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Literacy abilities of children and adolescents with Williams Syndrome.

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LITERACY ABILITIES OF CHILDREN AND ADOLESCENTS WITH

WILLIAMS SYNDROME

By

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DEDICATION

This dissertation is dedicated to my life partner, Igor Pereira, without whose support, tolerance, encouragement, patience, partnership, and love I would not have succeeded. To my parents, Graciela Maria Greiner and Eduardo Moura de Magalhães, for always believing in me and for providing me with an amazing childhood, great education, and support that allowed me to achieve my academic goals. To my sister Isabela, for her kind words and deep friendship even when we were so far away physically. Our long and deep conversations always gave me energy to keep moving and helped me know I was where I was supposed to be. To my sister Bianca for her easy-going personality and for her easy smile and happiness. To my dear friend Daniela Teixeira Goncalves, for her support, help, friendship, and for always believing in me. Our conversations helped me during so many hard times. I am very grateful to all who crossed my path and gave me the strength and support that helped me to be here today.

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ABSTRACT

LITERACY ABILITIES OF CHILDREN AND ADOLESCENTS WITH WILLIAMS

SYNDROME

Caroline Greiner de Magalhães

November 01, 2021

In this dissertation I report findings from two studies of the literacy abilities of

children with Williams syndrome (WS), a genetic disorder associated with intellectual

disability. I had two overarching goals: 1) evaluate the applicability of theories of reading

and spelling acquisition developed based on typically developing children to children

with WS; and 2) provide results that would inform the development of targeted

assessments and interventions.

In Study 1, individual differences in phonological awareness, visual spatial

perception, vocabulary, overall intellectual abilities, and reading instruction approach

(systematic phonics vs. other approaches) for sixty-nine 6 – 7-year-olds (most of whom

were in kindergarten) were used to predict word-reading abilities three years later.

Multiple regression analyses indicated that Time 1 reading instruction method,

phonological awareness, and visual-spatial perception each explained significant unique

variance in word reading at Time 2. A systematic phonics approach was associated with

significantly better performance than other reading-instruction approaches. Results of a

simple mediation analysis indicated that vocabulary at Time 1 indirectly influenced Time

2 word reading through its effect on Time 1 phonological awareness.

In Study 2, relations between spelling, word reading, and vocabulary abilities and method of reading instruction were investigated for eighty 9 – 17-year-olds. Spelling and reading abilities were highly correlated. Students taught to read using systematic phonics instruction had significantly higher spelling scores than those taught to read using other approaches. Spelling ability contributed significant unique variance to word-reading ability, beyond the effects of phonological awareness, vocabulary, and reading instruction method.

Overall, the results from Study 1 and 2 indicated that the word-reading and spelling abilities of students with WS vary widely but on average are well below the mean for same-aged children in the general population. Variations in overall intellectual ability did not play a central role in accounting for individual differences in word reading and spelling. Findings from this dissertation provide support for the universality of theoretical models of reading development developed based on typically developing children and suggest that educational approaches known to be effective for children who are having difficulty learning to read are likely to be appropriate for children with WS.

Keywords: Williams syndrome, word-reading ability, spelling ability, reading instruction method, systematic phonics instruction, intellectual disability

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CHAPTER I

GENERAL INTRODUCTION

The ability to read and spell is foundational for the engagement of individuals in contemporary society, allowing for acquisition of knowledge and success in school and the workplace, in addition to access to leisure activities (Castles et al., 2018; Pan et al., 2021; World Literacy Foundation, 2018). Given the critical importance of literacy skills, limited reading (Hendren et al., 2018; National Reading Panel, 2000) and spelling (Galuschka et al., 2020; Pan et al., 2021) abilities in adolescence and adulthood have been associated with a variety of negative outcomes including lower educational attainment, limited job opportunities, and increased problems with psychosocial adjustment and mental health. These concerns are shared by caregivers of individuals with intellectual disability (ID), who consider that difficulty with reading results in the greatest limitation in life outcomes (Koritsas & Iacono, 2011).

Individuals with Williams syndrome (WS), a genetic disorder associated with mild to moderate ID, typically present with a wide range of reading and spelling abilities. Reading (Mervis, 2009; Mervis et al., 2021) and spelling difficulties (Dessalegn et al., 2013; Howlin et al., 1998; Udwin et al., 1987, 1996) are very common. The first report of performance on academic achievement tests was provided by Pagon and colleagues (1987), who presented data for nine individuals aged 10 – 20 years. Despite the small sample, their data showed a large range of word reading and spelling ability, with the

oldest individual able to read and spell only a few words and a ninth grader able to decode at grade level, although her spelling ability was at the 4th grade level. Two larger-sample studies of adolescents and adults found that most individuals with WS could read at least a few words (75% in Howlin et al., 1998; 93% in Brawn et al., 2018) and that reading ability was significantly positively correlated with IQ. In addition, 27% of older adolescents and adults with WS met the criterion for functional literacy for both irregular and nonwords, and 20% for irregular words only (Brawn et al., 2018). For spelling ability, two larger-sample studies reported that 58% of children with WS (Udwin et al., 1987) and 74% of adults with WS (Howlin, et al., 1998) were able to meet the basal criterion on a standardized spelling test, which is far below the functional criterion used by Brawn et al. (2018). There are no published studies addressing the proportion of individuals with WS who meet the criteria for functional spelling. Understanding the cognitive predictors that contribute to the variability in the word reading and spelling abilities of individuals with WS is key to designing targeted assessments and interventions that would lead to better practices and optimized reading and spelling development, providing families with the necessary resources so individuals with WS can attain their full potential (Mervis & Greiner de Magalhães, in press).

In my dissertation, I report two studies that contribute to a broad understanding of the development of literacy abilities in children and adolescents with WS. I had two overarching goals: 1) evaluate the applicability of theories of reading and spelling acquisition developed based on typically developing (TD) children to children with WS, which will inform the science of literacy (e.g., Castles et al., 2018) more broadly; and 2) provide results that will inform the development of targeted assessments and

interventions. The studies were designed to investigate word reading ability, spelling ability, and their correlates for children and adolescents with WS. Similarities or differences among the correlates for TD children and children with WS were highlighted.

My dissertation includes four chapters. In Chapter 1, I provide a general introduction. I first briefly describe WS, the neurodevelopmental disorder that is the focus of this dissertation. Then, I present a brief overview of prior research on the word reading abilities and spelling abilities of TD children, individuals with ID, and individuals with WS. Finally, I briefly review the literature addressing the impact of method of reading instruction on the literacy abilities of TD children, individuals with ID, and individuals with WS. In Chapter 2, I report a study on the longitudinal cognitive, language, and instructional factors associated with early single-word reading ability for children with WS, and in Chapter 3, I report a study on the relations between spelling ability, word-reading ability, and method of reading instruction for children and adolescents with WS. In Chapter 4, I provide a general discussion of the findings of the two studies, including theoretical and educational implications, and briefly consider future directions.

Williams Syndrome

WS is a rare neurodevelopmental genetic disorder caused by a hemideletion of 25 – 27 protein-coding genes on chromosome 7q11.23 (Kozel et al., 2021). Ninety-five percent of individuals with WS have a classic deletion, which includes exactly these 25 – 27 genes (Mervis et al., 1999); the deletions of the remaining individuals with WS include either a subset of these genes or all of these genes plus additional genes. WS occurs in approximately 1 out of 7,500 live births and is equally likely to occur in boys

and girls (Strømme et al., 2002). WS is associated with mild to moderate ID, with a full range from severe ID to average intellectual ability (Mervis, 2009; Mervis & Klein-Tasman, 2000; Mervis & Velleman, 2011).

Cognitive Profile

The cognitive profile of children with WS is complex and there is considerable cognitive variability (Mervis & John, 2010). Relative to their overall intellectual ability, individuals with WS typically show strengths in concrete vocabulary, nonverbal reasoning, verbal short-term memory, and phonological processing. Relative to their overall intellectual ability, individuals with WS typically show weaknesses in spatial abilities and relational and conceptual language (Mervis & Greiner de Magalhães, in press; Mervis & John, 2010).

The pattern of relative strengths and weaknesses in intellectual ability is most clearly shown on the Differential Ability Scales-II (DAS-II; Elliott, 2007), which provides separate Verbal, Nonverbal Reasoning, and Spatial standard scores (SSs) based on performance on subtests that are normed four SDs below the general population mean. For 269 4 – 17-year-olds with classic WS deletions (see Mervis & Greiner de Magalhães, in press), mean General Conceptual Ability (GCA; similar to IQ), which is based on performance on the Verbal, Nonverbal Reasoning, and Spatial ability clusters, was 61.98 (SD = 12.94, range: 31 – 96). This overall score masks stark differences between performance on the Verbal cluster (M = 71.00, SD = 16.88, range: 30 – 107) and the Nonverbal Reasoning cluster (M = 75.04, SD = 14.76, range: 36 – 118) on the one hand and performance on the Spatial cluster (M = 53.16, SD = 13.04, range: 32 – 86) on the other. For 70% of the sample, Verbal SS did not differ significantly from Nonverbal

Reasoning SS. In contrast, Spatial SS was significantly lower than Verbal SS for 68% and significantly lower than Nonverbal Reasoning SS for 83%. For 89%, Verbal SS, Nonverbal Reasoning SS, or both were significantly higher than Spatial SS. At the other extreme, Spatial SS was significantly higher than Verbal SS for 3% and Nonverbal Reasoning SS for 0.4%.

Phonological Awareness. Previous studies of individuals with WS (Brawn et al., 2018; Levy et al., 2003; Mervis, 2009; Miezah et al., 2020) have demonstrated that although the phonological awareness abilities of individuals with WS vary widely, they are typically a strength relative to other abilities. Miezah and colleagues (2020) evaluated phonological awareness performance for 49 individuals with WS, ranging in age from 6 to 39 years (mean age = 19.95 years). On average, participants performed better on the phonological awareness cluster (mean SS = 94.36, *SD* = 21.03, range: 60 – 129) than on any other cluster on the Woodcock-Johnson Tests of Cognitive Abilities-III (WJ-III COG; Woodcock et al., 2001). The same pattern of findings was reported by Brawn et al. (2018). Most individuals (81%) scored in the low average to superior range on the phonological awareness task. For 60% of the sample, performance on the phonological awareness task was at least 1.5 *SD*s above the individual's mean SS across the other WJ-III COG domains, the definition of a relative strength used by Miezah et al. (2020).

