published in English and in a peer-reviewed journal between January 2000 – October 2020. The following journals were then hand searched for relevant titles*: Journal of Educational Research, Journal of Learning Disabilities, Journal of Literacy Research, Learning Disabilities Research and Practice, Augmentative and Alternative Communication, Journal of Developmental and Physical Disabilities, Research in Developmental Disabilities, Disability and Rehabilitation: Assistive Technology, Exceptional Children, International Journal of Disability, Development and Education, Education and Training in Autism and Developmental Disabilities, Research and Practice for Persons with Severe Disabilities*. Reference lists for each of the included studies and any systematic reviews identified in the above searches were checked for relevant titles, as were forward citations for each of the included studies (Lefebvre et al., 2022).

The first and second authors independently considered each of the identified studies for inclusion into the review. Article titles and abstracts were reviewed first for broad relevance to the topics of literacy instruction and CP. Only articles that were clearly not related to the target topics were excluded in this initial step. The remaining articles were then read in full and considered against the above-mentioned inclusion criteria. Agreement statistics based on the full-text review stage were almost perfect (κ = .898, $p < .001$) and the authors were able to reach consensus on all instances of disagreement.

2.3.3 Data Extraction

A data extraction form based on the methods of the NRP (NICHD, 2000), but expanded to capture details regarding participants' disabilities, was used by the first author to extract data from the included studies. This form summarised information on participant characteristics, research design, instruction characteristics, and outcomes. In the data extraction form and in the Results below, studies are grouped according to the skill instruction was designed to teach (i.e., phonics, sight-word recognition, reading fluency, reading comprehension, writing, and spelling skills). Studies encompassing three or more instruction targets were considered 'multicomponent' instruction. To ensure reliability of data extraction, the second author independently extracted data for a random selection (20%) of studies. There was an exceptionally high level of agreement (98%).

A standardised measure of effect size was used to quantify treatment effects. The Nonoverlap of All Pairs (NAP; Parker & Vannest, 2009) was computed using graphed data reported in single-subject design studies and extracted using the WebPlot-Digitizer data extraction software (Moeyaert et al., 2016; Rohatgi, 2018). NAP is an indicator of data overlap between phases in single-subject studies and was selected over other nonoverlap-based indices due to its superior external validity, efficiency, and accuracy. NAP can be interpreted using the following criteria: 0–0.65 indicates a weak effect; 0.66–0.92 indicates a medium effect; and 0.93–1.0 indicates a large effect (Parker & Vannest, 2009). NAP calculations were completed by the first author and checked by the second author. Any disagreements were discussed and resolved (e.g., interpretation of a tie versus overlap for a data point). The derived effect sizes are discussed in relation to individual studies and the range of treatment effects associated with each instruction subtype (i.e., phonics, sight-word recognition, reading fluency, reading comprehension, writing, spelling skills, multicomponent instruction).

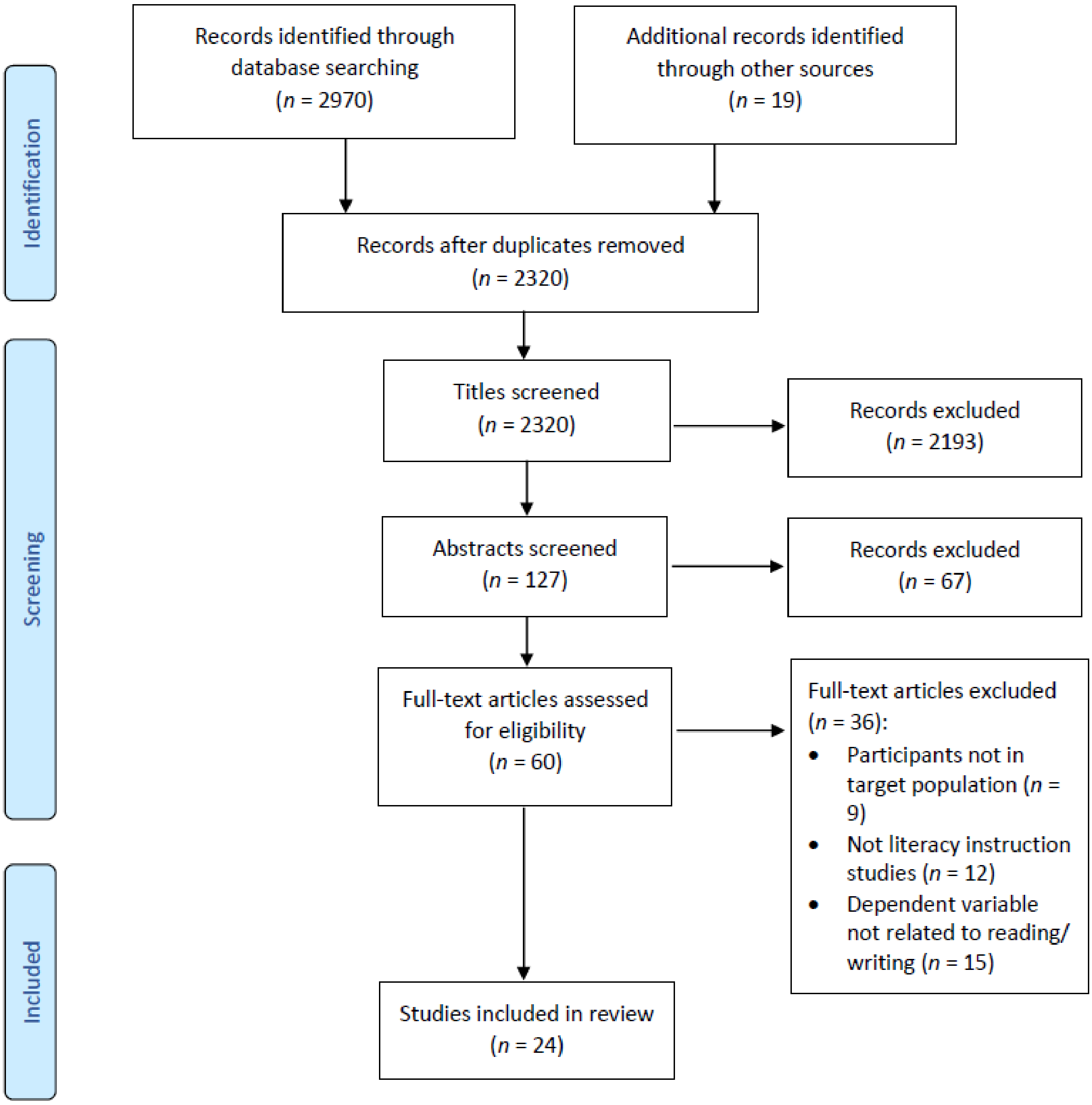
2.3.4 Analysis of Research Quality

The Mixed Methods Appraisal Tool (MMAT; Hong et al., 2018) was used to assess methodological rigour. The MMAT allows for critical appraisal of studies with different designs, enabling studies with diverse designs to be evaluated using the same quality rating scale. Studies were rated as 'yes', 'no', or 'can't tell' across five quality criteria questions pertaining to: (a) participant representativeness; (b) suitability of outcome and intervention measures; (c) completeness of outcome data; (d) accounting for confounders in study design and analysis; and (e) administering intervention as intended. Studies awarded a rating of 'yes' across all five questions were considered high quality; a rating of 'yes' across three to four questions were moderate quality; and a rating of 'yes' across two or fewer questions were low quality. Sub-criteria for each of the five quality questions were developed to ensure consistency in ratings. These sub-criteria were developed based on the quality indicators within single-subject research outlined by Horner et al. (2005). The first and second authors independently evaluated each of the included studies using the MMAT. Agreement statistics based on 'yes', 'no' and 'can't tell' ratings were substantial (κ = .709, p <.001). The authors were able to reach consensus on all instances of disagreement. For example, initial disagreements regarding the stability of baseline datapoints were resolved using standard criteria for baseline data stability (i.e., multiple data points, without significant trend or trend in the opposite direction than expected by intervention; Horner et al., 2005).

2.4 Results

Results of the search strategy are presented in Figure 2.1. A total of 2,970 potentially relevant articles were identified via the database search, with an additional 19 studies identified through journal and ancestry searches. Title and abstract screening revealed that 60 articles were related to the topics of literacy instruction and CP which were full-text reviewed. Thirty-six articles were not included based on the following reasons: nine did not involve participants within the target population, 12 were not literacy instruction studies, and 15 did not report a dependent variable related to reading or writing outcomes (these studies reported increasing turn-taking during literacy activities: n = 2; listening comprehension during book reading: n = 2; communication during literacy activities: n = 4; access to literacy activities using assistive technology: n = 7). Twenty-four studies met criteria for inclusion into the review.

Figure 2.1 *Flow Chart of Search Strategy Based on PRISMA Flow Diagram*



The included studies report on the effects of instruction designed to teach phonics[1] (*n* = 9), sight-word recognition (*n* = 4), reading fluency (*n* = 2), reading comprehension (*n* = 1), spelling (*n* = 3), written expression skills (*n* = 4), or multicomponent instruction (*n* = 1) on the

[1] Five studies utilised phonemic awareness or spelling instruction alongside phonics instruction targeting letter-sound correspondence. Phonics was determined to be the primary instruction method in these studies as instruction related to a small subset of letter-sounds.

reading or writing skills of children with CP. These studies are summarised across three tables. Table 2.1 provides an overview of participant characteristics. Table 2.2 describes instruction characteristics and Table 2.3 describes instruction outcomes. Instruction outcomes and effect sizes relate only to participants with CP. Quality analysis ratings are given in Table 2.4. Across the tables and Results section, studies on reading instruction are considered first (RQ1) and writing instruction are considered second (RQ2). Quality ratings (RQ3) are discussed in relation to each method of reading and writing instruction.

2.4.1 Effects of Instruction on Reading

Sixteen studies reported on the effects of phonics, sight word recognition, reading fluency, or reading comprehension instruction on the reading skills of children with CP. These studies are outlined below and are grouped according to the skill instruction was designed to teach.

2.4.1.1 Phonics Instruction. Nine single-subject studies investigated phonics-based instruction (Ainsworth et al., 2016; Clendon et al., 2005; Coleman-Martin et al., 2005; Fallon et al., 2004; Heller et al., 2002; Johnston, Davenport, Kanarowski, Rhodehouse, & McDonnell, 2009; Millar et al., 2004; Swinehart-Jones & Heller, 2009; Truxler & O'Keefe, 2007) and included a total of 18 participants from 5 to 16 years of age. All children were minimally verbal, 15 were reported to use AAC, and six were reported to have an intellectual disability. Participants' baseline literacy skills ranged from the ability to identify one letter to reading significantly below grade level. One participant was reported to be reading at grade level at commencement of the study.

Table 2.1 *Participant Characteristics*

Primary target skill First author (year)	Total no. of partici-pants	% Participants with Cerebral Palsy	Gender	Age (years)	Communication	Other Characteristics
Phonics						
Ainsworth (2016)	8	13% CP	1 Female	13	Minimally verbal.	• Moderate intellectual disability (*n* = 1)
Clendon (2005)	2	100% CP	1 Female 1 Male	7-10	Minimally verbal. AAC – symbol board/book (*n* =2).	• Mild intellectual disability (*n* = 1) • Mobility: wheelchair (*n* = 2)
Coleman-Martin (2005)	3	33% CP	1 Female	11	Minimally verbal. AAC - SGD with direct access.	• Borderline intellectual disability
Fallon (2004)	5	40% CP	1 Female 1 Male	9-10	Minimally verbal.	Unspecified
Heller (2002)	3	33% Spastic athetoid quadriplegia CP	1 Female	16	Minimally verbal. AAC – SGD with direct access.	• Mild intellectual disability • Mobility: wheelchair
Johnston (2009)	2	50% CP	1 Male	5	Minimally verbal. AAC – symbol board.	• Severe developmental delay
Millar (2004)	3	67% Spastic quadriplegia CP	2 Female	7-10	Minimally verbal. AAC - SGD with switch scanning (*n* = 2).	• Mobility: wheelchair (*n* = 1); wheelchair + walker (*n* = 1)
Swinehart-Jones (2009)	4	50% Spastic quadriplegia CP; 25% Athetoid CP; 25% Mixed CP	2 Female 2 Male	6-12	Minimally verbal. AAC – SGD with switch scanning (*n* = 2); direct access (*n* = 2).	Unspecified
Truxler (2007)	4	100% CP	4 Unspecified	8-9	Minimally verbal. AAC – symbol book/ board (*n* = 4).	• Intellectual disability (unspecified) • Mobility: wheelchair (*n* = 4) • English second language (*n* = 2)
Sight-word Recognition						
Hetzroni (2002)	5	20% Spastic quadriplegia CP; 40% Spastic diplegia CP; 20% Spastic hemiparesis CP	2 Female 2 Male	5-8	Intelligible speech (*n* = 1). Minimally verbal (*n* = 3).	• Mobility: Walk with aid (*n* = 2); walk independently (*n* = 1) • Primary language: Hebrew
Holyfield (2019)	3	100% CP	2 Female 1 Male	5-12	Minimally verbal. AAC - low-tech symbols.	• Mobility: wheelchair (*n* = 3)
Mandak (2020)	1	100% CP	1 Female	16	Minimally verbal. AAC - SGD with direct access.	• Mobility: wheelchair
Tjus (2004)	50	22% CP	11 Unspecified	9-17	Unspecified	• IQ age equivalent = 5:6-9:0 years • Primary language: Swedish

Primary target skill First author (year)	Total no. of partici-pants	% Participants with Cerebral Palsy	Gender	Age (years)	Communication	Other Characteristics
Reading Fluency						
Coleman (2010)	4	100% CP	1 Female 3 Male	9-12	Intelligible speech.	• Behavioural disorder (*n* = 1) • Mobility: independent (*n* = 1), wheelchair (*n* = 3) • Race: African American (*n* = 4)
Heller (2007)	2	50% CP	1 Male	9	Intelligible speech.	• Mobility: wheelchair • Race: African American
Reading Comprehension						
Ip (2005)	5	40% Spastic quadriplegia CP; 20% Ataxic CP; 20% Spastic diplegia CP; 20% CP	3 Female 2 Male	11-13	Moderate speech intelligibility (*n* = 2). Low speech intelligibility (*n* = 1).	• Mild intellectual disability (*n* = 5) • Mobility: independent (*n* = 2); walk with aid (*n* = 2); wheelchair (*n* = 1) • Primary language: Chinese
Spelling						
Coleman-Martin (2004)	3	67% Spastic quadriplegia CP	2 Female	10-12	Intelligible speech (*n* = 1). Mild dysarthria (*n* = 1).	• Mild intellectual disability (*n* = 1) • Mobility: wheelchair (*n* = 2)
McCarthy (2015)	7	100% CP	2 Female 5 Male	5-11	Intelligible speech (*n* = 3). Minimally verbal (*n* = 4). AAC - SGD with direct access (*n* = 4).	• Mobility: wheelchair (*n* = 3); Walk with aid (*n* = 1)
Raghavendra (2007)	1	100% Spastic quadriplegia CP	1 Male	11	AAC - SGD with direct access and symbol board.	• Mild intellectual disability. • Mobility: wheelchair and walker
Written Expression						
Garrett (2011)	5	20% CP	1 female	17	Intelligible speech.	• Asperger's syndrome
Mezei (2005)	3	33% Spastic quadriplegia CP	1 Male	13	Mild dysarthria.	• Low average intelligence, ADD • Mobility: wheelchair • Race: Caucasian
Mezei (2012)	4	25% Left hemiparesis CP; 25% spastic quadriplegia CP	1 Female 1 Male	12	Mild dysarthria (*n* = 1).	• Mobility: independent (*n* = 1)
Tumlin (2004)	4	25% Spastic quadriplegia; 25% athetoid/ataxic CP	2 Male	16-18	Moderate dysarthria (*n* = 2).	• Mobility: wheelchair (*n* = 1); independent (*n* = 1)
Multicomponent						
Hanser (2007)	3	100% Spastic quadriplegic CP	1 Female 2 Male	7-13	Minimally verbal. AAC - SGD with direct access (*n* = 2) or switch scanning (*n* = 1).	• Moderate intellectual disability (*n* = 1).

Note. AAC = augmentative and alternative communication; SGD = speech generating device; ADD = attention deficit disorder; Minimally verbal = does not produce intelligible speech (Mei, Reilly, et al., 2020).

Table 2.2 *Characteristics of Instruction*

Primary target skill First author (year)	Program	Setting (delivery)	Instructor	Duration [a]	Intensity	Instruction outcomes
Phonics						
Ainsworth (2016)	Accessible Literacy Learning (ALL) Curriculum (Light & McNaughton, 2009).	Locations within school (small group)	Researcher	3 months	3 x 45-60 min sessions/week	Letter-sound correspondence.
Clendon (2005)	Researcher designed instruction: Based on key principles from phonological awareness intervention studies.	Quiet room in school	Researcher	7 - 11 hours	3 x 30 min sessions/week	Phonemic awareness, letter-sound correspondence.
Coleman-Martin (2005)	Nonverbal Reading Approach (NRA) across three conditions: (a) teacher instruction, (b) teacher and Computer-assisted Instruction (CAI), and (c) CAI.	1:1 delivery in quiet room (computer-based)	Teacher	Unspecified	23 sessions	Decoding.
Fallon (2004)	Researcher designed phonics instruction based on Carnine et al. (1997).	1:1 delivery	Researcher	Unspecified	2-3 x 30 min sessions/week, 14-24 sessions	Decoding.
Heller (2002)	Nonverbal Reading Approach (NRA).	School (Words presented on computer)	Teacher	Unspecified	26 sessions	Decoding.
Johnston (2009)	Researcher designed phonics instruction.	Preschool classroom (during free play)	Teacher	Unspecified	81 sessions (5 trials / session)	Letter sound correspondence, Spelling.
Millar (2004)	Researcher designed phonics instruction including modified writing workshop-type activity.	Quiet room in school (letter selection via adaptive keyboard)	Researcher	Unspecified	2-3 x 30-45 min sessions/ week	Letter-sound correspondence, Initial letter identification.
Swinehart-Jones (2009)	Nonverbal Reading Approach (NRA) with motoric indicators/movements to parallel decoding steps.	1:1 delivery in classroom	Teacher	Unspecified	28 sessions	Decoding.
Truxler (2007)	Researcher designed phonics and phoneme awareness instruction (within context of book reading).	1:1 delivery in quiet room in school	Researcher	7 months	30 mins daily	Letter-sound correspondence, decoding.
Sight-word Recognition						
Hetzroni (2002)	Researcher designed instruction: Multimedia interactive software program for reading of a target book.	Clinic/quiet room at school (computer-based)	Researcher	Unspecified	2-3 sessions/week, 26-49 sessions	Single-word recognition.
Holyfield (2019)	AAC app programmed with video visual scene displays (VSDs) embedded with hotspots with the Transition to Literacy (T2L) feature.	Home / classroom (based on AAC app)	Researcher	10 weeks	2 x 20 min sessions/week	Single-word recognition.
Mandak (2020)	Video visual scene displays (VSDs) application with the Transition to Literacy (T2L) feature.	Quiet room in school (based on tablet)	Researcher	Unspecified	2 x 20-25 min sessions/week	Single-word recognition.
Tjus (2004)	DeltaMessages software (Nelson & Heimann, 1995).	Classroom (computer-based)	Teacher	2-4 months	3-6 sessions /month	Single-word recognition.

Primary target skill First author (year)	Program	Setting (delivery)	Instructor	Duration [a]	Intensity	Instruction outcomes
Reading fluency						
Coleman (2010)	Researcher designed instruction: Repeated reading with and without computer modelling, error correction, and performance feedback.	1:1 delivery in empty classroom (Computer-based)	Researcher	Unspecified	12-16 sessions	Reading fluency, reading comprehension.
Heller (2007)	Researcher designed instruction: a) Repeated reading (RR) with corrective feedback, and b) RR with corrective feedback alternating with unison readings.	Classroom-based (1:1 delivery)	Teacher	b) 3 weeks	a) 10 sessions; b) 19 sessions	Passage reading accuracy, reading fluency.
Reading Comprehension						
Ip (2005)	The metacognitive training program (Cole & Chan, 1990).	Classroom-based (whole class delivery)	Teacher	8 weeks	2 x 30 min sessions/week	Reading comprehension.
Spelling						
Coleman-Martin (2004)	Researcher designed spelling instruction utilising Constant Time Delay (CTD) procedure.	Classroom (small group-based)	Teacher	Unspecified	40-50 sessions	Spelling accuracy.
McCarthy (2015)	Researcher designed spelling instruction (computerised sounding out of pseudowords).	Quiet room in home/ clinic/day care (Computer-based)	Researcher	2 weeks	5 x 30 min sessions	Spelling accuracy.
Raghavendra (2007)	Systematic replication of Schlosser et al. (1998), involving three instructional conditions: (a) speech only, (b) speech-print, (c) print only.	Quiet room in school (AAC device based)	Speech Pathologist	a) 40 weeks	a) 1 x session/ week; b) 29 x daily sessions	Spelling accuracy.
Written Expression						
Garrett (2011)	Researcher designed instruction: using voice to text software (Dragon Naturally Speaking).	Quiet room in school (computer-based)	Researcher	Unspecified	20 sessions	Writing fluency, Spelling accuracy, Passage length.
Mezei (2005)	Instruction utilising Co:Writer 1400 word prediction software (Johnston, 1992) for writing of first drafts.	Classroom (computer-based)	Teacher	Unspecified	28 sessions	Writing fluency, spelling accuracy.
Mezei (2012)	Instruction utilising Co:Writer 1400 word prediction software (Johnston, 1992) for writing of first drafts.	1:1 in classroom (computer-based)	Teacher	Unspecified	25 sessions	Writing fluency, spelling accuracy, passage length.
Tumlin (2004)	Instruction utilising Co:Writer 1400 word prediction software (Johnston, 1992) for writing of first drafts.	Classroom (Computer-based)	Teacher	Unspecified	20 sessions	Writing fluency, spelling accuracy.
Multicomponent						
Hanser (2007)	Literacy Through Unity: Word Study program (Erickson & Hanser, 2007)	Home or school (on AAC device)	Parent, teacher	6 weeks	5 x 45-60-min sessions/week	Single-word recognition, spelling.

Note. VC = vowel consonant; CVC = consonant vowel consonant; AAC = augmentative and alternative communication.

[a] Information on instruction duration and intensity including session duration, number of sessions per week, total number of sessions noted where available.

Table 2.3 *Instruction Outcomes*

Primary target skill First author (year)	Design of study	Analyses	Outcome variables	Gains reported	Outcome statistics	Evidence of generalisation	Evidence of maintenance
Phonics							
Ainsworth (2016)	Multiple-baseline across groups	Visual analysis	Letter-sound correspondence	Yes	NAP = .94	No	No
Clendon (2005)	Single-subject pre-test/post-test design	Visual analysis & descriptive statistics	1) Letter-sound correspondence 2) Phoneme manipulation	1) Yes 2) Yes	1) Descriptive statistics 2) Descriptive statistics	1) No 2) Yes	1) No 2) No
Coleman-Martin (2005)	Multiple-conditions design with drop-down baselines	Visual analysis	Words read correctly	Yes, for all 3 instruction conditions	NRA with teacher only (NAP = .94), teacher and CAI (NAP = 1.0), CAI only (NAP = 1.0).	Yes, for all three instruction conditions	No, for all three instruction conditions
Fallon (2004)	Multiple-probe-across-subjects design	Visual analysis	Words read correctly	Yes	NAP = 1.0	Yes	Yes
Heller (2002)	Multiple baseline probe design	Visual analysis	Words read correctly	Yes	NAP = 1.0	No	No
Johnston (2009)	Within-subject, multiple-baseline probe design	Visual analysis	1) Letter-sound correspondence 2) Words spelled correctly	1) Yes 2) Yes	1) NAP = 1.0 2) NAP = 1.0	1) Yes 2) Yes	1) Yes 2) Yes
Millar (2004)	Multiple probe across subjects design	Visual analysis	1) Letter-sound correspondence 2) Initial letter identification	1) Yes 2) Yes	1) NAP = .88 - 1.0 2) NAP = 1.0	1) No 2) Yes	1) Yes 2) Yes
Swinehart-Jones (2009)	Changing-criterion design	Visual analysis	Words read correctly	Yes	NAP = .98 - 1.0	Yes	Yes
Truxler (2007)	Multiple baseline across subjects design	Visual analysis	1) Letter-sound correspondence 2) Word identification	1) Yes 2) Inconsistent	1) NAP = .79 - 1.0 2) NAP = .00 - .95	1) No 2) No	1) Inconsistent 2) No
Sight-word Recognition							
Hetzroni (2002)	a) Multiple probe; b) Multiple baseline	Visual analysis	Words read correctly	Yes	NAP = .86 - .92	No	Yes
Holyfield (2019)	Multiple baseline across participants	Visual analysis	Words read correctly	Yes	NAP = .85 - .92	No	Yes
Mandak (2020)	Multiple-probe across word-sets design	Visual analysis	Words read correctly	Yes	NAP = .95 - 1.0	Yes	No
Tjus (2004)	Quasi-experimental design	t-tests	Words read correctly	Yes	$t(49) = -1.76$, $p < 0.05$	No	No
Reading Fluency							
Coleman (2010)	Changing criterion design	Visual analysis	Words read correctly per minute	Inconsistent	NAP = .09 - .95	Inconsistent	No

Primary target skill First author (year)	Design of study	Analyses	Outcome variables	Gains reported	Outcome statistics	Evidence of generalisation	Evidence of maintenance
Heller (2007)	Alternating treatment design	Visual analysis & descriptive statistics	Words read correctly per minute	Yes	Descriptive statistics (no baseline phase for NAP calculation)	Yes	No
Reading Comprehension							
Ip (2005)	Multiple-case, single-subject AB design	Visual analysis	Reading comprehension	Inconsistent	NAP = .55 - .81	No	No
Spelling							
Coleman-Martin (2004)	Multiple baseline with probes design	Visual analysis	Words spelled correctly	Yes	NAP = .79 - .95	No	Yes
McCarthy (2015)	AB single-subject design	Visual analysis	Correct consonants/vowels in spelling	Inconsistent	NAP = .17 - 1.0	No	No
Raghavendra (2007)	Alternating treatment design; intrasubject direct replication	Visual analysis	1) Words spelled correctly 2) Letters spelled correctly	1) Yes 2) Yes	1) NAP = .80 - .97 2) NAP = .83 - 1.0	No	Inconsistent
Written Expression							
Garrett (2011)	Alternating treatment design	Visual analysis & descriptive statistics	1) Words written per minute 2) Recall of meaning 3) Words spelled correctly	1) Yes 2) Yes 3) Yes	1) Descriptive statistics 2) Descriptive statistics 3) Descriptive statistics	1) No 2) No 3) No	1) No 2) No 3) No
Mezei (2005)	Withdrawal design	Visual analysis & descriptive statistics	1) Words written per minute 2) Words spelled correctly	1) Yes 2) Yes	1) NAP = .73 - .88 2) Descriptive statistics	1) No 2) No	1) No 2) No
Mezei (2012)	Alternating treatment design	Visual analysis	1) Words written per minute 2) Words spelled correctly	1) Yes 2) Yes	1) NAP = .70 - .86 2) NAP = .99 - 1.0	1) No 2) No	1) No 2) No
Tumlin (2004)	Reversal design	Visual analysis & descriptive statistics	1) Words written per minute 2) Words spelled correctly	1) Inconsistent 2) Yes	1) NAP = .47 - .79 2) Descriptive statistics	1) No 2) No	1) No 2) No
Multicomponent							
Hanser (2007)	Non-concurrent multiple baseline	Visual analysis & descriptive statistics	1) Number of letters used in spelling 2) Words read correctly	1) Yes 2) Yes	1) NAP = .62 - .69 2) Descriptive statistics	1) Yes 2) Yes	1) Yes 2) No

Note. Visual analysis refers to interpretation of the level (mean performance), trend (rate of increase or decrease in performance), and variability of performance during baseline and intervention conditions in single-subject design studies using graphed data (Parsonson & Baer, 1978).

Table 2.4 *Quality Analysis*

Primary target skill First author (year)	**3.1 Are the participants representative of the target population?** [a]	**3.2 Are measurements appropriate regarding both the outcome and intervention (or exposure)?** [b]	**3.3 Are there complete outcome data?** [c]	**3.4 Are the confounders accounted for in the design and analysis?** [d]	**3.5 During the study period, is the intervention administered (or exposure occurred) as intended?** [e]	**Total**	**Quality rating**
Phonics							
Ainsworth (2016)	N	Y	Y	Y	Y	4	Moderate
Clendon (2005)	N	N	N	N	N	0	Low
Coleman-Martin (2005)	N	Y	Y	Y	Y	4	Moderate
Fallon (2004)	Y	Y	Y	Y	Y	5	High
Heller (2002)	Y	Y	Y	Y	Y	5	High
Johnston (2009)	N	Y	Y	Y	Y	4	Moderate
Millar (2004)	Y	Y	Y	Y	Y	5	High
Swinehart-Jones (2009)	Y	Y	Y	Y	Y	5	High
Truxler (2007)	N	Y	N	Y	Y	3	Moderate
Sight-word recognition							
Hetzroni (2002)	N	Y	Y	Y	Y	4	Moderate
Holyfield (2019)	Y	Y	Y	Y	Y	5	High
Mandak (2020)	N	N	Y	N	Y	2	Low
Tjus (2004)	N	N	N	N	N	0	Low
Fluency							
Coleman (2010)	N	Y	Y	Y	Y	4	Moderate
Heller (2007)	N	Y	Y	N	Y	3	Moderate
Reading Comprehension							
Ip (2005)	N	N	Y	N	N	1	Low
Spelling							
Coleman-Martin (2004)	N	Y	Y	Y	Y	4	Moderate
McCarthy (2015)	N	Y	Y	N	N	2	Low
Raghavendra (2007)	N	N	Y	Y	N	2	Low
Written Expression							
Garrett (2011)	N	Y	Y	N	Y	3	Moderate
Mezei (2005)	Y	Y	Y	Y	Y	5	High
Mezei (2012)	Y	Y	Y	Y	Y	5	High
Tumlin (2004)	Y	Y	Y	N	Y	4	Moderate
Multicomponent							
Hanser (2007)	Y	N	Y	N	N	2	Low

Note. Quality criteria questions based on Mixed Methods Appraisal Tool (MMAT; Hong et al., 2018) 'Quantitative non-randomised studies'; Y = yes (criteria met); N = no (criteria not met).

[a] Description of participants (age, gender, method of communication, type of CP/motor abilities); process for participant selection (inclusion/exclusion criteria, recruitment).

[b] Independent variable stated; dependent variable stated; measures for dependent variable stated and appropriate; reliability measures for ≥20% of sessions with ≥80% accuracy.

[c] Complete outcome data for ≥90% of data points; <5% dropout rate.

[d] Study design that meets standards for experimental design (single-subject designs based on Byiers et al., 2012); sufficient description of baseline condition to allow replication; ≥3 baseline data points; stable baseline data points).

[e] Independent fidelity measures for ≥20% of sessions with ≥80% accuracy

2.4.1.1.1 Instruction Characteristics. Five studies investigated methods designed to improve children's letter-sound knowledge. This involved children identifying a letter corresponding to a sound produced by an instructor (Ainsworth et al., 2016; Johnston et al., 2009; Millar et al., 2004) or identifying pictures based on the initial sound or letter said by an instructor (Clendon et al., 2005; Truxler & O'Keefe, 2007). All forms of instruction involved a small target set of sounds, ranging from three to six letters per study. Targets were selected based on unfamiliarity to children and following published recommendations (e.g. Carnine et al., 1997). The letters 's', 't', 'a', and 'm' were the most common targets of instruction, each included across three different studies. Three studies extended letter-sound instruction to include blending, decoding, or spelling words containing only the small set of target letters (Johnston et al., 2009; Millar et al., 2004; Truxler & O'Keefe, 2007).

Four studies investigated methods designed to teach decoding skills and utilised similar instruction protocols. All involved systematic direct instruction, starting initially with modelling and guided practice (i.e., instructor modelled each sound whilst tracking each letter in the word, then blended the sounds together to say the word). Children were encouraged to follow along and say the sounds 'in their heads', before being provided opportunities to decode independently. In three studies, systematic instruction was identified as the Nonverbal Reading Approach (NRA; Coleman-Martin et al., 2005; Heller et al., 2002; Swinehart-Jones & Heller, 2009). In the study by Coleman-Martin et al. (2005), the NRA was delivered across three conditions: (a) teacher only; (b) teacher and computer-assisted instruction (CAI – computer modelled the decoding steps aloud); and (c) CAI only. Swinehart-Jones and Heller (2009) taught children to use an individualised motoric movement (e.g. blinking) to provide an observable behaviour that they were using each step of the NRA decoding process. Fallon et al. (2004) investigated instruction designed to improve phonemic awareness skills (initial sound identification and auditory blending of sounds) alongside direct instruction for decoding skills using 14 target letters. The focus of this study was on single word reading skills and phonemic awareness activities were used to consolidate sound awareness for the 14 target letter sounds. All studies targeting decoding skills utilised real words.

2.4.1.1.2 Instruction Outcomes. Phonics instruction outcomes were evaluated using measures of letter-sound knowledge and word-level decoding. Letter-sound knowledge measures required children to match spoken sounds to printed letters (from a choice of three to four letters or selection on a keyboard). Word-level decoding measures involved children reading printed “decodable” words (e.g., pill, stamp) and either pointing to the corresponding picture or selecting the target word from a choice of four similar words (e.g. selecting the target word ‘think’ from a choice of ‘think’, ‘thank’, ‘link’, ‘thing’).

Visual analysis for the studies targeting letter-sound knowledge showed consistent gains associated with medium to large effect sizes (NAP = .79–1.0) for all 10 participants. Studies investigating transfer of letter-sound correspondence to spelling skills reported strong effects (NAP = 1.0; Johnston et al., 2009; Millar et al., 2004), whilst transfer to decoding skills was varied with strong effects for one participant (NAP = .95) and weak effects for three (NAP = .00; Truxler & O'Keefe, 2007). Instruction focusing on decoding skills was associated with strong effects for all eight participants (NAP = .94–1.0), with consistent gains in the number of words read correctly throughout instruction. Nine participants demonstrating strong effects across the studies were able to generalise their skills to spell/decode untrained words. Gains were maintained up to 2 months post instruction in three studies (Fallon et al., 2004; Johnston et al., 2009; Millar et al., 2004). Generalisation of target letters/words to new contexts was not demonstrated in Clendon et al. (2005), though was demonstrated for both participants in Fallon et al. (2004).

2.4.1.1.3 Quality Ratings. Four studies on phonics instruction received a high-quality rating, four received a moderate rating, and one received a low rating. Moderate ratings were due to insufficient participant descriptions across studies and incomplete outcome data in one study (Truxler & O'Keefe, 2007). The low rating was due to methodological limitations across all quality criteria, including use of a pre-experimental study design at risk of bias and no measures of fidelity or inter-rater agreement (Clendon et al., 2005).

2.4.1.2 Sight-Word Recognition Instruction. Three single-subject studies (Hetzroni & Schanin, 2002; Holyfield et al., 2019; Mandak et al., 2020) and one group study (Tjus et al., 2004) investigated instruction aimed at increasing sight-word recognition. Nineteen participants aged 5 to 17 years were included across the four studies. Seven children were minimally verbal and one child communicated verbally. Four children were reported to use AAC. The communication methods of 11 children in the group study were not reported. At baseline, most participants were classified as 'non-readers', with one participant demonstrating some basic word reading and spelling skills (fewer than 50 words).

2.4.1.2.1 Instruction Characteristics. Sight-word instruction involved teaching participants to read high frequency words automatically and accurately within motivating contexts, with all studies utilising technology in instruction. Target words were selected based on academic vocabulary relevant to the participant (Mandak et al., 2020), vocabulary of high interest (Holyfield et al., 2019), or vocabulary naturally occurring in instruction materials (Hetzroni & Schanin, 2002; Tjus et al., 2004). Holyfield et al. (2019) and Mandak et al. (2020) used a Transition to Literacy (T2L) feature enabled within video visual scene displays in an AAC app. Participants selected hotspots within videos to activate the T2L feature (target word would expand on the screen, remain static for 3 seconds accompanied by voice output, then shrink), with a total of five to 40 exposures to the target word per session. Hetzroni and Schanin (2002) used a researcher designed computer-based interactive book with switch activated words within the text. Tjus et al. (2004) utilised a computer-based language and literacy program where participants selected words to create sentences and received auditory and visual feedback once completed (written text spoken aloud and video animation of sentence).

2.4.1.2.2 Instruction Outcomes. Sight word instruction outcomes were evaluated in terms of children's ability to read sets of printed high-frequency words which had been targeted during instruction. Visual analysis of graphed data from the single-subject studies indicated steady and consistent gains in sight-word recognition during instruction for all participants. These gains were associated with medium to large effect sizes (NAP = .85–

1.0). A statistically significant gain in word reading was noted over the instruction period in the group study ($p < 0.05$); however, effect sizes were not reported (Tjus et al., 2004). Participants in the two studies that utilised a multiple-probe across word-sets design (Hetzroni & Schanin, 2002; Mandak et al., 2020) demonstrated faster acquisition of target words as they progressed through the program, resulting in fewer instruction sessions and exposures to target words in each new word set. Maintenance data was reported for three participants, with gains maintained up to 3 weeks post instruction. The participant in Mandak et al. (2020) demonstrated generalisation to reading target words in different contexts. Tjus et al. (2004) reported a decrease in single word reading between the training phase and follow-up period (approximately 2.5 months), although this difference was not statistically significant.