Mervis (2009) reported that the mean T-score on the DAS-II Phonological Awareness subtest for 55 children with WS (mean age = 8.89 years, range = 6.03 – 12.90) was 40.24 (*SD* = 13.28, range: 10 – 62), which was higher than the mean T-score for any of the DAS-II core subtests. (For the general population, the mean T-score is 50, with a SD of 10.) A similar pattern was reported by Levy et al. (2003) for twenty 12 – 20-

year-olds (mean age = 16.5 years) with WS tested on the Comprehensive Test of Phonological Processing (Wagner et al., 1999); mean SSs for the three phonological awareness tasks (Elision, $M = 70.50$, $SD = 12.97$; Segmenting Words, $M = 81.50$, $SD = 11.01$; Segmenting Nonsense Words, $M = 77.00$, $SD = 11.17$) were considerably higher than the sample's mean composite IQ of 57.05 (SD = 12.99) on the Kaufman Brief Intelligence Test (KBIT; Kaufman & Kaufman, 1990).

Visual-spatial Perception. Visuospatial skills encompasses both visuospatial perceptual skills – a more basic ability that allows someone to interpret visual information about where objects are in space, including how the objects are oriented – and visuospatial construction. In order to solve a block design task, an individual needs to be able to "see an object or picture as a set of parts" (i.e., visuospatial perception) "and then construct a replica of the original from these parts" (i.e., visuospatial construction) (Mervis et al., 1999, *p*. 1222). These abilities are important for everyday life, being required for activities such as packing a suitcase in a manner that takes the best advantage of the space available, constructing three-dimensional structures with blocks, drawing, completing jigsaw puzzles, and making a bed. Relevant to literacy development, being able to identify the orientation of features that make up a letter (e.g., *d* and *b*) requires visuospatial perceptual skills. Committing letter reversal errors when reading or spelling a word is a problem that results from difficulty with visuospatial perception.

Individuals with WS have a hallmark weakness in visuospatial construction ability as evidenced by their difficulty with block design and copying tasks (both included in the Spatial cluster of the DAS – see Cognitive Profile section above). Consistent with their weaknesses in spatial abilities, although children with WS recognize

simple objects in their canonical positions as well as chronological age matched TD

children do, children with WS perform significantly worse that chronological age

matched TD children when the objects are presented in an unusual position, varying in

their orientation or viewed from above or below (Landau et al., 2006; see Farran &

Formby, 2012; Landau & Ferrara, 2013 for reviews of visual perception and visuospatial

cognition in WS).

  Vocabulary. WS also is associated with a specific pattern of relative strength and

weakness in vocabulary ability: Concrete vocabulary (names for objects, actions, and

descriptors) is considerably stronger than relational vocabulary (e.g., labels for spatial,

temporal, and quantitative concepts). On the Peabody Picture Vocabulary Test-4 (PPVT-

4; Dunn & Dunn, 2007), an assessment of concrete vocabulary, mean SS for 129 4 – 17-

year-olds was 81.84 (SD = 15.04, range: 40 – 124), with 83% earning SSs ≥ 70 and 8%

earning SSs ≥ 100 (Mervis & John, 2010). Similarly, mean Expressive Vocabulary Test-2

(EVT-2; Williams, 2007) SS was 79.43, with 83% earning SSs ≥ 70 and 6% earning SSs

≥ 100 (Mervis & John, 2010). Relational vocabulary performance was considerably

weaker, with SSs on the Test of Relational Concepts (Edmonston & Litchfield Thane,

1988) very similar to SSs on the DAS Pattern Construction subtest and ~30 points below

PPVT SSs (Mervis & John, 2008). This pattern is consistent with Walsh's (2003)

argument that spatial, temporal, and quantitative processing are controlled by a common

magnitude system located in the intraparietal sulcus – a region that, in individuals with

WS is characterized by reduced gray matter and reduced functional connectivity to dorsal

stream regions (Gregory et al., 2019).

Importance of Functional Literacy for Adaptive Skills

The United Nations Educational, Scientific and Cultural Organization (UNESCO) defines functional literacy as the level of literacy that enables individuals to engage in activities in their community, successfully performing everyday tasks that depend on basic reading skills (UNESCO, 2021). Acquiring functional literacy is therefore foundational to promoting the inclusion of those with ID in society (Brawn et al., 2018). Along these lines, based on a large parental survey exploring perceptions of parents of children with ID related to their child's literacy instruction in schools, Wakeman and colleagues (2021) reported that 93% of their sample (parents of children in grades K – 12, $N = 211$) thought it was very important for their child to learn to read, and 85% thought that whether or not their child developed literacy would have an impact on their life outcomes.

According to the American Association on Intellectual and Developmental Disabilities and the Diagnostic and Statistical Manual of Mental Disorders-5, ID has its onset before the age of 18 years and is characterized by significant limitations in general intellectual functioning (IQ < 70–75) and adaptive behavior. The word reading ability of individuals with ID varies widely from not able being to read at all to reading at grade level (see Pezzino et al., 2019 for a review). It is not uncommon for individuals with ID to not earn high school diplomas or to not be engaged in post-secondary education or employment (Wagner et al., 2005). However, achieving these benchmarks is important for the inclusion of individuals with ID in the society. Considering the fundamental role of having adequate literacy skills to earning high school diplomas or engaging in post-secondary education or employment, studies that provide information that can be used to

identify or design more targeted assessments and literacy interventions for individuals with ID are crucial. Functional literacy is important for a wide range of daily activities, for full engagement with one's community, and for a wide range of occupations. Accurate spelling is important for obtaining and keeping jobs that routinely require written communication and can affect a person's ability to secure employment even in occupations that do not require that the employee communicate in writing on a daily basis (Pan et al., 2021).

The adaptive functioning of individuals with WS is well below age-expectations; overall adaptive level typically is in the mild to moderate disability range, with the full range extending from average for the general population to severe disability (see Brawn & Porter, 2018 for a systematic review). Fisher et al. (2020) reported that of 114 adults with WS (M = 27.99 years) only 6% lived independently. One crucial component of adaptive behavior is community living skills, the individual's ability to function independently in the world outside the home. Even after controlling for IQ, word-reading ability was positively associated with the community living skills of adolescents and adults with WS (Brawn et al., 2018). Greiner de Magalhães and Mervis (2021) further demonstrated the importance of word reading ability for community living skills for 59 children with WS (M age = 13.14 years, SD = 3.08, range: 9.00 – 17.98). In their study, word-reading skills contributed significant unique variance to community living skills as measured by parent report, even after the effects of chronological age, sex, overall intellectual ability, and emotion regulation were taken into account. Considering the clear importance of literacy skills for children with WS, in my dissertation, I investigated the word reading and spelling abilities of children and adolescents with WS.

Predictors of Word Reading Ability: Typically Developing Children, Individuals with Intellectual Disability, and Individuals with Williams Syndrome

Phonological Awareness

Phonological awareness is the ability to consciously attend to and manipulate the sounds in speech. This ability is clearly important for learning to read and spell in an alphabetic orthography (Ehri, 1992, 2014). It is well-known (e.g., Melby-Lervåg et al., 2012; Muter et al., 2004; Wagner et al., 2019) that for TD children, word reading ability is strongly related to phonological awareness. The discovery of this association has been described as one of the greatest scientific successes in the history of cognitive developmental psychology (Stanovich, 1987). Phonological awareness skills are central to word-level reading because children learning to read in an alphabetic writing system need to learn the arbitrary sound-grapheme correspondences that capture the sound sequences in the speech stream (Castles et al., 2018; Kilpatrick, 2020). Phonological awareness also has been shown to be strongly associated with word reading accuracy for individuals with ID of mixed or unknown etiology (Pezzino et al., 2019; Saunders & DeFulio, 2007; Wise et al., 2010), Down syndrome (Cardoso-Martins & Frith, 2001; Lemons & Fuchs, 2010), and fragile X syndrome (Randel et al., 2015).

Similar findings have been consistently found for individuals with WS (Brawn et al., 2018; Laing et al., 2001; Levy et al., 2003; Levy & Antebi, 2004; Menghini et al., 2004; Mervis et al., 2021; Steele et al., 2013). For a sample of fifteen 9 – 27-year-olds with WS (mean age = 15.1 years), Laing et al. (2001) reported moderate to strong correlations between single word reading and both phoneme awareness and rhyme awareness, although neither of these relations remained significant after controlling for

chronological age and overall intellectual ability. For a sample of twenty 12 – 20-year-olds (mean age = 16.5 years) with WS, Levy and colleagues (2003) reported that the correlation between phonological awareness (as measured by a deletion task) and single word reading remained significant even after controlling for nonverbal reasoning ability. Levy and Antebi (2004) reported significant and moderate correlations between word reading ability and phonological awareness for seventeen 11 – 22-year-olds (mean age = 16.2 years) with WS who spoke Hebrew and were able to read at least a few words. Menghini et al. (2004) found similar results for sixteen 10 – 30-year-olds (mean age = 17.7 years) with WS who spoke Italian. A strong correlation between early phonological awareness for twenty-six 5 – 8-year-olds English speakers with WS and word reading one year later was reported by Steele et al. (2013). Brawn et al. (2018) reported strong and significant correlations between phonological awareness and all three word reading measures investigated in their study (regular words, irregular words, and pseudowords). Finally, Mervis et al. (2021) reported a strong correlation between phonological skills (measured by a composite of phonological awareness and verbal short-term memory) and word reading (measured by a composite of real words and pseudowords) for seventy 9-year-olds with WS.

Visual-spatial Perception

There have been relatively few studies investigating the role of visual-spatial perception skills in successful reading acquisition (Dessalegn et al., 2013; Treiman et al., 2014). Visual-spatial perception is clearly important for discriminating the letters of the alphabet, many of which (e.g., *b* and *d* , *M* and *W*) differ only with regard to the orientation of their features (see Kaufman, 1980; Lachmann & Geyer, 2003 for reviews).

There is evidence that visual-spatial perception is correlated with reading ability, particularly among young children. Fisher et al. (1985) found that visual-spatial perception skills, as measured by a matching to sample task in which children were asked to identify the figure that was turned in the same direction as the sample, was significantly correlated word recognition in a sample of kindergarteners, even after IQ was partialled out. Lee et al. (1986) found that visual-spatial perception was a unique significant predictor of reading abilities for English-speaking first graders even after accounting for the effect of individual differences in visual-spatial construction, auditory memory, and general knowledge. McBride-Chang et al. (2011) demonstrated that Spanish-speaking kindergarten children with high visual-spatial perception skills performed significantly better on word reading than children with low visual-spatial perception skills. Finally, in a longitudinal study, visual-spatial perception, measured before children began to read, significantly predicted word reading approximately 10 months later for English-speaking children (Ho & Bryant, 1999). After controlling for chronological age and IQ, Ho and Bryant (1999) found that performance on spatial orientation tasks differentiated children who were able to read from children who were not able to read 10 months later.

To the best of my knowledge, no previous studies have investigated the relation between visual-spatial perception ability and word reading for individuals with ID. As mentioned earlier, individuals with WS have relatively poor visual-spatial skills (see Landau & Ferrara, 2013 for a review). Dessalegn et al. (2013) suggested that these individuals' difficulties in visual-spatial perception might be particularly detrimental for reading acquisition. They based their claim on the results of a case-study of two 16-year-

olds with WS who differed markedly in word reading ability despite a similar overall level of phonological awareness skills and similar IQs on the KBIT. In contrast, they showed striking differences in visual-spatial perception tasks with non-alphabetic stimuli (including line and object orientation) with the poorer reader performing worse than the better-able reader. In line with these findings, the poorer reader showed a larger number of letter orientation and letter ordering errors than the better-able reader on the reading and spelling tasks (e.g., writing "baily" for "daily" and reading "never" for "nerve"). She also performed relatively more poorly on visual discrimination tasks involving reversed letters (e.g., *b* and *d*) or strings of letters (e.g., *bad*, *dad*).

Vocabulary

Vocabulary also plays an important role in word reading, with beginning readers struggling more with reading words that are not part of their oral vocabulary (Nation & Snowling, 2004). There are at least three reasons for this relation. First, vocabulary contributes to the development of phonological awareness. According to the Lexical Restructuring Model (Metsala & Walley, 1998) as vocabulary size increases, children's phonological representations need to become more segmented to allow for the differentiation of words that are phonologically similar. According to this model, phoneme awareness emerges as a consequence of vocabulary development. Second, to become a skilled reader, the child must bond meanings to spellings and pronunciations (Ehri, 2020). Third, vocabulary knowledge is essential to decipher words whose spelling does not follow the most common letter-sound correspondences (Nation & Snowling, 2004; Ricketts et al., 2007). Along these lines, vocabulary ability has been associated with word reading for TD children (Hjetland et al., 2019; Hulme et al., 2015; Nation &

Snowling, 2004; Steele et al., 2013; Zhao et al., 2017), individuals with ID of mixed or unknown etiology (Conners et al., 2001; Pezzino et al., 2019), Down syndrome (Cardoso-Martins et al., 2009), fragile X syndrome (Adlof et al., 2015), and WS (Laing et al., 2001; Steele et al., 2013). For example, Cardoso-Martins and colleagues (2009) reported strong correlations (Spearman rhos ranging from .58 to .78) between receptive vocabulary and the literacy abilities considered in their study for nineteen 10 – 19-year-olds with Down syndrome. Similarly, Laing et al. (2001) reported a correlation of .56 between single word reading and vocabulary for fifteen 9 – 27-year-olds with WS. In addition, Steele et al. (2013) reported a correlation of .58 between vocabulary of twenty-six 5 – 8-year-olds with WS and single word reading measured 12 months later.

In some studies, the effect of vocabulary on word reading ability has been found to be significant even after controlling for other relevant skills, such as nonverbal abiltiy and phonological skills (Nation & Snowling, 2004; Ouellette & Beers, 2009; Ricketts et al., 2007). However, other studies investigating the effect of vocabulary on word reading for TD children did not find a significant effect after accounting for other relevant skills such as initial word reading, phoneme awareness, and letter name/sound knowledge (Muter et al., 2004) or chronological age (Steele et al., 2013).

Given the strong correlation typically found between vocabulary and phonological skills (e.g., Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002), it is important to consider the possibility that vocabulary influences the development of word reading indirectly, via its effect on phonological awareness. Along these lines, Storch and Whitehurst (2002) evaluated the predictors of reading ability over the first four years of elementary school in a longitudinal study involving 624 TD

children. Oral language skills (as measured by receptive vocabulary, expressive vocabulary, and conceptual knowledge) in kindergarten significantly predicted concurrent code-related skills (including a phonological awareness measure), which in turn predicted Grade 1 word reading. In other words, code-related skills mediated the relation between oral language and word reading, indicating an indirect relation between oral language and word reading through code-related skills (see Hjetland et al., 2019 for similar results). Hulme et al. (2015) also found similar results for children at familial risk of dyslexia. The possibility of an indirect effect of vocabulary on word reading via phonological processing has not been considered for children with ID or WS.

Overall Intellectual Ability

Overall intellectual ability has a weak but consistent relation with word reading abilities for TD children (Hayiou-Thomas et al., 2006). However, as demonstrated by Melby-Lervåg et al.'s (2012) meta-analysis, this relation is probably due to the underlying links between IQ and other abilities that are critical for word reading such as phonological skills and oral language. Relatedly, IQ accounted for only a small amount of unique variance in predicting response to reading intervention for TD struggling readers in Stuebing et al.'s (2009) meta-analysis. Similarly, children with ID with higher IQs also tend to have better word reading skills than children with lower IQs (see Pezzino et al., 2019 for a review). However, consistent with findings for TD children, once IQ is controlled, cognitive abilities such as phonological awareness, verbal short-term memory, and rapid naming skills account for a significant amount of the variability in word reading of children with ID. Pezzino et al. (2019) concluded that individuals with ID are able to attain word reading skills using these cognitive abilities despite their low IQ.

For individuals with WS, word reading ability is also significantly correlated with IQ, with reported correlations ranging from moderate (Brawn et al., 2018) to strong (Laing et al., 2001; Levy & Antebi, 2004)). Similar to individuals with ID, when participants with WS were split into groups based on IQ, the higher-IQ group(s) had better single-word reading abilities than the lower-IQ group (Howlin et al., 1998; Levy et al., 2003; Udwin et al., 1987). No previous study including individuals with WS has investigated if overall IQ contributes to word reading above and beyond the effects of phonological and vocabulary skills.

Spelling Abilities: Typically Developing Children, Individuals with Intellectual Disability, and Individuals with Williams Syndrome

Accurate spelling is an important component of skilled writing, an ability contributing to academic and professional success (Cutler & Graham, 2008; Daffern et al., 2017). Learning to spell also enhances understanding of the alphabetic system, which is important for development of the ability to read by recoding letters or letter units into their corresponding sounds, which is key to securing accurate orthographic representations of words in memory (Graham & Santangelo, 2014). For this reason, learning to spell also contributes to the development of fluent reading. Improving spelling ability is not only important for literacy development in general, but spelling correctly also allows for more effective communication, and presence of spelling errors can lead to lower evaluation of the credibility and qualifications of the writer, negatively impacting, for example, hiring decisions (Pan et al., 2021). Nevertheless, despite its importance, relative to the research on word reading, the spelling abilities of TD children (Treiman, 2017) and of individuals with ID (Lindström & Lemons, 2021) is less often investigated.

Word spelling in English is inherently more difficult than word reading because the approximately 40 unique phonemes in English are symbolized by about 70 graphemes (letters/letter combinations) (Ehri, 2000). Therefore, if an individual has an orthographic representation of "*cofee*" for "*coffee*", they may be able to read the word correctly but would not spell it accurately (Galuschka et al., 2020). Despite the complexity of English spelling, in recent years, a number of schools have eliminated spelling tests and dropped traditional methods of explicit spelling instruction (Pan et al., 2021).

Given that spelling and word reading rely on similar processes, that is, the understanding of the alphabetic principle and the establishment of grapheme and phoneme relations, it is not surprising that there should be a strong relation between them. According to Ehri's Phase Theory (Ehri, 1987, 2005) children go through four phases during reading development: pre-alphabetic, partial alphabetic, full alphabetic, and consolidated alphabetic. With regard to spelling development (Ehri, 2020), children in the pre-alphabetic phase rely primarily on visual and contextual cues, spelling, for example, W^BC for *giraffe*. In this spelling, instead of using letters to represent sounds, the child is using letters to represent the nonalphabetic visual characteristics of a giraffe (e.g., having a long neck). In the partial alphabetic phase, children know most letter sounds and can use this knowledge to spell phonetically, for example, *JRF* for *giraffe*. In the full alphabetic phase, children can form phoneme-grapheme connections and can spell from memory. To reach the consolidated alphabetic phase, in which the child is able to read automatically by sight, children need to accumulate a large number of written words in their lexical memory that are consistently bonded to their pronunciations and meanings (Ehri, 2020). Similarly, according to Perfetti's (Perfetti & Hart, 2002) lexical quality

hypothesis, the stored mental representation of a word (lexical quality) includes the individual's spelling of the word, its phonology, and its meaning. According to this hypothesis, before acquiring higher lexical representations, the child has partial orthographic representations (as in the example above, spelling "*coffee*" as "*cofee*"). These orthographic representations become precise when the child is able to spell the represented words accurately (e.g., correct spelling of "*coffee*"). Improving orthographic representations based on experience with print is key for the transition from novice to expert reader, allowing for rapid and automatic recognition of individual words (Castles et al., 2018).

Consistent with these theories, spelling ability and reading ability are strongly correlated for TD children (Ehri, 2000; Pan et al., 2021; Peterson et al., 2021). Given this strong correlation, it is not surprising that systematic spelling instruction also improves reading as demonstrated by the results of Graham and Santangelo's (2014) meta-analysis that children who received explicit and systematic instruction in spelling also had significantly better reading skills than children who received no or unrelated (e.g., math) instruction or incidental approaches to improving spelling. Strong correlations between spelling and word reading performance also have been found for children with mild to moderate ID of non-specific etiology ($r = .94$; Henry & Winfield, 2010) and children with DS (rs ranging from .82 to .86; Byrne et al., 2002; Cardoso-Martins et al., 2009; Loveall & Conners, 2013).