2.4.1.2.3 Quality Ratings. One study received a high-quality rating and one received a moderate rating. Two received a low rating due to insufficient reliability measures, using a study design at risk of bias or unstable baseline data, and unclear representativeness of participants, including unreported type of CP, lack of inclusion criteria, and no description of participant recruitment.

2.4.1.3 Reading Fluency Instruction. Two single-subject design studies investigated instruction designed to improve oral reading fluency (Coleman & Heller, 2010; Heller et al., 2007), including five participants aged 9 to 12 years. All participants had intelligible speech and communicated verbally. At baseline, participants read at least two grades below expected level. Reading fluency ranged from 10-18 words correct per minute.

2.4.1.3.1 Instructions Characteristics. Both studies investigated researcher designed protocols utilising variations of repeated reading in instruction. Heller et al. (2007) conducted two case studies involving the same participant. The first examined ‘unison reading’ and the second compared the ‘unison reading’ protocol with ‘repeated reading’ in an alternating treatment design. ‘Repeated reading’ involved three readings of the same passage with feedback and practice of errors after reading. ‘Unison reading’ followed the same format with the addition of unison readings (teacher and student reading together), interspersed between each of the repeated readings. Coleman and Heller (2010) utilised a similar protocol of instruction with three oral readings with error correction opportunities interspersed with two computer-modelled readings.

2.4.1.3.2 Instruction Outcomes. Reading fluency instruction outcomes were evaluated in terms of the number of words read correctly per minute (i.e., reading rate). Graphed data showed improved oral reading rates between the first and last reading in each session under all instruction protocols. In Heller et al. (2007), reading rates were found to be higher in the final reading under the 'unison reading' condition compared with 'repeated reading'. Transfer of reading fluency gains on novel passages were associated with strong effects for one participant in Coleman and Heller (2010), with the remaining participants demonstrating weak effect sizes (NAP = .09–.95).

2.4.1.3.3 Quality Ratings. Both reading fluency instruction studies received moderate ratings using the MMAT due to limitations in participant description across both studies (description of participant recruitment and inclusion/exclusion criteria), and an experimental design that did not meet standards in Heller et al. (2007).

2.4.1.4 Reading Comprehension Instruction. Only one study investigated reading comprehension instruction (Ip & Lian, 2005). This study involved five children aged 11 to 13 years with bilateral CP. All participants communicated verbally and had mild intellectual disability. Participants' general academic functioning was approximately equivalent to a second grade level at baseline.

2.4.1.4.1 Instruction Characteristics. Reading comprehension instruction utilised an existing program, modified to meet the academic level of study participants. Instruction covered eight topics to support text comprehension, with two sessions per topic: (a) deleting redundant information; (b) deleting trivial information; (c) locating the topic sentence in a paragraph and (d) passage; (e) rating order of importance of sentences; (f) identifying implicit main idea in paragraphs and (g) passages; and (h) review. Participants were explicitly taught to ask themselves three questions, which differed based on topic, for each of the eight topics.

2.4.1.4.2 Instruction Outcomes. Reading comprehension instruction outcomes were assessed in terms of children's ability to answer comprehension questions relating to eight short texts. Answers to these questions could be found explicitly in text (text explicit questions), inferred by combining information from various parts of the text (text implicit questions), or inferred by considering the passage as a whole and relating it to one's prior knowledge of the text topic (script implicit questions). Analysis of trend lines across the graphed data indicated that the program had a positive effect on reading comprehension skills for 80% of the participants. Reading comprehension gains were associated with weak to medium effect sizes (NAP = .55–.81).

2.4.1.4.3 Quality Ratings. The study investigating reading comprehension instruction rated as low quality on the MMAT due to limitations in participant representativeness (unreported inclusion criteria and selection of participants), unclear reliability and fidelity measures, and use of a pre-experimental (AB) study design.

2.4.2 Effects of Instruction on Writing

Seven studies reported on the effects of spelling and written expression instruction on the writing skills of children with CP. These studies are outlined below and are grouped according to the skill instruction was designed to teach.

2.4.2.1 Spelling Instruction. Three single-subject studies investigated instruction targeting spelling skills (Coleman-Martin & Heller, 2004; McCarthy et al., 2015; Raghavendra & Oaten, 2007), including 10 participants from 5 to 12 years of age. Five participants communicated verbally and five were minimally verbal and used AAC. Two participants had mild intellectual disability. At baseline, the majority of participants had limited spelling skills, ranging from the ability to spell one word to spelling skills two to four grades below expected skill level.

2.4.2.1.1 Instruction Characteristics. All three studies on spelling involved some form of phonics instruction linking letters and sounds, either with or without an AAC device. Two studies utilised assistive technology in instruction, with most participants typing responses during instruction on a laptop or AAC device. McCarthy et al. (2015) investigated computerised sounding-out of pseudowords in instruction (a digitised voice elongated and segmented target words into individual phonemes). Raghavendra and Oaten (2007) utilised a speech generating AAC device (SGD) to implement a 'copy-write-compare' method of spelling instruction under three instructional conditions: (a) speech (auditory feedback from SGD); (b) speech-print (both auditory and orthographic feedback); and (c) print (only orthographic feedback from device screen). Coleman-Martin and Heller (2004) utilised a modified constant-delay procedure with near-errorless learning, where if participants did not attempt to write the word within 5 seconds they were provided with a prompt (written word accompanied by the teacher pointing and verbalising each letter).

2.4.2.1.2 Instruction Outcomes. Spelling instruction outcomes were reported as either whole words spelled correctly, or correct use of individual letters within words in spelling attempts, with Raghavendra and Oaten (2007) reporting on both. Studies reported gains in spelling accuracy for all 10 participants following instruction. Gains were associated with medium to strong effects for most participants (NAP = .79–1.0), except for two participants in McCarthy et al. (2015) who performed lower during the instruction phase for use of consonants in spelling (NAP = .17 –.30). These participants both demonstrated gains associated with strong effect sizes for use of vowels in spelling during instruction (NAP = .97–1.0). The participant in Raghavendra and Oaten (2007) achieved spelling gains more quickly under the 'print' condition, where they received only orthographic not auditory feedback.

2.4.2.1.3 Quality Ratings. One study was rated as being of moderate quality due to lack of description of participant selection. Two studies were rated as low quality due to insufficient reliability and fidelity measures and use of a pre-experimental (AB) study design.

2.4.2.2 Written Expression Instruction. Four studies investigated instruction aimed at written expression (Garrett et al., 2011; Mezei & Heller, 2005, 2012; Tumlin & Heller, 2004), including six participants aged 12 to 18 years. All participants communicated verbally. Literacy skills at baseline ranged from three years below grade level to average on measures of reading and spelling, and spelling errors ranged from 6-13% on written work.

2.4.2.2.1 Instruction Characteristics. All written expression instruction utilised assistive technology designed to address the impact of motor impairments on text transcription skills, specifically writing fluency and spelling, during first draft writing. Regular word processing software was compared with word prediction software in three studies and speech-to-text software in one study (Garrett et al., 2011). In all four studies, training in using the target assistive software was provided in small groups across multiple sessions prior to instruction. Instruction sessions in all studies involved participants planning their responses to a given topic before being instructed to begin their first draft writing with a focus on getting their thoughts written down.

2.4.2.2.2 Instruction Outcomes. Written expression instruction outcomes were assessed using measures of spelling accuracy (i.e., percentage of correctly spelled words) and writing fluency (i.e., number of characters written per minute). Word prediction software was associated with increased spelling accuracy for all five participants. Graphed data for spelling accuracy was presented in one study (Mezei & Heller, 2012), indicating strong effects (NAP = .99–1.0). Descriptive statistics from Garrett et al. (2011) indicate that writing accuracy was higher using conventional word processing software (average 96.3%) compared with speech-to-text (average 92.5%). Gains in writing fluency when using the word prediction software were inconsistent across participants and were associated with weak to medium effect sizes (NAP = .47–.88). Participants in two studies demonstrated increased length of drafts when using the assistive technology compared with conventional word processing software (Garrett et al., 2011; Mezei & Heller, 2012).

2.4.2.2.3 Quality Ratings. Two studies were rated as having high methodological quality using the MMAT (Mezei & Heller, 2005, 2012). Two were rated moderate due to unstable baseline data (Tumlin & Heller, 2004) or study designs at risk of bias (Garrett et al., 2011).

2.4.3 Effects of Multicomponent Instruction on Reading and Writing

One single-subject study investigated multicomponent literacy instruction (Hanser & Erickson, 2007). The study involved three participants aged 7 to 13 years. All participants were minimally verbally and used AAC. One participant had moderate intellectual disability. Participants could identify most letters, read a few single words, and had some basic spelling skills at baseline.

2.4.3.1 Instruction Characteristics. Multicomponent literacy instruction sought to teach three reading/writing skills using a pre-existing program, Literacy Though Unity: Word Study (Erickson & Hanser, 2007), designed to integrate literacy instruction with communication intervention using the participants' AAC device. The program cycled through 25 lessons for each of three target skills: (a) word recognition; (b) spelling; and (c) communication, with one lesson per skill in a three-lesson cycle. Communication lessons targeted sequencing icons for expressive communication. A range of strategies were utilised in literacy instruction, including phoneme-by-phoneme and phonics-by-analogy strategies.

2.4.3.2 Instruction Outcomes. Graphed data shows variability in children's use of individual letters for spelling outside of instruction sessions during the instruction period, with weak-medium effect sizes (NAP = .62–.69). Pre- post-data indicates gains in developmental spelling scores for all three participants with an average increase of 13 points (points based on phonemes correct). Descriptive statistics indicate an average gain of 20% (five words) on word identification tasks from pre- to post-instruction. Maintenance of skills was inconsistent across participants five weeks post instruction.

2.4.3.3 Quality Ratings. The study investigating multicomponent instruction received a low-quality rating. This rating was due to insufficient reliability and treatment fidelity measures and unstable baseline data.

2.5 Discussion

This review considered research on the effects of literacy instruction for children CP. Most of the 24 included studies utilised single-subject designs and involved very small samples of children with varying communication and literacy skills. Literacy instruction was associated with gains in reading and writing for all participants, with medium to large effect sizes reported for at least one participant in all studies allowing calculation of effect size. Seven studies received a high-quality rating on appraisal of methodological quality and seven studies received a low-quality rating. The small sample sizes and varied quality ratings suggest that these results should be interpreted with caution.

2.5.1 Implications for Practice

The overarching aim of this review was to help guide clinicians, educators and families in their literacy instruction choices when working with school-aged individuals with CP. Results shed light on the suitability of instruction targeting specific reading and writing skills and multicomponent literacy instruction, as well as measures to promote access to literacy instruction for school-aged individuals with CP. Results also highlight to researchers the need to improve practices when evaluating interventions in this field.

2.5.1.1 Single Component Instruction. Results from the included studies show that instruction designed to teach phonics, reading fluency, reading comprehension, written expression, or spelling skills in isolation are effective in improving closely associated outcomes for children with CP. Several of these studies found improvements to novel contexts (i.e., stimuli unseen during instruction), suggesting that gains generalised beyond the reading and writing materials presented during instruction. There was also evidence that improvements in phonics skills were maintained for weeks following instruction. Though limited, this provides some promising evidence to educators and clinicians about the benefits of explicit literacy instruction for children with CP.

Studies investigating ‘sight-word recognition’ instruction reported gains with medium to large effect sizes; however, these results relate to participants’ recognition of words directly targeted in instruction. Sight-word recognition is frequently used with children with disabilities as a method of literacy instruction, particularly with individuals who use AAC (Browder et al., 2006; Mandak et al., 2018). Instruction focusing solely on sight-word recognition does not support individuals to develop the strategies necessary to decode new and unseen words and become skilled readers. Research recommends that sight-word recognition instruction should only be included as part of a more comprehensive literacy instruction program (Browder et al., 2012).

2.5.1.2 Multicomponent Instruction. The NRP recommends that reading instruction should integrate five key skills in explicit instruction: phonemic awareness, phonics, reading fluency, vocabulary, and reading comprehension (NICHD, 2000). None of the studies included in this review considered instruction targeting all the Big Five (comprehensive instruction). Only the study by Hanser and Erickson (2007) considered instruction targeting three or more skills (phonics, spelling, sight word recognition and communication). Participants in this study showed improvements in communication, word identification, and spelling skills following instruction. This study was rated as low quality though provides some promising evidence to educators and clinicians for integrating literacy instruction with existing AAC methods in order to promote both functional literacy and communication skills. Given the critical role of language skills in literacy acquisition (Snow, 2020), and the function of AAC in providing children a means to demonstrate their knowledge and understanding during literacy activities (Capp, 2017), both skills are fundamentally important for literacy development in children with CP.

2.5.1.3 Promoting Access to Instruction. Three studies received a high-quality rating and also reported strong effects for all participants (Fallon et al., 2004; Heller et al., 2002; Swinehart-Jones & Heller, 2009). These studies all investigated phonics instruction with minimally verbal participants and provided alternate response means to allow children to demonstrate their decoding skills. Across all studies, participants improved their ability to decode targeted words, but only two children showed evidence of generalised improvements using non-target words. These results indicate that children with CP can increase their single word reading skills when provided with explicit, systematic phonics instruction. However, this instruction needs to be accessible and allow children to demonstrate, and receive feedback on, their decoding skills. This is in line with the Universal Design for Learning (UDL) framework which proposes that educators can reduce barriers to learning by providing multiple ways for children to demonstrate their understanding (Capp, 2017).

Children used assistive technology to access instruction or demonstrate their understanding in some of the included studies. Fifteen studies incorporated assistive technology in instruction, including speech generating AAC devices, alternate access methods (e.g., switches, adaptive keyboards), and software programs. Studies utilising assistive technology reported improvements in decoding, reading fluency, sight-word recognition, spelling, and written expression. The high number of studies incorporating technology in instruction is in line with previous research suggesting that assistive technology is a crucial component of literacy instruction for individuals with severe speech and physical impairments (Stauter et al., 2019; Zascavage & Keefe, 2004), as well as research on reading instruction for typically developing children recommending inclusion of technology and educational software (NICHD, 2000). Although recognised as crucial to literacy instruction for children with CP, assistive technology is not always accessible to the children who require it. The cost and general lack of availability of assistive technology can impact implementation, as can family, educator, and peer acceptance of the technology and availability of trained support persons (Parette & Scherer, 2004; Zascavage & Keefe, 2004).

The World Health Organisation (2016) list priority assistive products that should, as a minimum, be available in all countries, many of which support access to education. Whilst funding systems for assistive technology differ based on country, De Witte et al. (2018) present an international framework providing guidance to ensure availability and accessibility of assistive technology. This framework highlights the many mainstream technologies with features enabling them to be used as assistive devices, several of which were used successfully to support access to literacy instruction throughout studies in this review (e.g., tablet computers with speech to text or AAC apps). Additionally, De Witte et al. (2018) emphasise the importance of including the assistive technology user throughout the decision-making process to ensure the technology meets their needs, supports their participation, and to reduce the likelihood of technology abandonment.

2.5.2 Limitations of the Review

Several methodological limitations were highlighted in the quality analysis conducted in this review. The most common limitation across studies was incomplete participant descriptions. Thorough participant description is a key quality indicator within single-subject research as it allows others to identify similar individuals (Horner et al., 2005). Only 11 studies described CP motor sub-type and topography for participants. No study described the functional ability level of participants using the functional classification systems for mobility, manual ability, and communication used with individuals with CP (e.g. Gross Motor Function Classification System [GMFCS]; Palisano et al., 1997). Additionally, only three studies directly reported participants' race (Coleman & Heller, 2010; Heller et al., 2007; Mezei & Heller, 2005). As there is a higher prevalence of CP in indigenous children and children of colour (Australian Cerebral Palsy Register Group, 2018; Maenner et al., 2016), and these children experience literacy learning difficulties at a higher rate than the general population (Wigglesworth et al., 2011), this demographic information should be provided. A further three studies reported instruction taking place in a language other than English (Hetzroni & Schanin, 2002; Ip & Lian, 2005; Tjus et al., 2004). Limited exploration of literacy instruction for children with CP who are bilingual, or in languages other than English, are a further limitation of the current literature and an important factor for future research to consider in a global context (Bailey & Arciuli, 2022).

Incomplete description of instruction characteristics, lack of independent outcome measures, and variability in instructors were additional limitations. Many studies did not report duration of instruction phases or time and frequency of instruction sessions. Without these details, it is difficult to compare instruction methods and draw conclusions regarding instruction efficiency. Studies frequently made use of researcher designed pre-post assessment measures, or probe tasks containing items directly targeted in instruction. This may be due to lack of standardised assessments for individuals with communication and/or motor impairments and future research is needed to develop rigorous assessment tools that are accessible to children with diverse abilities. Additionally, there was variability in instructors across studies: instruction was delivered by teachers in 12 studies (including one study where instruction was delivered by a teacher or parent), by researchers in 11 studies, and by a speech pathologist in one study. Implementation and intervention fidelity are important factors in developing evidence-based interventions as these measures ensure that interventions are administered as intended and can be replicated (Horner et al., 2005). Implementation fidelity refers to the training and coaching instructors receive to deliver interventions and intervention fidelity refers to the actual administration of the intervention (Barton & Fettig, 2013). Eighteen of the included studies reported on some form of fidelity. Only two of 12 studies involving teacher-delivered instruction reported implementation fidelity (i.e., how teachers were trained to deliver instruction: Hanser & Erickson, 2007; Johnston et al., 2009). Of these 12 studies, nine reported intervention fidelity. Nine of the 11 studies involving researcher-delivered instruction reported intervention fidelity. However, instruction delivered by researchers may not accurately represent what is occurring in the classroom setting for children with CP (Kim et al., 2012).

The search dates we used in the current review were restricted to results over a 20-year period. This decision was made in order to capture the most up-to-date research. This review may also be subject to publication bias as only literature published in peer reviewed journals was included. We did not search for relevant unpublished research. In addition, this review only investigates the impact of direct literacy instruction practices on the literacy skills of children with CP and does not explore the wider literacy experiences of these children (e.g., the quantity and quality of time spent engaging in literacy activities at home/school). This broader social context undoubtedly has an impact on literacy learning for children with CP and warrants further research.

2.5.3 Future Research

Although this review provides some promising findings that certain instruction methods can be effective in promoting the literacy skills of children with CP, this data is based largely on single-subject studies with low quality designs. Single-subject research can be valuable in determining 'what works for who' within heterogeneous populations such as CP, and future research should focus on utilising single-subject studies with higher quality designs along with high-quality group studies to confirm efficacy of instruction methods. Future research should also explore the impact of factors such as time spent in instruction and accessibility of instruction on literacy outcomes for children with CP. It would also be valuable to use qualitative methods to learn more about the lived experiences of literacy instruction by children with CP.

Although a range of literacy sub-skills were explored in this review, few studies focused on instruction designed to express or derive meaning from text; that is, written expression and reading comprehension. Reading studies tended to focus on reading accuracy (i.e., phonics, sight-word recognition, reading fluency), with no studies focusing on vocabulary and only one study exploring instruction designed to improve reading comprehension skills. As outlined in the SVR, both decoding and language comprehension skills are essential to becoming a skilled reader (Gough & Tunmer, 1986). Given that many children with CP experience impairments in language and communication (Mei, Reilly, et al., 2020), future research needs to examine instruction targeting reading comprehension skills. In addition, the studies included in our review that examined writing instruction tended to focus on accuracy (i.e., spelling and text transcription skills). There was some focus on 'planning' but other writing processes such as revising, text-organisation, self-evaluation, or other language-based aspects of text transcription, such as lexical-choice or syntax, were not explored (for a review of the cognitive processes of writing see: Flower & Hayes, 1981; Koppenhaver & Williams, 2010). Future research on writing instruction should focus on the writing process for children with CP to provide guidance to educators and clinicians supporting these children. Koppenhaver and Williams (2010) provide comprehensive recommendations for future research on writing instruction involving individuals with complex communication needs.

Evidence-based literacy instruction for typically developing children supports comprehensive instruction; however, only one study identified in this review investigated multicomponent instruction. As mentioned, no studies evaluated comprehensive instruction that included each of the Big Five. This is in line with previous reviews which have found that research on literacy instruction for children with developmental disabilities typically focuses on teaching isolated literacy sub-skills (Alquraini & Rao, 2020; Arciuli & Bailey, 2021; Bailey & Arciuli, 2020; Browder et al., 2006). Recent educational policies and reports (e.g. *The*

Reading Framework report by the UK Department for Education, 2021a; and the *Right to Read: Public inquiry into human rights issues affecting students with reading disabilities* by the Ontario Human Rights Commission, 2022) state that children with disabilities require the same high-quality, explicit, and comprehensive literacy instruction as is offered to typically developing children. However, results of this systematic review indicate that comprehensive literacy instruction practices are not currently being investigated for children with CP.

Future research should also include representative samples of participants with diverse motor and communication abilities. In the current review, children with limited verbal communication accounted for most participants in instruction targeting phonics, sight-word recognition, and spelling skills and were not included in any studies on reading fluency or reading comprehension. This may be due to these instruction methods typically requiring a verbal response to measure outcomes. For example, reading fluency is defined by the NRP as the ability to read text with speed, accuracy, and proper expression. Considering that approximately 80% of children with CP have delayed or disordered speech production (Mei, Reilly, et al., 2020), verbal reading fluency may be a particular area of difficulty for many children with CP. Future research needs to investigate instruction methods and outcome measures that allow all children to participate in literacy instruction and demonstrate their abilities. This includes a focus on assistive technology in literacy instruction for children with CP, particularly given frequent new developments in technology. For example, no studies identified in this review explored the use of eye-gaze technology in literacy instruction, even though this technology offers individuals with significant motor impairments independent and direct access to literacy materials (Karlsson et al., 2021).

Analyses of large, longitudinal datasets have revealed that expectations of children with disabilities are particularly important in their academic success (O’Donnell et al., 2022). Factors such as parent, educator, and clinician expectations need to change to ensure that implementation of literacy instruction involving children with CP has the greatest chance of success (Koppenhaver, 2000; Zascavage & Keefe, 2004). This should be a focus for future research.

2.6 Conclusion

Our search of the literature identified 24 articles related to literacy instruction and children with CP. Almost all studies utilised single-subject research designs and only seven studies received a high quality rating. Most studies investigated instruction targeting literacy skills in isolation, with no study investigating comprehensive instruction involving all five key skills recommended by the NRP. This review highlights gaps in the research involving children with CP that explore literacy instruction methods known to be effective for typically developing children and other groups of children (e.g., children with autism). These gaps impact educators' and clinicians' ability to learn about and provide evidence-based literacy instruction to children with CP, which may, at least partially, account for the poorer literacy outcomes documented for this group. We propose that systematic and explicit literacy instruction utilising evidence-based principles can be effective for children with CP; however, further research is greatly needed. Future studies should focus on investigating comprehensive literacy instruction and should make use of high-quality research designs. Additionally, future research should explore environmental factors that may affect literacy instruction for children with CP, including addressing the impact of stigma and stakeholders' expectations of literacy for individuals with CP.

CHAPTER 3 EMPIRICAL STUDY 1

Computer-assisted Literacy Instruction via Telepractice for a Child with Cerebral Palsy: A Case Study

3.1 Abstract

Background: Many children with cerebral palsy (CP) underachieve in reading, yet no studies have investigated comprehensive literacy instruction for these children (known to be the gold standard for typically developing children). We explored the use of a freely available comprehensive literacy web application (ABRACADABRA), supplemented by parent-led shared book reading (SBR), on the reading skills of one child with CP.

Method: This case study involved one 8-year-old girl with spastic quadriplegic CP. Reading outcomes were measured using standardised assessments administered at three timepoints: baseline (T1), after a 6-week no-intervention control phase (T2), and following 6-weeks of intervention (T3). Each week over the intervention phase, a speech pathologist delivered three 60-minute sessions via telepractice using the comprehensive literacy program, and parents completed two 15-minute SBR activities. Quantitative data from standardised assessments, as well as qualitative analysis of parent SBR logs, were used to capture outcomes.

Results: No changes to reading skills were made over the control phase (between T1 and T2 assessments). At the final assessment (T3), the participant was able to correctly identify more letter-sound correspondences and read some words on standardised measures independent of the intervention. There was an increase in the participants' ability to use a phonological decoding strategy at T3 compared with T2. Engagement with the parent-led SBR program was high.

Conclusion: Children with CP can make gains in their reading skills over a relatively short period of time when provided with intensive, high-quality, evidence-based comprehensive literacy instruction across multiple settings. Future research needs to explore comprehensive literacy instruction with a larger sample of children with CP with diverse abilities.

3.2 Introduction

Children with cerebral palsy (CP) are more likely to experience poorer literacy outcomes than their typically developing peers (Larsson et al., 2009; Peeters, Verhoeven, de Moor, & van Balkom, 2009; Wotherspoon et al., 2023). This can be true, even when intellectual abilities, language, and communication skills are within the average range (Critten et al., 2019; Micheletti et al., 2023; Wotherspoon et al., 2023). This case study explores a computer-assisted literacy program, ABRACADABRA (hereafter referred to as ABRA; Centre for the Study of Learning and Performance [CSLP], 2019), supplemented by parent-led shared book reading (SBR), for one child with CP. ABRA is a free multimedia web-based literacy application designed based on the recommendations of the National Reading Panel (NRP; National Institute of Child Health and Human Development [NICHD], 2000) and other evidence on effective reading instruction. ABRA consists of distinct modules that can be used flexibly to target reading skills and sub-skills based on an individual's needs. The program has been used extensively with typically developing children, demonstrating positive effects on phonemic awareness, phonics, reading fluency, listening comprehension, reading comprehension, and vocabulary skills (Abrami et al., 2020). ABRA has also been used with autistic children (Arciuli & Bailey, 2019, 2021; Bailey et al., 2017; Bailey et al., 2022), though has not previously been used with children with CP.

3.2.1 Cerebral Palsy and Literacy

The systematic review presented in Chapter 2 outlined the existing literature on literacy instruction for children with CP, published from 2000 to 2020. This review highlighted that no previous studies have explored comprehensive literacy instruction for children with CP (instruction incorporating the NRP Big Five: phonemic awareness, phonics, reading fluency, vocabulary, and reading comprehension). Comprehensive literacy instruction is considered high-quality and evidence-based and warrants exploration with this group.

Prior literacy instruction research involving children with CP has been delivered face-to-face. No studies have explored literacy instruction delivered via telepractice for children with CP. Telepractice permitted remote delivery of instruction and intervention during the global pandemic, and provides a flexible, convenient, and accessible service delivery option longer-term (Kwok et al., 2022). Other services for children with CP have been explored via telepractice, such as intensive speech intervention (e.g., Korkalainen et al., 2023), clinical swallowing evaluation (Kantarcigil et al., 2016), and language intervention (Micheletti et al., 2021). These studies all indicate that children with CP can participate in services delivered via telepractice, with high levels of parent satisfaction, and that these services can improve speech and language functioning for children with CP.

3.2.1.1 Impact of Motor Speech Impairments on Literacy Development. Motor speech impairments are common in children with CP and are typically the result of an injury to the brain impacting the muscles involved in speech (Pennington, 2012). Motor speech impairments include: (a) childhood apraxia of speech (CAS), a disorder of motor planning for speech; and (b) dysarthria, a disorder of motor execution, which impacts the strength, range of movement, tone, and accuracy of speech movements (Iuzzini-Seigel et al., 2022). Mei, Reilly, et al. (2020) conducted a population-based registry study of children with CP aged 4 to 6 years and found that 82% had speech difficulties. This included 78% with dysarthria, 54% with articulation difficulties, 43% with phonological errors, and 17% with features of CAS. Many children demonstrated mixed speech presentations and speech difficulties were associated with language impairment.

Children with motor speech impairments are more likely to experience literacy difficulties (Ferreira et al., 2007; Gillon & Moriarty, 2007; Peeters, Verhoeven, de Moor, & van Balkom, 2009). This is due to the combined impact of motor planning and phonological impairments which can hinder a child’s developing linguistic system (Gillon & Moriarty, 2007; Marquardt et al., 2002). Few studies have specifically explored the relationship between motor speech impairments and literacy for children with CP. Ferreira et al. (2007) explored the literacy skills of 12 children with speech and physical impairments, including nine children with CP (diagnosed with dysarthria or apraxia). Ferreira et al. (2007) found that two-thirds of the children with CP and motor speech impairments performed very poorly on reading and related measures. Three children with CP and motor speech impairments performed relatively higher on the reading measures, though still performed below their typically developing peers. This included reading more slowly and having difficulty reading longer sentences. As far as we are aware, no other studies have investigated the relationship between reading and motor speech impairments for children with CP, though this has been explored in other populations.

Amongst other populations, studies have compared the reading and related skills of children with CAS to other groups, including children with developmental speech sound disorders or reading disorders. Overall, these studies have found that children with CAS present with poorer receptive and expressive language, phonological awareness, letter-sound knowledge, word reading, reading comprehension, and spelling skills than children with developmental speech sound disorders or children with reading disorders and no speech sound difficulties (Lewis et al., 2004; McNeill et al., 2009c; Miller & Lewis, 2022; Miller et al., 2019). Studies exploring combined speech and literacy intervention for children with CAS (not specific to CP) have primarily focused on phonological awareness skills (McNeill et al., 2009a; Moriarty & Gillon, 2006). These studies have demonstrated that, following a relatively short but intensive integrated speech and phonological awareness intervention, some children are able to make gains in their speech, phonemic awareness, letter-sound knowledge, and decoding skills on independent standardised measures (McNeill et al., 2009a; Moriarty & Gillon, 2006). Follow-up studies indicate that gains were maintained for most children at 6 months post-instruction (McNeill et al., 2009b, 2010). Studies have not explored literacy instruction for children with CAS outside of phonological awareness interventions.

3.2.1.2 Parent-led Shared Book Reading Interventions. SBR is an evidence-based approach to support language and literacy development in young children, including children with developmental disabilities (Boyle et al., 2019; Noble et al., 2019; Pillinger & Vardy, 2022; What Works Clearinghouse, 2010). Many SBR programs involve training parents to use specific strategies during shared reading, such as asking questions and expanding on children’s responses (e.g., Sim & Berthelsen, 2014). Few studies have explored SBR as a mode of intervention for children with CP. Akamoglu and Meadan (2019) explored SBR as a context for parent-implemented communication intervention in a single-case study involving two preschool children, including one child with CP. Prior to intervention, parents received training on strategies to be used before, during, and after reading, as well as strategies to encourage communicative behaviours from their child during SBR. Following the parent-implemented intervention, the child's frequency of communicative initiations (i.e., use of words and gestures) during SBR increased. Measures of emergent or conventional literacy were not collected. Target SBR strategies were implemented by parents with a high level of fidelity. We are not aware of any studies that have explored parent-led SBR for school-aged children with CP or that have explored the impact of SBR on direct literacy skills for these children.

Methods of parent training and home programs have received a lot of attention within intervention research for children with CP (Novak et al., 2009; Pennington et al., 2004; Pennington et al., 2009; Whittingham et al., 2011). Home programs are delivered by a parent in the home environment, with guidance, training, and support provided by a clinician (Novak & Berry, 2014). Home programs may be provided in isolation or in addition to an intervention program. Studies have demonstrated that parent training and home programs can be an effective method of service delivery to facilitate skill development in children with CP (see umbrella review by Novak et al., 2020). Much of the research on home programs for children with CP has explored motor skill development (e.g., Beckers, Geijen, et al., 2020; Novak et al., 2009), and parent training interventions have explored oral language and/or AAC-based communication outcomes (e.g., Pennington et al., 2004; Pennington et al., 2009; Whittingham et al., 2011). Compliance with home programs is generally high. A systematic review by Beckers, Geijen, et al. (2020) identified 56% to 99% compliance from families across studies of home programs for upper limb skills in children with CP. Parents' engagement and compliance with home programs is likely driven by a desire to maximise their child's progress and from seeing their child progress towards their goals (Novak, 2011; Piggot et al., 2002). Parents report feeling more confident, and finding programs easier to implement, when they are provided with effective coaching from professionals (Beckers, Geijen, et al., 2020). Although some studies have explored parent implemented home programs for handwriting (Novak et al., 2009), other aspects of literacy, such as reading, have not been explored in parent training or home programs for children with CP.

3.2.2 Case Study Research

Single participant research is described using different terms throughout the literature, including *n* of 1, case study research, case reports, and single-case experimental designs (SCED) amongst others (Horner et al., 2005; Krasny-Pacini & Evans, 2018; Riley et al., 2017; Vance & Clegg, 2012). There are notable differences between SCEDs and case study research. SCEDs are designed to demonstrate causality between an independent and dependent variable, involve repeated measurement of outcome variables at multiple time points, include replication within or between subjects, and traditionally include visual analysis of graphed data (Horner et al., 2005; Maggin et al., 2018; Tate et al., 2016). Meanwhile, case study research is more descriptive in nature, reports an in-depth investigation of the effects of an intervention with an individual, including the clinical presentation, intervention, and outcomes, and “incorporates multiple streams of data combined in creative ways” (Alpi & Evans, 2019, p. 1). Case study research can, but does not always, include an element of experimental control (Vance & Clegg, 2012). If thoroughly reported, the depth and detail of either single-subject methodology can provide valuable information to determine whether an intervention or approach may be applicable to an educator and clinicians’ own setting (Horner et al., 2005; Riley et al., 2017; Vance & Clegg, 2012).

Case study research has a pivotal role in establishing and advancing knowledge within heterogeneous populations, such as children with developmental disabilities. According to Vance and Clegg (2012), robust and rigorous case study research can make a valuable contribution to the limited evidence base on educational and clinical interventions for child language and communication. Case studies are particularly useful for piloting novel interventions or applying an existing evidence-informed intervention to a new population (Krasny-Pacini & Evans, 2018). Case studies have a long history in speech pathology research (e.g., McNeill et al., 2009b; Vance & Clegg, 2012) and have been used frequently in research involving individuals with heterogeneous presentations, such as CP (e.g., Beckers, Stal, et al., 2020; Korkalainen et al., 2022). Case study research has played an important role in reading and education research, particularly special education (e.g., Horner et al., 2005; Maggin et al., 2018; Singh et al., 2021), and SCED research has formed the basis of some important recommended practices in education, including recommendations from the NRP (e.g., the reading fluency subsection of the NRP report; NICHD, 2000).

3.2.3 The Current Study

This case study explores the impact of comprehensive literacy instruction, delivered via telepractice and supplemented by a parent-led SBR program, on the reading skills of one 8-year-old girl with spastic quadriplegic CP. The aims of this single-case study were to: (a) detail the engagement and performance of a child with CP in online literacy sessions; (b) describe the feasibility, engagement, and outcomes of a home-based parent-led SBR program; and (c) report and explore any changes in this child's reading skills following participation in the program. In line with the definition of case study research provided by Alpi and Evans (2019), our case study explores multiple sources of data and evidence. As such, we report both quantitative and qualitative data, including results from standardised assessments, data collected during instruction sessions, and qualitative analysis of parent SBR logs.

3.3 Method

This research was approved by the Flinders University Human Research Ethics Committee and was part of a larger project on online literacy instruction for children with developmental disabilities conducted during the COVID-19 pandemic, including children with autism and children with Down syndrome. Results for the study involving children with Down syndrome are reported in Chapter 4 and have been published in the *British Journal of Educational Psychology* (Murphy et al., 2023). Results for the study involving children with autism are reported separately (Bailey et al., 2022).

A recruitment flyer was circulated through organisations and clinics supporting children with developmental disabilities Australia wide. The eligibility criteria were: (a) 6-12 years of age; (b) diagnosis of cerebral palsy; (c) no major hearing or vision impairments; (d) able to communicate verbally at sentence level; (e) able to identify at least one letter of the alphabet; (f) able to demonstrate sustained attention to task for at least 15-minutes; (g) living in Australia; (h) use English as their first language; (i) access to a computer with a webcam and internet. Only one child with CP responded to the advertisement and met the above criteria. Both the child and her parents provided written consent prior to participating in the study.

3.3.1 Participant

Sarah (pseudonym) was an 8-year-old Caucasian girl with spastic quadriplegic CP. She was classified as Level III on the Gross Motor Function Classification System, meaning she walked using an aid and used a wheelchair for longer distances (Palisano et al., 2008). She was able to handle most objects, though with reduced speed and quality (Manual Ability Classification System Level II; Eliasson et al., 2006). Sarah communicated verbally, though had reduced speech intelligibility (Viking Speech Scale Level III; Pennington et al., 2013). She presented with features of dysarthria, including imprecise articulation and decreased intelligibility over longer phrases, and had a diagnosis of CAS from her local speech pathologist (per parental report). We observed that she had characteristics consistent with a CAS diagnosis (e.g., inconsistent speech sound errors, articulatory groping); however, no formal motor speech assessment was conducted. A measure of single word articulation was completed at baseline (The Quick Screener; Bowen, 1996) and Sarah presented with 34% consonants correct. Sarah could communicate effectively with familiar and unfamiliar communication partners, though at a slower rate (Communication Function Classification System Level II; Hidecker et al., 2011), and had receptive and expressive language difficulties. Sarah was in her second year of schooling (Grade 1) and attended a mainstream public school. She spoke English as her primary language and was not exposed to any other languages at home. Her parents reported no hearing impairment and functional vision when wearing her glasses. Prior to commencing the study, Sarah engaged in reading with her parents at home daily.

3.3.2 Measures

3.3.2.1 Baseline Descriptive Measures. The following assessments were administered at baseline to gather descriptive information (see Table 3.1 for results of these assessments):

3.3.2.1.1 Nonverbal Intelligence. Nonverbal cognitive ability was assessed using the Raven's 2 Progressive Matrices Clinical Edition – Digital short form (Raven's 2; Raven et al., 2018). Sarah was shown an incomplete matrix and was asked to point to the correct piece from a choice of five to complete the pattern. The test-retest reliability of the Raven's 2 digital short form is good for both a normative sample (*r* = .80) and a sample of individuals with intellectual disabilities (*r* = .82).

3.3.2.1.2 Phonological Short-Term Memory. Sarah's phonological short-term memory (STM) was measured using the Number Repetition subtest of the Clinical Evaluation of Language Fundamentals – Fourth Edition (CELF-4; Semel et al., 2003). This test required Sarah to repeat a series of numbers in the same order as presented. Both forward and backwards digit spans, progressively increasing in length, were included. The test-retest reliability for this subtest shows good consistency (*r* = .78), as per the assessment manual.

3.3.2.1.3 Phonological Awareness. The Elision subtest of the Comprehensive Test of Phonological Processing – Second Edition (CTOPP-2; Wagner et al., 2013) was administered to measure Sarah's ability to segment and manipulate sounds in words. Sarah was asked to delete individual syllables or phonemes in words to create a new word. Test-retest reliability for this subtest, as reported in the assessment manual, shows good consistency (*r* = .73).

3.3.2.1.4 Adaptive Ability. The 'Parent/Caregiver Rating Form' of the Vineland Adaptive Ability Scales – Second Edition (VABS-2; Sparrow et al., 2005) was administered to evaluate adaptive ability. Sarah's parents responded to questions regarding Sarah's ability to demonstrate specific behaviours in the domains of communication, daily living skills, and socialisation. Based on the normative sample, the test-retest reliability for the VABS-2 Adaptive Behaviour Composite score shows good consistency (r = .92).

Table 3.1 *Scores on Baseline Descriptive Measures*

Measure	Standard/ scaled score	Percentile rank	Descriptive classification
Nonverbal IQ	76	5	Very low
Phonological STM	1	0.1	Very poor
Phonological awareness	1	<1	Very poor
Adaptive behaviour domains:			
Communication	72	3	Moderately low
Daily living skills	74	4	Moderately low
Socialisation	94	34	Adequate
Adaptive Behaviour Composite Score [a]	78	7	Moderately low

Note. Nonverbal IQ = Raven's 2 Progressive Matrices Clinical Edition digital short form; Phonological Short-Term Memory (STM) = Clinical Evaluation of Language Fundamentals – Fourth Edition (CELF-4), Number Repetition subtest; Phonological awareness = Comprehensive Test of Phonological Processing – Second Edition (CTOPP 2), Elision subtest; Adaptive behaviour = Vineland Adaptive Ability Scales – 2nd edition (VABS-2).
[a] Based on scores for the adaptive behaviour domains: Communication, Daily Living Skills, and Socialisation

3.3.2.2 Literacy Outcome Measures. A battery of standardised assessments were administered to measure literacy outcomes at three assessment timepoints (baseline, pre-instruction, and post-instruction). The literacy outcome measures are described in Table 3.2.

Table 3.2 *Literacy Outcome Measures*

Measure	Assessment tool	Descriptor
Word-level reading accuracy	Word Reading subtests of the Wide Range Achievement Test – 4th Edition (WRAT-4; Wilkinson & Robertson, 2006)	This subtest contains two sections: 'Letter Reading' and 'Word Reading'. In 'Letter Reading', children are asked to name 15 capital letters. In 'Word Reading', children read words of progressively increasing difficulty aloud. The internal consistency for this subtest is high (r = .92).
Supplementary measure of word-reading accuracy	Castles and Coltheart Test – 2nd Edition (CC-2; Castles et al., 2009)	Children read aloud a series of words of increasing difficulty, including real words with regular or irregular spellings, and nonwords. Words are presented in mixed order. This test has acceptable internal consistency (r_s = .90).
Passage-level reading accuracy	Reading Accuracy Composite score from the Neale Analysis of Reading Ability – 3rd Edition (NARA-3; Neale, 1999)	Children read aloud passages of text. These passages increase in length and complexity. Test-retest statistics show good consistency (r = .95).
Passage-level reading comprehension	Reading Comprehension Composite score from the NARA-3 (Neale, 1999)	After reading aloud the passages above, children are asked a series of set questions about the text. Test-retest statistics show good consistency (r = .93)
Functional reading comprehension	Test of Everyday Reading Comprehension (TERC; McArthur et al., 2012)	Children view pictures of text encountered during everyday situations (e.g., a road sign, a recipe) and respond to set questions about the text. This test has high inter-rater reliability (r = .99)

Note. Reliability statistics are based on the normative sample (as reported in assessment manuals).

3.3.3 Procedure

This study involved two phases: a 6-week no-intervention control phase and a 6-week instruction phase. Standardised assessments were administered at: baseline (T1), pre-instruction (T2), and post-instruction (T3).

3.3.3.1 Baseline Assessment. Sarah participated in a 90-minute baseline assessment via Zoom (a videoconferencing platform) with the researcher (a speech pathologist). She was seated at a desk with a laptop computer and supported by her father.

The descriptive measures and literacy outcome measures previously described were administered via screensharing. The measure of adaptive ability was completed with Sarah's parents at a separate time.

3.3.3.2 No-intervention Control Phase. Following the baseline assessment, Sarah entered a 6-week control phase where she did not engage with the researcher. This period of time without intervention provided a measure of how Sarah's literacy skills were progressing without specific intervention.

3.3.3.3 Pre-instruction Assessment. Prior to beginning intervention, Sarah again completed the literacy outcome measures. These were administered in the same way and in the same order as the baseline assessment.

3.3.3.4 Instruction Phase. The instruction phase ran over 6-weeks and involved two components: (a) three 60-minute online ABRA sessions per week via Zoom with the researcher, outside of school hours (for Sarah this included one before school session, one after school session, and one session on a Saturday each week); and (b) two 15-minute parent-led SBR activities per week. Each of these components is further explained below. Sarah was supported by her father for all online sessions and parent-led SBR activities. Prior to beginning the instruction phase, Sarah was sent an iPad with the Fitzroy Readers apps (Fitzroy Programs and Greygum Software, 2016). The Fitzroy readers are an Australian graded decodable book series.

3.3.3.4.1 Online ABRA sessions. The ABRACADABRA LITE version of the ABRA program (CSLP, 2019) was used in this study (https://literacy.concordia.ca/abra/en/). All online sessions followed the same session structure: (a) 15-minutes of word-level activities in ABRA targeting phonemic awareness or phonics skills; (b) 20-minutes of passage-level activities in ABRA targeting reading comprehension skills and/or decoding within passage-level text; (c) 15-minutes of clinician-led SBR targeting generalisation of reading skills; and (d) a 10-minute reward activity. A visual schedule containing the day's activities was used to support engagement in sessions. During the online ABRA sessions the researcher screen-shared the ABRA program so that Sarah could click and engage directly with the activities.

Due to her fine motor difficulties, Sarah needed support to click and use the computer mouse accurately.

Results from the baseline and pre-instruction assessments were used to set goals and determine appropriate ABRA activities and start points. During each session, the researcher recorded task accuracy data (number of items correct/incorrect and prompts required). Following each session, the researcher reviewed Sarah's goals and activity level difficulty in line with this data. For further details on ABRA learning activities, see the ABRACADABRA Teacher Guide (CSLP, 2019).

3.3.3.4.2 Parent-led Shared Book Reading. Sarah's father participated in a 60-minute online training session regarding the SBR procedures at the beginning of the instruction phase. Sarah's father was asked to use the following procedures in SBR (these same strategies were also used in the clinician-led SBR within ABRA sessions): (a) take turns reading pages; (b) ask a question every second page (three levels of questions from concrete to more abstract were introduced); (c) support decoding of words using a template provided; and (d) end SBR with a review of content read (ask three questions about the text and revise the words recorded on the decoding errors template). Sarah's father was asked to fill in a brief reading log, including details on reading accuracy and reading comprehension, following each SBR session. The researcher met with Sarah's father for approximately 15-minutes each week to review the SBR log and set the target decodable books and question level for the upcoming week.

3.3.3.5 Post-instruction Assessment. Following the 6-week instruction phase, the literacy outcome measures were again administered via Zoom to Sarah.

3.4 Results

Sarah participated in 17 online ABRA sessions over the 6-week instruction phase and completed all parent-led SBR activities. Outcomes for standardised literacy assessments, ABRA telepractice sessions, and parent-led SBR are detailed below.

3.4.1 Outcomes on Standardised Assessments

Sarah's raw scores at each assessment timepoint are shown in Table 3.3. Sarah was at the very early stages of learning to read and had limited responses on the passage-level assessments. Her responses on the word-level reading measures are analysed in the sections below.

Table 3.3 *Raw Scores on Baseline, Pre-, and Post-Instruction Standardised Literacy Outcome Measures*

Measure	Baseline	Pre-instruction	Post-instruction
Word-level reading accuracy (primary)[a]	0	0	2
Word-level reading accuracy (supplementary)	0	0	0
Passage-level reading accuracy	0	0	0
Passage-level reading comprehension	0	0	0
Functional reading comprehension	0	0	0

[a]The WRAT-4 requires children to say the letter name in the Letter Reading subsection of the Word Reading subtest to be scored correct. Sarah correctly produced some letter-sound correspondences at all timepoints, though was unable to correctly name any letters at any assessment timepoint.

3.4.1.1 WRAT-4 Letter Reading Subsection. Sarah was unable to correctly name letters on the WRAT-4 'Letter Reading' subsection at any assessment timepoint, though produced some correct letter-sound correspondences (see Table 3.4). Sarah was inconsistent with the letter-sounds she correctly produced at each assessment. She consistently labelled four letters across the three timepoints: A, O, S, P.

Table 3.4 *WRAT-4 Letter-Sound Correspondences Correctly Produced at Baseline, Pre-, and Post-Instruction*

Assessment timepoint	No. of letter-sounds correct	Letter-sounds correctly produced									
Baseline	6	A	O	S	H	P	I				
Pre-instruction	6	A	O	S	-	P	-	B	E		
Post-instruction	8	A	O	S	-	P	I	B	-	T	U

Note. There are 15 letters in the 'Letter Reading' subsection of the WRAT-4 Word Reading subtest.

3.4.1.2 WRAT-4 Word Reading Subsection.

Sarah was unable to correctly read any words on the WRAT-4 assessment at T1 and T2. She attempted to decode two words at T1 and no words at T2. At T3 she read ‘cat’ and ‘in’ correctly and attempted to decode an additional three words unsuccessfully. She initially decoded ‘cat’ correctly (i.e., ‘c-a-t’), though said “mat”. She self-corrected on her second attempt.

3.4.1.3 Decoding Attempts on the CC-2.

Sarah did not correctly read any words on the CC-2 at any timepoint. However, there was a significant difference in decoding attempts between T2 and T3 (see Table 3.5). Although some words were accurately decoded in the T3 assessment, Sarah was unable to produce the target word correctly (e.g., ‘m – i – s – t’ was said as “with”; ‘j – o – p’ was said as “op”). Of the 15 words attempted on the CC-2 at T3, Sarah only attempted to blend nine words after decoding.

Table 3.5 *Comparison of Decoding Attempts on the CC-2 at Pre-Instruction Versus Post-Instruction*

	Decoding attempts			
	Pre-instruction		Post-instruction	
Target word	**Sounds decoded**	**Graphemes correct**	**Sounds decoded**	**Graphemes correct**
bed	/b – æ/	1	/b – ɪ – b/	1
good	/e/	0	/ɒ – p/	0
norf	/m/	0	/n – ɒ – f/	2
gop	/p/	1	/j – ɒ – p/	2
wolf	-	0	/w – ɒ – l – f/	2
long	-	0	/l – ɒ – n – f/	2
work	/w – e – p/	1	/w – ɒ – f/	1
mist	/m/	1	/m – ɪ – s – t/	4
hest	-	0	/w – æ – s – t/	2
roft	/æ – s – l/	0	/f – ɒ – f/	2
free	-	0	/f – ɒ/	1
eye	/e/	0	/w – e – ɪ – k/	0
take	/e/	0	/æ – f – e/	0
give	-	0	/æ – ɪ – w – e/	1
blick	/b – e/	1	/f – æ – l – ɪ – k/	3
Total graphemes correct		5		23

3.4.2 Online ABRA sessions

Table 3.6 details the frequency of word- and passage-level ABRA activities completed by Sarah during the online sessions. Accuracy data for word-level activities is displayed in Table 3.7.

Figure 3.1 displays Sarah's accuracy with decoding target consonant-vowel-consonant (CVC) words within the clinician-led SBR activity in ABRA sessions. Fourteen different texts were utilised across sessions (three texts were read twice). As such, decoding targets differed across sessions. Sarah had two to three target words per book for sessions one to nine (Sarah was prompted to read the target word each time it occurred in the book). From session 10 onwards, she had four to six target words per book. Sarah's reading accuracy varied across sessions, though there was an upward trend line for decoding accuracy over the program. Variation across sessions was likely due to several factors, including familiarity with target sounds in CVC words, fatigue, attention and engagement, and impact of her motor speech disorder.

Table 3.6 *Frequency of ABRA Activities Completed During Sessions*

Word level activities:	No. of times completed	Passage level activities:	No. of times completed
Letter-sound search	3	Tracking	6
Word counting	1	Accuracy	1
Same word	1	Prediction	4
Animated alphabet	6	Summarising	6
Letter-bingo	3	Story elements	3
Word matching	3		
Word families	5		
Auditory blending	4		
Basic decoding	6		

Note. Only the activities completed by Sarah are included in the table. ABRA includes other activities not outlined above.

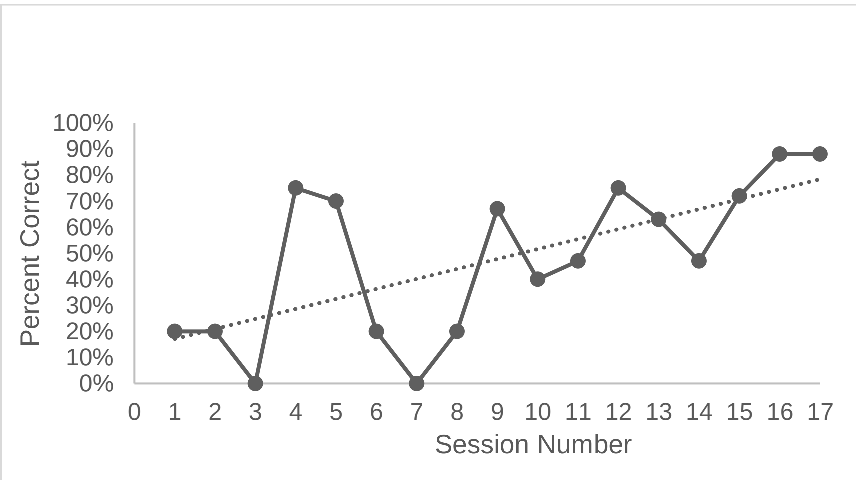
Table 3.7 *ABRA Word-Level Activity Accuracy Data*

Activity	Session level (accuracy)	Session level (accuracy)	Session level (accuracy)	Session level (accuracy)	Session level (accuracy)	Session level (accuracy)
Letter-sound	Session 1 Lvl 1 (60%)	Session 2 Lvl 1 (70%)	Session 12 Lvl 1 (67%)			
Word counting	Session 2 Lvl 1 (50%)	-	-			
Same word	Session 3 Lvl 1 (86%)	-	-			
Animated alphabet [a]	Session 7 Letters: g, l, h, t, a	Session 9 Letters: w, s, h, a, l, n	Session 11 Letters: g, y, a, d	Session 14 Letters: l, t, m, f, a, b, l, v	Session 15 Letters: t, e, o, a	Session 17 Letters: l, t, m, l, j
Letter-bingo	Session 1 Uppercase (0%)	Session 3 Lowercase (66%)	Session 6 Uppercase (78%)			
Word matching	Session 8 Lvl 1 (67%)	Session 10 Lvl 1 (67%)	Session 15 Lvl 1 (89%)			
Word families	Session 5 Lvl 1 (67%)	Session 6 Lvl 1 (67%)	Session 9 Lvl 1 (83%)	Session 12 Lvl 1 (100%)	Session 16 Lvl 1 (67%)	
Auditory blending	Session 4 Lvl 1 (100%)	Session 5 Lvl 1 (67%)	Session 10 Lvl 1 (100%) Lvl 2 (33%)	Session 16 Lvl 2 (33%)		
Basic decoding	Session 4 Lvl 1 (0%)	Session 7 Lvl 1 (30%)	Session 11 Lvl 1 (30%)	Session 13 Lvl 2 (50%)	Session 15 Lvl 2 (80%)	Session 17 Lvl 2 (50%)

Note. Lvl = ABRA activity level.

[a] This activity is not based on accuracy and was used to practise letter identification.

Figure 3.1 *Accuracy for Target CVC Words Read in Decodable Readers*



3.4.3 Review of Researcher Session Notes

3.4.3.1 Engagement and Access. Sarah's engagement in ABRA sessions was generally good, though her wariness of making errors impacted her willingness to attempt some tasks. Modifications were made to assist Sarah's access to ABRA, such as increasing the cursor size on the screen, trialling touch screen access (which reduced accuracy compared with the computer mouse), and full assistance with using the mouse (e.g., Sarah pointed to items on the screen and her father selected). As sessions progressed, Sarah's accuracy with using the mouse independently improved, though she remained unable to complete some activities.

3.4.3.2 Print and Sound Awareness. Sarah's print awareness increased throughout the program. For example, in session 13 she said, *"what's this?"* whilst pointing to a low frequency letter ('z') and in session 16 she asked, *"what's this?"* whilst pointing to a question mark in the text. Prior to these observations, Sarah had not made any explicit comments about the text except for a select few highly familiar letters (e.g., *"/d/ for daddy name"*).

3.4.3.3 Support Needs. Blending was recorded in Sarah's session notes as an area of difficulty during online ABRA sessions. She was often able to decode a word correctly, though was unable to blend a word until she heard the sounds decoded aloud by an adult. Sarah's speech inconsistencies were also noted to impact her performance in ABRA sessions.

3.4.4 Analysis of SBR Logs

Sarah's father returned eight completed SBR logs (in some weeks Sarah's father completed the set books twice and returned two logs). Comments in the logs indicated that Sarah's father was following the SBR protocol and providing both decoding and comprehension support. Examples of comments regarding reading accuracy and reading comprehension are included in Table 3.8.

Table 3.8 *Examples of Reading Accuracy and Comprehension Comments from SBR Log*

Reading component	Example quotes from reading log
Reading accuracy	• "*(Sarah) is blending letters to form words really well*" • "*sounding out became better as the book progressed*" • "*still need to work on "u". "e" and "I" were pretty good, "the" was a sight word that she read well*"
Reading comprehension	• *"Comprehension is improving and quite good and consistent for Level 1 + 2. (Sarah) is answering and trying well with 'what happens next?'"* • *"Questions answered well when fresh. Harder to recollect at end of story but she did well with some cues"* • *"Her memory is good when asked a question about the page just read"*

Along with comments on reading accuracy and comprehension, review of comments in the log revealed four main themes:

3.4.4.1 Consistency/Repetition. Sarah's father often reflected on consistency and repetition of reading activities as being crucial to Sarah's reading success: *"Repetition is the key to get good concentration and recognition"* and *"Consistent reading is vital - she was a bit rusty with her vowels - I didn't read for 2 days (sorry)"*. Repetition and consistency were particularly key for tasks that were more difficult: "*More words to sound out does tire (Sarah) out a bit, but the repetition is important too*".

3.4.4.2 Speech Sound Errors. Sarah's father noted that Sarah's speech sound errors impacted her reading accuracy. For example, Sarah "a*lways notices /d/ was for dad but sometimes said /b/*" and she "*recognises words but says wrong letters at times*". He commented on strategies she used to assist with producing sounds during reading: *"(Sarah) uses her phonics signing to demonstrate a letter that she knows but is having trouble to get out"*.

3.4.4.3 Engagement. Sarah's engagement in SBR sessions was regularly noted in the SBR logs: "*She was really engaged on all pages tonight*" or "*Needs me to keep her focused*". When engagement was high, Sarah's reading accuracy increased: "*When (Sarah) concentrates on letters, she continues to improve with forming the word*".

3.4.4.4 Strategies and Support. One of the most frequently commented on aspects in the log was the support that Sarah's father was providing: "*I do constantly need to remind her to focus on the words. Then she does pretty well*", as well as specific strategies used during SBR, such as "*I need to tell her to 'sound it out' and not just guess*" and *"bringing the word into big font caught her attention and works well"*. Her father recognised the need to increase her independence with reading: *"Tonight she looked at me a lot for help. I needed to remind her that 'she can do it' without looking at me for help/answers"*, though acknowledged the support she still required: "*If I let her read on her own she 'guesses' again. So direction/focus is really needed"*.

3.5 Discussion

This single-case study evaluated use of ABRA delivered via telepractice, supplemented with a parent-led SBR program, for one child with CP during the COVID-19 pandemic. The child in this study, Sarah, made modest gains in her letter-sound knowledge and word reading skills following participation in the intensive literacy program. Findings in relation to each of the study's aims are discussed below.

3.5.1 Engagement and Performance in Online Sessions

The first aim of this study was to detail the engagement and performance of a child with CP in online literacy instruction sessions. Consistent with previous telepractice research exploring speech and language interventions for children with CP (e.g., Korkalainen et al., 2023; Micheletti et al., 2021), Sarah was able to participate in the target number of sessions via telepractice and made some skill gains. Previous studies involving telepractice delivery of interventions to children with CP have involved the child interacting verbally with the clinician online (Korkalainen et al., 2022), or interacting with their parent under guidance from the clinician (Micheletti et al., 2021), and have not required the child to directly access the computer in any way. In the current study, the child with CP was required to use the computer mouse to engage directly with the ABRA program during telepractice sessions. Many mainstream technologies contain features that can increase accessibility for individuals with diverse access needs (De Witte et al., 2018). Such features were utilised in this study and were an important factor in Sarah's successful participation in the online sessions (e.g., modifying the display of the cursor on the screen). However, despite modifying her existing home technologies to the best of our abilities, some ABRA activities were not accessible for Sarah's motor abilities. For example, Sarah was unable to click and drag items in some games which reduced her independence with these activities. Inclusive design is an important factor that should be considered in the design of educational programs to ensure they are accessible to children with a diverse range of skills and needs (González et al., 2013).

While Sarah was able to engage in all online sessions, her performance varied greatly (see Table 3.7 and Figure 3.1). Sarah's performance day to day was likely influenced by several factors, including levels of motivation and engagement, self-confidence, self-concept and self-efficacy, fatigue, and the impact of her motor speech impairment. CAS is a disorder of speech motor planning which, amongst other factors, results in inconsistency of speech sound production across words and multiple repetitions of words (ASHA, 2007; Iuzzini-Seigel et al., 2022). It is likely that Sarah's difficulties planning, sequencing, and executing the motor movements for speech affected her verbal decoding of words and accounted for at least some of the variability observed in her reading performance across sessions. Variability in task performance between sessions has been documented in previous literacy studies involving children with CP (e.g., Clendon et al., 2005; Fallon et al., 2004; Hanser & Erickson, 2007; Ip & Lian, 2005; McCarthy et al., 2015).

3.5.2 Parent-Led SBR

The second aim of this study was to describe the feasibility, engagement, and outcomes of a home-based parent-led SBR program for a child with CP. Sarah's father engaged extremely well in the home-based SBR program. Although only guided to complete two 15-minute activities per week, Sarah's father reported completing the activities almost daily some weeks. This is consistent with previous research on home-programs for children with CP which have found high levels of parent engagement (Beckers, Geijen, et al., 2020). Sarah's father frequently commented on Sarah's progress and his own confidence in providing appropriate support for her participation and reading accuracy in SBR. It is likely that key features of our SBR program contributed to the success of the parent-implemented SBR in this study. These features included appropriate and engaging materials, the SBR log to document progress, and regular parent coaching and support from a clinician (Novak & Berry, 2014).

3.5.3 Reading Outcomes

3.5.3.1 Reading Accuracy. Finally, this study aimed to explore any changes in reading skills for a child with CP following participation in the online literacy program. There were limited changes to Sarah's raw scores on standardised assessments across the three assessment timepoints, other than a small increase in the number of words read correctly on the primary measure of word-level reading accuracy (WRAT-4) post-instruction. In-depth review of Sarah's responses on the word-level assessments indicated that Sarah made some modest gains in her letter-sound knowledge and decoding skills post-instruction, when compared with her performance at baseline and pre-instruction. These findings are consistent with previous research which indicates that children with CP can make gains in their phonics skills over a relatively short period of time when provided with explicit phonics instruction (Ainsworth et al., 2016; Clendon et al., 2005; Coleman-Martin et al., 2005; Fallon et al., 2004; Heller et al., 2002; Millar et al., 2004; Swinehart-Jones & Heller, 2009).

Despite gains in decoding skills within the instruction sessions, Sarah did not generalise these skills to read any real or nonwords correctly on the supplementary measure of word-level reading accuracy (CC-2; Castles et al., 2009). Sarah demonstrated a substantial increase in the total number of graphemes correctly decoded at post-instruction, (gain of 18 graphemes correctly decoded when compared to pre-instruction); yet, she was unable to accurately read any words on this test. Similarly, within instruction sessions, Sarah's notes indicated that she was often able to decode words accurately, though had difficulty blending the sounds to produce a word. Inconsistent speech sound production in children with CAS can result in indistinct or inaccurate internalised phonological representations of words, making it difficult for children to accurately retrieve phonological forms (Marion et al., 1993; Marquardt et al., 2002). Such difficulties can affect children's reading skill development and potentially impacted Sarah's ability to accurately produce words she had verbally decoded (Carroll & Snowling, 2004; Elbro et al., 1998). Children with CP also often experience working memory and executive function deficits (Micheletti et al., 2023; Sakash et al., 2018) and indeed, Sarah's phonological memory and nonverbal intelligence were very poor as measured in the baseline assessment. These factors all likely impacted Sarah's ability to independently store, recall, and blend phonemes whilst actively decoding a word.

3.5.3.2 Reading Comprehension. Sarah did not make any changes to her reading comprehension scores on the standardised reading comprehension measures and achieved a score of zero at all three assessment timepoints. This is not surprising given that Sarah was at the very early stages of learning to read and decode words. Potentially, a measure of word-level reading comprehension or listening comprehension may have returned different results. The focus of early reading instruction is on developing phonemic awareness and phonics skills, as well as providing exposure to rich vocabulary and oral language through texts (Department for Education, 2023). Only once children develop the ability to read words with some level of automaticity and fluency does reading comprehension become the focus of instruction (Castles et al., 2018). Sarah's SBR logs indicated that she made small but meaningful changes in her comprehension of short decodable texts (whether read to her or by her) and her ability to answer questions of increasing difficulty. By the second half of the program, Sarah was beginning to respond to questions requiring prediction or reasoning. Ability to answer such questions are important for facilitating ongoing cognitive and linguistic development and future reading comprehension (Hogan et al., 2011).

3.5.4 Clinical Implications

There are several clinical implications of this single-case study. Firstly, some children with CP can participate in interventions via telepractice that require engaging with both a clinician and shared stimulus material on the screen (such as a multimedia literacy program). This has important implications for increasing access to education and clinical services for children with CP who may be more likely to miss school due to associated complex medical conditions. Secondly, parents see the value in home-based reading activities and can be effective partners in delivering a reading intervention to their child. Sarah's parents saw reading as a priority and were motivated to help Sarah to develop these skills. Motivation to complete home program activities is likely higher when aligned with parents' goals for their child (Novak, 2011; Novak & Berry, 2014). Lastly, children with CP and motor speech impairments can benefit from comprehensive literacy instruction, including some programs that are designed for typically developing children. Further research is required to determine the optimal period of intervention and specific support needs within instruction for children with CP.

3.5.5 Limitations and Future Research

Case study research provides valuable information on topics with a limited evidence base, particularly for heterogeneous populations such as children with CP; however, the generalisability of results are limited (Vance & Clegg, 2012). One of the benefits of case study methodology is the comprehensive participant description and level of detail regarding performance and responsiveness to intervention at the individual level. This detail can be used by educators and clinicians to identify similar individuals (Horner et al., 2005). This level of detail is unique to this chapter of this thesis and is not feasible within group-based studies. Future research should seek to include a larger sample of participants and employ high-quality randomised controlled trial or single-case experimental designs. Future research should also include children with CP with diverse motor and communication abilities.

Another limitation is the relatively short timeframe in which this study was conducted. We note that this length of time and intensity is similar to other literacy studies involving children with CP (five sessions per week over 6-weeks, Hanser & Erickson, 2007; two 30-minute sessions per week over 8-weeks, Ip & Lian, 2005) or children with CAS (two sessions per week over two 6-week intervention blocks, McNeill et al., 2009c; three 45-minute sessions per week over 3-weeks, Moriarty & Gillon, 2006). Six-weeks of intervention, albeit intensive, was likely an inadequate amount of time to make substantial changes to Sarah's reading skills considering the severe nature of her speech impairment and limited reading and phonological awareness skills at baseline. Children with lower nonverbal intelligence, such as Sarah, may require additional time to learn and integrate new skills due to slower processing skills (Miller et al., 2001). We note no objective measure of oral language skills as an additional limitation of this study.

Sarah's motor speech impairment was likely a significant factor impacting outcomes in this study. Although research around literacy interventions for children with CAS exists, it is relatively limited when compared to other populations of speech difficulties (e.g., developmental speech sound disorders or phonological disorders; Loudermill et al., 2021). Over 15 years ago, Gillon and Moriarty (2007) called for more research on literacy interventions for children with CAS. Since this time, there has been an increase in longitudinal and descriptive studies regarding the literacy skills of children with CAS; yet, very few literacy intervention studies have been conducted and these have primarily focused on phonological awareness skills (e.g., McNeill et al., 2009a; McNeill et al., 2009b, 2010). While this case study does not explore combined speech and literacy intervention, it adds to the sparse knowledge base on response to literacy instruction for children with CAS, an important topic which requires research attention. Future research should investigate methods of intervention that develop speech, language, and literacy skills in cooperation.

3.6 Conclusion

As far as we are aware, this is the first study of comprehensive literacy instruction via telepractice for a child with CP, and the first study to investigate a parent-implemented SBR program, targeting direct reading skills, for a child with CP. Session performance data and outcomes on standardised assessments suggest that the intensive ABRA and parent-led SBR program was successful in developing this child's letter-sound knowledge and ability to use a phonological decoding strategy. However, a longer intervention period was likely required to consolidate these skills. We hope that the results of this single-case study can inform further research in the field of literacy for children with CP, including children with motor speech impairments, an area that has received limited research attention.

CHAPTER 4 EMPIRICAL STUDY 2

Murphy, A., Bailey, B., & Arciuli, J. (2023). ABRACADABRA literacy instruction for children with Down syndrome via telepractice during COVID-19: A pilot study. *British Journal of Educational Psychology*, 93(1), 333-352. https://doi.org/10.1111/bjep.12558

4.1 Abstract

Background: COVID-19 has resulted in some educators and allied health practitioners transitioning to online delivery of literacy instruction. As far as we are aware, no studies have investigated online delivery of comprehensive literacy instruction for children with Down syndrome.

Aims: In this pilot study, we explore the efficacy of online delivery of ABRACADABRA (a free literacy web application) for children with Down syndrome, alongside supplementary parent-led shared book reading, during the COVID-19 pandemic.

Sample: Six children with Down syndrome, aged 8 to 12 years, participated in this within-participants design study.

Methods: Participants acted as their own controls with outcome variables measured at three timepoints: baseline, pre-instruction, and post-instruction. Children participated in 16-18 hours of one-to-one literacy instruction online over a 6-week instruction phase, along with twice weekly parent-led shared book reading activities.

Results: Outcomes from standardised assessments revealed statistically significant improvements in word- and passage-level reading accuracy skills over the instruction phase (pre-instruction to post-instruction) compared with the no-instruction control phase (baseline to pre-instruction). Improvements in reading comprehension skills were inconsistent across assessment measures and statistical analyses.

Conclusion: Children with Down syndrome can benefit from comprehensive literacy instruction delivered via telepractice. Our study provides critical initial evidence of successful service delivery during a global pandemic and beyond.

4.2 Introduction

Learning to read has been described as a "basic and essential human right" (Ontario Human Rights Commission, 2022, p. 2), with literacy skills providing a foundation for educational success and positive long-term life outcomes in vocational, social, and health domains. Children with neurodevelopmental disabilities have an increased likelihood of literacy difficulties (Browning, 2002; van Bysterveldt & Gillon, 2014) and may fall even further behind without access to usual education and therapy services. The COVID-19 pandemic has adversely impacted many children's access to their usual education and intervention services during critical periods of learning and development, potentially affecting acquisition of essential skills, such as literacy. Some children have been able to access education and clinical services online during the pandemic via telepractice. Telepractice is the use of a technology medium to connect professionals and clients at a distance in real-time to engage in assessment or intervention services (American Speech-Language-Hearing Association, n.d.-a). The efficacy of many interventions via telepractice is unknown, particularly reading and spelling interventions (Furlong et al., 2021). Even less is known about the efficacy of literacy interventions delivered via telepractice for children with neurodevelopmental disabilities, such as Down syndrome[2] (Pierson et al., 2021; Valentine et al., 2021). The current research was designed as a pilot study to explore the efficacy of literacy instruction delivered via telepractice for English-speaking children with Down syndrome.

4.2.1 Literacy and Children with Down Syndrome

Children with Down syndrome vary in their reading profiles. Most develop some conventional reading and writing skills, with the level of literacy attained linked to phonological awareness, oral language, and cognitive skills, along with the quality of instruction they receive, stakeholder expectations, and a range of other school related factors experienced by children with disabilities (Arciuli & Emerson, 2020; Arciuli et al., 2019;

[2] Down syndrome is a neurodevelopmental genetic disorder caused by a full or extra partial copy of chromosome 21, which alters development and causes developmental and intellectual disabilities (American Psychiatric Association, 2022; Bull, 2020).

Boudreau, 2002; Cologon, 2013; O'Donnell et al., 2022; Ratz, 2013). According to the Simple View of Reading (SVR), successful reading is the result of both decoding and linguistic comprehension skills (Gough & Tunmer, 1986). Many children with Down syndrome have difficulties with the various component skills that underpin decoding and comprehension in this model, such as short-term memory, phonemic awareness, and receptive language (Lemons & Fuchs, 2010; Næss et al., 2011). These difficulties, combined with relative strengths in visual processing skills, led some to believe that children with Down syndrome might benefit from reading instruction focused on recognition of whole words, rather than decoding using letter-sound relationships (Buckley & Bird, 1993; Byrne et al., 2002; Hodapp & Fidler, 1999). Contemporary research has provided evidence to the contrary and there is now general consensus that, as with all beginning readers, phonological awareness and phonics skills play an essential role in literacy development for children with Down syndrome (Cupples & Iacono, 2002; Lemons & Fuchs, 2010; Lim et al., 2014). Although less researched than phonological awareness and reading accuracy, reading comprehension is a considerable area of difficulty for many children with Down syndrome (Boudreau, 2002; van Bysterveldt & Gillon, 2014). A longitudinal study by Byrne et al. (2002) followed 24 children with Down syndrome over two years and found that children made steady progress with reading accuracy, but limited progress with reading comprehension over this time. These limited gains in reading skills may be, to some extent, due to ineffective or poorer quality literacy instruction (Cologon, 2013).