Correlations between spelling and word reading have not been reported for individuals with WS. Little is known about the spelling abilities of individuals with WS, which have been addressed in only six studies (Howlin et al., 1998; Laing et al., 2001;

Pagon et al., 1987; Polse, 2013; Udwin et al., 1987, 1996) and one case comparison report (Dessalegn et al., 2013). All but one (Polse, 2013) of these studies reported spelling age or grade equivalent instead of SSs. In general, findings indicated that spelling abilities of individuals with WS vary widely. Some of these studies suggested that spelling age or grade equivalents were younger than reading age or grade equivalent (Howlin et al., 1998; Udwin et al., 1987). In addition, age equivalents were related to IQ (Howlin et al., 1998). However, results from these studies are difficult to interpret due to psychometric problems with the use of age equivalents or grade equivalents rather than SSs (Mervis & Robinson, 2005). In particular, age or grade equivalents are not measured on an interval scale and are not comparable across different assessments or even across subtests from a single assessment. No study to date has reported correlations between spelling ability and reading ability for individuals with WS.

Impact of Method of Reading Instruction on Literacy Abilities: Typically Developing Children, Individuals with Intellectual Disability, and Individuals with Williams Syndrome

Two seminal meta-analyses that investigated the effectiveness of various approaches for teaching children to read based on the research literature provided evidence of a moderate positive impact of phonemic awareness (Ehri, Nunes, Willows, et al., 2001) and phonics instruction (Ehri, Nunes, Stahl, et al., 2001) on word reading for TD children relative to instruction based on approaches that included less phonics or no phonics. Similar results have been consistently demonstrated for spelling, with systematic phonics instruction having a positive effect on spelling (see Galuschka et al., 2020; Graham et al., 2018 for meta-analysis). The findings from these meta-analyses (see also

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Castles et al., 2018) indicate that TD children who are taught to read using systematic phonics instruction, which involves teaching letter-sound relations systematically, have significantly better word reading, spelling, and reading comprehension abilities than children who are taught to read using literacy approaches that do not teach letter-sound relations systematically.

Similar to the meta-analytic findings for TD children, findings from a recent meta-analysis of beginning reading interventions for children and adolescents with ID ranging in age from 4 – 21 years (Reichow et al., 2019) indicated that relative to instruction-as-usual, interventions including elements of phonological awareness, letter sound instruction, and decoding had a moderate positive effect on word reading (see also Sermier Dessemontet et al., 2021). Importantly, phonological awareness and systematic phonics-based instruction have been demonstrated to be feasible for those with fragile X syndrome (Adlof et al., 2018) and Down syndrome (LeJeune et al., 2018). Consistent with those findings, according to Stuebing et al.'s (2009) meta-analysis, IQ accounts for only a small amount (1% to 3%) of unique variance in struggling readers' response to reading intervention.

Systematic phonics instruction also has a positive relation with word reading and reading comprehension for children and adolescents with WS. This relation has been investigated in only two studies (Mervis, 2009; Mervis et al., 2021). In both studies, the children who were taught to read using a systematic phonics approach had significantly higher SSs on both word reading and reading comprehension than did children taught to read with other approaches, even after taking into account differences in overall IQ. No

studies have been reported that investigated the relation between systematic phonics instruction and spelling for individuals with ID or individuals with WS.

Despite the strong research evidence supporting systematic phonics-based instruction for both TD children (Castles et al., 2018; Ehri, 2020; Ehri, Nunes, Stahl, et al., 2001) and children with ID (Reichow et al., 2019), a substantial proportion of early childhood teachers do not think that explicitly teaching the relations between letters and sounds to children, or using systematic phonics instruction, is the best approach to teaching children how to read (Campbell, 2020; see also Cunningham & O'Donnell, 2015). In addition, a substantial proportion of early childhood teachers do not themselves know phonics (Brady et al., 2009; Campbell et al., 2014; Cunningham et al., 2004, 2009; Nelson & Machek, 2007; Porter et al., 2021), which in combination with their negative beliefs, makes it even less likely they will use this approach to teach their students. As clearly highlighted by Castles et al. (2018) and Seidenberg et al. (2020), although there is substantial research supporting the systematic phonics approach, this knowledge is not always translated into the schools. This problem clearly spills over into the instruction that is given to children with ID. For example, Wakeman and colleagues (2021) reported that only 44% of the 82 parents of elementary school-aged children with ID in their study thought their child was receiving phonics/decoding instruction, and only 46% of the 9-year-olds with WS in the Mervis et al. (2021) study were receiving systematic phonics instruction.

Dissertation Studies

The two studies included in this dissertation focus on the literacy abilities of children and adolescents with WS. In Study 1 (Chapter 2), I investigated the longitudinal

predictors of word reading ability for young children with WS. In Study 2 (Chapter 3), I

investigated the spelling ability of children and adolescents with WS and relations with

word reading ability and method of reading instruction.

CHAPTER II

LONGITUDINAL PREDICTORS OF WORD READING FOR CHILDREN WITH

WILLIAMS SYNDROME

Acquiring functional literacy is foundational to promoting the inclusion of

individuals in society (Castles et al., 2018). For individuals with intellectual disabilities

(ID), functional word-reading ability provides increased opportunities for independent

living and employment and enhances quality of life (Lindström & Lemons, 2021). The

word-reading abilities of adolescents and adults with ID vary widely, from not being able

to read at all to reading at the level expected for age (see Pezzino et al., 2019 for a

review). Understanding the early predictors of this variability is foundational to providing

guidance for targeted assessments and interventions that promote the development of

functional literacy.

In the present study, we examined the relations of cognitive, language, and

reading instruction factors to early word-reading ability in children with Williams

syndrome (WS), a rare genetic disorder associated with mild to moderate ID (Kozel et al.,

2021). Similarly to what has been found for individuals with ID in general, reading

ability ranges widely in WS, with only about 30% of older teenagers and adults attaining

functional literacy (Brawn et al., 2018). The results of previous studies suggest that the

factors that have been associated with word-reading ability in typically developing

children also are associated with word-reading ability in WS (e.g., Brawn et al., 2018;

Mervis et al., 2021). To begin to identify early predictors of word-reading ability in children with WS, longitudinal studies are needed. The findings from the only published longitudinal study (Steele et al., 2013) are difficult to interpret due to serious methodological concerns. In the present paper, we report findings from a longitudinal study addressing the contributions of early cognitive, language, and instructional factors previously shown to be important for TD children to later word-reading ability in a relatively large sample of similar aged children with WS. All cognitive and language variables included were measured by age-appropriate standardized assessments.

Predictors of Word Reading for Typically Developing Children and Individuals with Intellectual Disability

Phonological Awareness

Phonological awareness is the ability to consciously attend to and manipulate speech sounds. This ability is clearly important for learning to read in an alphabetic orthography (Ehri, 2014) and is strongly associated with concurrent word reading for TD children (see Melby-Lervåg et al., 2012 for a meta-analysis). Longitudinal correlations between early phonological awareness and later word-reading ability for TD children range from moderate to strong (Caravolas et al., 2013; Clayton et al., 2020), remaining significant even after controlling for word reading at Time 1 (Steele et al., 2013). When measured prior to or at the start of formal schooling, phonological awareness and letter knowledge (which are strongly correlated) are usually the best predictors of how well children will learn to read during the first years of instruction (Ehri, Nunes, Willows, et al., 2001).

The results of cross-sectional studies have also shown that phonological awareness is strongly correlated with word reading for children (Saunders & DeFulio, 2007) and adults (Wise et al., 2010) with ID of mixed etiology, children with Down syndrome (DS; Cardoso-Martins & Frith, 2001; see Næss, 2016 for a meta-analysis), and children with fragile X syndrome (see Randel et al., 2015 for a systematic review). Significant longitudinal correlations also have been reported for children with DS (Lemons & Fuchs, 2010; Steele et al., 2013) and individuals with ID of mixed etiology (Pezzino et al., 2019).

Visual-spatial Perception

Visual-spatial perception skill is important for discriminating the letters of the alphabet, many of which (e.g., *b* and *d*, *M* and *W*) differ only in the orientation of their features. Given that changes in orientation are in general irrelevant for identifying objects in the world (Treiman et al., 2014), it is not surprising that most young TD children, at some point, confuse letters that represent reversals (see Lachmann & Geyer, 2003 for a review). Although the contribution of visual-spatial perception ability to word reading ability is much less often studied than the contribution of phoneme awareness, findings from several studies document that visual-spatial perception is significantly correlated with reading ability, particularly among young children. For example, Fisher et al. (1985) found that visual-spatial perception was significantly correlated with printed word recognition in a sample of kindergarteners, even after controlling for IQ. Lee et al. (1986) found that visual-spatial perception was a significant unique predictor of word-reading abilities for English-speaking first graders even after accounting for individual differences in visual-spatial construction, auditory memory, and general knowledge.

McBride-Chang et al. (2011) demonstrated that Spanish-speaking kindergarten children with high visual-spatial perception skills performed significantly better on word reading than children with low visual-spatial perception skills. In a longitudinal study, Ho and Bryant (1999) found that visual-spatial perception, measured before the participants began to read, significantly predicted word reading approximately 10 months later. To the best of our knowledge, no previous studies have investigated the relation between visual-spatial perception ability and reading for individuals with ID.

Vocabulary

Previous studies have reported cross-sectional (e.g., Zhao et al., 2017) and longitudinal (Hjetland et al., 2019; Steele et al., 2013; Storch & Whitehurst, 2002) correlations between oral language skills, including vocabulary, and word-reading abilities for TD children. Cross-sectional (Boudreau, 2002; Hulme et al., 2012) and longitudinal (Hulme et al., 2012; Steele et al., 2013) correlations between vocabulary and word reading also have been found for children with DS. However, these simple correlations do not necessarily result in a unique effect of vocabulary on word reading for TD children after accounting for the effect of variations in other relevant skills such as initial word reading, phoneme awareness, and letter-name/sound knowledge (see Muter et al., 2004) or chronological age (see Steele et al., 2013).