Much of the contemporary research on evidence-based early reading instruction is founded on the recommendations of a large-scale meta-analysis conducted by the United States National Reading Panel (NRP; National Institute of Child Health and Human Development [NICHD], 2000). According to the NRP, there are five key components essential for effective early reading instruction (phonemic awareness, phonics, reading fluency, vocabulary, and reading comprehension) and comprehensive instruction involves integration of all five of these skills. The recommendations of the NRP are based on studies that included many children without disabilities; however, it is now well accepted that these principles apply to all beginning readers. A recent report commissioned by the UK government on learning to read emphasised that this same high-quality evidence-based literacy instruction should be provided to all children, including children with developmental disabilities (Department for Education, 2021a). Yet, studies investigating literacy instruction for children with Down syndrome have often focused on isolated reading sub-skills or a combination of a few sub-skills during instruction, such as phonological awareness and decoding skills (Snowling et al., 2008). These studies have demonstrated that children with Down syndrome can make gains in their reading skills when provided with appropriate instruction. For example, Lemons et al. (2012) compared the effectiveness of two commercially available reading intervention programs targeting phonological awareness and decoding skills, Road to Reading and Road to the Code (Blachman et al., 2000; Blachman & Tangel, 2008), in a study involving 15 children with Down syndrome aged 5 to 13 years. Children demonstrated improvements on researcher designed measures of word reading accuracy following decoding intervention; however, limited gains were made in phonological awareness skills. A study by Lim et al. (2019) utilised the *Making Up Lost Time in Literacy* program (MULTILIT, 2007), an evidence-based program for improving reading and spelling skills in low progress readers. The study involved 15 children with Down syndrome from 9 to 17 years of age. Children demonstrated significant improvements on standardised measures of phonological awareness, word reading accuracy, and spelling following instruction. Studies, such as that reported by Burgoyne et al. (2012), have explored instruction targeting both vocabulary and word reading skills. This study was a randomised controlled trial involving 57 children with Down syndrome aged between 5 and 10 years. Compared with the control condition, children who received the intervention made statistically significant gains on measures of letter-sound knowledge, phoneme blending, single word reading, and taught expressive vocabulary. Intervention effects in this study did not transfer to other measures of literacy (e.g., spelling) or standardised receptive and expressive language assessments. As far as we know, there have been no previous studies that have considered comprehensive literacy instruction for children with Down syndrome.

4.2.2 Computer-Assisted Instruction

There is growing evidence that computers may provide an appropriate medium for literacy instruction involving children with Down syndrome (e.g., Næss et al., 2022; Nakeva von Mentzer et al., 2021). Computer-assisted instruction (CAI) utilising multimedia technology provides information via both visual and auditory modes which can reduce cognitive load during learning activities (Mayer & Moreno, 2003). This mode of instruction may be particularly well-suited to children with Down syndrome, given their profile of cognitive difficulties (Davis, 2008). Some studies involving children with Down syndrome that have compared computer-assisted literacy or mathematics instruction with paper-based versions of the same material support this theory, with children demonstrating larger academic gains after receiving computer-based instruction (Felix et al., 2017; Ortega-Tudela & Gómez-Ariza, 2006). Studies have explored computer-assisted reading instruction for isolated skills, including phonics (Nakeva von Mentzer et al., 2021) and oral vocabulary (Næss et al., 2022); however, no studies have explored computer-assisted comprehensive literacy instruction. Here, we explored the use of a freely available evidence-based comprehensive literacy program delivered via computer, ABRACADABRA (Centre for the Study of Learning and Performance [CSLP], 2021).

4.2.2.1 ABRACADABRA. ABRACADABRA (hereafter ABRA; CSLP, 2021) is an interactive web application designed to improve children's reading and spelling skills. The program incorporates best practice recommendations on reading instruction from the NRP to target skills within the areas of alphabetics, reading fluency, reading comprehension, and writing. A recent meta-analysis by Abrami et al. (2020) identified 17 studies published between 2008 and 2017 on the effectiveness of ABRA-based literacy instruction for Kindergarten to Grade 3 children. Collectively, these studies involved 7,388 children located across five countries. Included studies compared ABRA-based instruction with business-as-usual classroom literacy instruction in randomised controlled trials or quasi-experimental studies. All studies found positive effects for ABRA-based instruction compared with regular reading instruction on various standardised literacy measures. Statistically significant effects were found for phonics, phonemic awareness, reading comprehension, and listening comprehension. Analysis revealed that ABRA-based instruction resulted in the largest gains for struggling-readers and that these children were able to maintain gains post-instruction.

The children included in the meta-analysis above were primarily typically developing. Children with neurodevelopmental disabilities, specifically autism, were represented in only one study (Bailey et al., 2017). The benefits of ABRA for children with autism have been replicated across several other studies not included in the meta-analysis (Arciuli & Bailey, 2019, 2021; Bailey et al., 2017). This evidence, combined with previous research exploring CAI for children with Down syndrome, suggests that ABRA may be an effective comprehensive literacy intervention for children with neurodevelopmental disabilities more generally.

4.2.3 Shared Book Reading

ABRA delivery should include non-computerised extension tasks alongside ABRA computer activities to generalise skills learned in the program (CSLP, 2019). Shared book reading (SBR) is one way to generalise skills targeted within ABRA into enjoyable everyday literacy tasks. SBR is an interactive experience where an adult and child engage in joint reading of a text. Parent-led SBR has become part of the daily routine for many families with recognised benefits in cognition, language, and literacy development (Burgoyne & Cain, 2020; Cutler & Palkovitz, 2020). Clinician-led SBR may be used as part of an intervention session to explicitly target a child’s language or literacy skills (Kaderavek & Justice, 2002; Yorke et al., 2018).

Several studies have explored parent-led SBR with children with developmental disabilities. Arciuli et al. (2013) explored SBR between mothers and their children with autism and found that mothers corrected print-based errors more frequently than they provided contextual information to support reading comprehension during SBR. A recent study by Pierson et al. (2021) investigated the effects of parent coaching via telepractice to deliver a SBR intervention for children with developmental disabilities, including one child with Down syndrome. For the parent of the child with Down syndrome, parent training via telepractice had strong to very strong effects for implementation of the target SBR procedures; however, no changes were observed in the child's ability to respond to comprehension questions following intervention. Lim et al. (2018) investigated the SBR behaviours of children with Down syndrome and their parents before and after participating in intervention using the MULTILIT program (MULTILIT, 2007). This study found that children were able to generalise the skills learned during intervention to a naturalistic SBR context with their parents. These studies suggest that SBR may be a valuable context to support generalisation of skills targeted in literacy intervention for children with developmental disabilities, and that parents may benefit from additional support to maximise SBR experiences.

Indeed, studies investigating the home literacy environments of children with Down syndrome have found that parents value SBR and engage in these activities with their children at a similar frequency to parents of typically developing children (Ricci, 2011; Westerveld & van Bysterveldt, 2017). Yet, Trenholm and Mirenda (2006) observed that during SBR parents tended to ask their children with Down syndrome to find or label pictures, with few parents asking higher-level questions requiring more abstract thinking and reasoning. Cognitive reasoning skills are essential for developing text comprehension skills (NICHD, 2000) and studies have found that children with Down syndrome are capable of responding to these questions during SBR (Engevik et al., 2016). Given the importance of SBR for all beginning readers, we provided training to parents and included supplementary parent-led SBR in our pilot study.

4.2.4 The Current Study

The current study is the first to explore telepractice delivery of comprehensive literacy instruction for children with Down syndrome, a practice which many educators and clinicians have been keen to adopt as a result of the COVID-19 pandemic. Our study was designed to pilot a hybrid model of ABRA instruction for children with Down syndrome alongside supplementary parent-led SBR. Our pilot study was guided by these specific research questions:

1. Can a hybrid ABRA + parent-led SBR program be delivered via telepractice to children with Down syndrome?
2. What is the impact of a hybrid ABRA + parent-led SBR program delivered via telepractice on the word-level and passage-level reading accuracy skills of children with Down syndrome?
3. What is the impact of a hybrid ABRA + parent-led SBR program delivered via telepractice on the reading comprehension skills of children with Down syndrome?

4.3 Method

A within-participants design consisting of two phases and three assessment time points was used. Participants acted as their own controls with outcome variables measured at baseline, after a 6-week no-intervention control phase and, finally, following a 6-week instruction phase. We opted for this design in view of the modest sample, strict timeline, and previous studies that have utilised the same approach (e.g., Cologon et al., 2011; Lim et al., 2019). The aim was to deliver at least 16 hours of ABRA instruction to each child during the intervention phase. All assessment and instruction sessions were delivered by a speech-language pathologist experienced in working with children with neurodevelopmental disabilities.

4.3.1 Participants

This research project was approved by the Flinders University Human Research Ethics Committee. Participants were recruited via circulation of a flyer online and through organisations and clinics supporting children with neurodevelopmental disabilities Australia wide. Both guardians and children provided written consent prior to taking part in the study.

Participants were eligible to participate if they met the following criteria: (a) 6-12 years of age; (b) diagnosis of Down syndrome; (c) no major hearing or vision impairments; (d) able to communicate verbally at sentence level; (e) able to identify at least one letter of the alphabet; (f) able to demonstrate sustained attention to task for at least 15-minutes; (g) currently living in Australia; (h) use English as their first language. These eligibility criteria were set to ensure children were able to engage and demonstrate their abilities during online assessment and instruction sessions.

Six children with a diagnosis of Down syndrome were eligible to participate. Participants ranged from 8 years 6 months to 12 years 9 months at baseline and were situated in a variety of locations around Australia. Participant demographics are summarised in Table 4.1. Participants all spoke English as their primary language. Three children were reported to experience mild hearing loss (participants 1, 2, 4), though none required hearing aids. Three children had mild vision impairment requiring glasses (participants 1, 4, 6). Most children presented with mild articulation errors. Additionally, Participant 6 presented with a mild stutter. All children were supported by their mothers during instruction and parent-led SBR activities. At baseline, five parents reported engaging in daily SBR at home with their child using readers sent home from school or fiction/non-fiction books of interest. The parent of participant 5 reported engaging in SBR once a week using a reader sent home from school.

Table 4.1 *Participant Baseline Scores and Demographics*

Participant	Age at baseline	Gender	School	Grade	Phono-logical STM[a]	Phonological awareness[b]	Adaptive ability[c]	Nonverbal IQ[d] (descriptive classification)	% Consonants correct[e]
Child 1	8;6	F	MS	2	0.1	<1	7	23 (Low average)	80%
Child 2	12;9	M	MS	5	0.1	<1	1	0.3 (Extremely low)	61%
Child 3	8;7	M	MS	2	5	5	39	1 (Extremely low)	97%
Child 4	8;10	F	MS	3	0.1	1	10	25 (Average)	87%
Child 5	11;3	M	SS	5	0.1	<1	1	5 (Very low)	87%
Child 6	9;5	F	MS	3	0.1	<1	4	19 (Low average)	83%

Note. Scores expressed as percentile rank. MS = mainstream; SS = special school; STM = short term memory.

[a]Clinical Evaluation of Language Fundamentals – 4th Edition, Number Repetition subtest

[b]Comprehensive Test of Phonological Processing – 2nd Edition, Elision subtest

[c]Vineland Adaptive Ability Scales – 2nd Edition, Adaptive Behaviour Composite Score

[d]Raven's 2 Progressive Matrices Clinical Edition digital short form

[e]The Quick Screener

4.3.2 Measures

A battery of standardised assessments was used to gather baseline information and measure outcomes, all independent of the ABRA program. Measures were administered in the same order for all participants at all timepoints, individually via the Zoom video conferencing platform. Note that known speech errors were not penalised when scoring any of the below reading assessments.

4.3.2.1 Descriptive Measures

4.3.2.1.1 Phonological Short-Term Memory. The Number Repetition subtest of the Clinical Evaluation of Language Fundamentals – Fourth Edition (CELF-4; Semel et al., 2003) was administered as a measure of phonological short-term memory (STM). In the current sample, the number repetition subtest had an acceptable level of internal consistency[3] (Cronbach's α = .61).

4.3.2.1.2 Phonological Awareness. The Elision subtest of the Comprehensive Test of Phonological Processing – Second Edition (CTOPP-2; Wagner et al., 2013) was administered to measure children's ability to segment and manipulate sounds within words. For the children in our sample, the CTOPP-2 had a high level of internal consistency (Cronbach's α = .96).

4.3.2.1.3 Adaptive Ability. The Vineland Adaptive Ability Scales – Second Edition (VABS-2; Sparrow et al., 2005) was administered to evaluate adaptive ability in the domains of communication, daily living skills, and socialisation. For the children in our sample, the VABS-2 had a high level of internal consistency (Cronbach's α = .96).

4.3.2.1.4 Nonverbal Intelligence. The Raven's 2 Progressive Matrices Clinical Edition - Digital short form (Ravens 2; Raven et al., 2018) was used to measure nonverbal cognitive ability. Digital administration does not provide scores to allow the internal consistency with this group of children to be calculated. According to Raven et al. (2018), the assessment has good test-retest reliability both when used with a broad, normative sample

[3] All internal consistency statistics are based on baseline administration.

(r = .80) and a sample of individuals with intellectual disabilities (r = .82).

4.3.2.1.5 Speech Sound Production. The Quick Screener (Bowen, 1996) was used to provide a sample of children's use of speech sounds in single words. Percent Consonants Correct (%CC) was calculated by dividing the number of consonants the child produced correctly with the total number of consonants in the 44 words.

4.3.2.2 Literacy Outcome Measures

4.3.2.2.1 Word-Level Reading Accuracy. Two assessments were used to measure word-level reading accuracy. The primary measure was the Word Reading subtest of the Wide Range Achievement Test – Fourth Edition (WRAT-4; Wilkinson & Robertson, 2006) which measured participants' ability to accurately decode letters and words. The maximum possible raw score for this test is 70. For the children in our sample, the WRAT-4 had high internal consistency (Cronbach's α = .84).

The Castles and Coltheart Test – Second Edition (CC-2; Castles et al., 2009) measured participants' ability to read aloud real words with regular or irregular spellings, as well as nonwords, and was selected as a supplementary measure of word reading accuracy due to utilising an online test interface. The maximum possible raw score for this test is 120. For the children in our sample, the overall internal consistency for the CC-2 was high (Cronbach's α = .94).

4.3.2.2.2 Passage-Level Reading Accuracy. The Reading Accuracy Composite score from the Neale Analysis of Reading Ability – Third Edition (NARA-3; Neale, 1999) was used to measure participants' ability to accurately read passage-level text. The maximum possible raw score for this test is 100. In the current sample, the reading accuracy composite had high internal consistency (Cronbach's α = .89).

4.3.2.2.3 Passage-Level Reading Comprehension. Participants' ability to understand passage-level text was assessed using the Reading Comprehension Composite score from the NARA-3 (Neale, 1999). After reading passages aloud, participants were asked a series of set questions related to the text. The maximum possible raw score for this test is 44. In the current sample, the reading comprehension composite had high internal consistency (Cronbach's α = .82).

4.3.2.2.4 Functional Reading Comprehension. The Test of Everyday Reading Comprehension (TERC; McArthur et al., 2012) was administered to assess participants' ability to derive meaning from pictures of text encountered during everyday tasks (e.g. party invitation, cooking instructions). The maximum possible raw score for this test is 20. For the children in our sample, the TERC had low internal consistency (Cronbach's α = .50).

4.3.3 Procedure

4.3.3.1 Baseline Assessment. All participants completed a baseline assessment comprising both the descriptive and literacy outcome measures previously described. Literacy measures were administered in the order outlined above. Two descriptive measures were administered prior to the literacy measures (CELF-4 and CTOPP-2 subtests) and two following the literacy measures (Raven's 2 and The Quick Screener). Assessment sessions took place via Zoom over 60 to 90 minutes. Guardians participated in a separate session with the researcher to complete the measure for adaptive ability.

4.3.3.2 No-intervention Control Phase. Immediately following the baseline assessment, participants entered a 6-week wait-control phase. Children continued their business-as-usual therapy and literacy activities and received no instruction from the researcher during this 6-week period.

4.3.3.3 Pre-instruction Assessment. At the end of the 6-week control phase, and prior to the start of the instruction phase, children participated in a pre-instruction assessment session online. Children completed the same battery of literacy outcome measures administered during the baseline assessment.

4.3.3.4 Instruction Phase. During the 6-week instruction phase, children participated in three 60-minute online ABRA sessions per week via Zoom with the researcher (supported by a parent-guardian), along with two 15-minute parent-led SBR sessions each week. Participants were provided with the Fitzroy Readers apps, (graded decodable books; Fitzroy Programs and Greygum Software, 2016), on an iPad which were used for both clinician- and parent-led SBR.

4.3.3.4.1 ABRA sessions. The online ABRACADABRA Lite version (CSLP, 2021) was used in this study. To begin sessions, parents joined a Zoom meeting sent by the researcher. The researcher screen shared the ABRA program and participants had screen control to click and engage directly with ABRA activities. All participants were able to operate a computer mouse, though some required assistance to operate the mouse accurately (e.g., to click and drag items). The researcher verbally repeated prompts or instructed the child to click to hear a repetition in instances where the computer audio was disrupted. A visual timetable showing the day's activities set by the researcher was used to orientate children to the session. Each instruction session followed the same structure: (a) 15-minute word-level ABRA activities (targeting phonemic awareness, phonics, high-frequency word identification, or word spelling skills); (b) 20-minute passage-level ABRA activities (targeting reading fluency or comprehension skills within the ABRA interactive stories); (c) 15-minute clinician-led SBR activity (targeting generalisation of skills from earlier computer activities); and (d) 10-minute reward activity (e.g. computer game selected by the child).

Activities within ABRA are levelled for different degrees of difficulty, from early reading skills to more advanced, and target four key literacy abilities: (a) alphabetics; (b) reading fluency; (c) reading comprehension; and (d) writing. The program has a flexible structure, permitting highly individualised literacy instruction. Pre-instruction assessment data was used to set individual goals for participants and determine appropriate activities and starting levels. Task accuracy data was recorded by the researcher during sessions (number of correct and incorrect/prompted responses). Following each session, goals and activity level difficulty were reviewed in line with this data. A performance criterion of 65-85% accuracy was used to determine appropriate instruction content and difficulty. Skill mastery was set at 85% accuracy for word-level activities over three sessions. Children progressed to the next activity level or a different activity following mastery. Children were encouraged to make use of the ABRA embedded support when needed (e.g., selecting 'help' during word level activities, clicking on words in ABRA texts for decoding support). For further details on ABRA learning activities and program interactivity, see the ABRACADABRA Teacher Guide

(CSLP, 2019).

4.3.3.4.2 Parent-led Shared Book Reading. Prior to the instruction phase, parents attended a one-on-one 60-minute information session with the researcher where they received training on SBR strategies. Parents were encouraged to use four primary strategies during SBR: (a) take turns reading; (b) ask a question every second page; (c) support decoding of reading errors; and (d) end SBR with a recap of content read. Parents were provided with two resources to accompany SBR sessions: (a) a 'reading comprehension' resource outlining three levels of question difficulty and examples of each; and (b) a 'reading error' worksheet to support decoding of words read in error. These same resources and strategies were used during clinician-led SBR in the ABRA sessions. Parents completed a reading log documenting estimated reading accuracy, example errors, and comprehension accuracy after each SBR session. Reading logs from the previous week were reviewed weekly during a 15-minute consultation session between the parent and researcher and were used to guide setting texts and strategies for the following week.

4.3.3.5 Post-instruction Assessment. Immediately following the 6-week instruction period, children again completed the battery of literacy outcome measures following the same procedures as the baseline and pre-instruction assessment.

4.3.4 Implementation Fidelity

Two instruction sessions for each participant were selected at random and video recorded for fidelity assessment. A co-author with knowledge of the ABRA program viewed the recorded instruction sessions and rated procedural integrity using a fidelity rating form outlining the standard protocol of ABRA instruction sessions. Video recordings for fidelity purposes were voluntary and one parent did not provide consent for recording of their child's instruction sessions.

4.3.5 Data Analysis

A one-way repeated measures analysis of variance (ANOVA) was conducted to analyse the possible effects of the ABRA + parent-led SBR program on participants' reading skills. The within-subject factor was Time, given that all participants were assessed at three time points (baseline vs. pre-instruction vs. post-instruction), and the dependent variable was raw scores for each of the literacy outcome measures. Our interpretation of effect sizes (η_p^2) was guided by the following reference points: .01 was considered a small effect size, .06 a medium effect size, and .14 a large effect size (Richardson, 2011). ANOVAs were conducted using SPSS version 27. For data that did not meet assumptions for parametric analysis, non-parametric analyses were conducted using Friedman's ANOVA with post hoc analysis using Wilcoxon signed-rank tests. In view of the multiple tests an adjusted alpha of .01 was used.

4.4 Results

All participants completed at least 16 ABRA sessions over the 6-week instruction period. Participants 2 and 6 participated in a total of 17 sessions, and participants 1 and 5 participated in 18 sessions. See Table 4.2 for examples of ABRA activities completed most frequently by participants.

Descriptive statistics for each of the outcome measures at all time points are presented in Table 4.3 and individual participants' raw scores are shown in Table 4.4. The assumption of normality was not met for one variable[4]. For all other variables, assumptions for repeated measures ANOVA were met.

[4] Baseline scores on the measure of functional reading comprehension were not normally distributed and therefore failed to meet assumptions for parametric analysis. As such, we used non-parametric equivalent analyses to evaluate this measure. Both parametric and non-parametric results are reported in the interests of transparency and in view of disagreement about whether some parametric analyses, including ANOVA, are robust despite violations of assumptions (Glass et al., 1972).

Table 4.2 *Example ABRA Activities by Module with Percent of Sessions Each Participant Completed Activity*

Module Example activities	**Description**	**Child 1**	**Child 2**	**Child 3**	**Child 4**	**Child 5**	**Child 6**
Alphabetics (*n* = 17)							
Word families	Identify initial letters to create target words	17%	12%	-	6%	6%	24%
Blending train	Blend sounds from the computer and say the word	6%	18%	6%	19%	11%	12%
Auditory blending	Blend sounds from the computer and select the corresponding image	33%	24%	25%	25%	39%	35%
Auditory segmenting	Match target full word to segmented version	17%	12%	13%	25%	6%	-
Basic decoding	Decode written word and match to corresponding image	50%	65%	56%	56%	61%	47%
Word changing	Substitute letters in a word to create a new target word	-	12%	25%	6%	6%	-
Fluency (*n* = 6)							
High frequency words	Read high-frequency words before the timer runs out	-	12%	-	-	28%	-
Expression	Listen to a passage and identify if it was read with correct expression. Then read the same passage with expression	-	-	19%	31%	-	24%
Accuracy	Listen to a passage, then read the same text with no errors	-	47%	-	-	11%	-
Comprehension (*n* = 6)							
Prediction	Answer questions to predict events in a story	33%	29%	31%	31%	33%	29%
Comprehension monitoring	Identify the word on a page that does not make sense	39%	24%	38%	38%	39%	35%
Sequencing	Place images of story events in correct order after reading	22%	12%	19%	19%	17%	18%
Summarising	Answer questions during a story about key events	28%	-	13%	-	17%	12%
Story elements	Respond to multiple choice questions about key story events	22%	24%	31%	25%	6%	29%
Writing (*n* = 2)							
Spelling words	Type a word to dictation	-	-	13%	-	-	-

Note. Only the most frequently completed activities across participants are described in the table. Some additional activities included in ABRA were completed, though are not outlined above. Data in parentheses indicate total number of ABRA activities in this module.

Table 4.3 *Mean Raw Scores for Baseline, Pre-, and Post-Instruction Outcome Measures*

Measure	**Baseline**		**Pre-instruction**		**Post-instruction**	
	Mean	***SD***	**Mean**	***SD***	**Mean**	***SD***
Word-level reading accuracy (primary measure)	25.83	3.55	27.00	3.46	29.50	3.51
Word-level reading accuracy[a] (supplementary measure)	21.67	11.29	24.50	12.65	32.00	10.26
Passage-level reading accuracy	16.50	6.35	15.83	9.54	26.50	9.55
Passage-level reading comprehension	3.17	2.56	3.00	2.10	4.83	2.14
Functional reading comprehension	0.67	1.03	1.17	1.60	2.83	2.14

Note. Word-level reading accuracy (primary measure) = Wide Range Achievement Test (WRAT-4), Word Reading subtest; Word-level reading accuracy (supplementary measure) = Castles and Coltheart Test - 2nd edition (CC-2); Passage-level reading accuracy and reading comprehension = Neale Analysis of Reading Ability (NARA-3); Functional reading comprehension = Test of Everyday Reading Comprehension (TERC).

[a]Based on combined CC-2 raw scores for reading of regular, irregular, and nonwords.

Table 4.4 *Individual Participant Raw Scores on Baseline, Pre-, and Post-Instruction Outcome Measures*

Measure	Child 1	Child 2	Child 3	Child 4	Child 5	Child 6
Word-level reading accuracy (primary):						
Baseline	24	24	27	28	21	31
Pre-instruction	26	25	29	32	22	28
Post-instruction	30	27	31	34	24	31
Word-level reading accuracy (supplementary):						
Baseline	22	10	31	28	6	33
Pre-instruction	26	12	36	35	6	32
Post-instruction	33	22	44	41	18	34
Passage-level reading accuracy:						
Baseline	19	16	23	13	6	22
Pre-instruction	17	6	30	15	5	22
Post-instruction	25	21	35	36	11	31
Passage-level reading comprehension:						
Baseline	1	6	6	2	0	4
Pre-instruction	1	2	6	3	1	5
Post-instruction	6	4	7	5	1	6
Functional reading comprehension:						
Baseline	0	0	2	2	0	0
Pre-instruction	1	0	2	4	0	0
Post-instruction	3	3	4	6	0	1

Note. Word-level reading accuracy (primary measure) = Wide Range Achievement Test (WRAT-4), Word Reading subtest; Word-level reading accuracy (supplementary measure) = Castles and Coltheart Test - 2nd edition (CC-2); Passage-level reading accuracy and reading comprehension = Neale Analysis of Reading Ability (NARA-3); Functional reading comprehension = Test of Everyday Reading Comprehension (TERC).

4.4.1 *Word-Level Reading Accuracy*

There was a statistically significant effect of time on word reading accuracy, F (2, 10) = 11.347, p = .003, η_p^2 = .694. Pairwise comparisons confirmed that significant improvements in word reading accuracy occurred from pre- to post-instruction (p = .002), but not from baseline to pre-instruction (p = .817). These results indicate that participants' word reading accuracy did not increase over the 6-week no-intervention control phase, by contrast with statistically significant improvement over the 6-week instruction phase where they received the ABRA + parent-led SBR program.

The supplementary measure of word reading accuracy (CC-2) was analysed based on combined raw scores for overall word reading accuracy. Results again indicated a statistically significant main effect of time on participants' word reading accuracy F (2, 10) = 24.017, p <.001, η_p^2 = .828. No significant difference was found in participants' scores between baseline and pre-instruction (p = .218), with a significant difference occurring from pre- to post-instruction (p = .009).

4.4.2 *Passage-Level Reading Accuracy*

Analysis revealed a statistically significant effect of time on participants' passage-level reading accuracy, F (2, 10) = 10.992, p = .003, η_p^2 = .687. Post hoc tests revealed that there was no significant improvement in passage-level reading accuracy from baseline to pre-instruction (p = 1.000), however a significant improvement occurred from pre- to post-instruction (p = .024).

4.4.3 *Passage-Level Reading Comprehension*

There was no statistically significant effect of time on participants' passage-level reading comprehension, F (2, 10) = 3.033, p = .093, η_p^2 = .378. As evident in Table 4.3, participants demonstrated a slight increase in mean raw scores following instruction, however this was not statistically significant. Pairwise comparisons revealed no statistically significant difference from baseline to pre-instruction (p = 1.000) or pre-instruction to post-instruction (p = .143).

4.4.4 Functional Reading Comprehension

A statistically significant effect of time was found for the measure of functional reading comprehension, F (2, 10) = 11.780, p = .002, η_p^2 = .702. Pairwise comparisons confirmed that statistically significant improvements occurred in participants' functional reading comprehension from pre- to post-instruction (p = .032), with no statistically significant difference over the no-intervention control phase (p = .609). Non-parametric analysis confirms a statistically significant increase in participant's functional reading comprehension over time, χ^2 (2) = 9.294, p = .010; however, no statistically significant difference occurred from either baseline to pre-instruction or from pre- to post-instruction.

4.4.5 Parent-led Shared Book Reading

Our review of SBR logs indicated the following: (a) the difficulty of texts set for participants increased as the program progressed; (b) parents recorded more reading errors in logs during later weeks; (c) most children started with either Level 1 or Level 2 comprehension questions (concrete questions based on explicit information in pictures/text); (d) by the end of the program, all parents were asking Level 3 questions (questions requiring abstract thinking and reasoning); and (e) parents inconsistently commented on their child's compliance and engagement in SBR sessions. Where parents did report reduced compliance, reasons included: child was tired, child not interested in story content, child reluctant to use specific reading strategies (e.g., sounding out, tracking words in the text, responding to questions).

4.4.6 Fidelity

Procedural fidelity for ABRA implementation was high, as can be seen in Table 4.5. Engagement and adherence were also high for parent-led SBR sessions and logs. All participants completed two parent-led SBR sessions each week (and documented in the reading logs) to achieve the target of 12 SBR sessions, except for Participant 5 who completed only eight parent-led SBR sessions. Parents commented on both accuracy and comprehension skills with varied detail in SBR logs, indicating they were following the guidelines set for SBR activities and providing both decoding and comprehension support.

Table 4.5 *Procedural Fidelity*

Fidelity items	Mean rating	*SD*
Researcher is familiar with the ABRA program/lesson content	5	0
Lesson has clear goals and objectives	5	0
Lesson is planned ahead of time	5	0
Lesson content is balanced (i.e., alphabetics, word and text activities)	5	0
Lesson includes introduction	5	0
Lesson includes demonstration	5	0
Researcher monitors child's navigation of the program	5	0
Lesson includes conclusion	4.5	1.58
Lesson includes ABRA and non-ABRA activities	5	0
Lesson includes computer and non-computer activities	5	0
Researcher manages student behaviour	4.3	1.06
Learning environment is appropriately organised	5	0
Lesson is at least 30mins in duration	5	0

Note. Each fidelity checklist item rated as: 1 = Strongly disagree; 2 = Disagree; 3 = Neutral; 4 = Agree; 5 = Strongly agree.

4.5 Discussion

Here we conducted the first ever evaluation of comprehensive literacy instruction via telepractice for children with Down syndrome. We evaluated whether it was possible to deliver a hybrid ABRA + parent-led SBR program via telepractice, as well as the impact of this program on the reading abilities of children with Down syndrome.

4.5.1 Reading Accuracy

Our results revealed children made statistically significant gains in their single word reading accuracy and passage-level reading accuracy as a result of the hybrid ABRA + parent-led SBR intervention. Effect sizes were large for both word- and passage-level reading accuracy, although these effect sizes must be interpreted with caution given the small sample in this pilot study. All measures used were independent of the ABRA program, indicating that children were able to generalise reading skills learned throughout the instruction phase. These finding are in line with some previous studies exploring phonics-based reading instruction for children with Down syndrome which have reported gains in word reading accuracy on independent assessment measures (e.g. Cologon et al., 2011; Lim et al., 2019). Our study adds to the existing evidence that children with Down syndrome can generalise their word-level reading skills to untaught, independent, standardised materials.

4.5.2 Reading Comprehension

Our results revealed that children did not make statistically significant gains in their conventional reading comprehension skills. There was a slight increase in mean raw scores from pre- to post-instruction for passage-level reading comprehension skills; however, this increase was not statistically significant. Children made gains in their functional reading comprehension skills which were statistically significant on some analyses; however, these gains were not reliably significant across statistical tests. Given that the assessment of functional reading comprehension scores had the lowest internal consistency and a floor effect at baseline for most participants, it may not be as reliable as the other assessments in our battery.

The nature of our reading comprehension measures may also have affected outcomes. The conventional measure of passage-level reading comprehension we used requires a higher level of literacy and related language processing as children read texts of increasing difficulty and respond to both literal and inferential questions using oral language. In contrast, the functional reading comprehension measure requires a lower level of literacy and related language processing because children view images containing smaller amounts of fragmented text, though are still required to respond to questions using oral language. It is possible that the children in our study were unable to make significant gains on the conventional passage-level reading comprehension test due to increased oral language skill demands, by comparison with our more functional test. Some previous studies have shown that children with Down syndrome make slower progress with reading comprehension skills, likely as a result of oral language difficulties (Boudreau, 2002; Byrne et al., 2002). Unfortunately, our protocol did not include a measure of general receptive and/or expressive language skills; however, all parents reported that their child had language difficulties requiring additional support. A more sensitive measure of reading comprehension, such as sentence level comprehension, may have yielded different results. Though, it is also possible that 6-weeks of instruction is not long enough to allow consolidation of conventional reading comprehension skills.

4.5.3 Speculative Analysis

Analysis of individual participants' response to intervention may provide some valuable information on the impact of intervention for specific profiles within this group (Horner et al., 2005). For example, Participant 5 in our study had the lowest raw scores for word- and passage-level reading accuracy at both baseline and pre-instruction and made the largest gains in raw scores for reading accuracy from baseline to post-instruction. Additionally, this participant was the only child not attending a mainstream school and his family reported engaging in SBR infrequently prior to the study. These factors may have meant that this child was more responsive to the hybrid ABRA + parent-led SBR intervention than some of the other children. Despite making the greatest gains in reading accuracy, Participant 5 did not make any gains in reading comprehension. Participant 1 made the largest gains in passage-level reading comprehension raw scores from baseline to post-instruction. This participant had one of the highest nonverbal intelligence scores but also had the most difficulty engaging in telepractice sessions at the beginning of the program. It is possible that these factors contributed to Participant 1's reading comprehension gains. Further research is needed to follow up these possibilities.

4.5.4 Practical Implications

4.5.4.1 Parent-led Shared Book Reading. Collaborative implementation of instruction across people and settings has the potential to increase instruction dosage and generalisation of skills to new contexts. This can be particularly important for children with Down syndrome, some of whom have difficulty generalising reading skills across contexts (e.g. Lim et al., 2018; Nakeva von Mentzer et al., 2021). The impact of parent-led SBR in addition to the ABRA sessions is unclear in the current study. However, comments in reading logs indicated that parents felt more confident supporting both their child's reading accuracy and reading comprehension skills, and that this positively impacted their child's reading performance. For example, two parents commented that their child would now attempt to answer higher-level questions, even if not correctly, where previously they would not respond. Children's difficulty with higher-level language questions is in line with previous studies exploring reading comprehension and children with Down syndrome (Nash & Heath, 2011). Consistent with previous research (e.g., Trenholm & Mirenda, 2006), our study demonstrates that parents did benefit from support in this area.

4.5.4.2 Telepractice. The current study is one of the first to investigate comprehensive literacy instruction for children with neurodevelopmental disabilities via telepractice (see Bailey et al., 2022, for a study of online comprehensive literacy instruction for children with autism), and the first to investigate online comprehensive literacy instruction for children with Down syndrome. There are currently too few studies to draw strong conclusions regarding the efficacy of literacy instruction via telepractice. A recent rapid review investigating literacy assessment and instruction via telepractice by Furlong et al. (2021) identified nine relevant studies and concluded that this mode of instruction delivery can be feasible and engaging, and that online literacy assessment can be equivalent in quality to face-to-face assessment. Our study supports the findings of Furlong et al. (2021), demonstrating that literacy assessment and instruction via telepractice was acceptable to both children with Down syndrome and their parents and also effective in achieving literacy gains.

Although our study was shorter in overall length of intervention (6-weeks in the current study compared with 8 to 18-weeks in the studies identified by Furlong et al., 2021), the session length and intensity resulted in a similar overall dosage to that reported in many previous studies of literacy instruction via telepractice. The acceptability of this high dosage of instruction over a relatively short period of time to families in this study was likely facilitated by the convenience of telepractice. Children in our study were able to achieve 16-18 hours of instruction over 6-weeks, speaking to one of the many benefits of telepractice as a mode of service delivery. A recent meta-analysis by Roberts et al. (2022) suggests that increased dosage of one-to-one literacy instruction leads to more positive literacy outcomes. Telepractice as a mode of service delivery for literacy instruction has the potential to increase children's access to one-to-one services. Indeed, in the current study telepractice facilitated inclusion of children with Down syndrome from all over the country to participate in intensive literacy instruction. Our study adds to the small but promising evidence base, suggesting telepractice can be a practical and engaging mode of service delivery for comprehensive literacy assessment and instruction.

4.5.5 Limitations and Future Research

While this study presents some promising results, there are several limitations that warrant consideration. First, this pilot study included a small sample which, although similar in size to previous studies involving children with Down syndrome (e.g., Cologon et al., 2011 [*n* = 7]; Cupples & Iacono, 2002 [*n* = 7]; Lemons et al., 2018 [*n* = 6]; Lemons et al., 2015 [*n* = 5]), makes it difficult to draw firm conclusions and generalise results to the broader population. In particular, we found inconsistent results regarding gains in reading comprehension in this study. It is possible that a larger sample size may have led to greater statistical power to detect improvements in this skill on our conventional measure of reading comprehension. In addition, the small sample does not allow for statistical control of additional variables in the analysis. This is potentially relevant given that Burgoyne et al. (2012) found that, for children with Down syndrome, chronological age and number of instruction sessions predicted outcomes following literacy instruction. Second, as mentioned, our study was conducted over a relatively short period of time at high intensity. The 16-18 hours of intervention delivered to each participant over 6-weeks in the current study falls short of the recommended ABRA administration time (2 hours per week for at least 13 weeks; Abrami et al., 2020). Third, it is possible that practice effects may have influenced our results given that participants completed the same assessments at three timepoints.

However, we note that participants were not provided with any feedback on their performance during the assessments and little change was seen between participants' baseline and pre-instruction assessment scores. Finally, some participants' behaviour and attention differed across assessment sessions resulting in some variation in performance within participants. This variability in test performance has been reported previously in studies involving children with Down syndrome (e.g., Nakeva von Mentzer et al., 2021).

This pilot study provides proof of concept for the feasibility and efficacy of online computer-based comprehensive literacy instruction for children with Down syndrome that warrants further research. As well as addressing the above limitations, future research should aim to build on the current findings and provide more rigorous evidence by employing a randomised controlled trial study design and including a larger sample size to ensure children with a greater range of ages and abilities are included. Future studies could also investigate the effects of ABRA delivered within the context of an inclusive online or face-to-face classroom or small-group learning setting for children with Down syndrome (Vousden et al., 2022). Additionally, future research could aim to tease apart the relative contributions of ABRA instruction and parent-led SBR on children's reading outcomes. Future studies should investigate methods of online literacy instruction that are accessible to children with Down syndrome with a diverse range of abilities (e.g., children with hearing impairments, children who do not communicate verbally).

4.6 Conclusion

As far as we are aware, no previous study has investigated comprehensive literacy instruction for children with Down syndrome via telepractice, even though this is now recognised as critical during a global pandemic and beyond. The ABRA literacy web application has not previously been explored with children with Down syndrome, via online delivery or otherwise. Our results suggest that children with Down syndrome made significant gains in word- and passage-level reading accuracy skills as a result of online ABRA instruction and supplementary parent-led SBR, and that this method of service delivery was acceptable to both children and parents. We hope that this pilot study encourages more research in this area, especially during the current global pandemic where many children with neurodevelopmental disabilities have been under supported with regards to education and health services.

CHAPTER 5 EMPIRICAL STUDY 3

An Effectiveness Trial of ABRACADABRA Literacy Instruction for Children with Autism during the COVID-19 Pandemic

5.1 Abstract

Introduction: This study explored literacy instruction for children with autism during the of the COVID-19 pandemic in an area of relative socioeconomic disadvantage.

Method: Fifty-nine children with autism (5 to 12 years) participated in a baseline assessment before being assigned to one of two instruction conditions or a control group. Instruction group participants received 13-weeks of literacy instruction using ABRACADABRA (a free online literacy web application), with one instruction condition receiving additional supplementary shared book reading. Instruction was initially delivered to children face-to-face in small groups; however, government mandated stay-at-home orders issued mid-way through the study necessitated a change to one-to-one instruction delivery via telepractice, with associated participant attrition leaving a final sample of 47 participants.

Results: Children who participated in instruction made statistically significant gains in their nonword reading skills from pre- to post-instruction with a large effect size, relative to control group participants. There were no other statistically significant results at the conservative alpha level utilised. However, effect sizes for all other reading outcome measures were similar to previous research using ABRACADABRA instruction for children with autism (medium effects for word reading accuracy and reading comprehension and large effect sizes for passage reading accuracy; Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022).

Conclusion: Further research on literacy instruction delivered via telepractice for children with autism is greatly needed. These findings contribute to the scarce knowledge base of literacy instruction for children with autism and the impact of the COVID-19 pandemic on this group.

5.2 Introduction

Learning to read is a fundamental human right and is associated with positive academic performance, employment opportunities, social and mental health outcomes (Hendren et al., 2018; Ontario Human Rights Commission, 2022; Singh, 2013). Yet, children with developmental disabilities, such as autism, often receive poor quality or ineffective literacy instruction due, at least in part, to lower expectations regarding their 'potential' for learning and limitations in educational resources (Arciuli & Bailey, 2021; Arciuli & Emerson, 2020; Bailey & Arciuli, 2020; O'Donnell et al., 2022; Whalon & Hart, 2011). The COVID-19 pandemic has further affected these children's access to their usual literacy learning opportunities (Dickinson et al., 2020; Marella et al., 2022). Here we report on a study conducted in an area of relative socioeconomic disadvantage during the COVID-19 pandemic. Our study was initially designed to investigate the effects of a computer-based literacy program delivered in person to small groups of children with autism, including the value-added effects of supplementary shared book reading (SBR). Due to government mandated stay-at-home orders issued midway through the study, instruction was moved online and delivered on a one-to-one basis to participants via telepractice. We explore each of the factors central to this study below.

5.2.1 Literacy and Children with Autism

Autism is a neurodevelopmental difference characterised by difficulties in social communication and restricted, repetitive patterns of behaviours or interests (American Psychiatric Association [APA], 2022). These core characteristics can occur alongside intellectual and oral language difficulties, which can subsequently impact literacy development (APA, 2022; Hendren et al., 2018). Children with autism[5] present with highly heterogeneous reading profiles, with some more likely to experience difficulties with reading comprehension than reading accuracy skills (Brown et al., 2013; McIntyre, Solari, Grimm, et al., 2017; Nation et al., 2006; Sorenson Duncan et al., 2021). For instance, a recent meta-

[5] Throughout this paper we deliberately use person-first and identity-first language interchangeably (Monk et al., 2022; Shakespeare, 2018).

analysis by Sorenson Duncan et al. (2021) found that across 26 studies involving 1,211 autistic children, word reading scores were higher on average than reading comprehension scores.

Reading comprehension is the ultimate goal of reading instruction and is essential for engaging meaningfully with texts. The Simple View of Reading (SVR) is a well-established theory for understanding the component skills that contribute to reading comprehension: decoding (recognising words in print) and linguistic comprehension (understanding spoken language; Gough & Tunmer, 1986; Hoover & Tunmer, 2018). Studies, including the meta-analysis by Sorenson Duncan et al. (2021), have shown that these same skills are equally important for reading comprehension in autistic children (Brown et al., 2013; Ricketts et al., 2013). Accordingly, children with autism require comprehensive literacy instruction that develops both their decoding and comprehension skills (Arciuli & Bailey, 2021; Whalon, 2018).

Literacy instruction based on the recommendations of the United States National Reading Panel (NRP) is widely considered to be evidence-based (National Institute of Child Health and Human Development [NICHD], 2000). The NRP states that effective early reading instruction should incorporate five key skills, known as the Big Five (phonemic awareness, phonics, reading fluency, vocabulary, reading comprehension). These recommendations are based on research focused on non-autistic children; however, Whalon et al. (2009), who reviewed research published prior to 2008, and Bailey and Arciuli (2020), who reviewed research published between 2009 and 2017, found that autistic children can make gains when provided with instruction targeting one or more of the NRP Big Five. Both reviews identified few studies that incorporated all of the Big Five in instruction. A recent report commissioned by the UK government cites a review by Arciuli and Bailey (2021) and concludes that all children, including children with autism and other developmental disabilities, should be provided with the same high-quality comprehensive literacy instruction (The reading framework; Department for Education, 2023). Within the Arciuli and Bailey (2021) review, the literacy web application ABRACADABRA (Centre for the Study of

Learning and Performance [CSLP], 2021) is discussed as an example of a promising comprehensive reading program for autistic children.

5.2.2 ABRACADABRA Literacy Instruction

ABRACADABRA (hereafter referred to as ABRA) is a free online interactive web application designed to develop the NRP Big Five (https://literacy.concordia.ca/abra/en/; CSLP, 2019). ABRA contains 33 activities, each with different complexity and difficulty levels, that target skills in the areas of alphabetics (phonemic awareness and phonics), reading fluency, reading comprehension (vocabulary and comprehension strategies), and writing (spelling). The program is modular and can used flexibly to provide highly individualised literacy instruction when facilitated by a knowledgeable instructor. A recent meta-analysis by Abrami et al. (2020) investigated 17 studies published between 2008 and 2017 on the effects of ABRA literacy instruction, involving a total of 7,388 children. This analysis showed positive effects for ABRA instruction on phonemic awareness, phonics, reading fluency, reading comprehension, listening comprehension, and vocabulary knowledge skills.

Only one study included in this meta-analysis demonstrated that ABRA instruction can be effective for children with developmental disabilities. In this study, Bailey et al. (2017) investigated the effects of ABRA instruction delivered one-to-one in person by a clinician to autistic children, aged 5 to 11 years. Children who participated in ABRA (26 hours of instruction over 13-weeks) achieved statistically significant gains with large effect sizes post-instruction, relative to children in the control group, for word- and passage-level reading accuracy ($\eta_p^2 = .41$ for both measures) and reading comprehension skills ($\eta_p^2 = .32$). A more recent study by Arciuli and Bailey (2019) that fell outside of the search dates for Abrami et al.'s meta-analysis explored ABRA delivered in person, facilitated by educators to small groups of autistic children aged 5 to 8 years in a school setting. Compared to a control group, children in the instruction condition (20 hours of ABRA instruction over 9-weeks) again made statistically significant gains in word- and passage-level reading accuracy with large effect sizes following instruction ($\eta_p^2 = .30$ and $\eta_p^2 = .18$ respectively). Gains in reading comprehension were not statistically significant (medium effect size; $\eta_p^2 = .08$).

ABRA's effects on reading comprehension skills in autistic children have been inconsistent. It is possible that reading comprehension outcomes were impacted by differences across these studies in instruction duration (26 hours over 13-weeks vs. 20 hours over 9-weeks), instructor (clinician vs. teacher), and/or mode of delivery (individual vs. group-based). Learning within a group-based setting requires social communication skills. Social communication is a key support need for children with autism and can play a critical role in reading comprehension (McIntyre et al., 2018; McIntyre, Solari, Grimm, et al., 2017; Ricketts et al., 2013). Some research suggests that social-communication differences can predict reading comprehension difficulties in autistic children, independently of word reading and language comprehension skills (McIntyre et al., 2018; Ricketts et al., 2013). Indeed, Arciuli and Bailey (2019) found that reading comprehension gains following group-based ABRA delivery were associated with children's social communication skills. In most education settings, literacy instruction is primarily delivered to children in whole class/group-based contexts. As such, methods of instruction that maximise reading comprehension gains for autistic children during group-based instruction require further exploration.

5.2.3 Shared Book Reading

SBR has potential to increase reading comprehension gains for autistic children within an individual and group-based setting (Clendon et al., 2014; Fleury et al., 2021; Weadman et al., 2022). Whilst many types of SBR exist, SBR is typically defined as an interactive experience where an adult and child engage in joint reading of a text (Noble et al., 2019). A systematic review of SBR interventions for autistic children by Boyle et al. (2019) found positive effects of SBR on skills fundamental to reading comprehension (e.g., listening comprehension, expressive language). These positive effects were found across a range of interventionists, settings, instruction dosages, and adult SBR behaviours, suggesting that SBR is a robust literacy instruction method.

SBR can also have language and literacy benefits when applied in small groups (Henry & Solari, 2020). One well established method of group-based SBR is cooperative learning (Klingner & Vaughn, 1998; Robert & Robert, 1995; Vaughn et al., 2011). Cooperative learning is an evidence-based reading comprehension strategy recommended by the NRP where children work in small groups and support each other to use strategies to promote reading comprehension (NICHD, 2000). Several studies have reported reading comprehension gains for autistic children who have engaged in cooperative learning (e.g., Boardman et al., 2016; Reutebuch et al., 2015). In a systematic review by Tárraga-Mínguez et al. (2020), the second most utilised method of reading comprehension instruction for children with autism was cooperative or shared reading, and all studies exploring these methods reported reading comprehension gains following instruction. The current study aimed to investigate the effects of such SBR methods for autistic children as an adjunct to the ABRA program within face-to-face group-based literacy instruction. However, the COVID-19 pandemic forced our study to move online and, as such, to also explore these methods within a one-to-one context via telepractice.

5.2.4 Face-to-face versus Literacy Instruction via Telepractice for Children with Autism

Few studies exploring literacy instruction via telepractice have involved autistic children. Recently, Henry et al. (2023) explored a SBR intervention delivered via telepractice targeting reading-related skills. In this study, autistic children aged 5 to 9 years participated in twice weekly sessions via telepractice for 11-weeks. Half of the children received an additional parent-led SBR component. Following instruction, children across both groups made gains in their listening comprehension skills, but not in their oral vocabulary or narrative retell ability. Bailey et al. (2022) recently investigated literacy instruction delivered via telepractice for children with autism and reported direct reading outcomes. In this study, Bailey et al. (2022) investigated one-to-one delivery of ABRA instruction by a clinician via telepractice to autistic children aged 5 to 12 years. Instruction was supplemented by parent-led SBR. Quantitative analyses showed no statistically significant gains in reading skills for children who received instruction (16 hours over 8-weeks) compared with the control group, with small to large effect sizes (word-reading accuracy [$\eta_p^2 = .01$]; combined word and nonword reading accuracy [$\eta_p^2 = .09$]; passage-reading accuracy [$\eta_p^2 = .14$]; reading comprehension [$\eta_p^2 < .01$]). This study fell short of the recommended amount of ABRA instruction (2 hours per week for at least 13-weeks; Abrami et al., 2020). Qualitative data indicated that parents were positive about the ABRA program via telepractice, with some noting gains in their child's reading skills or confidence. Both Henry et al. (2023) and Bailey et al. (2022) reported poorer outcomes for literacy and literacy-related skills following instruction for autistic children delivered via telepractice when compared with face-to-face versions of the same intervention (i.e., Bailey et al., 2017; Henry & Solari, 2020).

5.2.5 The Current Study

Our study was initially designed to test the effects of small-group face-to-face literacy instruction, delivered by clinicians, on the reading skills of autistic children in an area of relative socioeconomic inequity (as per the 'Index of Relative Socio-economic Disadvantage', a broad Australian-based socio-economic index that encapsulates various details about the economic and social conditions of individuals and households in a specific area, including income, number of people without qualifications, and number of people in low skilled occupations; Australian Bureau of Statistics, 2021). Our primary aim was to refine methods of reading comprehension instruction in a group setting while delivering the recommended amount of ABRA instruction. However, due to stay-at-home orders issued mid-way through the study these plans changed. We had an ethical duty to continue providing services to research participants and therefore moved instruction online. This unexpected change offered a unique opportunity to explore the effects of literacy instruction under the same 'real world' conditions impacting all educational and clinical settings at the time. As such, our results speak to the effectiveness of literacy instruction for autistic children delivered across multiple modalities during the COVID-19 pandemic as opposed to the efficacy study which was our initial intention.

Given the changes to the initial study design, the aims of this study were to: (a) determine the feasibility of clinic-based small group literacy instruction; (b) explore the value-added effects of clinician-led SBR during ABRA-based instruction; and (c) investigate the benefits of supplementary ABRA-based instruction for children with autism during the COVID-19 pandemic. We hypothesised initially that children in instruction conditions would achieve gains in their reading accuracy and reading comprehension skills, with children participating in SBR activities achieving greater gains in reading comprehension than children who did not participate in SBR. We had no precedent for how the change to instruction modality midway through the study, and the additional confounding factors associated with the pandemic, would impact study participation and outcomes.

5.3 Method

This project was approved by the Flinders University Human Research Ethics Committee. Participants were recruited via flyers sent to local clinics and community organisations supporting children with autism across the metropolitan area of Western Sydney, and through professional networks via email and social media. In addition, the project was advertised via our industry partner, The Luke Priddis Foundation. Guardians and children provided written consent prior to participation.

The study was conducted over two phases and utilised participatory research methods (Fletcher-Watson et al., 2019). Phase one involved focus group consultation with key stakeholders to develop a model of group-based literacy instruction. This phase involved three focus groups comprising: a parent group (four parents of autistic children), clinician group (three allied health clinicians working with children with autism), and an adolescent group (three autistic adolescents). Key recommendations from the three focus groups related to: (a) barriers to participation in literacy instruction; (b) strategies for promoting attendance at the instruction sessions; (c) optimising the start of each session; (d) assisting children through transitions; (e) assisting access to computers; (f) supporting participation in shared reading activities; (g) selection of reading materials for SBR activities; (h) encouraging peer-to-peer interactions; (i) how to deal with missed instruction sessions; and (j) how to support children who become tired or dysregulated during sessions. The method and findings of Phase One will be reported separately and are summarised in Appendix A.

Phase two was conducted as a quasi-experimental study in which children were assigned to one of three conditions: ABRA instruction only (ABRA), ABRA instruction plus shared book reading (ABRA+SBR), no-instruction control group (NI). We intended Phase two instruction group participants to participate in 13-weeks of face-to-face, small-group literacy instruction working with a researcher in a quiet room of a clinic or school. However, at the end of the seventh instruction week the New South Wales government issued stay-at-home orders which prohibited such gatherings. Uncertain of how long these orders would be in place, the literacy instruction sessions were put on hold for a period of five weeks before a decision was made to move them online. The final six weeks of instruction took place via telepractice (hosted on the Zoom platform), with participants working one-to-one with a researcher. See Supplementary Material A at the end of this chapter for a summary of the planned and modified study methodology.

5.3.1 Participants

Eligibility for phase two was based on the following criteria: (a) 5-12 years of age; (b) autism diagnosis based on the Autism Diagnostic Observation Schedule, Second Edition (ADOS-2; Lord et al., 2012); (c) able to complete a standardised vocabulary test; (d) able to identify at least one letter of the alphabet; (e) able to demonstrate sustained attention to shared interaction for 15-minutes; and (f) no serious hearing or vision impairments**.** Sixty participants completed the pre-instruction assessment. One participant did not meet the criteria for autism according to the ADOS-2 assessment and was excluded from the study, leaving 59 eligible children (43 male, 16 female).

The sample was drawn from predominantly lower socioeconomic areas in Western Sydney. See Table 5.1 for a summary of sociodemographic characteristics across the sample. Children attended a variety of school placements, including mainstream (*n* = 35), specialist class within a mainstream school (*n* = 21), special school (*n* = 1), and home school (*n* = 2). All children spoke English as their first language; however, six were exposed to a language other than English at home. Many participants had diagnoses in addition to autism, the most prevalent being Attention-deficit/hyperactivity disorder (ADHD) or attention-deficit disorder (ADD; *n* = 33).

Table 5.1 *Sociodemographic Characteristics of the Study Sample*

Variable	*n*	%
Parental highest level of education		
Left before end of high school	11	19%
Higher school certificate (Year 12)	9	15%
Graduate certificate or diploma	22	37%
Trade school	2	3%
Bachelor's degree	10	17%
Not reported	5	8%
Parental employment status		
Employed (full-time or part-time)	27	46%
Unemployed	16	27%
Unable to work	10	17%
Retired	1	2%
Not reported	5	8%
Annual household income		
$0 - $29,999	9	15%
$30,000 - $49,999	9	15%
$50,000 - $69,999	5	8%
$70,000 - $89,999	6	10%
$90,000 - $109,999	2	3%
$110,000 - $129,999	6	10%
$130,000 - $149,999	2	3%
$150,000 - $209,99	4	7%
$210,000 - $249,999	3	5%
Not reported	13	22%

To ensure similar ability levels across the three experimental groups a matched design was used where the sample was divided into groups of three participants (triplets) who were of a similar age and shared similar adaptive ability, nonverbal intelligence, vocabulary, and reading skills. For each triplet, participants were randomly assigned to one of the three experimental conditions (ABRA, ABRA+SBR, NI). Twenty children were initially allocated to the ABRA only group, 20 to ABRA+SBR, and 19 to NI[6]. One member from each of the ABRA and ABRA+SBR groups opted out of the study prior to the start of instruction and two ABRA group members opted out of the study after attending a single instruction session. Due to the move to online instruction and stay-at-home orders, a further eight

[6] Two children allocated to the ABRA+SBR group were reassigned to the control group due to parent request and the control group participant from each triplet substituted into the instruction group in their place.

participants chose to withdraw from the study in the seventh week of instruction (three ABRA, two ABRA+SBR, three NI). A total of 47 participants completed the study (14 ABRA, 17 ABRA+SBR, 16 NI).

5.3.2 Measures

A battery of standardised assessments was administered to obtain information on participant demographics, literacy, and related skills. Baseline assessments were administered in person in a quiet room at the school or clinic. Post-instruction literacy outcome measures were administered via telepractice, one-to-one on Zoom.

5.3.2.1 Literacy Related Measures

5.3.2.1.1 Adaptive Ability. The Comprehensive Interview form of the Vineland Adaptive Behaviour Scales – Third Edition was used to assess adaptive ability in the domains of Communication, Daily Living Skills, and Socialisation (Vineland-3; Sparrow et al., 2016). For children aged 6 years or younger, questions regarding motor skills were also administered. Individuals with autism comprised 0.9% of the normative sample for the Vineland-3. Based on the normative sample, correlations between the Vineland-3 Adaptive Behaviour Composite scores and previous versions of the Vineland were high (r = .64-.87). For the children in our sample, the Vineland-3 had high internal consistency (Cronbach's α = .99).

5.3.2.1.2 Nonverbal Intelligence. The Test of Nonverbal Intelligence – Fourth Edition was administered to measure cognitive ability (TONI-4; Brown et al., 2010). The TONI-4 is a language-free measure of intelligence, where children are required to point to a picture to complete a matrix. Children with intellectual or other disabilities comprised 3% of the normative sample for the TONI-4. Correlations between the TONI-4 and other established criterion measures of intelligence are high (r = .73-.79). In this study, the TONI-4 had high internal consistency (Cronbach's α = .94).

5.3.2.1.3 Phonological Awareness. The Phonological Awareness composite of the Comprehensive Test of Phonological Processing – Second Edition was used to measure phonological awareness skills (CTOPP-2; Wagner et al., 2013). This composite comprised the ‘Elision’ and ‘Blending Words’ subtests for all participants. Children 6-years and under also completed the ‘Sound Matching’ subtest, and children older than 6-years completed the ‘Phoneme Isolation’ subtest. Across the subtests, children were asked to identify and manipulate sounds in words. Children with disabilities comprised 5% of the normative sample for the CTOPP-2. Correlation between the Phonological Awareness composite and other phonological awareness measures indicate a robust level of validity (r = .64-.82). The CTOPP-2 had high internal consistency (Cronbach’s α = .95) for the children in our sample.

5.3.2.1.4 Vocabulary. The Peabody Picture Vocabulary Test – Fifth Edition was administered to measure receptive vocabulary (PPVT-5; Dunn 2019). Participants were required to point to a series of line drawings depicting individual words read aloud. Individuals with autism comprised 0.7% of the normative sample for the PPVT-5. Correlations between PPVT-5 scores and other measures of vocabulary provide evidence of validity (r = .61-.85). In the current sample, the PPVT-5 had high internal consistency (Cronbach’s α = .98).

5.3.2.2 Literacy Outcome Measures

5.3.2.2.1 Word-Level Reading Accuracy. The Word Reading subtest of the Wide Range Achievement Test – Fifth Edition was used to measure participants’ ability to read aloud letters and real words (WRAT-5; Wilkinson & Robertson, 2017). The maximum possible raw score for this test is 70. Children with intellectual disabilities comprised 1% of the normative sample for the WRAT-5, with individuals from other special groups excluded. As per the assessment manual, correlations between Word Reading subtest scores and other established measures of letter and word reading accuracy indicate validity (r = .79-.83). Internal consistency for the WRAT-5 was high for the children in our sample (Cronbach’s α = .97).

5.3.2.2.2 Nonword Reading Accuracy. The Word Attack subtest of the Woodcock-Johnson Tests of Achievement Fourth Edition – Australasian Adaptation was administered to measure participants’ ability to decode words (WJ-IV; Schrank et al., 2014). The maximum possible raw score for this test is 32. Children with autism or other disabilities were not described in the normative sample of the WJ-IV. Correlations between the WJ-IV Tests of Achievement and other similar measures provide evidence of validity (r = .75-.83). The WJ-IV had high internal consistency (Cronbach's α = .94) for the children in this study.

5.3.2.2.3 Passage-Level Reading Accuracy. Children's ability to accurately read aloud passage-level text was assessed using the Reading Accuracy Composite score from the Neale Analysis of Reading Ability – Third Edition (NARA-3; Neale, 1999). Children read aloud text passages of increasing length and complexity. The maximum possible raw score for this test is 100. The test manual does provide information on the number of children with disabilities or autism included in the normative sample. As per the assessment manual, correlations between other established reading measures and the composite scores on the NARA-3 provide evidence of validity (r = .70-.77). The NARA-3 Reading Accuracy Composite had high internal consistency in this study (Cronbach's α = .89).

5.3.2.2.4 Passage-Level Reading Comprehension. Two assessments were utilised to measure participants' ability to understand passage-level text. The primary measure was the Reading Comprehension Composite score from the NARA-3. Children were asked set questions after reading each of the previously described passages aloud. As noted above, correlations between the NARA-3 composite scores and other established reading measures were strong. In the current sample, the reading comprehension composite of the NARA-3 had high internal consistency (Cronbach's α = .95). The York Assessment of Reading for Comprehension – Australian Edition was administered as a supplementary measure of reading comprehension (YARC; Snowling et al., 2012). Again, children were asked a set of specified questions after reading a passage of text aloud. The YARC manual does not state the number of children with autism, or other disabilities, in its normative sample. Based on the normative sample, internal consistency values for reading comprehension on individual passages of the YARC – Form A are low[7] (Cronbach's α = .48-.59). Internal consistency for

[7] According to the YARC manual, this "reflects the fact that comprehension is a multi-faceted construct and our estimates of it are based on quite a small number of comprehension questions for each passage" (Snowling et al., 2012, p. 100).

the YARC was high in the current study (Cronbach's α = .96). Both assessments were administered starting from passage level one for all participants, meaning the maximum possible raw score for the NARA-3 was 44 and the maximum possible raw score for the YARC was 56.

5.3.3 Procedure

The procedure below describes Phase Two of the study.

5.3.3.1 Pre-instruction Assessment. Pre-instruction assessments were conducted across two 90-minute sessions with breaks provided as necessary. In the first session, parents provided demographic information using a standard questionnaire and completed the measure of adaptive ability, and participants engaged in assessments to confirm autism diagnosis and test nonverbal intelligence. In the second session, participants completed vocabulary, phonological awareness, and literacy assessments, including those used as outcome measures. Assessments were administered in the same order for all participants.

5.3.3.2 Instruction Phase. Participants allocated to the NI control group continued their business-as-usual school and therapy activities over the instruction period and received no intervention from the researchers over this time. Instruction group participants attended two instruction sessions per week with a researcher over 13-weeks, as well as continuing their business-as-usual learning activities. For instruction weeks 1 to 7, sessions were delivered across two sites (school and clinic) both based in Western Sydney (school-based sessions were conducted during school hours and clinic-based sessions took place after school). Instruction was implemented in the same way across the two sites. Participants attended sessions at only one site and were placed into groups of two to four children with similar ages and skills on baseline literacy measures. All sessions took place in a quiet room and the same researchers (both experienced speech pathologists) provided instruction at both sites. There was a 5-week break between instruction weeks 7 and 8 due to increasing restrictions owing to COVID-19. Weeks 8 to 13 of instruction were delivered one-to-one via telepractice (on Zoom) at a time convenient for participants (note: most children were not attending face-to-face schooling at this time due to stay-at-home orders). The same researchers administered the instruction online via telepractice. See Supplementary Material A for a breakdown of the modality of instruction sessions over the 13-weeks. Based on recommendations from the phase one focus groups, we implemented an external reward system for session completion (children received stickers on a chart and worked towards a prize of their choosing) and set group rules to facilitate engagement with instruction materials.

5.3.3.2.1 ABRA only sessions. All sessions, both face-to-face and via telepractice, followed the same session structure and involved approximately: (a) 15-minutes of word-level ABRA activities targeting alphabetics, high-frequency word identification, or spelling skills; (b) 20-minutes of passage-level ABRA activities targeting reading fluency or comprehension skills (including vocabulary) within the ABRA interactive texts; and (c) a 10-minute reward activity. For the group-based sessions, each child worked independently on a touchscreen laptop with headphones to complete the word-level activity. Participants then gathered around a large monitor to collaboratively complete the passage-level activity using a single computer. For sessions via telepractice, the researcher screen-shared the ABRA program and gave participants screen control to enable them to directly access and engage with the word- and passage-level activities in ABRA.

Pre-instruction assessment data was used to set individual learning goals for participants and to determine appropriate activity start points for individuals (for further details regarding ABRA activities and goal setting see: CSLP, 2019). Word-level ABRA activities range in level of difficulty and complexity. This meant that participants could complete activities at their individual level during group sessions. Participants' task accuracy data (number of items correct/incorrect) were recorded during instruction and reviewed following each session. For the face-to-face group sessions, this data was recorded electronically by the ABRA program. For individual telepractice sessions, task accuracy data was recorded manually by the researchers. A performance criterion of 65-85% accuracy was used to determine appropriate activities and level of difficulty. A mastery level of 85% accuracy over three sessions was set to determine progression to the next activity level or a different skill. This mastery level and performance criterion were used for all word-level activities and for passage-level activities during individual sessions. An average of the group's performance was taken for passage-level activities during group sessions to determine appropriate activity level of difficulty.

5.3.3.2.2 ABRA + SBR sessions. Participants in the ABRA+SBR group completed the same protocol as outlined above in the ABRA only sessions, with the addition of a 15-minute SBR activity prior to the reward activity. SBR activities were conducted using the Fitzroy Reader iPad application (Fitzroy Programs and Greygum Software, 2016). During the face-to-face group-based sessions, children took turns with other participants to read aloud a page in the book. In one-to-one sessions via telepractice, children alternated reading a page aloud with the researcher. Children were encouraged to ask and answer questions with the researcher and/or child group members throughout SBR. They were provided with visual prompts for generating basic or more complex questions based on their ability level. A simplified visual checklist of strategies to be used before, during, and after reading was implemented during SBR (modified based on: Klingner & Vaughn, 1998 and Reutebuch et al., 2015) and children were encouraged to independently monitor their use of specific reading comprehension strategies introduced during ABRA activities (e.g., predicting, comprehension monitoring, summarising).

5.3.3.3 Post-instruction Assessment. Post-instruction assessments were conducted at the end of the instruction period and took place one-to-one via Zoom (in the same order as the pre-instruction assessment). Assessments were administered in the same way as the pre-instruction assessment, except that participants viewed PDF versions of the materials on their computer screens instead of the paper-based stimuli.

5.3.4 Implementation Fidelity

Both researchers administering the instruction sessions were speech pathologists with many years' experience working with children with developmental disabilities, including children with autism. The researchers had both received training on the ABRA program for previous intervention studies and had experience prior to this study administering similar instruction protocols of ABRA and SBR. A random selection of thirty-six sessions across experimental conditions and instruction modalities were recorded for fidelity assessment (approximately 7% of the 549 total sessions conducted). This included 17 ABRA only sessions (4 group sessions) and 19 ABRA + SBR sessions (4 group sessions). These recordings, along with the corresponding session plan, were reviewed by either a researcher or research assistant on the project with knowledge of the ABRA program. The ABRA Fidelity Rating Form was used alongside the SBR Fidelity Rating Form (as appropriate to the experimental condition) to evaluate the quality of literacy instruction and adherence to the instruction protocol (see Tables 5.2 and 5.3 for fidelity checklist items, ratings, and details of rating scale).

Table 5.2 *Means and Standard Deviations for ABRA Implementation Fidelity Ratings*

Fidelity item	*M*	*SD*	Range
Researcher is familiar with the ABRA program/lesson content	4.86	0.35	4 – 5
Lesson has clear goals and objectives	5	0	5
Lesson is planned ahead of time	5	0	5
Lesson includes word- and text-level activities	5	0	5
Lesson includes introduction	5	0	5
Lesson includes demonstration	5	0	5
Researcher monitors child's navigation of the program	4.86	0.35	4 – 5
Lesson includes conclusion	4.78	0.96	0 – 5
Children's behaviour doesn't interfere with learning	4.56	0.73	2 – 5
Researcher uses ability level differentiation when appropriate	4.72	0.61	3 – 5
Researcher prepares learning environment prior to the lesson	4.86	0.35	4 – 5
Lesson includes at least 10mins of word-level ABRA activities	4.67	0.76	2 – 5
Lesson includes at least 15mins of text-level ABRA activities	4.97	0.17	4 – 5
Researcher reflects on lesson planning and implementation and modifies future lessons accordingly	4.97	0.17	4 – 5

Note. Each item was rated using a six-point scale: 0 = not applicable, 1 = strongly disagree, 2 = disagree, 3 = neutral, 4 = agree, 5 = strongly agree.

Table 5.3 *Means and Standard Deviations for SBR Implementation Fidelity Ratings*

Fidelity item	*M*	*SD*	Range
Lesson includes at least 15mins non-ABRA SBR activities	4.47	1.12	2 – 5
Learning environment is appropriate for SBR	4.89	0.32	4 – 5
SBR activity is planned ahead of time	5.00	0	5
SBR activity includes decoding and comprehension strategies	4.95	0.23	4 – 5
SBR activity involves input from child/ren	5.00	0	5
Researcher facilitates discussion/interaction during SBR	5.00	0	5
Child/ren's behaviour doesn't interfere with SBR activity	4.37	0.60	3 – 5
Researcher uses ability level differentiation when appropriate	5.00	0	5
Researcher models target SBR strategies during activity	4.95	0.23	4 – 5
Child/ren are encouraged to decode words originally read incorrectly	4.74	0.45	4 – 5
Child/ren are encouraged to reflect on their own reading errors	4.58	1.17	0 – 5
Children are encouraged to ask/answer at least one question during reading	5.00	0	5
SBR activity includes discussion of at least one reading comprehension strategy at each stage of reading (before, during, after)	4.95	0.23	4 – 5
Researcher reflects on lesson planning and implementation and modifies future lessons accordingly	4.89	0.46	3 – 5

Note. Each item was rated using a six-point scale: 0 = not applicable, 1 = strongly disagree, 2 = disagree, 3 = neutral, 4 = agree, 5 = strongly agree.

5.3.5 Data Analysis

One-way ANOVAs were used to determine whether there were any statistically significant differences in ability level across the three groups at baseline. The between-subjects factor was group (ABRA vs ABRA+SBR vs NI) and the dependent variables were age, adaptive ability, nonverbal intelligence, vocabulary, phonological awareness, and all reading measures. The one-way ANOVA is robust and able to withstand some violations of normality whilst still providing valid results (Glass et al., 1972); however, in instances where there were violations, the equivalent non-parametric analyses were also completed.

A series of two-way mixed ANOVAs were conducted to analyse the effects of instruction on participants' reading skills. The within-subjects factor was time (pre- vs post-instruction), and the between-subjects factor was group (ABRA vs ABRA+SBR vs NI). The dependent variables were word-level reading accuracy, nonword reading accuracy, passage-level reading accuracy, and passage-level reading comprehension (all based on raw scores). Given the number of tests that were conducted, a more conservative alpha of .01 was adopted. Where there was a statistically significant two-way interaction, simple main effects were investigated using separate one-way ANOVAs for group and separate repeated measures ANOVAs for time. Two-way mixed ANCOVAs using adaptive ability, phonological awareness, nonverbal intelligence, and receptive vocabulary as covariates were also conducted. Effect sizes were interpreted as follows: η_p^2 of .01 corresponded to a small effect size, .06 a medium effect size, and .14 a large effect size (Richardson, 2011). Where an alpha of .01 was not reached but a large effect size was found, this result was further explored using separate repeated measures ANOVAs. For variables that violated assumptions of the two-way mixed ANOVA, non-parametric analyses were also conducted using the Kruskal Wallis *H* test.

5.4 Results

5.4.1 Instruction Adherence and Fidelity

Of the 31 instruction group participants who participated in both the face-to-face and online sessions, all but three attended the target number of 26 instruction sessions (one ABRA group member completed 20 sessions, two ABRA+SBR group members completed 16 and 23 sessions respectively). Reasons for session non-attendance included school suspensions and session refusal. Sessions were considered ‘complete’ if participants completed all scheduled activities set by the researcher or ‘partially complete’ if participants did not complete one of the set activities for the session. Sessions in which participants did not fully complete two or more of the planned activities were considered ‘incomplete’. Of the 787 instruction sessions[8], 738 were ‘complete’ (94%), 39 sessions were ‘partially complete’ (5%), and 10 sessions were ‘incomplete’ (1%). Implementation fidelity was high for both ABRA instruction delivery and SBR (see Tables 5.2 and 5.3). Review of fidelity ratings

[8] A total of 549 sessions were conducted in the study. The number 787 includes each child’s participation data separately during group sessions.

across instruction modalities indicates that the researchers were less able to monitor children's navigation of the ABRA program and provide ability level differentiation for activities during the face-to-face group sessions than during the individual sessions via telepractice. However, implementation fidelity ratings remained high and did not differ substantially between face-to-face and telepractice instruction delivery. This high level of fidelity was likely due to several factors, including the researchers' prior training and knowledge of the ABRA program and level of clinical skills and experience. These factors potentially influenced the careful adherence to the instruction protocol and the quality of instruction delivery, including responsiveness, differentiation, and dosage within instruction sessions. Instruction delivered by researchers, rather than teachers or community-based clinicians, may have contributed to the high level of fidelity. Fidelity is an important factor in research with practical implications for program effectiveness and outcomes.

5.4.2 Instruction Outcomes

Table 5.4 shows baseline scores by group for participants who completed both the pre- and post-instruction assessments (*n* = 47). One-way ANOVAs revealed no statistically significant difference between the three groups for any literacy or related skills at baseline. As can be seen in Table 5.4, baseline scores for each measure varied considerably within groups, reflecting the broad inclusion criteria. Children tended to perform below the age and year-of-schooling norms for typically developing children on standardised measures of word reading accuracy (mean percentile rank = 26.60, *SD* = 30.18), nonword reading accuracy (mean percentile rank = 18.25, *SD* = 20.25), passage-level reading accuracy (mean percentile rank = 17.39, *SD* = 22.20), and passage-level reading comprehension (mean percentile rank = 15.39, *SD* = 22.00). Baseline scores by group for all participants who completed the pre-instruction assessment (*n* = 59) can be seen in Supplementary Material B at the end of this chapter. The number of participants who withdrew across conditions was too small to conduct any formal statistical analyses. However, the participants in intervention conditions who withdrew had generally higher reading scores than the control group, suggesting that participant withdrawal was not selectively biased in favour of the intervention condition.