Findings from longitudinal studies of TD children have indicated that vocabulary influences the development of word reading indirectly, via its effect on phonological awareness. In this manner, phoneme awareness emerges as a consequence of vocabulary development. Storch and Whitehurst (2002) evaluated the predictors of reading ability over the first four years of elementary school. Oral language, as measured by receptive

vocabulary, expressive vocabulary, and conceptual (relational) vocabulary in kindergarten, concurrently predicted code-related skills, including phonological awareness, which in turn predicted Grade 1 word reading. Hjetland et al. (2019) reported similar findings in a study examining the longitudinal predictors of reading comprehension in Norwegian-speaking children. Hulme et al. (2015) found that for both TD children and children at high risk for reading disability, oral language (including vocabulary) measured in preschool predicted phonological awareness one year later, which in turn predicted word-reading skills at 5.5 years. This indirect effect of vocabulary on word reading through its effect on phonological processing is consistent with the Lexical Restructuring Model (Metsala & Walley, 1998). According to this model, as vocabulary size increases, the child's phonological representations become more segmented to allow for the differentiation of words that are phonologically similar.

Overall Intellectual Ability

Overall intellectual ability has a weak but consistent relation with word-reading abilities for TD children (Hayiou-Thomas et al., 2006). When children with ID are divided into groups based on IQ, the higher-IQ group typically demonstrates better word-reading skills than the lower-IQ group (see Pezzino et al., 2019 for a review). At the same time, phonological awareness has been found to predict word-reading abilities above and beyond IQ for both TD children (Greiner de Magalhães et al., 2021) and children with ID (Pezzino et al., 2019). Relatedly, IQ accounts for only a small amount of unique variance in predicting response to reading intervention for TD struggling readers (see Stuebing et al., 2009 for a meta-analysis). Additionally, some children with ID have age-appropriate word-reading skills (e.g., Share et al., 1989; Stuebing et al., 2009).

Reading Instruction Method

There is a large body of evidence demonstrating that systematic phonics instruction, which involves teaching letter-sound relations systematically, is a significantly more effective method to teach word reading to TD children than approaches that do not teach these relations systematically (e.g., Castles et al., 2018; Ehri, Nunes, Stahl, et al., 2001). Similarly, students with ID receiving systematic phonics instruction made significantly greater progress in word reading than students receiving either the type of instruction typically provided by their district (Allor et al., 2014; Hunt et al., 2020) or whole-word instruction (Browder et al., 2012). This pattern held for both children with mild ID (Allor et al., 2014) and children with moderate to severe ID (Browder et al., 2012; Hunt et al., 2020).

Predictors of Word Reading: Individuals with Williams Syndrome

WS is caused by a hemideletion of 25 – 27 genes on chromosome 7q11.23 (Kozel et al., 2021). Individuals with WS typically have mild to moderate ID, although the full range is from severe ID to average intellectual ability (Mervis & John, 2010). Relative to their overall intellectual ability, individuals with WS typically show strengths in concrete vocabulary, nonverbal reasoning, verbal short-term memory, and phonological processing but weaknesses in spatial abilities and relational and conceptual language (Mervis & Greiner de Magalhães, in press).

The literature on reading abilities of individuals with WS is limited (see Mervis, 2009, and Brawn et al., 2018, for comprehensive reviews). Previous cross-sectional studies have generally shown that factors that contribute to individual differences in word-reading ability for TD children also contribute to individual differences in the

word-reading abilities of individuals with WS (e.g., Mervis et al., 2021). For example, strong correlations have been found between word-reading skills and phonological awareness (Brawn et al., 2018; Laing et al., 2001; Levy et al., 2003; Levy & Antebi, 2004; Menghini et al., 2004; Mervis et al., 2021). Levy et al. (2003) reported that the correlation between phoneme awareness and word reading remained significant even after controlling for nonverbal reasoning ability. Mervis et al. (2021) reported that phonological processing skills contributed significant unique variance to word-reading ability even after the effects of reading instruction method, vocabulary, nonverbal reasoning, visual-spatial construction, verbal working memory, and rapid naming were taken into account.

Visual-spatial perception has been considered in two studies of the word-reading abilities of adolescents or adults with WS. Brawn et al. (2018) found a significant moderate correlation between concurrent visual-spatial perception abilities and word reading. Dessalegn et al. (2013) reported a case-study of two 16-year-olds with WS who differed markedly in word-reading ability despite similar overall phonological awareness skills and IQ. The stronger reader performed strikingly better than the weaker reader on visual-spatial perception tasks focused on line orientation, design orientation, and discrimination of mirror-image letters (e.g., *b* and *d*). On word- and nonword-reading tasks, 10% of the weaker reader's errors (but only 1% of the stronger reader's errors) involved misperception of letter orientation (e.g., reading "drooch" for "brooch").

Previous studies have reported moderate correlations between vocabulary and concurrent single-word reading ability for individuals with WS (Laing et al., 2001; Levy et al., 2003; Mervis et al., 2021). However, vocabulary did not contribute unique variance

to word-reading skills after accounting for the contribution of reading instruction method, phonological skills, nonverbal reasoning, visual-spatial skills, verbal working memory, and rapid naming (Mervis et al., 2021).

For individuals with WS, word-reading ability also is significantly associated with overall intellectual ability. Correlations between single-word reading and overall intellectual ability vary from moderate (Brawn et al., 2018) to strong (Laing et al., 2001; Mervis et al., 2021). When participants with WS were split into groups based on IQ, the higher-IQ group(s) had significantly better single-word reading abilities than the lower-IQ group(s) (Howlin et al., 1998; Levy et al., 2003; Udwin et al., 1987). However, Mervis et al. (2021) found that the correlation between word reading and reading instruction method was significantly stronger than the correlation between word reading and overall intellectual ability. In addition, the correlation between word reading and reading instruction method remained significant and strong even after controlling for overall intellectual ability. In contrast, overall intellectual ability was only weakly correlated with word reading after controlling for reading instruction method. As suggested by the results discussed above (e.g., Dessalegn et al., 2013; Mervis et al., 2021), it is unlikely that overall IQ is the main factor determining word-reading performance in WS.

Only one study has considered the cognitive predictors of word-reading ability for children with WS longitudinally. Steele and colleagues (2013) assessed 26 children with WS twice, first when they were 5 – 8 years of age (mean age = 6.6 years) and then 12 months later. Single-word reading raw scores improved from Time 1 to Time 2, and Time 2 word reading correlated strongly with Time 1 phonological awareness, letter

name/sound knowledge, and receptive vocabulary raw scores. The correlation between Time 1 vocabulary and Time 2 word reading remained significant after controlling for Time 1 word reading. Parallel analyses were not performed for Time 1 phonological awareness or Time 1 letter-name/sound knowledge. Instead, both Time 1 word reading and Time 1 verbal mental age equivalent were controlled, after which the correlations between Time 1 phonological awareness or Time 1 letter knowledge with Time 2 word reading were no longer statistically significant. It is difficult to interpret these findings, however. No explanation was provided either for how verbal mental age was determined or the reason for controlling for it. Furthermore, there are serious psychometric problems with the use of age equivalents (AEs) in statistical analyses (see Brawn et al., 2018; Mervis & Robinson, 2005). In addition, although raw scores rather than standard scores (SSs) were used, chronological age was not controlled for, and the sample size was relatively small.

Current Study

Reading ability varies widely in WS, ranging from inability to read any words to reading at the level expected for age (e.g., Howlin et al., 1998; Laing et al., 2001; Mervis et al., 2021; Pagon et al., 1987). Given the positive outcomes associated with literacy ability for adolescents and adults with WS (Brawn et al. 2018), identification of the factors that affect the later word-reading abilities of children with this syndrome is crucial. As described earlier, methodological problems with the only previous study addressing the longitudinal predictors of single-word reading ability in children with WS (Steele et al., 2013) make the findings difficult to interpret. In the present study, we

included a considerably larger sample of children with WS and used age-appropriate standardized assessments, with performance measured by SSs or T-scores.

Our goal was to determine if previously identified longitudinal predictors of word-reading ability in TD children also predict later word-reading ability in children with WS. To address this goal, children with WS were evaluated at two time points. At Time 1, when the children were 6 or 7 years old, they completed assessments of several cognitive and language abilities previously found to affect word-reading achievement in TD children and/or children with WS. The primary instructional approach used to teach word reading also was determined. Children's word-reading abilities were assessed on average three years later, at Time 2.[1]

The following predictors were considered: phonological awareness, visual-spatial perception, overall intellectual ability, vocabulary, and reading instruction method. Based on prior cross-sectional or longitudinal findings for TD children (e.g., Ehri, Nunes, Stahl, et al., 2001; Ehri, Nunes, Willows, et al., 2001; Greiner de Magalhães et al., 2021; Melby-Lervåg et al., 2012) or children with WS (Brawn et al., 2018; Mervis et al., 2021; Steele et al., 2013), we predicted that both phonological awareness and reading instruction method would significantly predict later word-reading ability in children with WS. We also expected to find a significant effect for visual-spatial perception, which has been found to be significantly related to word-reading ability for both young TD children (e.g., Fisher et al., 1985; Ho & Bryant, 1999; Lee et al., 1986) and individuals with WS (Brawn et al., 2018; Dessalegn et al., 2013).

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[1] The primary instructional approach at Time 2 was the same as at Time 1 for all of the participants.

Based on prior findings for TD children (e.g., Muter et al., 2004) and children with WS (Mervis et al., 2021), we predicted that individual differences in vocabulary at Time 1 would not contribute uniquely to word-reading ability at Time 2, after accounting for the other predictors. At the same time, based on the results of Storch & Whitehurst (2002), we predicted that vocabulary at Time 1 would predict word-reading abilities at Time 2 indirectly, via its effect on Time 1 phonological awareness.