Table 5.5 provides mean raw scores by group for each outcome measure at pre- and post-instruction assessment. The assumption of normality was not met for one outcome measure (NARA passage-level reading comprehension). For this reason, we report both parametric and non-parametric results for this measure below. All other variables met the assumptions for parametric analysis, including no significant outliers and approximately normal distribution (based on Shapiro-Wilk test of normality and skewness and kurtosis values). ANCOVAs with adaptive ability, phonological awareness, nonverbal intelligence, and receptive vocabulary as covariates did not change the pattern of statistical significance for any measures.

5.4.2.1 Word-level Reading Accuracy. Analysis of the word-level reading accuracy data found a significant main effect of Time, $F(1, 44) = 49.20$, $p < .001$, $\eta_p^2 = .53$, indicating improvement in word-level reading accuracy from pre- to post-instruction across the sample. The Time by Group interaction was not statistically significant, suggesting that word-level reading accuracy gains did not vary between the instruction and control groups, $F(2, 44) = 2.57$, $p = .088$, though the effect size fell within the medium range, $\eta_p^2 = .11$.

5.4.2.2 Nonword Reading Accuracy. For nonword reading accuracy, the main effect of Time was statistically significant, $F(1, 44) = 32.01$, $p < .001$, $\eta_p^2 = .42$, suggesting an improvement in skills from pre- to post-instruction assessment for the sample as a whole. A significant Time by Group interaction was also identified, $F(2, 44) = 6.60$, $p = .003$, with a large effect size, $\eta_p^2 = .23$, showing that gains in nonword reading accuracy varied between the instruction and control groups. There was no simple main effect of group at post-instruction, $F(2, 44) = 1.48$, $p = .240$, $\eta_p^2 = .06$. Repeated measures ANOVAs revealed that there were statistically significant simple main effects of Time for the ABRA group, $F(1, 13) = 9.98$, $p = .008$, $\eta_p^2 = .44$, and ABRA+SBR group, $F(1, 16) = 51.36$, $p < .001$, $\eta_p^2 = .76$, but not for the NI group, $F(1, 15) = .45$, $p = .514$, $\eta_p^2 = .03$.

Table 5.4 *Scores for Pre-Instruction Baseline Measures by Group for Participants Remaining at Completion of Study*

	ABRA only (*n* = 14)			ABRA+SBR (*n* = 17)			NI control (*n* = 16)					
Measure	*M*	*SD*	Range	*M*	*SD*	Range	*M*	*SD*	Range	*F(2, 44)*[c]	*p*	η_p^2
Age [a]	102	22.79	72 -139	106	25.59	66 - 145	103	23.67	60 - 143	.114	.893	.005
Adaptive ability [b]	73.00	9.59	60 - 94	73.24	11.75	48 - 102	74.31	9.57	48 - 90	.068	.934	.003
Nonverbal intelligence	19.86	6.54	8 - 29	22.18	7.96	9 - 38	21.69	9.36	7 - 40	.340	.714	.015
Vocabulary	144.71	26.38	94 - 192	143.65	28.34	98 - 183	135.6	37.53	47 - 194	.395	.676	.018
Phonological awareness [b]	83.36	18.98	56 - 110	78.94	25.03	12 - 112	78.38	13.10	50 - 96	.279	.758	.013
Word-level reading accuracy	28.00	12.60	12 - 49	27.53	13.50	7 - 46	24.75	16.50	0 - 54	.219	.804	.010
Nonword reading accuracy	14.93	5.98	5 - 23	13.82	7.63	1 - 26	12.88	7.68	2 - 25	.304	.739	.014
Passage-level reading accuracy	27.64	27.11	0 - 83	29.82	26.62	0 - 87	25.19	25.79	0 - 77	.126	.882	.006
Reading comprehension	8.79	9.28	0 - 29	10.71	9.89	0 - 33	8.25	8.74	0 - 26	.316	.731	.014

Note. Adaptive ability: Vineland Adaptive Behavior Scale – 3rd edition (Vineland-3), Adaptive Behaviour Composite standard score; Nonverbal intelligence: Test of Nonverbal Intelligence – 4th Edition (TONI-4); Vocabulary: Peabody Picture Vocabulary Test – 5th Edition (PPVT-5); Phonological awareness: Comprehensive Test of Phonological Processing – 2nd edition (CTOPP-2), phonological awareness composite score; Word-level reading accuracy: Wide Range Achievement Test – 5th Edition (WRAT-5), Word Reading subtest raw score; Nonword reading accuracy: Woodcock-Johnson Tests of Achievement – 4th Edition (WJ-IV), Word Attack subtest; Passage-level reading accuracy and reading comprehension: Neale Analysis of Reading Ability – 3rd edition (NARA-3), Accuracy and comprehension composite raw scores.

[a] age reported in months. [b] scores for adaptive ability and phonological awareness are based on composite score. All others based on raw scores. [c] *F* (2, 43) for adaptive ability.

Table 5.5 *Mean Raw Scores Pre- and Post-instruction for Each Outcome Measure by Group*

Measure	ABRA only (*n* = 14)			ABRA+SBR (*n* = 17)			NI control (*n* = 16)		
	M	*SD*	Range	*M*	*SD*	Range	*M*	*SD*	Range
Pre-instruction									
Word-level reading accuracy	28.00	12.60	12 - 49	27.53	13.50	7 - 46	24.75	16.50	0-54
Nonword reading accuracy	14.93	5.98	5 - 23	13.82	7.63	1 - 26	12.87	7.68	2-25
Passage-level reading accuracy	27.64	27.11	0 - 83	29.82	26.62	0 - 87	25.19	25.79	0-77
Reading comprehension	8.79	9.28	0 - 29	10.71	9.89	0 - 33	8.25	8.75	0-26
Supplementary – reading comprehension	14.14	11.49	0 - 39	17.29	13.43	0 - 38	11.81	11.91	0-36
Post-instruction									
Word-level reading accuracy	31.64	11.21	15 - 52	33.24	12.97	11 - 57	27.44	16.69	0-55
Nonword reading accuracy	17.07	7.16	7 - 26	17.59	7.47	3 - 30	13.38	8.02	0-27
Passage-level reading accuracy	36.29	27.98	0 - 91	42.00	30.92	0 - 93	28.44	27.41	0-90
Reading comprehension	13.43	10.75	0 - 34	16.06	12.44	0 - 37	10.81	11.23	0-35
Supplementary – reading comprehension	18.79	13.96	0 - 46	20.94	14.44	0 - 41	15.93	14.55	0-41

Note. Word-level reading accuracy: WRAT-5, Word Reading subtest; Nonword reading accuracy: WJ-IV, Word Attack subtest; Passage-level reading accuracy and reading comprehension: NARA-3, Reading accuracy and comprehension composite raw scores; Supplementary reading comprehension: YARC, Reading comprehension raw score. Zero scores represent true zeros, where participants were unable to correctly respond to any items on that measure. Children who scored zeros were approximately evenly distributed between experimental conditions (three ABRA group members, four ABRA+SBR members, five NI group members). Participants consistently scored zero on measures across assessment timepoints, with the exception of two participants in both the ABRA and ABRA+SBR groups who registered higher scores at post-assessment, and one child in NI group scored zero on one of the post-assessments after achieving a score of 1 in the pre-assessment.

5.4.2.3 Passage-Level Reading Accuracy. The main effect of Time revealed a statistically significant change in mean passage-level reading accuracy scores from pre- to post-instruction, $F(1, 44) = 35.04$, $p < .001$, $\eta_p^2 = .44$. There was no statistically significant Time by Group interaction at our conservative alpha level, $F(2, 44) = 3.86$, $p = .028$, though the effect size was large, $\eta_p^2 = .15$. Given the large effect size, this result was explored further. Repeated measures ANOVAs revealed statistically significant simple main effects of Time for the ABRA group, $F(1, 13) = 12.81$, $p = .003$, $\eta_p^2 = .50$, and ABRA+SBR group, $F(1, 16) = 28.04$, $p < .001$, $\eta_p^2 = .64$, but not for the NI group, $F(1, 15) = 1.99$, $p = .18$, $\eta_p^2 = .12$.

5.4.2.4 Passage-Level Reading Comprehension. For both reading comprehension measures, analysis showed a statistically significant main effect of Time (NARA-3: $F[1, 44] = 34.32$, $p < .001$, $\eta_p^2 = .44$; YARC: $F[1, 43] = 25.83$, $p < .001$, $\eta_p^2 = .38$), indicating that reading comprehension scores increased between pre- and post-instruction assessment. However, there was no statistically significant interaction between instruction Group and Time on either measure of reading comprehension abilities (NARA-3: $F[2, 44] = 1.43$, $p = .250$, $\eta_p^2 = .06$ [medium effect]; YARC: $F[2, 43] = .16$, $p = .855$, $\eta_p^2 = .01$ [small effect]). Non-parametric analysis confirmed that median reading comprehension pre-post-difference scores on the NARA-3 were not statistically significant between groups, χ^2 (2) = 4.249, $p = .120$.

Given the link between social-communication and reading comprehension skills in autistic children, we conducted correlational analyses exploring the relationship between children's scores on the Socialisation domain of the Vineland-3 (Sparrow et al., 2016) and pre-post-difference scores on the primary reading comprehension measure (NARA-3; Neale, 1999). In view of violations of normality and linearity assumptions, non-parametric analyses (Spearman's *r*) were used. There was a statistically significant positive correlation between socialisation scores and reading comprehension for the ABRA+SBR group, $r_s(15) = .539$, $p = .026$. There was no statistically significant correlation between socialisation and reading comprehension scores for the ABRA only group ($r_s(11) = .146$, $p = .634$) or control group (r_s (14) = -.223, $p = .406$). Across all groups, there were no statistically significant correlations between socialisation scores and word, nonword, or passage-level reading accuracy scores.

5.5 Discussion

This study explored the effects of ABRA instruction for children with autism during the COVID-19 pandemic. As a result of government mandated stay-at-home orders issued midway through the study, the method of instruction delivery changed from face-to-face small group instruction to one-to-one sessions delivered via telepractice. This modification, though necessary, was unprecedented and undoubtedly had a substantial impact on outcomes, both due to the methodological deviation as well as the adverse effects of the pandemic (Baweja et al., 2022; Di Renzo et al., 2020; Pellicano et al., 2022). The effects of instruction, practical implications, and lessons learned are discussed below.

Although reading gains were statistically significant between groups on only one outcome measure, most outcomes were associated with medium to large effect sizes. These effect sizes are similar to what has been found in previous studies of ABRA instruction for children with autism (Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022). This study ended up being underpowered for several unforeseen reasons, including being conducted in the middle of a pandemic, in a geographic location of socioeconomic inequity, and involving a special population, intersecting factors that contributed to participant attrition. In light of this, statistical significance, or lack thereof, should not be interpreted in isolation and should be considered within the context of sample size, meaningful effect sizes, and prior research (Abelson, 1995; Betensky, 2019; Kyle et al., 2013; Wasserstein & Lazar, 2016; Wasserstein et al., 2019).

5.5.1 Effects of Instruction

Our analyses show that children who participated in the intervention conditions made statistically significant gains compared with the control group on our measure of nonword reading skills. It appears that these gains in phonics skills were further facilitated by the SBR activity, with gains on the nonword reading measure greater for children in the ABRA+SBR condition compared with the ABRA only condition (larger effect size and greater gain in mean scores as seen in Table 5.5). In the three previous studies of ABRA and autistic children (Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022) and in the current study, passage-level reading accuracy gains have consistently shown large effect sizes. Although passage-level reading accuracy gains were not statistically significant at our conservative alpha level in this study, the effect sizes for this skill are similar to what has been found in previous studies.

In the current study, gains in reading comprehension skills for children in the ABRA+SBR condition were positively correlated with socialisation skills, indicating that children in this group with higher socialisation adaptive abilities made greater reading comprehension gains following instruction. Previous research indicates that social communication skills are associated with reading comprehension for autistic children (McIntyre et al., 2018; Ricketts et al., 2013), and Arciuli and Bailey (2019) found a link between children's socialisation skills and reading comprehension gains in their study of group-based ABRA delivery. The different findings across the ABRA and ABRA+SBR conditions in this study potentially reflect the higher demands placed on children's social communication skills with the SBR activity (initially group-based and then one-to-one with the researcher online), compared with only the computer-based ABRA instruction. Future studies could examine the effects of socialisation skills on literacy gains within one-to-one versus group-based literacy instruction for autistic children and could explore the quality of children's social interactions within these settings, as well as during computerised and non-computerised activities.

5.5.2 Lessons Learned

5.5.2.1 Participant Attrition. Given that previous ABRA studies involving autistic children have not reported any attrition (Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022), it is noteworthy that 12 participants withdrew from the current study. This attrition is largely due to the pandemic related disruptions, though there may be other considerations. Participants with a range of abilities chose to leave the study (see Supplementary Material C at the end of this chapter); however, those with lower socioeconomic characteristics were more likely to withdraw. Almost half of the families across the entire sample who reported an annual household income of $0-$29,999 (AUD) were amongst the participants who withdrew from the study (44%), as were almost half of the parents who described themselves as unemployed (40%). Furthermore, we observed site-based attrition patterns with all instruction group participants who withdrew in the first half of the study being from the clinic and all who withdrew following the lockdown being from the school. These findings have important implications for engaging with areas of socio-demographic inequity, both during a pandemic and beyond, as well as considerations regarding the feasibility and accessibility of some interventions.

5.5.2.2 Pandemic Effects. Researchers have started to explore the learning loss for children as a result of school closures and home learning throughout the pandemic (e.g., Engzell et al., 2021). While some studies suggest there were no differences in children's literacy development during the pandemic (Gore et al., 2021), most suggest that lockdowns and school closures had a significantly negative impact on children's reading abilities. Studies estimate approximately 2 to 8 months of learning loss on reading progress over the pandemic, with some suggesting a higher learning loss for children from lower socioeconomic backgrounds (Aurini & Davies, 2021; Betthäuser et al., 2023; Department for Education, 2021b; Government of Alberta, 2021; Ludewig et al., 2022; Molnár & Hermann, 2023). Interestingly, our results indicate that children across both intervention conditions and the control condition (where children received remote schooling only and did not participate in additional literacy instruction with the research team) made gains in their reading skills over the course of this study.

5.5.2.3 Changes to Planned Methodology. The shift to telepractice instruction and assessment midway through our study was the only option available to us. Some studies suggest that literacy assessment and intervention via telepractice is largely equivalent to face-to-face; however, these studies have not included children with autism (Furlong et al., 2021; Hodge et al., 2018). Systematic reviews exploring non-literacy based assessment and intervention for autistic children via telepractice again suggest that online services are comparable to face-to-face; yet, the overwhelming majority of studies have been parent/teacher mediated interventions, with very few involving an autistic child actively engaging with a clinician online (Ellison et al., 2021; Sutherland et al., 2018). Emerging studies exploring the experience of autistic individuals throughout the pandemic suggest that whilst some have had positive experiences with the transition to online service delivery, many report struggling with this service modality, with some finding the additional work of interpreting social communication cues via text, phone, and video calls difficult and tiring (Fatehi et al., 2023; Pellicano et al., 2020; Pellicano et al., 2022; Simpson & Adams, 2023).

5.6 Limitations and Conclusions

There are several clear limitations to the current study, many of which have been outlined above. Given deviations from the original study protocol and the resulting small sample size that was underpowered for a study of this design, our results should be interpreted with caution. The high level of attrition is a limitation, though not unexpected given the effects of the pandemic, socio-demographic area in which the study took place, and support needs of our child participants (Justice et al., 2011; Sanders et al., 2021; Yi & Dixon, 2021). As discussed, this resulted in children from lower-income families being more likely to withdraw, leaving a potentially biased final sample. Future research needs to investigate ways to keep children and families from lower socioeconomic areas, who are already at risk of more adverse health and literacy outcomes, engaged with educational and clinical services during challenging times.

Our findings contribute to the scarce knowledge base of literacy instruction for autistic children during the pandemic. These findings may have implications for telepractice delivery of literacy instruction to children with autism living in remote settings beyond the pandemic. We hope that our findings can facilitate discussion in the research community and support future research and next steps in the field.

5.7 Supplementary Material A

Figure 5.1 *Planned Study Methodology*

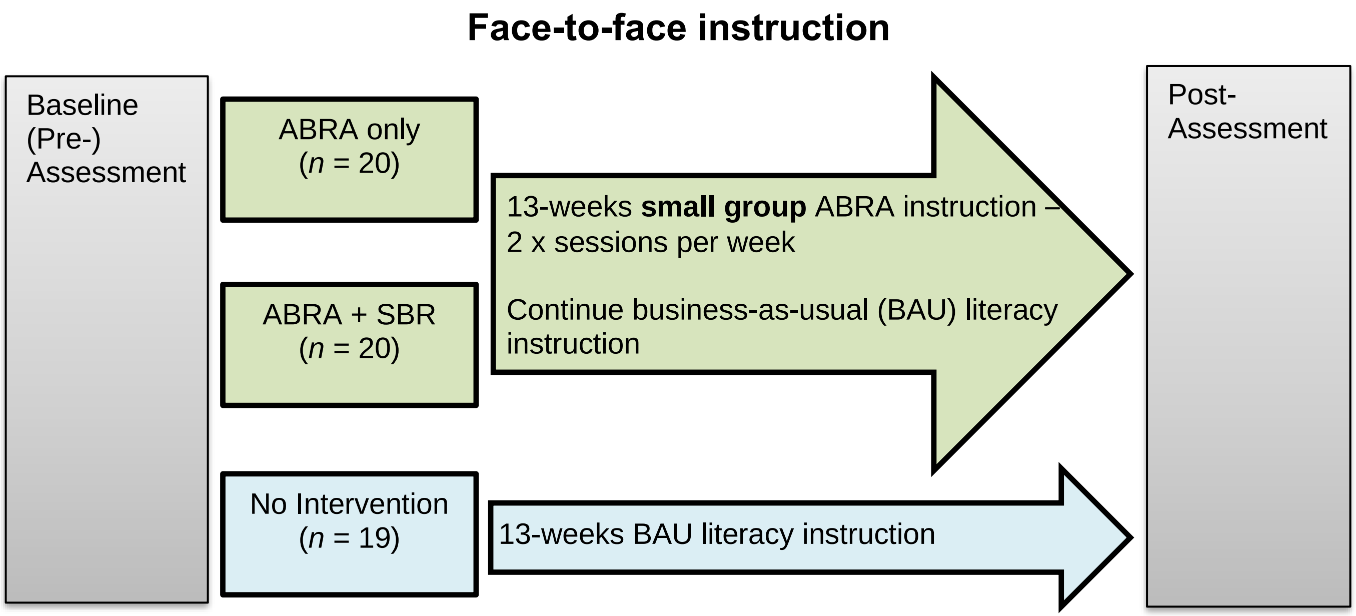
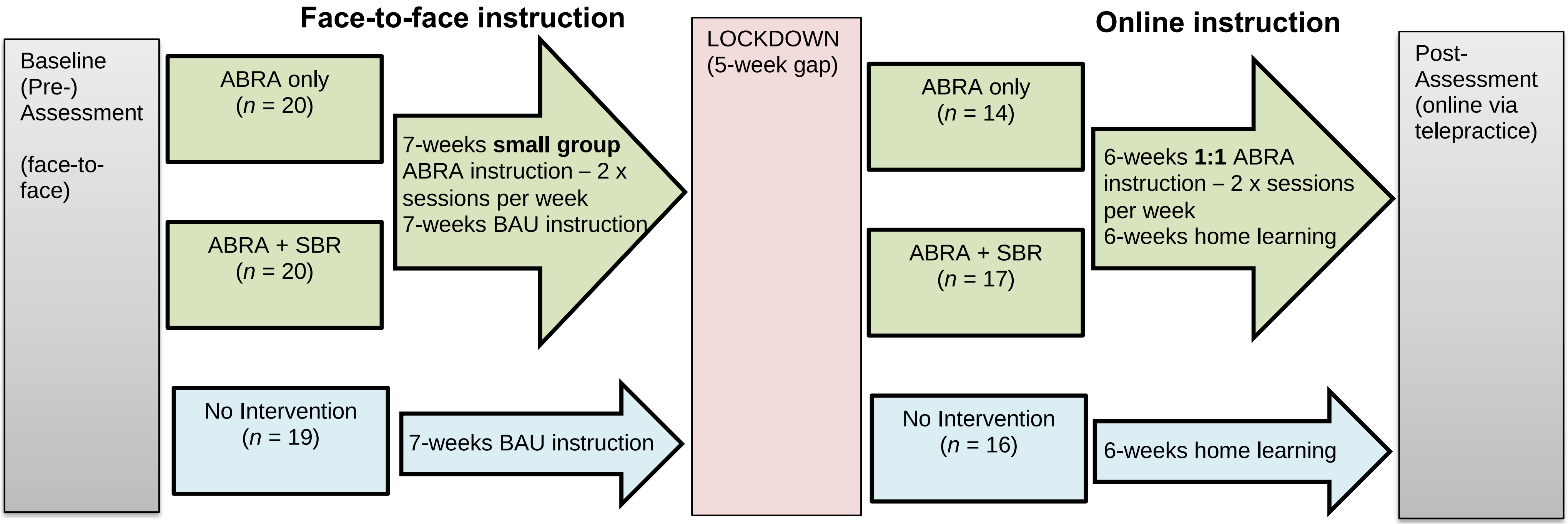


Figure 5.2 *Modified Study Methodology*



5.8 Supplementary Material B

Table 5.6 *Scores for Pre-Instruction Baseline Measures by Group (All Participants at Baseline)*

	ABRA only instruction (*n* = 20)			ABRA+SBR instruction (*n* = 20)			NI control (*n* = 19)					
Measure	*M*	*SD*	Range	*M*	*SD*	*Range*	*M*	*SD*	Range	$F(2, 56)^c$	*p*	η_p^2
Age[a]	104	24.67	66 – 150	108	24.85	66 – 145	104	23.69	60 – 143	.168	.845	.006
Adaptive ability[b]	71.47	11.78	59 – 94	76.00	14.76	48 – 115	74.26	8.76	48 – 90	.721	.491	.027
Nonverbal intelligence	18.40	7.21	3 – 29	21.95	7.67	9 – 38	22.32	8.96	7 – 40	1.460	.241	.050
Vocabulary	138.60	33.78	67 – 192	140.85	28.65	98 – 183	136.63	34.67	47 – 194	.083	.921	.003
Phonological awareness[b]	76.90	19.19	50 – 110	74.70	26.68	12 – 112	77.11	13.88	50 – 96	.082	.921	.003
Word-level reading accuracy	27.00	14.26	0 – 49	27.85	13.33	7 – 46	25.26	15.83	0 – 54	.144	.866	.005
Nonword reading accuracy	13.95	7.19	0 – 24	14.15	7.71	1 – 26	12.89	7.29	2 – 25	.160	.852	.006
Passage-level reading accuracy	26.65	25.08	0 – 83	30.35	27.62	0 – 87	24.63	24.058	0 – 77	.250	.779	.009
Reading comprehension	8.55	8.55	0 – 29	10.80	10.05	0 – 33	8.05	.25	0 – 26	.522	.596	.018

Note. Adaptive ability: Vineland Adaptive Behavior Scale – 3rd edition (Vineland-3), Adaptive Behavior Composite standard score; Nonverbal intelligence: Test of Nonverbal Intelligence – 4th Edition (TONI-4); Vocabulary: Peabody Picture Vocabulary Test – 5th Edition (PPVT-5); Phonological awareness: Comprehensive Test of Phonological Processing – 2nd edition (CTOPP-2), phonological awareness composite score; Word-level reading accuracy: Wide Range Achievement Test – 5th Edition (WRAT-5), Word Reading subtest raw score; Nonword reading accuracy: Woodcock-Johnson Tests of Achievement – 4th Edition (WJ-IV), Word Attack subtest; Passage-level reading accuracy and reading comprehension: Neale Analysis of Reading Ability – 3rd edition (NARA-3), Accuracy and comprehension composite raw scores.

[a] age reported in months [b] scores for adaptive ability and phonological awareness are based on composite score. All others based on raw scores. [c] F (2, 52) for adaptive ability.

CHAPTER 6 DISCUSSION AND CONCLUSIONS

The primary purpose of this thesis is to provide a clearer understanding of effective reading instruction methods and models of service delivery for children with developmental disabilities to help guide policies and practices that aim to improve literacy outcomes for these children. In this chapter, I discuss the results of the empirical studies reported in Chapters 3 to 5, including the impact of the global COVID-19 pandemic on this research. These findings have important implications for parents, educators, clinicians, researchers, and policy makers, which are explored towards the end of the chapter. This chapter ends with a discussion of the research limitations and considerations for future research.

6.1 Systematic Review

The second chapter of this thesis was a systematic review on the effects of different methods of literacy instruction on the reading and writing skills of children with cerebral palsy (CP). This review was necessary as children with CP regularly underachieve in reading (Critten et al., 2019, 2023; Micheletti et al., 2023; Wotherspoon et al., 2023), yet no existing studies have systematically explored literacy instruction methods specifically for this group. The findings from this systematic review indicated that instruction designed to teach phonics, reading fluency, reading comprehension, written expression, or spelling skills in isolation were effective in improving closely associated outcomes for children with CP. This review highlighted several issues with the state of research on literacy instruction for children with CP. Firstly, included studies primarily explored instruction targeting literacy skills in isolation, and only one study explored multicomponent literacy instruction. No studies explored comprehensive literacy instruction (instruction incorporating the National Reading Panel's (NRP) Big Five; National Institute of Child Health and Human Development [NICHD], 2000). Secondly, new technologies which allow individuals with significant speech and motor impairments independent access to literacy materials and instruction (e.g., eye-gaze technology) were not included in any studies. Thirdly, many studies were of lower quality, and all but one study utilised single-subject research designs. Finally, included studies focused on developing reading accuracy skills, with only one study exploring methods to develop reading comprehension skills. The findings of this review go some way towards explaining the poorer literacy outcomes documented for children with CP. The case study presented in this thesis was a small but necessary next step towards including children with CP in research exploring comprehensive methods of literacy instruction.

6.2 Outcomes of Empirical Studies

The three empirical studies reported in this thesis were all designed to extend prior research on literacy instruction for children with developmental disabilities. As no studies had previously explored comprehensive literacy instruction for children with CP or Down syndrome, these studies were small-scale and exploratory as a first step towards larger studies in the future. Based on previous research, we hypothesised that the children in our studies would achieve gains in both reading accuracy and reading comprehension skills following instruction. Contrary to our hypotheses, the most consistent gains made across the empirical studies involving children with CP, Down syndrome, or autism were in skills that support reading accuracy, namely, phonics skills (letter-sound knowledge and decoding). Study findings are discussed further in the sections below, as are the potential impacts of the pandemic on study outcomes. Key features and differences between the three studies are summarised in Table 6.1 to contextualise the discussion that follows in this chapter.

Table 6.1 *Summary of Key Study Features*

	Empirical study 1: CP	**Empirical study 2: Down syndrome**	**Empirical study 3: Autism**
Participant Characteristics:			
No. of participants	1	6	59 (47 in final sample)
Age of participants (year; months)	8;0	Range: 8;6 – 11;3 (mean = 9;8)	Range: 5;0 – 12;5 (mean = 8;7)
IQ[a]	PR = 5	PR range: 0.3 – 25 (mean PR = 12.22, *SD* =11.36)	PR range: 3 – 86 (mean PR =40.42, *SD* = 22.86)
Adaptive ability[b]	PR = 2	PR range: 1 – 39 (mean PR = 10.33, *SD* = 14.47)	PR range: <1 – 84 (mean PR = 8.0, *SD* = 13.90)
Baseline reading abilities[c]	Word reading accuracy (PR = 0.1)	Word reading accuracy (mean PR = 11.50, *SD* = 8.50)	Word reading accuracy (mean PR = 26.60, *SD* = 30.18)
	Reading comprehension (unable to complete task)	Reading comprehension (mean PR = 2.50, *SD* = 4.23)	Reading comprehension (mean PR = 15.39, *SD* = 22.00)
Participant SES[d]	Index of relative socioeconomic advantage and disadvantage = medium	Index of relative socioeconomic advantage and disadvantage = medium to high	Index of relative socioeconomic advantage and disadvantage = low to medium
Participants with dual diagnoses	Dyspraxia	None reported	ADHD/ADD (*n* = 33); intellectual disability (*n* = 6); anxiety (*n* = 3); epilepsy (*n* = 2)
Study characteristics:			
Study design	Case study	Repeated measures within-participants design	Quasi-experimental study
Program	ABRA + Clinician-led SBR	ABRA + Clinician-led SBR	(a) ABRA + Clinician-led SBR;

	Empirical study 1: CP	Empirical study 2: Down syndrome	Empirical study 3: Autism
			OR (b) ABRA only
SBR texts	Fitzroy readers app	Fitzroy readers app	Fitzroy readers app
Supplementary instruction	2 x 15-min parent-led SBR/week	2 x 15-min parent-led SBR/week	None
Instructor	One speech pathologist	One speech pathologist	Two speech pathologists
Duration	6-weeks	6-weeks	13-weeks (delivered over 18-weeks due to a 5-week gap midway with no instruction)
Intensity	3 x 60-min sessions per week	3 x 60-min sessions per week	2 x 60-min sessions per week
Total hours of instruction	17 hours	16 to 18 hours	26 hours
Instruction mode	TP	TP	Face-to-face (7-weeks) then via TP (6-weeks)
Instruction delivery	1:1	1:1	Small group (7-weeks) then 1:1 (6-weeks)
Changes to instruction delivery	No	No	Yes
Unexpected interruption in instruction sessions	No	No	Yes (5-weeks of no instruction between week 7 and week 8)
Analyses	Basic descriptive statistics, visual analysis, and qualitative analysis	Statistical analysis (one-way repeated measures ANOVA)	Statistical analysis (two-way mixed ANOVA)
Study outcomes[e]:			
Word reading accuracy			
Measure	WRAT-4 (Word Reading subtest)	WRAT-4 (Word Reading subtest)	WRAT-5 (Word Reading subtest)
Result	Gains of 2 LSC and 2 words read	(η_p^2 = .69)**	(η_p^2 = .11)

	Empirical study 1: CP	**Empirical study 2: Down syndrome**	**Empirical study 3: Autism**
Nonword reading accuracy [f]			
Measure	CC-2	CC-2	WJ-IV (Word Attack subtest)
Result	Gain of 18 graphemes correct	(η_p^2 = .83)**	(η_p^2 = .23)**
Passage-level reading accuracy			
Measure	NARA-3	NARA-3	NARA-3
Result	Score of 0	(η_p^2 = .69)**	(η_p^2 = .15)*
Reading comprehension			
Measure	NARA-3	NARA-3	NARA-3
Result	Score of 0	(η_p^2 = .39)	(η_p^2 = .06)
Secondary measure of reading comprehension			
Measure	TERC	TERC	YARC
Result	Score of 0	(η_p^2 = .70)**	(η_p^2 = .01)
Parent-led SBR	All target parent-led SBR sessions completed.	All target parent-led SBR sessions completed for all but one participant.	None

Note. ** = statistically significant result at conservative alpha of $p < .01$; * = statistically significant result at $p < .05$. ADD = attention-deficit disorder; ADHD = attention-deficit/hyperactivity disorder; CC-2 = Castles and Coltheart Test – 2nd edition; LSC = letter-sound correspondence; NARA-3 = Neale Analysis of Reading Ability – 3rd Edition; PR = percentile rank; SBR = shared book reading; SES = Socioeconomic Status; TERC = Test of Everyday Reading Comprehension; TP = telepractice; WJ-IV = Woodcock-Johnson Tests of Achievement – 4th Edition; WRAT-4 = Wide Range

Achievement Test – 4th Edition; WRAT-5 = Wide Range Achievement Test – 5th Edition; YARC = York Assessment of Reading for Comprehension – Australian Edition.

[a] IQ based on 'Raven's 2 Progressive Matrices Clinical Edition digital short form' in Chapter 3 and 4 studies and 'Test of Nonverbal Intelligence – 4th Edition' in Chapter 5 study.

[b] Adaptive ability based on 'Adaptive Behavior Composite score' from Vineland Adaptive Behavior Scale – 2nd edition in Chapter 3 and 4 studies and Vineland Adaptive Behavior Scale – 3rd edition in Chapter 5 study.

[c] Word reading accuracy based on Word Reading subtest from WRAT-4 or WRAT-5. Reading comprehension based on Reading Comprehension Composite score from the NARA-3.

[d] Based on Australian Bureau of Statistics (2021) census of population and housing socioeconomic indexes for areas.

[e] Effect sizes were interpreted as: η_p^2 of .01 corresponded to a small effect size, .06 a medium effect size, and .14 a large effect size (Richardson, 2011).

[f] Nonword reading accuracy combined with real-word reading accuracy in CP and Down syndrome studies.

As is clear in Table 6.1, the empirical studies in this thesis differed in several important ways related to participant characteristics, study characteristics, and study outcomes. Notably, the sample size of the study involving children with autism was substantially larger than the other two studies, but this study was markedly disrupted by the COVID-19 pandemic. In terms of participant characteristics, key differences related to the lower socioeconomic status of the participants with autism, the number of autistic participants with dual diagnoses, and the lower nonverbal intelligence scores for the participants with Down syndrome, by comparison with the other groups. There was a similar average age of participants across the studies, though the autism study included younger participants than the other two studies. Despite this, the average baseline word reading accuracy and reading comprehension skills for the children with autism was substantially higher than for the child with CP or children with Down syndrome. Key differences in study characteristics related to the different study design used in each chapter, differences in instruction intensity and duration, inclusion of parent-led shared book reading (SBR) in some studies, and differences in mode and method of instruction delivery. Due to the numerous differences across the studies, study results are not directly comparable. Important similarities and differences in study outcomes are discussed in the coming sections to highlight key findings for children with diverse developmental disabilities.

6.2.1 Reading Accuracy

6.2.1.1 Word-level Reading Accuracy. Children with CP, Down syndrome, or autism made modest to large gains in skills that support word reading accuracy, specifically phonics skills (letter-sound knowledge and decoding skills) following participation in ABRA literacy instruction (plus clinician- and/or parent-led SBR for most children). These gains were statistically significant on both measures of word-level reading accuracy for children with Down syndrome, and statistically significant on only the measure of nonword reading accuracy for children with autism. All outcome measures were independent of the ABRA and SBR program, indicating that most children were able to generalise their decoding skills. These findings align with previous studies which have found that children with Down syndrome or autism can demonstrate gains in word reading accuracy on standardised assessment measures (e.g., Bailey et al., 2017; Cologon et al., 2011; Grindle et al., 2013; Lim et al., 2019). Very few literacy instruction studies involving children with CP have used standardised assessments to measure reading outcomes, with most studies utilising researcher designed measures within single-subject research designs (as outlined in the systematic review in Chapter 2). The very modest gain made by the participant with CP on the independent measure of word reading accuracy in our case study may be meaningful in this context.

A significant factor in the word-level reading accuracy outcomes for the participant with CP was a motor speech impairment. Motor speech impairments, specifically childhood apraxia of speech (CAS), are common in children with developmental disabilities (Shriberg et al., 2019), particularly children with CP (Mei, Reilly, et al., 2020) or Down syndrome (Wilson et al., 2019), but also children with autism (some studies suggest up to 64% of autistic children with communication delays have CAS, Tierney et al., 2015; whilst others suggest no increased prevalence, Shriberg et al., 2011). All of the children with Down syndrome in our study had articulation or phonological impairments, though none presented with a diagnosed motor speech impairment. We did not collect a measure of speech sound accuracy for the autistic children in this research; however, in the initial demographic survey, 42% of parents reported that their child had some difficulty with production of speech sounds. No parents reported a motor speech diagnosis for their child with autism. Given the confounding impact of motor speech impairments on literacy outcomes (Ferreira et al., 2007; Gillon & Moriarty, 2007; Miller et al., 2019; Stein et al., 2020; Zaretsky et al., 2010) and the high incidence of these impairments amongst children with developmental disabilities (Mei, Reilly, et al., 2020; Shriberg et al., 2019; Tierney et al., 2015; Wilson et al., 2019), this is a factor that deserves consideration and exploration in future reading research involving children with developmental disabilities.

6.2.1.2 Passage-level Reading Accuracy. Gains in passage-level reading accuracy were inconsistent across the studies. Our participant with CP did not have the skills necessary to participate in passage-level reading assessment. The children with Down syndrome made statistically significant gains in their passage-level reading accuracy skills with a large effect size (η_p^2 = .69). The autistic children made gains in passage-level reading accuracy that were not statistically significant at our conservative alpha level, though were consistent with a large effect size (η_p^2 = .15). As previously discussed, this effect size is similar to those reported in previous studies of ABRA and children with autism (e.g., Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022), and may be clinically relevant in this context.