Method

Participants

The final sample included 69 children (31 girls, 38 boys) with genetically-confirmed classic-length deletions of the WS region. The participants ranged in age from 6.01 to 7.87 years ($M = 6.53$, $Mdn = 6.37$, $SD = 0.54$) at Time 1 and from 9.01 to 10.79 years ($M = 9.47$, $Mdn = 9.30$, $SD = 0.44$) at Time 2. Except for one child whose native language was Chinese but was fluent in English at Time 1, all participants were native speakers of English. Sixty-one of the participants were also included in the cross-sectional study reported in Mervis et al. (2021). For 55 of them, the Time 2 assessment in the present study is the same assessment that was included in Mervis et al. (2021). Participants lived in 23 different U. S. states, representing all U.S. census regions (26.1% Northeast, 42.0% South, 20.3% Midwest, 7.2% West) and two Canadian provinces (4.3%). The distribution of participants' racial/ethnic background was: 78.3% White non-Hispanic, 8.7% White Hispanic, 1.4% African-American non-Hispanic, 5.8% multiracial non-Hispanic, and 5.8% multiracial Hispanic. Eighteen of the participants' mothers (26.1%) did not have a bachelor degree; the remaining 51 (73.9%) had earned at least a bachelor degree.

Children were included in the present study if they had a classic WS deletion, had completed an assessment when they were between 6.00 and 7.99 years of age, had completed another assessment between ages 9.00 and 10.99 years, and both assessments had included all measures used in this study. The time between assessments ranged from 1.99 to 3.43 years (mean = 2.93 years, *Mdn* = 3.01, *SD* = 0.33). Some children were assessed at both 6 and 7 years and/or both 9 and 10 years. For these children, the set of assessments that was closest to three years apart was used. Two children who met the inclusion criteria were excluded, one because he was nonverbal so could not complete the reading assessment and one because he also had fetal alcohol spectrum disorder. Data collection for Time 1 began in June 2007 and ended in November 2017. Data collection for Time 2 began in July 2010 and ended in February 2020.

The participants' median grade in school at Time 1 was Kindergarten with a range from the winter of Pre-kindergarten to the winter of 2nd grade. At Time 2, median grade was 3rd, with a range from the summer after 1st grade to 5th grade. Primary classroom placement at Time 2 was in a mainstream class for 44 children (19 with reading instruction primarily in the mainstream, 25 with reading instruction primarily in a resource room or other special education classroom) and in a special education (self-contained) class for 22 children (all with reading instruction in a special education classroom). The three remaining children were homeschooled.

Measures

Time 1: Independent Variables

Intellectual Ability. The Differential Ability Scales-II (DAS-II; Elliott, 2007) Early Years version was used to evaluate children's overall intellectual ability. The DAS-

II estimates a child's General Conceptual Ability (GCA; similar to Full-Scale IQ) based on performance on subtests measuring verbal, nonverbal reasoning, and spatial abilities. Performance is reported as a SS (general population mean = 100, SD = 15). The IRT-based internal consistency coefficients for 6- and 7-year-olds in the norming sample were .95 and .96, respectively.

Phonological Awareness. The DAS-II includes a supplemental Phonological Processing subtest which assesses knowledge of the sound structure of the English language and the ability to manipulate sounds. Four types of skills are assessed: rhyming, blending, deletion, and phoneme identification and segmentation. The Phonological Processing subtest yields an overall T-score (general population mean = 50, SD = 10). The IRT-based internal consistency coefficients for 6- and 7-year-olds in the norming sample were .94 and .90, respectively.

Visual-spatial Perception. The DAS-II Early Years Matching Letter-like Forms subtest is a supplemental subtest that assesses visual discrimination and awareness of the spatial orientation of asymmetric letter-like figures, measuring the child's ability to discriminate between different orientations of the same form. In this subtest, the child is shown a target figure (an asymmetric letter-like form resembling either an English or a Greek letter) and asked to indicate the identical match from the six figures shown below the target. The five distracters are transpositions of the original figure: a reversal, a 180° rotation, a 180° rotation and reversal, a 45° rotation, and a 315° rotation. The original figure remains visible while the child is making a selection. This subtest yields an overall T-score. For TD children, the rate of errors on a similar matching letter-like forms task used by Gibson et al. (1962) was strongly correlated with the number of errors made in

matching real letters ($r = .87$). The IRT-based internal consistency coefficients for 6- and 7-year-olds in the norming sample were .77 and .64, respectively.

Vocabulary. The Peabody Picture Vocabulary Test-4 (PPVT-4; Dunn & Dunn, 2007) is a measure of single-word receptive vocabulary. The Expressive Vocabulary Test-2 (EVT-2; Williams, 2007), which was co-normed with the PPVT-4, is a measure of single-word expressive vocabulary. Vocabulary ability was measured by a composite based on the mean of each child's PPVT-4 and EVT-2 SSs. For the present participants, the correlation between PPVT-4 and EVT-2 SSs was $r = .84$, $p < .001$. Split-half internal consistency for 6- and 7-year-olds in the norming sample ranged from .94 – .95 for the PPVT-4 and from .90 – .95 for the EVT-2.

Reading Instruction Method. The primary approach to teaching reading to each child was classified as Systematic Phonics (hereafter, Phonics) or Other following the procedure described in Mervis et al. (2021). The primary reading instruction approach was Phonics for 35 (50.7%) participants and Other for 34 (49.3%) participants. Mean chronological age was 6.54 years ($SD = 0.60$) for the Phonics group and 6.52 years ($SD = 0.48$) for the Other group at Time 1 and 9.47 years ($SD = 0.46$) for the Phonics group and 9.46 years ($SD = 0.41$) for the Other group at Time 2. The two groups did not differ significantly in chronological age at either Time 1, $t(67) = 0.18$, $p = .855$, Cohen's $d = 0.04$; or at Time 2, $t(67) = 0.16$, $p = .876$, Cohen's $d = 0.02$. A Mann-Whitney U test indicated that the distribution of grade did not differ significantly as a function of Reading Instruction Method group either at Time 1 (Mdn = Kindergarten, IQR = Kindergarten – Kindergarten for each group), $z = 0.32$, $p = .752$, Cohen's $d = 0.08$, or at

Time 2 (Mdn = 3rd grade, IQR = 2nd grade – 3rd grade for each group), z = 0.20, p = .844,

Cohen's d = 0.05.

Time 2: Dependent Variable

Single-word Reading. Word reading was measured by the Wechsler Individual

Achievement Test-III (WIAT-III; Wechsler, 2009) Basic Reading Composite SS. The

WIAT-III Basic Reading Composite includes two subtests, one measuring single real-

word reading (Word Reading) and one measuring pseudoword decoding (Pseudoword

Decoding). The standardized ceiling rule of four consecutive failed items leads to

discontinuation of each subtest. For the present participants, the correlation between

Word Reading SS and Pseudoword Decoding SS was r = .90, p < .001. According to the

WIAT-III technical manual, split half internal consistency for both Word Reading and

Pseudoword Decoding was .98 for the 9-year-olds in the norming sample and .97 for the

10-year-olds. Split half internal consistency for Basic Reading Composite was .99 for the

9-year-olds in the norming sample and .98 for the 10-year-olds.

Procedure

The study protocol was reviewed and approved by the university's Institutional

Review Board. Parents or legal guardians of all participants provided written informed

consent and participants provided oral or written assent. Children completed the

standardized measures at the senior author's laboratory as part of a larger two-day

assessment. All measures were administered by trained doctoral students or research

assistants and scored according to the standardized procedures detailed in the assessment

manuals.

Results

Data were analyzed using IBM SPSS v. 27.

Performance on Standardized Assessments

Descriptive statistics for all measures are provided in Table 1. There was considerable variability, with scores on each measure ranging from average or above average for the general population to moderate-severe disability. As indicated in the Introduction, there are serious psychometric concerns regarding AE scores (e.g., Brawn et al., 2018; Mervis & Robinson, 2005). However, as AEs are the only statistical measure provided in most of the prior studies of the reading abilities of individuals with WS, nonparametric descriptive statistics for the WIAT-III Word Reading and Pseudoword Decoding subtests AEs are provided in Table 2 for comparison, along with the corresponding nonparametric descriptive statistics for SSs on the same measures.

To compare children's real word-reading ability to their pseudoword-reading ability, a dependent *t*-test comparing WIAT-III Word Reading SS (floored at the lowest possible Pseudoword Decoding SS for children in the Time 2 age range) to WIAT-III Pseudoword Decoding SS was conducted. Mean SSs were 75.59 (*SD* = 13.73) for Word Reading and 74.88 (*SD* = 13.08) for Pseudoword Decoding. The difference was not statistically significant, $t(68) = 1.00$, $p = .320$, Cohen's $d = 0.05$.

Table 1

Descriptive Statistics for Time 1 and Time 2 Measures

Variable	Mean	Median	SD	Range
Time 1				
DAS-II Phonological Processing T	41.29	45.00	13.43	10 – 66
DAS-II Matching Letter-like Forms T	36.26	37.00	13.94	10 – 72
DAS-II General Conceptual Ability SS	67.33	69.00	13.64	31 – 96
Vocabulary SS	86.57	88.00	12.58	45.5 – 122
Time 2				
WIAT-III Basic Reading composite SS	74.52	73.00	13.68	52 – 109
WIAT-III Word Reading SS	75.01	75.00	14.53	50 – 112
WIAT-III Pseudoword Decoding SS	74.88	73.00	13.08	59 – 107

Note. N = 69. DAS-II = Differential Ability Scales-II; T = T-score; SS = standard score; WIAT-III = Wechsler Individual Achievement Test-III.

Table 2

Descriptive Statistics for WIAT-III Age Equivalents and Standard Scores

Variable	Median	Interquartile Range	Range
Age Equivalent			
Word Reading	6.80 yrs	6.00 – 7.80 yrs	< 6.00[a] – 13.00 yrs
Pseudoword Decoding	6.40 yrs	< 6.00[a] –7.40 yrs	< 6.00[a] – 12.40 yrs
Standard Score			
Word Reading	75.00	61.50 – 86.00	50 – 112
Pseudoword Decoding	73.00	63.00 – 86.50	59 – 107
Basic Reading Composite	73.00	63.00 – 84.50	52 – 109

Note. N = 69. Age equivalents are not available for Basic Reading Composite. WIAT-III = Wechsler Individual Achievement Test-III; yrs = years.
[a] Lowest possible age equivalent.

Multiple Regression Analyses

Maternal Education level was not significantly correlated with Basic Reading SS ($r = .05$, $p = .657$). Therefore, Maternal Education was not included in the multiple regression models. To investigate the longitudinal predictors of single-word reading for 9–10-year-olds with WS, two multiple regression analyses were performed with Basic Reading SS at Time 2 as the dependent variable. All continuous predictors were centered on the sample mean.

For the first multiple regression analysis, four independent variables were included in the model: phonological awareness (as measured by Time 1 Phonological Processing T-score), visual-spatial perception ability (as measured by Time 1 Matching Letter-like Forms T-score), intellectual ability (as measured by Time 1 GCA), and Time 1 reading instruction method. Given Steele et al.'s (2013) finding of a significant longitudinal effect of vocabulary on word reading, an additional multiple regression analysis was performed. For this analysis, Time 1 Vocabulary SS was included along with Phonological Processing T-score, Matching Letter-like Forms T-score, and reading instruction method. Given the very high correlation between GCA and Vocabulary SS ($r = .83$), GCA was not included in the second regression analysis.

All assumptions of multiple linear regression analyses were met. Cohen's f^2 was used to measure effect size (0.02 = small effect, 0.15 = medium, 0.35 = large; Cohen, 1988). Pearson correlations ($\alpha = .01$) among the variables included in the regression analyses are reported in Table 3. All correlations were statistically significant.

The results of the first analysis are presented in Table 4. Phonological Processing T-score (medium effect), Matching Letter-like Forms T-score (small effect), and reading

instruction method (large effect) made significant independent contributions to the variance in Basic Reading SS. GCA did not explain significant unique variance in Basic Reading SS after accounting for the other predictors. After controlling for the effects of the remaining independent variables, a 1-point increase in Phonological Processing T-score resulted in a .30-point increase in Basic Reading SS. After controlling for the effects of the other independent variables, a 1-point increase in Matching Letter-like Forms T-score resulted in a .21-point increase in Basic Reading SS. Finally, after controlling for the remaining independent variables, Phonics instruction resulted in a Basic Reading SS 16.42 points higher than Other reading-instruction approaches.

The results for the second multiple regression analysis are reported in Table 5. Phonological Processing T-score (small effect), Matching Letter-like Forms T-score (small effect), and reading instruction method (large effect) made significant independent contributions to the variance in Basic Reading SS. Vocabulary SS did not explain significant unique variance in Basic Reading SS after accounting for the effect of individual differences in the other predictors.

Analyses excluding children who were not able to read at least one of the real words ($n = 3$) and/or pseudowords ($n = 11$) yielded the same pattern of results (see Appendix). In addition, as reported in the Appendix, regression analyses conducted separately for the two subtests included in the Basic Reading Composite (Word Reading and Pseudoword Decoding) resulted in the same pattern of findings as for the composite measure.

Table 3

Bivariate Correlations Among the Measures Included in the Regression Analyses

Measure	2	3	4	5	6	7	8
1. Reading instruction method	.40*	.37*	.41*	.34*	.78**	.75**	.77**
2. DAS-II Phonological Processing T		.52**	.72**	.72**	.62**	.61**	.60**
3. DAS-II Matching Letter-like Forms T			.70**	.65**	.55**	.53**	.55**
4. DAS-II General Conceptual Ability SS				.83**	.56**	.54**	.56**
5. Vocabulary SS					.53**	.51**	.54**
6. WIAT-III Basic Reading Composite SS						–	–
7. WIAT-III Word Reading SS							.90**
8. WIAT-III Pseudoword Decoding SS							

Note. N = 69. DAS-II = Differential Ability Scales-II; T = T-score; SS = standard score; WIAT-III = Wechsler Individual Achievement Test-III.
* *p* < .01. ** *p* < .001

Table 4

First Multiple Regression Analysis Predicting WIAT-III Basic Reading Composite

Standard Score

Predictor	B	t	p-value	95% CI for B	Semi-partial r	Cohen's f^2
Constant	66.19	51.21	< .001	[63.61, 68.77]		
Phonological Processing T	0.30	3.19	.002	[0.11, 0.48]	.20	0.16
Matching Letter-like Forms T	0.21	2.33	.023	[0.03, 0.38]	.15	0.09
General Conceptual Ability SS	-0.04	-0.38	.707	[-0.26, 0.18]	-.02	< 0.01
Reading instruction method	16.42	8.59	< .001	[12.60, 20.25]	.54	1.15

$R^2 = .75$, adjusted $R^2 = .73$, $F (4, 64) = 47.19$, $p < .001$

Note. N = 69. All continuous independent variables were centered on the sample mean. WIAT-III = Wechsler Individual Achievement Test-III; CI = confidence interval; T = T-score; SS = standard score.

Table 5

Second Multiple Regression Analysis Predicting WIAT-III Basic Reading Composite

Standard Score

Predictor	B	t	p-value	95% CI for B	Semi-partial r	Cohen's f^2
Constant	63.66	6.59	< .001	[44.37, 82.95]		
Phonological Processing T	0.26	2.78	.007	[0.07, 0.45]	.18	0.12
Matching Letter-like Forms T	0.18	2.15	.035	[0.01, 0.34]	.14	0.07
Vocabulary SS	2.56	0.27	.789	[-16.53, 21.65]	.02	< 0.01
Reading instruction method	16.37	8.58	< .001	[12.56, 20.18]	.54	1.15

$R^2 = .75$, adjusted $R^2 = .73$, $F (4, 64) = 47.12$, $p < .001$

Note. $N = 69$. All continuous independent variables were centered on the sample mean. WIAT-III = Wechsler Individual Achievement Test-III; CI = confidence interval; T = T-score; SS = standard score.

Mediation Analysis

To examine if vocabulary had an indirect longitudinal effect on word reading via phonological awareness, as has been demonstrated for TD children (e.g., Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002), a simple mediation analysis was conducted using ordinary least squares path analysis (Hayes, 2018). Results indicated that Time 1 Vocabulary SS indirectly influenced Time 2 Basic Reading SS through its effect on Time 1 Phonological Processing T-score. As can be seen in Figure 1 and Table S1, children who had higher Time 1 Vocabulary SSs had higher Time 1 Phonological Processing T-scores ($a = 0.77$), and children who had higher Time 1 Phonological Processing T-scores also had higher Time 2 Basic Reading SSs even after controlling for Time 1 Vocabulary SS ($b = 0.49$). A percentile bootstrap 95% confidence interval for the indirect effect ($ab = 0.37$) based on 5,000 bootstrap samples with seed number 25071990 was entirely above zero (0.18 to 0.61). There was no evidence that Time 1 Vocabulary SS significantly influenced Time 2 Basic Reading SS independent of its effect on Time 1 Phonological Processing T-score ($c' = 0.21$, $p = .169$). As reported in the Appendix, the same pattern of results was found when vocabulary ability was measured by a composite that incorporated performance on a measure of relational (conceptual) vocabulary along with performance on the PPVT-4 and the EVT-2. This composite was similar to the measure used by Storch and Whitehurst (2002).

Figure 1

Simple Meditation Model Predicting Time 2 WIAT-III Basic Reading Standard Score
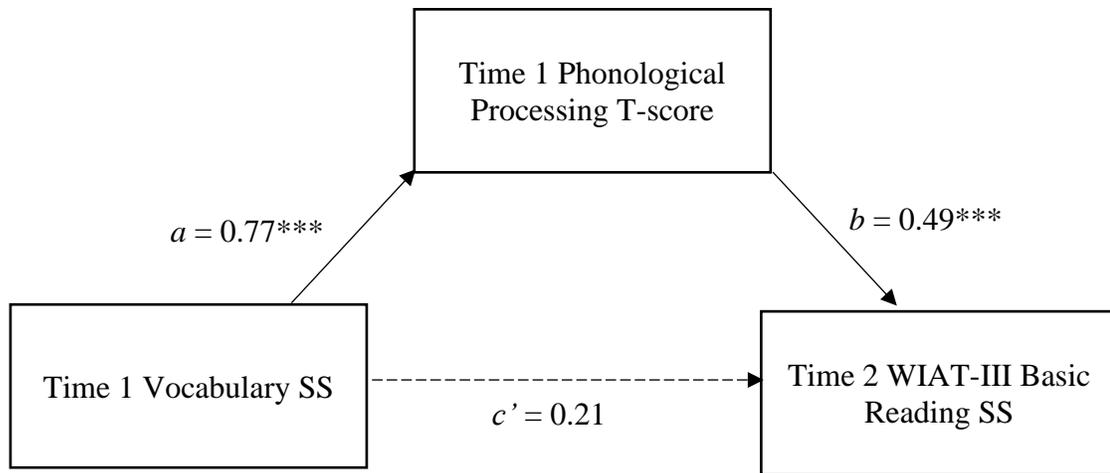
Note. This simple mediation model represents the indirect effect of Time 1 vocabulary standard score on Time 2 WIAT-III Basic Reading standard score through Time 1 Phonological Processing T-score. Statistics are unstandardized regression coefficients. Solid lines indicate statistically significant relations and broken lines indicate statistically nonsignificant relations. a = effect of X on the mediator; b = effect of the mediator on Y partialling out the effect of X; c' = direct effect of X on Y; SS = standard score; WIAT-III = Wechsler Individual Achievement Test-III.

*** $p < .001$

Discussion

The present study examined longitudinal predictors of word-reading ability in a relatively large sample of children with WS. Consistent with our predictions, method of reading instruction, phonological awareness, and visual-spatial perception at ages 6–7 years (Time 1) contributed significantly and uniquely to variance in word-reading ability at ages 9–10 years (Time 2), even after the effect of individual differences in overall intellectual ability or vocabulary was taken into account. Furthermore, the results of a simple mediation analysis suggested that individual differences in Time 1 vocabulary contributed significantly to Time 2 word-reading ability indirectly, via its effect on Time 1 phonological awareness. These results along with their theoretical and educational implications are discussed below.

Longitudinal Predictors of Word Reading Ability in Williams Syndrome

A previous cross-sectional study with an overlapping sample of 9-year-olds with WS (Mervis et al., 2021) demonstrated that the primary concurrent predictor of word-reading ability was reading instruction method, with a systematic phonics approach associated with considerably better performance than other reading instruction approaches. This finding is consistent with the literature for TD children (see Castles et al., 2018 for a review and Ehri, Nunes, Stahl, et al., 2001 for a meta-analysis). The present longitudinal finding that a systematic phonics approach at ages 6–7 years was associated with considerably better word-reading performance at ages 9–10 years, even after accounting for the effects of phonological awareness, visual-spatial perception, and either overall intellectual ability or vocabulary at age 6–7 years, extends the prior cross-sectional finding and provides longitudinal evidence for the importance of early

systematic phonics instruction. This finding is also consistent with the results of intervention studies for children with ID (Allor et al., 2014; Browder et al., 2012; Hunt et al., 2020).