Previous literacy instruction research involving children with Down syndrome has used word-level reading to measure reading accuracy (including real and nonwords), rather than passage-level reading measures (e.g., Baylis & Snowling, 2012; Burgoyne et al., 2013; Cologon et al., 2011; Cupples & Iacono, 2002; Felix et al., 2017; Goetz et al., 2008; Lemons et al., 2018; Lim et al., 2019; Nakeva von Mentzer et al., 2021). We are not aware of any literacy instruction studies that have utilised a standardised measure of passage-level reading accuracy with children with Down syndrome, giving little to compare our results to. Lim et al. (2018) used a non-standardised measure of passage-level reading accuracy within SBR before and after literacy instruction. Other studies have included measures of oral reading fluency, though not specific passage-level reading accuracy (e.g., Allor, Mathes, Roberts, Cheatham, et al., 2010; King, Rodgers, et al., 2022; Lemons et al., 2012). In a descriptive study involving 25 children with Down syndrome aged 5 to 13 years, van Bysterveldt and Gillon (2014) utilised the same passage-level reading accuracy measure as was used in our studies (Neale Analysis of Reading Ability - 3rd edition [NARA-3]; Neale, 1999). The children in the van Bysterveldt and Gillon (2014) study achieved similar raw scores on average to the children in our Down syndrome sample at baseline (average raw score of 14.3 [*SD* = 26.98] for children in the Van Bysterveldt and Gillon [2014] study and average raw scores of 16.50 [*SD* = 6.35] and 15.83 [*SD* = 9.54] at baseline and pre-instruction respectively for the children in our study). Following instruction, the mean raw score for passage-level reading accuracy increased to 26.50 (*SD* = 9.55) on the NARA-3 for the children in our study. This gain demonstrates the significant impact of the hybrid ABRA and parent-led SBR intervention on passage-level reading skills for our participants with Down syndrome.

6.2.2 Reading Comprehension

Limited reading comprehension gains were made across the studies in this thesis. The child with CP did not have sufficient word reading skills to participate in our reading comprehension measure. Nonetheless, this child made practical gains in her ability to respond to more complex questions in SBR. The children with Down syndrome did not make statistically significant gains in their conventional passage-level reading comprehension skills, though made gains in their functional reading comprehension skills which were inconsistently significant across statistical tests. The children with autism made gains in their passage-level reading comprehension skills that were not statistically significant, though were associated with a medium effect size. A medium effect size may indicate a clinically significant effect given that reading comprehension is a recognised area of need for children with autism (Abelson, 1995; Betensky, 2019; Brown et al., 2013; Kyle et al., 2013; Nation et al., 2006; Wasserstein & Lazar, 2016; Wasserstein et al., 2019; Wei et al., 2015).

There are many possible reasons why we did not see statistically significant gains in passage-level reading comprehension across the studies, many of which have been outlined in Chapters 3 to 5. The short timeframe of the study involving children with Down syndrome (6-weeks) was likely insufficient to allow children to integrate and make meaningful changes to reading comprehension skills. Children with developmental disabilities, particularly those with intellectual impairment, may require longer periods of time to process and consolidate new skills, including reading comprehension (Boudreau, 2002; Byrne et al., 2002; Miller et al., 2001). Methodological changes in the study involving autistic children almost certainly impacted reading comprehension outcomes. In addition, both studies which permitted statistical analyses were likely underpowered for different reasons, potentially impacting patterns of statistical significance for reading comprehension. The study involving children with Down syndrome had a modest sample size due to piloting of a novel intervention. The study involving autistic children was underpowered due to participant attrition likely due to being conducted during the pandemic in an area of sociodemographic inequity.

The nature of reading comprehension assessments may also have impacted outcomes. There is an extensive research base which suggests that reading comprehension is a complex skill that is difficult to measure with a single test (see Castles et al., 2018; Keenan & Meenan, 2014; Nation & Snowling, 1997; Perfetti & Stafura, 2014). Factors such as test format and length of text passages can impact performance on reading comprehension measures, as can the specific component skills that are being measured by the test (Keenan & Meenan, 2014; Nation & Snowling, 1997). The primary measure of conventional passage-level reading comprehension used in this thesis (NARA-3; Neale, 1999) has been shown to underestimate reading comprehension skills in children with poorer decoding skills or children who have difficulty responding to open-ended questions (Spooner et al., 2004). Despite this, the NARA-3 is widely regarded as a valid measure of reading accuracy and comprehension and has been used in several important studies involving children with developmental disabilities (e.g., Bailey et al., 2017; Nally et al., 2021; Nation et al., 2006; van Bysterveldt & Gillon, 2014). Many of the children across our sample had poor decoding skills and expressive language difficulties and these factors may have impacted their performance on the NARA-3. A non-oral measure of reading comprehension involving multiple choice responses may have returned different results.

6.2.3 Impact of the Pandemic

The COVID-19 pandemic resulted in an unprecedented global disruption to education, with more than 1.5 billion children worldwide affected by school closures (United Nations, 2020). Research globally was substantially impacted, with many researchers forced to alter or cease research projects (e.g., Myers et al., 2021; Yi & Dixon, 2021). This section first explores the impact of the pandemic and school closures on children's academic achievement generally and then the impact of these disruptions to children with developmental disabilities. This section ends with a discussion of the impact of the pandemic on the studies in this thesis.

6.2.3.1 Impact on Children's Learning Progress. Since the World Health Organisation declared the global pandemic in March 2020, numerous studies have attempted to assess the impact of school closures on children's learning globally. A recent meta-analysis by Betthäuser et al. (2023) found 42 studies from across 15 countries that explored children's learning progress over the first 2.5 years of the pandemic. The analysis indicated that, over the pandemic, children's learning progress slowed considerably, equating to a learning loss worth approximately 35% of a regular school year. Learning deficits occurred early in the pandemic and, although they persisted over time, these learning deficits did not increase. It is possible that as children, parents, and educators became more proficient at remote learning, they were able to prevent the occurrence of further learning loss. Learning deficits were larger for mathematics than for reading and did not differ significantly across grade levels. Existing educational inequalities were exacerbated, with children from disadvantaged socioeconomic backgrounds experiencing greater learning loss. High-income countries accounted for most studies in the meta-analysis, with few middle-income countries contributing research and no studies from low-income countries. The learning loss globally may have be even greater if evidence from low-resource countries were included (United Nations, 2020).

Few Australian studies have investigated learning progress throughout the pandemic. A study by Gore et al. (2021) was included in the meta-analysis by Betthäuser et al. (2023), and was one of the only studies to report learning progress for children throughout the early stages of the pandemic, in both mathematics and reading. A follow on study by Miller et al. (2024) fell outside of the search dates for the meta-analysis, though similarly reported no learning loss in reading or mathematics for Year 3 and 4 children in NSW during the second year of the pandemic (when compared with a matched cohort of students from 2019). Surprisingly, this study found that children from disadvantaged schools actually made an additional 3-months' gains in mathematics in 2021, compared with the 2019 cohort. Potentially, additional funding and resources provided to schools by the NSW government in 2021 (e.g., funding for targeted small-group teaching; NSW Government, 2020; a library of evidence-based lesson plans made available to teachers; NSW Government, 2021) helped to facilitate the learning gains found by Miller et al. (2024). Results from the annual National Assessment Program – Literacy and Numeracy (NAPLAN) in Australia similarly showed no decline in children's literacy or numeracy scores in 2021 when compared with the 2019 results (Australian Curriculum Assessment and Reporting Authority [ACARA], 2021). Only one Australian study reported some learning loss in students' reading skills (NSW Department of Education, 2020). This study found that in NSW from August to October 2020, Year 3 students were approximately 3 to 4 months behind in reading and Year 5 students were 2 to 3 months behind in reading and mathematics.

6.2.3.2 Impact on Children with Developmental Disabilities. Many studies found that children with disabilities were disproportionately impacted by the pandemic when compared to their peers with typical development (Dickinson et al., 2020; Mann et al., 2021; Marella et al., 2022). In addition to disrupted schooling, many of these children experienced disruptions to their usual therapy services. For example, in a survey of 3,502 caregivers for individuals with autism, 88% reported disruptions to speech pathology intervention at the beginning of the pandemic, yet only 50% reported accessing remote therapy services (White et al., 2021). These service disruptions resulted in deterioration in the behaviour and emotional wellbeing of some children with developmental disabilities (Zhang et al., 2022). Along with disruptions to schooling and therapy services, many children with disabilities also experienced disruptions to medical care, reduced access to assistive technologies, poorer sleep and eating habits, and decreased physical activity throughout the pandemic (Mann et al., 2021). Changes to usual schooling, clinical, and care supports, meant that families took on an even greater role in supporting their child with a disability, placing substantial strain on caregivers throughout the pandemic (Mann et al., 2021; Marella et al., 2022).

An Australian-based longitudinal study sheds further light on the lived experiences of children with disabilities and their families throughout the pandemic (Marella et al., 2022). This study highlighted key issues throughout the pandemic for children with disabilities, including: (a) communication barriers between parents and schools; (b) insufficient accommodations and supports for individual learning needs; (c) varied use and accessibility of digital technologies for remote learning; and (d) limited access to usual supports. An additional barrier faced was that "educators' low expectations of the capabilities of students with disability to engage with technology stymied opportunities for online remote learning" (p. 4). Many families also noted regression with children's academic and social skills and reduced motivation for learning during remote schooling. However, positives of learning from home were reported by some families. For example, for children with physical disabilities or impaired executive functioning "learning from home reduced the physical and psychological fatigue associated with getting ready for school" (p. 55).

Despite the significant impact of school closures and disrupted services on children with developmental disabilities, no empirical studies have explored learning progress for children with disabilities over the pandemic. This is in stark contrast to the numerous studies that have explored learning outcomes for cohorts of children with typical development (e.g., Betthäuser et al., 2023; NSW Department of Education, 2020). Studies that explore learning outcomes for children with developmental disabilities throughout the pandemic are needed in order to help prevent such disparities in the future. Such studies could assist in the development of interventions to support children with disabilities should emergency remote learning occur again (Mann et al., 2021).

6.2.3.3 Impact on our Studies. All three empirical studies in this thesis were conducted during the COVID-19 pandemic, meaning that additional external factors at the time likely impacted children's performance and outcomes. The studies involving the child with CP or children with

Down syndrome were conducted early on during the pandemic when telepractice was still relatively novel for children. This may have influenced children's engagement in sessions. In addition, participants in these studies were from different states across Australia, meaning they were subjected to different degrees and lengths of school closures based on their state's jurisdictions. This was not the case for the study involving autistic children, where all children were from the same state and geographic location and experienced the same strict pandemic induced restrictions. Australia's state government stay-at-home orders were stricter than many other countries and compliance was remarkably high (Chang et al., 2020). Additional, harsher, restrictions were in place for certain local government areas during the lockdowns, including the area of relative socioeconomic disadvantage where our study involving children with autism took place (Australian Bureau of Statistics, 2021).

Although we attempted to recruit a diverse sample of participants for the studies involving children with CP or Down syndrome, participants were primarily based in metropolitan areas and were from well-resourced families with a relatively good understanding of their child's support needs. These families volunteered to participate in a research study delivered via telepractice, indicating they had capacity to engage in a program delivered remotely. This is potentially why there was a high level of engagement from these families. In contrast, the families in the study involving autistic children signed up for a face-to-face clinic or school-based intervention study and were thrust into an online telepractice environment. As the autism study took place in an area of relative socioeconomic disadvantage, some participants were unable to continue in the study due to limited resources at home (eight participating families chose to withdraw from the study for various reasons following the transition to telepractice). For the families who were able to continue participating, one potential positive was that these families developed a sense of empowerment and confidence in using technology for learning. This was flagged as an area of need for families of children with disabilities during the pandemic (Marella et al., 2022).

Of the studies in this thesis, the study involving children with autism was most substantially impacted by the pandemic. This was due to the strict government mandated stay-at-home orders issued halfway through the study. These stay-at-home orders changed our study design midway, including significant changes to key instruction characteristics (from face-to-face to telepractice delivery of instruction and assessments and from group-based to one-to-one instruction). There was an unexpected 5-week gap where our participants received no literacy instruction from the research team while we waited to establish how long the stay-at-home orders would be in place. This gap in instruction is unprecedented in previous studies of ABRA and children with autism. It is unclear to what extent additional external factors at this time, such as the quality of remote learning and funding schemes put in place by the NSW government (as described in Section 6.2.3.1), impacted the children in our sample. Studies have reported that some autistic children had an increase in restrictive and repetitive behaviours and poorer self-regulation and cooperation skills during the pandemic lockdowns (Di Renzo et al., 2020; Morris et al., 2021; White et al., 2021). These behavioural disturbances were likely a result of changes to daily routines, lack of peer interaction, and unpredictable demands of the pandemic (Lee et al., 2021; Pellicano et al., 2022; White et al., 2021). The additional stress placed on children and families throughout the second half of our study cannot be underestimated and may have impacted some participants' ability to engage in, and benefit from, the online literacy instruction.

6.3 Instruction Methods

6.3.1 ABRACADABRA (ABRA)

Previous research has primarily explored ABRA with typically developing children (e.g., Abrami et al., 2020) and some studies have explored ABRA with autistic children (Arciuli & Bailey, 2019; Bailey et al., 2017; Bailey et al., 2022). The research presented in Chapters 3 and 4 of this thesis are the first to explore ABRA with children with other developmental disabilities, specifically children with Down syndrome and a child with CP.

6.3.1.1 ABRA and Reading Accuracy Skills. Our studies found that ABRA was effective at increasing the word- and passage-level reading accuracy skills of children with Down syndrome, with modest gains in letter-sound knowledge and decoding skills for a child with CP and children with autism. These findings align with previous research which has demonstrated that ABRA has robust effects on phonics skills across a range of instruction contexts and study designs (see the meta-analysis by Abrami et al., 2020). Yet, the recent large-scale effectiveness trial of computerised versus paper-based ABRA instruction in Year 1 mainstream classrooms by Bell et al. (2022) demonstrated some contrasting findings. This study involved 3,462 Year 1 students across 154 schools in the UK over the 2018 to 2019 school year. In this study, children who received the computerised version of ABRA made no additional progress in reading when compared to a business-as-usual control group (meanwhile, children who received the paper-based version of ABRA made an average of 2-months' additional reading progress). However, several factors related to how ABRA was delivered in this study may have contributed to these findings. Data from the Implementation and Process Evaluation (IPE) indicated that 79% of instructors delivering the computerised version of ABRA experienced technology issues, largely due to poor internet connections. This resulted in fewer instructors in the computerised ABRA version delivering the intended number of sessions, and likely impacted dosage and engagement within sessions. Additionally, more instructors delivering the paper-based version of ABRA reported adapting the protocol to cater to students' individual needs and provided additional time for struggling students. In contrast, instructors delivering the computerised ABRA were less likely to amend the programme. This may have contributed to the greater reading outcomes made by the paper-based ABRA group. Nevertheless, the findings of Bell et al. (2022) are noteworthy, given that we hypothesised ABRA would be beneficial for children with developmental disabilities due to factors specific to the computerised version (e.g., multimedia presentation of information, specific and immediate feedback, game-based). Future studies could explore differences in outcomes for a paper-based versus computerised version of ABRA for children with developmental disabilities.

The level of adult-child interaction in our studies may have impacted outcomes. Previous research has indicated that higher levels of adult-child interaction within computer-assisted instruction (CAI) leads to more positive reading outcomes (Abrami et al., 2020; Cheung & Slavin, 2013; McTigue et al., 2020; McTigue & Uppstad, 2019). A speech pathologist delivered literacy instruction to children across all the studies in this thesis, and there was a high level of interaction between the child and clinician within sessions. For most sessions via telepractice, children's parents were also present and engaged, providing a high level of adult feedback and interaction for children alongside the computer-assisted ABRA instruction and clinician online. Parents of children with CP or Down syndrome tended to be more involved in telepractice instruction sessions, compared with parents of autistic children, most likely due to their children needing more support with the physical skills required to access ABRA. This high level of parental involvement was likely a significant factor in successful delivery of ABRA to a child with CP and children with Down syndrome. Children in the autism study did not have parental involvement during the small-group face-to-face sessions at the clinic or school, but they experienced clinician and peer involvement in these sessions.

6.3.1.2 ABRA and Comprehension Related Skills. ABRA instruction did not result in significant gains in conventional passage-level reading comprehension skills for any of the children in our studies. Only the children with Down syndrome demonstrated statistically significant gains in their functional reading comprehension skills. ABRA targets the Big Five, though contains more activities focused on reading accuracy than reading comprehension skills (i.e., there are 17 activities targeting phonemic awareness or phonics skills and six targeting reading fluency, but only two targeting vocabulary and six targeting reading comprehension skills). All the activities link to different stories within the program to provide multiple adaptations of the same activity, including reading comprehension activities (e.g., the 'comprehension monitoring' activity can be practised within seven different stories). In addition, each of the 35 stories in ABRA provides exposure to rich vocabulary and narrative structure. However, ABRA activities do not explicitly target oral language skills. Research indicates that oral language comprehension is the biggest predictor of reading comprehension for children with CP, Down syndrome, or autism, more so than word recognition skills (Asbell et al., 2010; Dorman, 1987; Lucas & Norbury, 2014; Norbury & Nation, 2011; Roch et al., 2021; Roch & Levorato, 2009). Studies have also indicated that explicit teaching of language structures can be more effective than implicit teaching only for children with language delays (e.g., Calder et al., 2018; Finestack, 2018).

For children with documented oral language difficulties, such as the children in our samples, the activities in ABRA were potentially not sufficient to develop children's oral language skills and effect change on their reading comprehension. For example, of the two vocabulary activities in ABRA, one targets relatively basic vocabulary knowledge through word to picture matching (targeted at children learning English as an additional language) and the other develops background knowledge of vocabulary prior to reading a story. This second activity provides children with two meanings of a target word and then asks children to choose which sentence, from a choice of two, uses the word correctly. This activity places high demands on children's working memory to recall decontextualised word meanings and navigate through multiple steps in the activity, factors that are difficult for children with developmental disabilities and intellectual impairment. Potentially, ABRA could be enhanced to support children's oral language skills by expanding the suite of vocabulary activities. Additional activities could teach vocabulary within more contextualised situations and target depth of vocabulary understanding and use. Activities directly targeting oral language skills, such as morphology and syntax, could also be incorporated into the program. Currently many of the phonemic awareness and phonics activities in ABRA have multiple difficulty levels to cater to children with different abilities, yet activities related to reading comprehension skills do not provide such differentiation. Customisable difficulty levels could be included within the existing ABRA vocabulary and reading comprehension activities to make them more suitable for children with varied language abilities (e.g., by using simpler question types, providing multiple choice options, or using visual supports to scaffold children's oral language responses). In addition, children with documented oral language difficulties likely need ABRA to be supplemented with explicit language intervention to further develop their oral language skills and support reading comprehension.

6.3.1.3 Useability of ABRA for Children with Developmental Disabilities. Children with CP, Down syndrome, or autism generally engaged well with the ABRA program, though reading outcomes differed substantially. The modest to large gains in phonics skills across the three studies suggest that the program may have some value for children with developmental disabilities, though further research is warranted. ABRA is highly cost-effective and can provide equitable access to evidence-based literacy instruction for children with developmental disabilities. For example, McNally et al. (2016) found that the cost of the program delivered within mainstream schools in the UK per student over three years was £8.52 (GBP). This estimate was based on ABRA delivery within a mainstream school environment, delivered by a teaching assistant to small groups of children. ABRA delivered in a one-to-one context as it was in most of the studies in this thesis would be more resource intensive. ABRA delivered by a clinician would have an even higher cost estimate. However, as the program is designed to be used flexibly to meet an individual's needs, ABRA represents a time efficient and cost-effective resource for clinicians to use within intervention sessions. There are also the benefits of ABRA for children with developmental disabilities that are broadly associated with any CAI program, such as: (a) appealing graphics and simple game-like activities to increase engagement and motivation with learning tasks; (b) interactive interface which requires active engagement from children; (c) multimedia supports including visually cued instructions and auditory prompts; (d) high levels of repetition within activities; (e) immediate, specific, and structured feedback; and (f) individualised content and pace of learning activities.

6.3.1.3.1 Inclusive Design. Universal Design for Learning (UDL) is a set of principles for designing inclusive instructional materials that improve the learning process by meeting the diverse needs of students (Capp, 2017). The ABRA program partially aligns with the principles of UDL. Figure 6.1 describes the three primary principles of UDL and provides examples of how ABRA aligns with each of these principles. In light of the principles of UDL, it is interesting to consider the findings of Bell et al. (2022), where children who received paper-based ABRA outperformed children who received the computerised ABRA program. Most of the UDL features which make ABRA suitable and accessible to children with developmental disabilities would disappear if a paper-based version was applied with this group.

Figure 6.1 *Ways ABRA Aligns with the Principles of UDL* (based on Capp, 2017; Courey et al., 2013)

Principle 1
presenting information to learners using multiple formats

- animated computer modelling of target reading skills and activities;
- independent access to 'help' within activities which provides visually cued instructions (including audio and visual prompts);
- opportunities for repeated practice of target skills across word- and passage-level activities;
- options for texts to be read aloud by the computer.

Principle 2
providing a variety of ways for learners to express their knowledge

- interactive interface which provides alternate response options in some activities, e.g., choice of onscreen pictures/letters/ keyboard;
- option of mouse, touchscreen, or keyboard access to some tasks;
- structured feedback and graded prompts within activities allowing for varied levels of support.

Principle 3
stimulating learners' motivation and engagement in different ways

- engaging graphics;
- game-like and novel activities;
- overarching themes and characters throughout the program;
- customisable activity difficulty level;
- opportunities for autonomy and independence with activity completion, including self-regulated and self-paced learning.

Despite aligning with some of the principles of UDL, many of the ABRA activities are still geared towards verbal responses (e.g., answering questions within the texts [prediction activity], verbally producing a word after blending the phonemes [blending train activity]). These activities were difficult for some children with poor expressive language skills and lower intellectual abilities to access. These children required a high level of adult support and scaffolding to complete such activities (e.g., the clinician verbally offered children a choice of two responses to prediction questions). In addition, ABRA provides repeated audio prompts within activities until a child completes the required task. These prompts often did not provide adequate processing time for children with developmental disabilities to think about and then execute the task. As such, these prompts sometimes interrupted the child while they were attempting to complete the task or were receiving support from the clinician via telepractice. We also found that not all features of the ABRA program were physically accessible to children with CP or Down syndrome (e.g., clicking and dragging items, selecting small letters on the screen).

6.3.1.3.2 Can Children with Complex Communication Needs Access ABRA? In theory, children with complex communication needs who have access to augmentative and alternative communication (AAC) could engage with some ABRA activities. For example, children could use their speech generating AAC device to say the word they heard within auditory blending activities or to respond to prediction and summarisation questions within passage-level texts. However, many of the ABRA reading fluency activities would not be accessible to nonspeaking children without modification. This is an issue inherent with the definition of reading fluency, which is “the ability to read a text quickly, accurately, and with proper expression” (NICHD, 2000, p. 3-5). In practice, some of the reading fluency activities could be modified so that nonspeaking children could participate. For example, children who use AAC could still identify whether a page was read by the computer with the appropriate expression or not, though would be unable to complete the second part of the reading expression activity (reading the page aloud themselves using the proper expression). Children with severe speech and physical impairments who use eye-gaze technology could directly complete some of the activities in ABRA via eye-gaze computer access. Given the additional cognitive load involved, and to prevent fatigue, children who use AAC may require shorter instruction sessions of increased frequency to ensure a similar dosage of instruction. Future research could explore the feasibility of ABRA and other computer-based literacy programs with children who use AAC.

6.3.1.4 Summary. In summary, ABRA is an empirically supported and highly cost-effective tool for literacy instruction which shows some benefits for developing the reading skills of children with developmental disabilities. Although not all features of the program are well-matched for this group, we are not aware of any other freely available and evidence-based comprehensive literacy programs that offer comparable support, accessibility, and potential value for children with developmental disabilities. The program offers educators and clinicians a 'ready to use' tool for literacy instruction and intervention that can be implemented following minimal training. However, the program, or potentially how the program was used in our studies, does not seem as effective at developing reading comprehension skills as reading accuracy skills for children with developmental disabilities. Other programs, or a combination of programs, may be better suited to the needs of children with developmental disabilities and should be explored in future research.

6.3.2 Shared Book Reading

Several aspects of SBR were explored throughout this research. Clinician-led SBR is discussed first in this section, followed by parent-led SBR.

6.3.2.1 Clinician-led SBR. Clinician-led SBR was implemented in a similar way across the three studies in this thesis and was used as a non-computerised extension task to support children to generalise the skills targeted within ABRA. Slightly different procedures were used to optimise SBR in a small-group setting in the first half of the study involving autistic children. SBR methods that utilise routine procedures may be particularly suited to children with developmental disabilities as they provide a predictable context for learning that can support children's engagement (Akemoglu et al., 2020).The clinician-led SBR activities followed the same structure and utilised the same resources each session in our studies (some resources differed across studies, though were consistent within each individual study). Potentially this factor helped to facilitate children's engagement in the clinician-led SBR activities. Though, in the study involving autistic children, the SBR fidelity item "*Child/ren's behaviour doesn't interfere with SBR activity*" had the lowest mean fidelity rating, indicating that these procedures were not completely effective at engaging children (this fidelity item was still relatively highly rated, with a mean of 4.37 out of 5). Elements essential for effective SBR, including incorporating decoding and reading comprehension strategies and facilitating discussion and interaction with children during SBR had high fidelity ratings, indicating that the clinician-led SBR was implemented efficaciously. This same item regarding children's behaviour also had the lowest mean fidelity rating for the computerised ABRA activities which are designed to be motivating and engaging for children. Potentially embedding children's specific interests within these activities may have increased engagement and compliance for autistic children (Ninci et al., 2020; Solis et al., 2016).

Only a small number of children in this research (a group of the autistic children in Chapter 5) did not participate in any clinician-led SBR. These children participated in ABRA-only instruction and made statistically significant gains in their nonword decoding skills. Yet, their overall gains in nonword decoding and the associated effect size were smaller than for the group of children who participated in ABRA plus clinician-led SBR. It is unclear which aspects of the program facilitated the additional gains made by the children who participated in clinician-led SBR alongside ABRA. These gains may have been a result of the extra dosage of instruction (a further 15-minutes per session during the SBR activity) or a possible benefit of the extra decoding opportunities these children had within the SBR decodable texts. Future research could separate these variables to determine the value-added effects of supplementary decodable texts versus time in instruction.

In the study involving autistic children, there was a positive correlation between socialisation scores and reading comprehension gains for children who received supplementary clinician-led SBR within literacy instruction sessions. There was no correlation between socialisation scores and reading comprehension gains for the children who did not participate in SBR (i.e., children who received ABRA only). An association between social communication and reading comprehension skills for children with autism has been found in many studies across the literature (e.g., McIntyre, Solari, Grimm, et al., 2017; Norbury & Nation, 2011; Ricketts et al., 2013;

Tong et al., 2020). Yet, these studies have primarily been descriptive, with few exploring the relationship between social communication skills and reading comprehension gains within intervention studies. In their study of group-based ABRA instruction for autistic children, Arciuli and Bailey (2019) found that gains in reading comprehension were associated with socialisation skills. All of the children in the instruction group in Arciuli and Bailey (2019) participated in teacher-led SBR within instruction sessions. The study involving autistic children in this thesis is unique in separating out the 'ABRA' and 'ABRA plus SBR' instruction contexts. The correlation between reading comprehension gains and socialisation scores for only the children who participated in clinician-led SBR may be due to the high levels of social-interaction involved in this activity (i.e., a high level of interaction and discussion between the clinician and child regarding texts and, for the first half of the study, engaging in discussion with other child group members). This is in comparison to the children who participated in the computerised ABRA activities only (and no SBR), where, despite some level of peer and adult-child interaction, task instructions were primarily delivered via the computer. Potentially, instruction in only the computerised ABRA context did not place as many demands on children's social communication skills as the clinician-led SBR. Future research should seek to explore the association between computerised and non-computerised reading comprehension instruction and socialisation skills for children with autism.

6.3.2.2 Parent-led SBR. The two studies in this thesis that utilised parent-led SBR had high levels of parent engagement (the studies involving a child with CP or children with Down syndrome). All but one of the seven families involved in these studies completed the target number of home SBR activities. In their study involving 21 autistic children and their parents, Bailey et al. (2022) used similar parent-led SBR methods to the studies in this thesis. However, Bailey et al. (2022) found much lower levels of engagement with home-based SBR activities. The high levels of parental engagement in the studies in this thesis may have been due to the children's type of disability (CP or Down syndrome). Studies suggest that parents of children with Down syndrome experience lower levels of stress compared with parents of children with other developmental disabilities, particularly parents of children with autism (e.g., Cuzzocrea et al., 2016; Smith et al., 2014; Stoneman, 2007). Similarly, parents of children with CP have reported lower parental stress than parents of autistic children (Valicenti-McDermott et al., 2015). It is possible that the parents in our CP and Down syndrome studies had lower levels of parental stress which facilitated their capacity to engage in the home SBR activities. We did not include parent-led SBR methods in the study involving autistic children in this thesis, though noted that parents tended to be less involved in sessions via telepractice than the parents of children with CP or Down syndrome.

Previous research on parent-led SBR has largely focused on using this method to facilitate children's communication and language skills (e.g., Boyle et al., 2019; Noble et al., 2019; Towson et al., 2021). Few studies have explored how to upskill parents of children with developmental disabilities to support their child's reading accuracy or print-based skills within a SBR context (Biggs et al., 2023). Our studies found that parents of children with CP or Down syndrome responded positively to SBR coaching and were able to effectively implement strategies to support their child's decoding and text comprehension skills. The regular coaching and support from a clinician was likely key to successful implementation of the home-based SBR in the two studies (Novak & Berry, 2014). This collaborative implementation of literacy instruction across contexts and people has important implications for increasing reading instruction dosage for children and facilitating skill generalisation.

6.3.2.2.1 Limitations and Future Research Directions for Parent-led SBR. Although there were many positive outcomes associated with the parent-led SBR program, several aspects could be modified to potentially enhance this program in future studies. Firstly, we had no direct measure of quality or fidelity of SBR delivered by parents, only what parents self-reported in their SBR logs. Parents were provided with training in target SBR strategies; yet we cannot be sure of how parents implemented the procedures on their own with their child. Secondly, almost all parents reported engaging in SBR with their child daily prior to participating in the study. Future research could record parents' SBR behaviours before and after participating in a SBR program to provide a measure of change in parents' use of reading accuracy and reading comprehension-related supports. Finally, the levels of comprehension questions taught to parents in the SBR program were developed as a simplified version of well-established question levels (Blank's levels of questions; Blank et al., 1978; see Westby, 2017). These simplified question levels were used as they required minimal teaching and support for parents. However, much of the research around SBR for developing children's oral language comprehension skills has used dialogic reading (e.g., Mol et al., 2008; Pierson et al., 2021; Pillinger & Vardy, 2022; What Works Clearinghouse, 2010). Dialogic reading is a method of SBR that promotes active participation and dialogue between an adult and child using standardised procedures and specific prompts (Whitehurst et al., 1988). Dialogic reading has been successfully used in studies involving children with autism or Down syndrome (e.g., Fleury et al., 2014; Pierson et al., 2021; Whalon et al., 2015). Including dialogic reading strategies within SBR would require increased time for parent training and support, though may be beneficial in enhancing language and text comprehension skills for children with developmental disabilities. Dialogic reading has not been explored in SBR for older children with developmental disabilities who are able to read parts of the text themselves. Future research could explore such methods within parent- or clinician-led SBR programs.

A further limitation worth considering in future studies of parent-led SBR is the potential confounding factor of the gene-environment correlation. The gene-environment correlation refers to how a person's genotype influences their exposure to environments (i.e., genotypes shape an individual's personality, behaviour, and cognition which then influences how an individual interacts with their environment and how others interact with them; Hart et al., 2021). These factors are relevant to studies exploring parent-delivered interventions where genetic factors may potentially confound genuine environmental effects of the home-based intervention. The gene-environment correlation was beyond the scope of this thesis, though could be considered in future studies exploring parent-led interventions, such as parent-led SBR. In addition, the impact of the gender of the parent conducting SBR activities could be explored further in future research. In the studies in this thesis, the children with Down syndrome were all supported by their mothers for SBR, whilst the child with CP was supported by her father. The child with CP completed far more SBR activities over the 6-week intervention phase than any of the children with Down syndrome. Future research could explore the impact of parent gender on motivation and engagement for both parents and children in SBR.

6.4 Influence of Instruction Modalities

6.4.1 Individual and small group instruction

The studies in this thesis indicated that one-to-one literacy instruction, delivered via telepractice, was feasible and could be delivered with high levels of fidelity to children with CP, Down syndrome, and autism. The primary aim of the study involving children with autism was to refine methods of reading comprehension instruction for autistic children in a group setting. Unfortunately, due to the pandemic induced restrictions; we were unable to explore group-based instruction delivery past the midway point in this study. Implementation fidelity was high across both the group and individual instruction. Yet, instructors were less able to provide ability level differentiation for activities in group-based instruction, compared with individual instruction delivery. This is consistent with Tier-3, compared with Tier-2, interventions in the multi-tier intervention framework (Gersten et al., 2009), which allow for highly individualised and focused instruction.

Previous research indicates that small-group and one-to-one delivery of literacy instruction results in similar reading gains for low progress readers (Gersten et al., 2020; Miles et al., 2022; Neitzel et al., 2021; Vaughn et al., 2003). These studies were not, however, specific to children with developmental disabilities. Unfortunately, we were unable to determine the impact of group versus individual ABRA literacy instruction for children with autism in this research. Few studies investigating reading interventions for typically developing children have directly compared one-to-one and small-group delivery of the same intervention (e.g., Miles et al., 2022; Schwartz et al., 2012; Vaughn et al., 2003), with even fewer involving children with developmental disabilities. Given that small-group instruction is a more cost-effective delivery method and has shown similar reading gains for children with typical development (Neitzel et al., 2021), this requires attention in future research.

6.4.2 Telepractice

6.4.2.1 For Delivery of Literacy Instruction. Telepractice delivery of literacy instruction was feasible for most children with developmental disabilities in this research. All the participants with CP or Down syndrome, and all but three of the participants with autism, achieved the target number of instruction sessions. We note that eight families withdrew from the autism study following the transition from face-to-face to telepractice sessions. Socioeconomic inequity and limited resources were potential factors in these particular participants choosing to withdraw. These findings have important implications given that up to 70% of services for children with developmental disabilities transitioned to telepractice during the pandemic (Furlong & Serry, 2023; Kowanda et al., 2021; Zhang et al., 2022). As a result of the pandemic, many have called for more research on individual and small group tutoring via telepractice (e.g., Neitzel et al., 2021). This is because telepractice can offer more flexible service delivery options for educators, practitioners, and families (Kwok et al., 2022). The empirical studies in this thesis go a small way to starting to fill this research gap for children with developmental disabilities.

As far as we are aware, only two previous studies have explored delivery of multimedia literacy programs to children with developmental disabilities via telepractice (i.e., where children engage directly with a computer program on the screen at the same time as interacting with a facilitator online: Bailey et al., 2022; Hansen et al., 2023). Both studies were conducted during the COVID-19 pandemic and involved children with autism. The study by Bailey et al. (2022) which utilised ABRA has been described previously, with children in this study not achieving statistically significant gains in reading skills compared to a control group. Hansen et al. (2023) explored the Headsprout reading program (a commercially available multimedia literacy program developed based on the NRP Big Five) with 16 autistic children with reading delays. Approximately half of the children in this study received the intervention face-to-face and the remaining half received the intervention via telepractice (after the pandemic restrictions were introduced). Hansen et al. (2023) found no statistically significant gains in reading accuracy, reading fluency, or reading comprehension for the intervention group when compared with a control group. These studies add to the very limited evidence-base of interventions delivered via telepractice directly to autistic children (i.e., interventions that involve direct engagement between the child and clinician online rather than parent coaching. See systematic reviews by Ellison et al., 2021; and Sutherland et al., 2018). Together, the studies by Bailey et al. (2022), Hansen et al. (2023), and our autism study suggest that literacy intervention via telepractice is feasible, though potentially not equivalent to face-to-face literacy interventions for autistic children (e.g., studies involving face-to-face delivery of the ABRA or Headsprout programs for autistic children have reported statistically significant reading gains; Arciuli & Bailey, 2019; Bailey et al., 2017; Grindle et al., 2013; Grindle et al., 2021; Nally et al., 2021). Several factors may contribute to these differences as study designs have differed across face-to-face and telepractice delivery in all of these studies. Future research should seek to compare literacy instruction of similar intensity and duration across instruction modalities for children with autism.

In contrast, children with Down syndrome made statistically significant gains in their reading accuracy and functional reading comprehension skills following literacy instruction delivered via telepractice in our study. No previous studies have explored direct literacy instruction via telepractice for children with Down syndrome. Strengths in social skills associated with the Down syndrome phenotype may have facilitated engagement in telepractice sessions for these children, when compared to children with autism who find social communication challenging (Bull, 2020; Fidler, 2005; Reilly, 2009; Versaci et al., 2021). However, the difference in study design and consistency in instruction mode also likely affected outcomes for children with Down syndrome when compared to the study involving autistic children in this thesis.

6.4.2.2 For Parent Coaching. We found that parent coaching in SBR procedures via telepractice was effective for parents of children with CP or Down syndrome. This is consistent with previous studies that have explored parent training in SBR procedures via telepractice (e.g., Akemoglu et al., 2021; Dodge-Chin et al., 2022; Pierson et al., 2021). Unlike previous studies, the studies in this thesis are the first to explore direct reading outcomes for children with developmental disabilities following SBR training delivered to parents via telepractice. Our studies indicate that this mode of parent coaching was feasible and acceptable to parents of children with CP or Down syndrome and that parents were able to effectively implement the target SBR strategies.

6.5 Implications for policy and practice

6.5.1 Policy

6.5.1.1 Educational Policy. In recent years, policies and government reports on teaching reading have become more inclusive of children with developmental disabilities and diverse needs. These policies convey similar messages that, whilst children with disabilities may need modifications or additional time in instruction, they require access to the same evidence-based, high-quality, comprehensive literacy instruction as their peers. Table 6.2 provides examples of statements from four recent key government policies and reports around different aspects of reading instruction. This table illustrates positive changes across recent policies and reports, but does not comprehensively explore all new policies and government recommendations.

Table 6.2 *Examples of Statements from Recent Government Reports Demonstrating Emphasis on Inclusive Practices in Reading Instruction*

Area of Practice Report *Country*	**Examples Statements**
Assessment	
NSW DoE (2023a & 2023b) *Australia*	• “Students with disability require a diverse range of assessment approaches and appropriate adjustments to demonstrate their understanding.” (p. 5)
Individualised instruction	
NSW DoE (2023a & 2023b) *Australia*	• “Ensure teachers are making adjustments and differentiating learning to cater for the needs of all learners including EAL/D learners, students with disability and additional learning and support needs” (p. 8)
DfE (2023) *England*	• “Teaching should…take full account of each pupil’s individual strengths, weaknesses, knowledge and understanding, and profile of needs.” (p. 79)
Methods of instruction	
DfE (2023) *England*	• “Consensus is growing among academics and teachers that the best reading instruction for pupils with SEND is SSP, taught by direct instruction” (p. 77)
Accessibility	
OHRC (2022) *Canada*	• “A common accommodation for students with reading disabilities is assistive technology. This can be a device, piece of equipment, software or system that helps students access grade-level curriculum. Access to the curriculum means that students can take in and understand the material being taught in school, understand and complete assignments, and show what they have learned.” (p. 48)
DfE (2023) *England*	• “Some pupils may need additional strategies, such as for those who: have physical disabilities that affect their fine motor control for holding and manipulating objects, e.g. use of desktop manipulatives, alternative writing strategies; are pre- or non-verbal, e.g. use of alternative communication strategies, such as selecting their response from auditory choices anchored to visual symbols or place-markers; have both fine motor difficulties and are pre- or non-verbal, e.g. use of low- or high-tech eye gaze strategies.” (p. 79)
Expectations	
OHRC (2022) *Canada*	• “The assumption that some students – including students with disabilities – will never learn to read well is a form of ableism. It is used to justify maintaining systemic barriers instead of making changes we know will help all students learn to read.” (p. 14)
DfE (2023) *England*	• “Literacy is as important for these pupils as for their peers and teachers should be ambitious about teaching them to read and write.” (p. 77)

Note. DfE = Department for Education (2023); EAL/D = English as an Additional Language or Dialect; NSW DoE = NSW Department of Education (2023a, 2023b); OHRC = Ontario Human

Rights Commission (2022); SEND = special education needs and disabilities; SSP = systematic synthetic phonics

Most noteworthy of these four key reports is *The Reading Framework* from the UK Department for Education (2023) which includes an entire section on supporting reading skills for 'Pupils with special educational needs and disabilities'. Much of the evidence cited within this section of the report is from Australian-based research groups (e.g., Arciuli & Bailey, 2021; Castles et al., 2018; Cologon, 2013; Trembath et al., 2015), demonstrating how Australian research on literacy and children with developmental disabilities, including autism and Down syndrome, is influencing policy and practice globally. This report emphasises repeatedly that children with disabilities "have to navigate the same written language, unlock the same alphabetic code, learn the same skills, and learn and remember the same body of knowledge as their peers" (p. 77) and as such, these children require access to the same type of instruction. This is an important message for policy makers and consistent with the research presented in this thesis.

Recent changes to enhance inclusive literacy practices extend beyond educational policies into policies regarding graduate teacher training programs. A new report by the Teacher Education Expert Panel on improving initial teacher education programs, commissioned by the Australian Government, includes discussion of practices that can improve teaching and learning for children with disabilities (Scott, 2023). The report identifies that beginning teachers are underprepared in key areas, including teaching reading and supporting the needs of diverse learners in the classroom, amongst others. The report recommends that core content on supporting diverse learners, such as children with disabilities, should be mandated for accreditation of teacher training programs. This includes training in "evidence-based approaches to cater for specific needs and disabilities" (p. 103). This also includes ensuring educators understand that evidence-based practices for core content, such as teaching reading, "are highly effective for the vast majority of students, particularly those with additional needs or from disadvantaged backgrounds" (p. 103).

Changes to graduate teacher training programs are not unique to Australia, and a recent inquiry into human rights issues affecting students with reading disabilities by the Ontario Human Rights Commission (2022) similarly "makes recommendations about what pre-service and in-service teachers should learn about teaching reading and reading disabilities" (p. 6). Given the relatively sparse evidence base on teaching reading to children with developmental disabilities, as seen in the systematic review relating to CP in Chapter 2, it is not surprising that many teachers feel underprepared to support the reading skills of children with disabilities (Gesel et al., 2021; Peeters, Verhoeven, & de Moor, 2009; Ruppar et al., 2016). Changes to teacher training programs and policies around teaching reading are a good first step in ensuring that children with developmental disabilities are provided with high-quality literacy instruction.

Despite recent progress, some studies suggest that advances in research knowledge and new government policies may not yet have impacted classroom practice (e.g., Solis & McKenna, 2023) and significant challenges to inclusive education for children with disabilities in Australia continues (Poed et al., 2022). More work is needed to ensure that children with diverse needs are supported by well-designed and well-resourced policies that reduce barriers to learning and promote access to evidence-based reading instruction. It is our hope that research, such as the studies presented in this thesis, can promote interest in this topic and continue to inform policy initiatives that prioritise the needs and inclusion of children with disabilities.

6.5.1.2 Clinical Guidelines. Clinical practice guidelines from professional bodies are used to guide intervention practices for clinicians. The '*Practice guidelines for speech pathologists working in childhood and adolescent literacy*' by Speech Pathology Australia (SPA, 2021) does not give specific recommendations for children with disabilities. These guidelines assert that as reading relies on the same core set of skills for all children, the information is applicable to a range of children, including those with developmental disabilities. The guidelines are inclusive of children who communicate in different ways, noting that whilst 'speech' is used to refer to verbal communication, "this may be changed to suit the purposes of students who use alternative forms of communication" (p. 10). Yet, specific guidance on modifying literacy interventions for children who have complex communication needs is not provided. The American Speech-Language-Hearing Association (ASHA, n.d.) practice portal for written language disorders includes a section on literacy assessments for "children who are nonverbal or have limited speech (including users of AAC)" and includes examples of how to modify assessments to make them accessible for nonspeaking children. These ASHA guidelines also include a section on "intervention for children with complex communication needs" which encourages use of assistive technology to provide independent access to reading and writing activities. However, the ASHA guidelines do not provide further advice on specific literacy intervention methods for working with this group.

The SPA and ASHA clinical practice guidelines for literacy may help to explain why up to 79% of speech pathologists report that they do not feel they have the expertise to support the literacy skills of children who use AAC (Fallon & Katz, 2008). A recent survey of Australian speech pathologists does not present a more optimistic picture for literacy intervention generally. This study found substantial variability in clinicians' knowledge and skills in literacy constructs (Stephenson et al., 2023). In this study, only 60% of clinicians indicated a high level of confidence in using literacy assessments to inform reading intervention and 87% of clinicians indicated that speech pathologists do not receive adequate training at university to support children with reading difficulties. Clinicians in this study reported that they felt most confident in their ability to provide intervention for phonological and phonemic awareness skills and least confident to provide literacy intervention to children learning English as an additional language. Concerningly, almost one third of the clinicians surveyed felt that reading difficulties did not include oral language difficulties. It may be that additional work is required to bring clinicians' literacy-related knowledge, skills, and beliefs in line with evidence-based practices in order to increase the integrity of inclusive literacy support.

6.5.2 Practice

The findings in this thesis should encourage all educators and clinicians to use the science of reading in practice when working with children with developmental disabilities. Active steps should be taken to ensure that the literacy learning environment does not pose a barrier to children with disabilities when teaching reading. This includes applying the principles of UDL to learning materials. Educators and clinicians should: (a) consider that the same evidence-based principles for teaching children to read apply for all children, including children with developmental disabilities (this includes explicit and direct reading instruction); (b) utilise comprehensive literacy instruction and intervention incorporating the Big Five; (c) ensure that methods of assessment and instruction are accessible to children with a diverse range of skills and abilities; (d) take instruction at an individualised pace and include frequent repetition; (e) involve parents in literacy instruction, including training parents to use evidence-based strategies to support their child's reading accuracy and reading comprehension skills at home; and (f) be ambitious about all children's level of achievement when teaching children to read.

6.5.2.1 Alternative Service Delivery Models. There is currently a shortage of providers with the appropriate skills and expertise to support children with developmental disabilities in Australia (Australian Government, 2023). This shortage is even more pronounced in rural and remote areas (Australian Government, 2021). The demand for allied health clinicians in the disability sector is only projected to rise in coming years (Australian Government, 2023). In this context, findings from this research regarding the feasibility of parent implemented literacy supports, and literacy instruction delivered via telepractice, have important implications for clinical practice. Parent implemented interventions can reduce the frequency of face-to-face sessions required whilst ensuring a high 'dose' of practise opportunities or could be provided as an alternative to direct intervention for children on waiting lists. Telepractice can reduce travel for clinicians and result in more efficient service delivery, allowing more children access to supports. Unfortunately, we were unable to explore the effectiveness of group-based literacy instruction for autistic children in this research as originally planned. Group-based interventions are another service delivery option which can increase practitioners' efficiency in intervention delivery and should be explored in future research.

6.5.2.2 Specialised Supports in Instruction. Although the same scientific principles of learning to read apply to children with developmental disabilities, additional factors may need to be considered in practice to provide an optimal learning environment. Outlined below are some of the evidence-based strategies and specialised supports used throughout the studies in this thesis to support the engagement and learning of children with CP, Down syndrome, or autism:

- Routine procedures and a visual schedule were used across all studies to provide structure and predictability and support children's engagement in sessions. This strategy is well documented to support children with autism and other developmental disabilities to reduce anxiety and frustration, process information, ease transitions between tasks, and facilitate task completion and on-task behaviour (Foster-Cohen & Mirfin-Veitch, 2017; Knight et al., 2015; van Dijk & Gage, 2019).
- Additional visual supports were used in SBR with the autistic children in this research, including scripts to generate questions. These visuals were used to reduce the need for verbal instructions, support engagement and on-task behaviour, and serve as a prompt for children to self-monitor their use of reading comprehension strategies (Arciuli & Bailey, 2019; Whalon, 2018; Whalon & Hanline, 2008).
- Highly engaging instructional materials involving both visual and auditory stimuli were used within instruction sessions. Such supports may be effective in reducing working memory demands for children with developmental disabilities (Bailey et al., 2017; Loveall & Barton-Hulsey, 2021; Mayer, 2008; Mayer & Moreno, 2003). The ABRA program and the instructors themselves also provided high levels of repetition within and across sessions to support learning and working memory difficulties (Lemons et al., 2015).
- Culturally relevant texts were used in SBR (Australian-based decodable book series) to

support children’s engagement and interest in texts (Barber et al., 2018; Kim et al., 2016).

- SBR activities were designed to capitalise on the motivation and strengths in social interaction for children with Down syndrome (Loveall & Barton-Hulsey, 2021). Turn-taking visuals and clinician scaffolding were used to support the social-communication skills of autistic children during these highly socially demanding SBR activities (Arciuli & Bailey, 2019; Reutebuch et al., 2015; Whalon & Hanline, 2008).
- Reward contingencies were used across all sessions to encourage motivation and participation (e.g., reward activity at the end of sessions) and an external reward-based token system was used with the autistic children (Bailey et al., 2017; Solis et al., 2016; Whalon, 2018).

6.6 Limitations and Future Research

6.6.1 Methodology

The studies involving children with CP or Down syndrome were small-scale and of short duration. While such pilot studies are necessary to guide the design and implementation of larger scale efficacy studies, small scale studies may over-estimate effect sizes and caution should be taken when interpreting statistical results (Leon et al., 2011). In addition, the duration of these studies (16-18 hours of instruction over 6-weeks) fell short of the recommended ABRA administration time (2 hours per week for at least 13 weeks; Abrami et al., 2020). Future research involving children with CP or Down syndrome should include a larger sample over a longer duration and utilise a quasi-experimental or randomised controlled trial design to further investigate the benefits of these comprehensive literacy instruction methods. As previously discussed, the various pandemic-driven changes to methodology in the study involving autistic children compromised experimental control and were a clear limitation. This study, though not the effectiveness study that was originally planned, was a true reflection of education and clinical services in NSW at the time it was conducted.

Across the studies, instruction was delivered primarily in a one-to-one context and by researchers (speech pathologists) as first steps in investigating new literacy instruction methods. However, instruction delivered one-to-one by researchers is resource intensive and may not accurately translate to a school environment (Kim et al., 2012). Future studies should look to upscale these methods to utilise common service providers as instructors, such as teachers and speech pathologists in the community, and could explore use of these methods in the classroom environment. The same speech pathologist delivered instruction to all participants in the studies involving children with CP or Down syndrome, whereas two speech pathologists were involved in delivering instruction to the children with autism. It is possible that the different instructors in the autism study impacted outcomes. Due to the methodological changes in this study, including some children changing between clinicians in the transition from face-to-face to telepractice sessions, we were unable to examine these potential effects of clinician. As a standard protocol was set for instruction sessions, and fidelity across clinicians was rated highly in this study, we do not expect this to have had a significant impact on outcomes.

Additional limitations for each of the studies have been discussed in detail within each of the study chapters. Limitations associated with the reading comprehension outcome measure and parent-led SBR have been discussed in Sections 6.2.2 and 6.3.2.2.1 respectively. Further potential limitations that are relevant to all three empirical studies and could be considered in future research are: (a) no longitudinal follow-up of children's reading skills; and (b) no measure of what kind of literacy instruction children were receiving at school and during remote learning. Such details would provide further information regarding the efficacy and effects of instruction in this research.

In addition to larger-scale and carefully controlled experimental studies investigating literacy instruction for children with CP, Down syndrome, or autism, future research should include observational studies that explore classroom-based literacy instruction for these children. This could include type of instruction, time spent in instruction, and accessibility of instruction. As identified in Section 6.5.1, educational policies have become more inclusive of children with diverse needs in the classroom, yet studies indicate that recent research and policies have not yet completely influenced practice (e.g., Solis & McKenna, 2023). Future observational research of literacy instruction for children with developmental disabilities in the classroom could shed light on the state of implementation of new research and policies within applied settings. These observations could assist policy makers with priority areas for knowledge translation and upskilling of practitioners. Qualitative studies exploring the lived experiences of literacy instruction for adults and children with developmental disabilities should also be considered to add further depth to our understanding of this topic.

6.6.2 Participant Characteristics

Inclusion criteria across the three studies in this thesis captured only children who used verbal speech to communicate. This criterion was set due to the novel and exploratory nature of these studies and to capture a comparable sample of participants. Consequently, a large subgroup of children with CP, Down syndrome, or autism were excluded from this research. Children with complex communication needs experience considerably poorer literacy outcomes and research on literacy instruction for children who use AAC is severely lacking (Cheng & Chavers, 2023; Dahlgren Sandberg et al., 2010; Foley & Wolter, 2010; Koppenhaver, 2000; Yorke et al., 2021). Future research should explore accessible and efficacious literacy interventions that allow these children to demonstrate, and receive feedback on, their reading skills without requiring verbal responses. In addition, most standardised literacy assessments are not accessible for children with complex communication needs. This makes it difficult to accurately gather information on these children's literacy strengths and areas of need. Future research needs to explore and develop literacy assessments that are accessible and accurately measure the reading skills of children who do not speak.

Due to the nature of instruction in this study, English as a primary language was set as an inclusion criterion for the three studies. As such, outcomes of this research are based on a sample of primarily monolingual children. Literacy instruction for children with developmental disabilities and English as an additional language or dialect is an important factor for future research to explore (Arciuli & Bailey, 2021). In addition, most participants across the studies in this thesis attended mainstream schools. Only one child with Down syndrome and one child with autism attended a special school (though 36% of the sample of autistic children were in a specialist class within a mainstream school). This possibly reflects increased expectations of children with disabilities or increased supports for inclusive education within mainstream school settings in Australia. Another possibility is that the nature of our study and inclusion criteria drew a subsample of children with developmental disabilities representative of children with higher intellectual abilities and less significant support needs. Future research should seek to include children with diverse support needs.

Another consideration for future literacy instruction research is including children with dual diagnoses. Over half of our sample of autistic children had a dual diagnosis of ADD/ADHD, yet other dual diagnoses were largely unreported in our sample. As noted in Chapter 1, many children with CP, Down syndrome, or autism have dual diagnoses (e.g., Craig et al., 2019; DiGuiseppi et al., 2010; Moss et al., 2012; Påhlman et al., 2020; Warner et al., 2014). Based on the research findings in this thesis, we hypothesise that high-quality comprehensive literacy instruction involving the Big Five will also be effective for children with dual developmental disability diagnoses. However, different supports, accommodations, and levels of individualisation will likely be required in instruction, and future research should explore what such adjustments look like. Additional factors known to influence literacy development were outside the scope of this research, though should be explored for children with developmental disabilities in future research (e.g., the impact of factors such as self-efficacy, self-concept, and mental health, home literacy environments, and sleep, pain, and fatigue on literacy outcomes).

6.7 Conclusion

The research in this thesis sought a clearer understanding of effective reading instruction methods for children with developmental disabilities to help guide policies and practice and improve literacy outcomes for these children. Across the three empirical studies in this thesis, children with CP, Down syndrome, or autism made varied gains in their reading skills following participation in comprehensive computer-assisted literacy instruction (ABRA) and SBR. All of the studies were conducted during the COVID-19 pandemic, though the study involving children with autism was most significantly disrupted, including a change to the mode and method of literacy instruction delivery. One child with CP made modest gains in their letter-sound knowledge and decoding skills. Children with Down syndrome made gains in word- and passage-level reading accuracy skills and functional reading comprehension. Despite being conducted in less that ideal circumstances, the autistic children in our study made statistically significant gains in their nonword reading skills. The studies in this thesis add to the limited research base and substantiate recent policy recommendations that children with developmental disabilities require access to the same high-quality comprehensive literacy instruction as their typically developing peers. This research contributes new evidence to show that remote delivery of high-quality literacy instruction is feasible and can be effective for some children with developmental disabilities. This has the potential to increase the accessibility of some services and has important implications for policy and practice both during a pandemic and beyond.

This research contributes original empirical evidence for comprehensive literacy instruction delivered to groups of children with developmental disabilities which have not previously been explored (a child with CP and children with Down syndrome). It is hoped that by highlighting the importance of this area, future studies will continue to explore the promise of comprehensive literacy instruction for children with developmental disabilities. It is also our hope that research such as this, which demonstrates the capacity for children with disabilities to make meaningful literacy gains within relatively short periods of time, can assist in shaping more optimistic stakeholder views and literacy expectations of these children.

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APPENDIX A: EMPERICAL STUDY 3 - PHASE ONE METHOD AND FINDINGS

Phase One Method

Participants. Research flyers advertising the study were circulated via the researchers' professional networks via email and using the researchers' social media accounts and on websites and/or newsletters of local disability support organisations, and in hard copy using the notice boards of local community organisations. Recruits for three focus groups were targeted: (a) a group of adolescents with autism; (b) a group of parents of children with autism; and (c) a group of allied health clinicians working with children with autism.

A total of 10 volunteers took part in the focus groups. The three focus groups comprised three adolescents with autism aged 14 to 17 years (adolescent group), four parents of children with autism (parent group), and three allied health clinicians (all speech pathologists; clinician group).

Procedure. Each focus group was conducted as a 60-minute semi-structured discussion hosted at the Luke Priddis Foundation or online via Zoom. The primary aim of the meetings was to co-design clinician-led shared reading activities which would supplement ABRA instruction for some participants during the clinic-based ABRA sessions. Phase One participants were asked about various practicalities of group-based instruction for children with autism which would help the researchers' design of the instruction sessions in Phase Two (e.g. questions such as 'Do you prefer physical books or iPads for SBR?' and 'What supports might help children with autism to participate in group-based shared reading?')

The focus groups were audio recorded and later transcribed. Key recommendations were extracted and guided the design and delivery of instruction in Phase Two.

Phase One Key Recommendations

Barriers to participation in literacy instruction. Participants in all three focus groups highlighted time as the primary barrier to participation in the ABRA and SBR instruction sessions. In addition, parents noted competing priorities, such as other appointments and activities for siblings. The clinicians flagged that children may have difficulty acclimatizing to group-based instruction and unfamiliar peers and instructors.

Strategies for promoting attendance at the instruction sessions. Parents suggested that the researchers would need to be flexible when scheduling the sessions and work around families' schedules when timetabling. Both adolescent and clinician focus group members recommended that children be told what they will be doing during the instruction sessions in advance, with the clinicians recommending the use of social stories. Clinicians also recommended that parents be provided information regarding parking and travel to the foundation and working to establish routines with the Phase Two participants (e.g., instruction sessions should follow a routine structure).

Optimizing the start of each session. The adolescent group recommended that participants be offered food and a drink at the beginning of each session to help them transition and also in anticipation that some children may come to the session directly from school. Clinicians and parents highlighted the need to explain the activities and explicitly state what participants are expected to do during the session. Both groups also recommended that the researchers explain that participants will be given a reward at the end of each session and in the longer-term contingent on adhering to the session rules.

Assisting children through transitions. Participants in each of the focus groups reiterated that visual schedules may be of use, with adolescents explaining that these may be particularly useful when moving the groups between instruction tasks. All three groups also recommended specific resources to assist transitions, such as Time Timers. In addition, clinicians encouraged the researchers to give frequent verbal reminders of impeding transitions.

Assisting access to computers. Adolescents recommended that the researchers independently assess the technological needs of each participant in Phase Two. They also suggested that noise cancelling and noise limiting headphones might be useful for those participants with sensory sensitivities or those that have difficulty attending when there are competing demands (e.g., noise from other group members). Clinicians also mentioned headphones, saying that participants should be given the option of bringing their own.

Supporting participation in shared reading activities. Adolescents highly recommended the use of external rewards, such as additional play time or material rewards but also acknowledged that intrinsic motivation is important in the longer term. Parents recommended that the researchers explore children's interests and integrate these into the instruction sessions where possible. Clinicians recommended that rules be put in place to support compliance, such as marking each participants' seating position and making a uniform rule that one person may speak at a time.

Selection of reading materials for SBR activities. Adolescents recommended a few genres that may be of interest to the Phase Two participants but also stated that texts do not necessarily need to be matched to each participants' interests as long as they target appropriate skills (i.e., text is of an appropriate complexity). Clinicians also recommended that texts be selected on the basis of skills needing to be targeted, specifically recommending the use of decodable texts.

Encouraging peer-to-peer interactions. Adolescents highly recommended the use of group-based games to encourage interactions between Phase Two participants. Adolescents also said that short breaks during instruction and a reward activity at the end of the session would provide opportunities for interaction. Parents highlighted a need to select groups carefully such that groups comprise children with similar abilities and interests. Clinicians suggested that having rules around turn taking would be of benefit.

How to deal with missed instruction sessions. Parents recommended that Phase Two participants be offered "make-up" sessions online using Zoom. They also recommended that sessions be rescheduled to another time if needed. Clinicians encouraged the researchers to be in regular contact with parents and have protocols in place to address late arrivals.

How to support children who become tired or dysregulated during sessions. Adolescents highly recommended the use of breaks and providing participants time to themselves with the choice of rejoining the groups. Clinicians encouraged the researchers to establish both short (each session) and medium-term rewards (star charts) and have parents assist in the event that children become dysregulated.

APPENDIX B: ETHICS APPROVAL (STUDY 1 & STUDY 2)

6 July 2020

Flinders UNIVERSITY

HUMAN RESEARCH ETHICS COMMITTEE

APPROVAL NOTICE

Dear Prof Joanne Arciuli,

The below proposed project has been **approved** on the basis of the information contained in the application and its attachments.

Project No:	2069
Project Title:	Online literacy instruction for children with developmental disabilities
Primary Researcher:	Prof Joanne Arciuli
Email:	joanne.arciuli@flinders.edu.au
Approval Date:	06/07/2020
Expiry Date:	31/12/2020

Please note: *Due to the current COVID-19 situation, researchers are strongly advised to develop a research design that aligns with the University's COVID-19 research protocol involving human studies. Where possible, avoid face-to-face testing and consider rescheduling face-to-face testing or undertaking alternative distance/online data or interview collection means. For further information, please go to https://staff.flinders.edu.au/coronavirus-information/research-updates.*

RESPONSIBILITIES OF RESEARCHERS AND SUPERVISORS

1. **Participant Documentation**

Please note that it is the responsibility of researchers and supervisors, in the case of student projects, to ensure that:

- all participant documents are checked for spelling, grammatical, numbering and formatting errors. The Committee does not accept any responsibility for the above mentioned errors.
- the Flinders University logo is included on all participant documentation (e.g., letters of Introduction, information Sheets, consent forms, debriefing information and questionnaires – with the exception of purchased research tools) and the current Flinders University letterhead is included in the header of all letters of introduction. The Flinders University international logo/letterhead should be used and documentation should contain international dialling codes for all telephone and fax numbers listed for all research to be conducted overseas.
- the HREC contact details, listed below, are included in the footer of all letters of introduction and information sheets.

This research project has been approved by the Flinders University Human Research Ethics Committee (Project Number 2069). For more information regarding ethics approval of the project the Executive Officer of the Committee can be contacted by telephone on 8201 3116, by fax on 8201 2035 or by email human.researchethics@flinders.edu.au.

2. **Annual Progress / Final Reports**

In order to comply with the monitoring requirements of the *National Statement on Ethical Conduct in Human Research 2007 (updated 2018)* an annual progress report must be submitted each year on the **6th of July** (approval anniversary date) for the duration of the ethics approval using the HREC Annual/Final Report Form available online via the ResearchNow Ethics & Biosafety system.

Please note that no data collection can be undertaken after the ethics approval expiry date listed at the top of this notice. If data is collected after expiry, it will not be covered in terms of ethics. It is the responsibility of the researcher to ensure that annual progress reports are submitted on time; and that no data is collected after ethics has expired.

If the project is completed *before* ethics approval has expired please ensure a final report is submitted immediately. If ethics approval for your project expires please either submit (1) a final report; or (2) an extension of time request (using the HREC Modification Form).
For student projects, the Low Risk Panel recommends that current ethics approval is maintained until a student's thesis has been

submitted, assessed and finalised. This is to protect the student in the event that reviewers recommend that additional data be collected from participants.

First Report due date: 6 July 2021

Final Report due date: 31 December 2020

3. Modifications to Project

Modifications to the project must not proceed until approval has been obtained from the Ethics Committee. Such proposed changes / modifications include:

- change of project title;
- change to research team (e.g., additions, removals, researchers and supervisors)
- changes to research objectives;
- changes to research protocol;
- changes to participant recruitment methods;
- changes / additions to source(s) of participants;
- changes of procedures used to seek informed consent;
- changes to reimbursements provided to participants;
- changes to information / documents to be given to potential participants;
- changes to research tools (e.g., survey, interview questions, focus group questions etc);
- extensions of time (i.e. to extend the period of ethics approval past current expiry date).

To notify the Committee of any proposed modifications to the project please submit a Modification Request Form available online via the ResearchNow Ethics & Biosafety system. Please note that extension of time requests should be submitted prior to the Ethics Approval Expiry Date listed on this notice.

4. Adverse Events and/or Complaints

Researchers should advise the Executive Officer of the Ethics Committee on 08 8201-3116 or human.researchethics@flinders.edu.au immediately if:

- any complaints regarding the research are received;
- a serious or unexpected adverse event occurs that effects participants;
- an unforeseen event occurs that may affect the ethical acceptability of the project.

Yours Sincerely,

Andrea Mather

on behalf of

Human Research Ethics Committee
Research Development and Support
human.researchethics@flinders.edu.au
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Flinders University
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http://www.flinders.edu.au/research/researcher-support/ebi/human-ethics/human-ethics_home.cfm

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APPENDIX C: ETHICS APPROVAL (STUDY 3)

15 December 2020

Flinders UNIVERSITY

HUMAN RESEARCH ETHICS COMMITTEE

APPROVAL NOTICE

Dear Professor Joanne Arciuli,

The below proposed project has been **approved** on the basis of the information contained in the application and its attachments.

Project No:	2844
Project Title:	Literacy instruction for children with autism
Primary Researcher:	Professor Joanne Arciuli
Approval Date:	15/12/2020
Expiry Date:	20/12/2021

Please note: *Due to the current COVID-19 situation, researchers are strongly advised to develop a research design that aligns with the University's COVID-19 research protocol involving human studies. Where possible, avoid face-to-face testing and consider rescheduling face-to-face testing or undertaking alternative distance/online data or interview collection means. For further information, please go to https://staff.flinders.edu.au/coronavirus-information/research-updates.*

RESPONSIBILITIES OF RESEARCHERS AND SUPERVISORS

1. Participant Documentation

Please note that it is the responsibility of researchers and supervisors, in the case of student projects, to ensure that:

- all participant documents are checked for spelling, grammatical, numbering and formatting errors. The Committee does not accept any responsibility for the above mentioned errors.
- the Flinders University logo is included on all participant documentation (e.g., letters of Introduction, information Sheets, consent forms, debriefing information and questionnaires – with the exception of purchased research tools) and the current Flinders University letterhead is included in the header of all letters of introduction. The Flinders University international logo/letterhead should be used and documentation should contain international dialling codes for all telephone and fax numbers listed for all research to be conducted overseas.
- the HREC contact details, listed below, are included in the footer of all letters of introduction and information sheets.

This research project has been approved by the Flinders University Human Research Ethics Committee (Project Number 2844). For more information regarding ethics approval of the project the the Committee can be contacted by telephone on 8201 2543 or by email human.researchethics@flinders.edu.au

2. Annual Progress / Final Reports

In order to comply with the monitoring requirements of the *National Statement on Ethical Conduct in Human Research 2007 (updated 2018)* an annual progress report must be submitted each year on the anniversary of the approval date for the duration of the ethics approval using the HREC Annual/Final Report Form available online via the ResearchNow Ethics & Biosafety system.

Please note that no data collection can be undertaken after the ethics approval expiry date listed at the top of this notice. If data is collected after expiry, it will not be covered in terms of ethics. It is the responsibility of the researcher to ensure that annual progress reports are submitted on time; and that no data is collected after ethics has expired.

If the project is completed *before* ethics approval has expired please ensure a final report is submitted immediately. If ethics approval for your project expires please either submit (1) a final report; or (2) an extension of time request (using the HREC Modification Form).
For student projects, the Low Risk Panel recommends that current ethics approval is maintained until a student's thesis has been submitted, assessed and finalised. This is to protect the student in the event that reviewers recommend that additional data be collected from participants.

3. Modifications to Project

Modifications to the project must not proceed until approval has been obtained from the Ethics Committee. Such proposed changes / modifications include:

- change of project title;
- change to research team (e.g., additions, removals, researchers and supervisors)
- changes to research objectives;
- changes to research protocol;
- changes to participant recruitment methods;
- changes / additions to source(s) of participants;
- changes of procedures used to seek informed consent;
- changes to participant remuneration;
- changes to information / documents to be given to potential participants;
- changes to research instruments (e.g., survey, interview questions etc);
- extensions of time (i.e. to extend the period of ethics approval past current expiry date).

To notify the Committee of any proposed modifications to the project please submit a Modification Request Form available online via the ResearchNow Ethics & Biosafety system. Please open the project, then select the 'Create Sub-Form' tile in the grey Action Menu, and then select the relevant Modification Request Form. Please note that extension of time requests should be submitted prior to the Ethics Approval Expiry Date listed on this notice.

4. Adverse Events and/or Complaints

Researchers should advise the Executive Officer of the Ethics Committee on 08 8201-3116 or human.researchethics@flinders.edu.au immediately if:

- any complaints regarding the research are received;
- a serious or unexpected adverse event occurs that effects participants;
- an unforeseen event occurs that may affect the ethical acceptability of the project.

Yours sincerely,

Hendryk Flaegel

on behalf of

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http://www.flinders.edu.au/research/researcher-support/ebi/human-ethics/human-ethics_home.cfm

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APPENDIX D: FIRST PAGE OF PUBLISHED STUDIES

ASHA American Speech-Language-Hearing Association

LSHSS

Review Article

Exploring the Effects of Literacy Instruction for Children With Cerebral Palsy: A Systematic Review

Annemarie Murphy,[a] Benjamin Bailey,[a] and Joanne Arciuli[a]

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ABSTRACT

Purpose: Some children with cerebral palsy (CP) have difficulty acquiring conventional reading and writing skills. This systematic review explores the different types of literacy instruction and their effects on the reading and writing skills of children with CP.
Method: Relevant studies published between 2000 and 2020 were identified using electronic databases and terms related to CP and literacy. Data on participant characteristics, instruction characteristics, and instruction outcomes were extracted. A standardized measure of effect size was used to quantify reported treatment effects.
Results: The systematic search identified 2,970 potentially relevant studies, of which 24 met inclusion criteria. These studies included 66 children with CP aged 5–18 years. One of the included studies utilized a group research design, whereas the remaining used single-subject designs. Studies investigated literacy instruction methods designed to teach phonics, sight-word recognition, reading fluency, reading comprehension, spelling, or written expression skills, or multicomponent instruction (instruction methods encompassing three or more of these skills). Most instruction methods were associated with gains in reading and writing skills with medium to large effects; however, our analysis of methodological rigor suggests that these findings need to be interpreted with caution.
Conclusions: We propose that literacy instruction utilizing evidence-based principles can be effective for children with CP, provided instruction is accessible and allows children to demonstrate and receive feedback on their skills; however, further research is greatly needed. Clinical implications and priorities for future research are discussed.
Supplemental Material: https://doi.org/10.23641/asha.21357558

Cerebral palsy (CP) is the most common physical disability of childhood and can impact children's functional abilities in many ways (Rosenbaum et al., 2007; Sellier et al., 2020). Up to 80% of children with CP have communication or speech difficulties, with approximately one in four communicating nonverbally, one in two having an intellectual disability, and one in 25 having a severe hearing impairment (Mei et al., 2020; Novak et al., 2012). These cognitive and linguistic factors, along with children's broader social context and learning environments, can all influence literacy development. Literacy is essential for inclusion and access in society (National Commission on Writing, 2004). For individuals with complex communication needs, literacy provides access to independent and meaningful communication (Koppenhaver & Williams, 2010), supports friendships and social participation (Caron & Light, 2016), and facilitates self-advocacy, self-determination, and self-care (Kitson et al., 2021). Yet, literacy instruction practices are often not inclusive of children with severe speech and physical impairments, creating an "opportunity gap" where children are provided fewer opportunities to engage in quality literacy instruction than their typically developing peers (Wolter, 2016; Zascavage & Keefe, 2004). In addition, educators,

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ARTICLE

ABRACADABRA literacy instruction for children with Down syndrome via telepractice during COVID-19: A pilot study

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Abstract

Background: COVID-19 has resulted in some educators and allied health practitioners transitioning to online delivery of literacy instruction. As far as we are aware, no studies have investigated online delivery of comprehensive literacy instruction for children with Down syndrome.

Aims: In this pilot study, we explore the efficacy of online delivery of ABRACADABRA (a free literacy web application) for children with Down syndrome, alongside supplementary parent-led shared book reading, during the COVID-19 pandemic.

Sample: Six children with Down syndrome, aged 8–12 years, participated in this within-participants design study.

Methods: Participants acted as their own controls with outcome variables measured at three timepoints: baseline, pre-instruction and post-instruction. Children participated in 16–18 hrs of one-to-one literacy instruction online over a 6-week instruction phase, along with twice weekly parent-led shared book reading activities.

Results: Outcomes from standardized assessments revealed statistically significant improvements in word- and passage-level reading accuracy skills over the instruction phase (pre-instruction to post-instruction) compared with the no-instruction control phase (baseline to pre-instruction). Improvements in reading comprehension skills were inconsistent across assessment measures and statistical analyses.

Conclusion: Children with Down syndrome can benefit from comprehensive literacy instruction delivered via telepractice. Our study provides critical initial evidence of successful service delivery during a global pandemic and beyond.

KEYWORDS

ABRACADABRA, Down syndrome, literacy, reading, telepractice