Phonological awareness was longitudinally associated with word reading and continued to explain significant unique variance in this ability even after we accounted for the effects of visual-spatial perception, reading instruction method, and either overall intellectual ability or vocabulary. These results are consistent with previous cross-sectional (see Melby-Lervåg et al., 2012 for a meta-analysis) and longitudinal (Caravolas et al., 2013; Clayton et al., 2020) findings for TD children. Our findings also are in line with previously-reported bivariate associations between phonological awareness and word reading for individuals with WS both in cross-sectional studies (Brawn et al., 2018; Laing et al., 2001; Levy et al., 2003; Levy & Antebi, 2004; Menghini et al., 2004; Mervis et al., 2021), and in the only previous longitudinal study of the reading abilities of children with WS (Steele et al., 2013).

Visual-spatial perception also was longitudinally associated with word-reading ability for the children with WS in our sample. This finding is consistent with those of previous cross-sectional studies of individuals with WS (Brawn et al., 2018; Dessalegn et al., 2013). In the present study, this relation remained significant even after controlling for individual differences in phonological awareness, overall intellectual ability or vocabulary, and reading instruction method. These findings also are consistent with prior cross-sectional (e.g., Fisher et al., 1985; Lee et al., 1986; McBride-Chang et al., 2011) and longitudinal (Ho & Bryant, 1999) findings for TD children. Visual-spatial construction, which depends on visual-spatial perception, has also been associated with

word-reading skills for children with WS (Laing et al., 2001; Mervis et al., 2021). These results are likely due in part to individual differences in visual-spatial perception, which was not measured separately in those studies.

Broad language skills, including vocabulary, are clearly important for reading comprehension for both TD children (see Hjetland et al., 2019 for longitudinal findings) and children with WS (see Mervis et al., 2021 for cross-sectional findings). Previous longitudinal studies with TD children have suggested that vocabulary is also important for the development of word reading through its effect on the development of code-related skills, including letter-name/sound knowledge and phonological awareness (Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002). Vocabulary at Time 1 did not account for a significant amount of unique variance in word-reading ability at Time 2 after taking into account individual differences in Time 1 phonological awareness, visual-spatial perception, and reading instruction method. The absence of a significant direct effect is consistent with findings from prior longitudinal studies of TD children (Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002) and a prior cross-sectional study in an overlapping sample of children with WS (Mervis et al., 2021). At the same time, our mediation analysis results indicating a significant indirect effect of Time 1 vocabulary on Time 2 word reading through Time 1 phonological awareness replicated previous findings for TD children (Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002). Consistent with the Lexical Restructuring Model (Metsala & Walley, 1998), the contribution of vocabulary to reading abilities of children with WS is not restricted to reading comprehension; individual differences in vocabulary

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skill contributed to individual differences in the development of decoding skills through their effect on individual differences in phonological awareness.

Although overall intellectual ability was strongly correlated with word-reading ability, it did not explain significant unique variance in single-word reading after we took into account the effects of the other variables. These longitudinal findings suggest that limited overall intellectual ability, including IQs in the moderate disability range, does not preclude word-reading acquisition for children with WS. These results are consistent with previous cross-sectional findings for TD struggling readers (Stuebing et al., 2009), individuals with mild, moderate, or severe ID (Pezzino et al., 2019), and children with WS (Mervis et al., 2021).

Limitations

The results of the present study should be interpreted in the context of certain limitations. Most importantly, as word-reading ability was not measured at Time 1, we were not able to include this factor in the regression analyses. In addition, letter-sound knowledge, an important predictor of early reading ability (Clayton et al., 2020) also was not measured at Time 1. This concern is somewhat attenuated by the strong correlation between phonological awareness (which was measured at Time 1) and letter-sound knowledge (e.g., Clayton et al., 2020). Finally, despite efforts to enroll a diverse sample, most of the participants were White non-Hispanic and the majority of their mothers had completed at least a bachelor degree.

Theoretical and Educational Implications

Our findings that early phonological awareness and visual-spatial perception, along with systematic phonics instruction, contributed to later word-reading skills in

children with WS are consistent with Ehri's Phase Theory (Ehri, 1987, 2005), which was developed based on findings from studies of TD children. Ehri's theory highlights the importance of forming grapheme-phoneme connections for successful reading acquisition, an endeavor requiring both the ability to recognize and discriminate letter shapes and the ability to pay conscious attention to the sounds in speech. In addition, our finding of an indirect effect of early vocabulary on later word-reading skills is consistent with the Lexical Restructuring Model (Metsala & Walley, 1998), in which phoneme awareness emerges as a consequence of vocabulary development.

The educational implications of our findings are clear. First, our findings provide further support for the importance of systematic phonics instruction that incorporates systematic phonological and phonemic awareness training for teaching word reading to children with WS. Second, our results for visual-spatial perception suggest that children with WS are likely to benefit from preschool and early primary school programs that incorporate the use of embedded letter mnemonics to teach letter-sound correspondences, a procedure known to support the learning of letter-sound correspondences by TD children (e.g., Ehri, Nunes, Stahl, et al., 2001; Roberts, 2021; Shmidman & Ehri, 2010). In fact, given their marked visual-spatial difficulties, young children with WS should be particularly likely to benefit from the use of embedded letter-mnemonics to learn letter-sound correspondences (Mervis, 2009). Third, our results suggest that children with WS also would benefit from preschool programs that address vocabulary and other broad language skills that provide the foundation for acquisition of both code-related and reading comprehension skills (see Mervis et al., 2021 for findings for reading comprehension). Prior findings (Becerra & Mervis, 2019) for considerably younger

children with WS have indicated that 24-month expressive vocabulary size accounts for a significant amount of unique variance in 48-month expressive and receptive vocabulary sizes, even after accounting for age at onset of referential pointing and 24-month nonverbal reasoning ability. For this reason, speech-language intervention for infants and toddlers with WS, by supporting initial language development, also is crucial for providing important foundational skills that will allow them to benefit from later preschool and early primary school programs that directly support early literacy. In sum, integrated preschool and early primary school programs that focus on systematic phonics instruction that incorporates phonological and phonemic awareness training and embedded-letter mnemonics, accompanied by broad language instruction, can offer children with WS the opportunity to optimize their early reading abilities.

CHAPTER III

SPELLING ABILITIES OF SCHOOL-AGED CHILDREN WITH WILLIAMS

SYNDROME[2]

Accurate spelling is an important component of skilled writing, an ability that

contributes to both academic and professional success (e.g., Galuschka et al., 2020).

Learning about spelling also enhances knowledge of the alphabetic system, which is

crucial to the development of skilled reading. Nevertheless, despite its importance,

spelling ability has been investigated much less often than reading abilities, either for

typically developing (TD) children (Treiman, 2017) or for individuals with intellectual

disability (ID; Lindström & Lemons, 2021).

In the present study, we examined the relations between spelling ability, word-

reading ability, and method of reading instruction for school-aged children with Williams

syndrome (WS), a genetic disorder associated with mild to moderate ID. Spelling

difficulty is very common in individuals with WS. For example, in the largest study that

measured spelling ability ($N = 62$, ages 19 – 39 years), Howlin and her colleagues

(Howlin et al., 1998) reported that 26% of the participants were not able to score above

basal on a standardized spelling assessment. For the individuals who did meet the basal

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2021 and is currently in press.

criterion, mean spelling age equivalent (AE) was 7.6 years (range: 6.0 – 12.5 years). These findings suggest that at the end of formal schooling, the spelling ability of most individuals with WS is below the level of functional literacy.

To the best of our knowledge, the spelling abilities of individuals with WS have been addressed in only seven published studies (Dessalegn et al., 2013; Howlin et al., 1998; Laing et al., 2001; Pagon et al., 1987; Polse, 2013; Udwin et al., 1987, 1996). All of these studies had methodological limitations such as small sample size and/or the use of AE or grade-equivalent (GE) scores rather than standard scores (SSs). In the present study, we used SSs to measure the spelling abilities of a relatively large sample of school-aged children with WS.

Relations Between Spelling Ability and Reading Ability

In order to read accurately in an alphabetic orthography, the beginning reader must learn to translate letters or letter units (graphemes) into the sounds (phonemes) they represent in the pronunciation of words; conversely, in order to spell, phonemes need to be converted into their respective graphemes (Ehri, 2000; Treiman, 2017). Therefore, spelling and reading involve the same processes in the opposite order. Not surprisingly, spelling ability and word-reading ability are highly correlated for TD children, with correlations ranging from .50 to .90 (Pan et al., 2021). Relatedly, a confirmatory factor analysis examining the correlations among several reading, writing, and math abilities indicated that spelling clustered with basic reading; the latent trait correlation between word-reading accuracy and word-spelling accuracy was .96 (Peterson et al., 2021).

According to Ehri's (2020) Word Identity Amalgamation Theory, to become a skilled reader, one has to bond the orthography (spelling), phonology (pronunciation),

morphology, syntax, and semantics of a word together as a lexical unit. In oral language, most of these aspects already have been bonded. However, to complete the bonding process and allow for fluent reading, the child must bond spellings to pronunciations and meanings. According to Ehri (2020; see also Perfetti & Hart, 2002), decoding, which is best learned through systematic phonics instruction (Ehri, 2020; Moats, 2019), plays a crucial role in learning the orthography of words. As Share (1995, p. 173) stated, "This ability [decoding] represents the *sine qua non* of reading acquisition," as it provides a mechanism for forming accurate spellings in memory, obligatorily drawing the reader's attention to the identity and order of the letters and how they map onto sounds in the pronunciation of words. Ehri (2020) has described these grapheme-phoneme mappings as the glue that secures the spellings of individual words in memory. This mechanism helps explain why spelling ability and reading ability are so strongly correlated.

Given this strong correlation, it is not surprising that systematic spelling instruction improves reading. For example, based on their meta-analysis, Graham and Santangelo (2014) reported that children who received explicit and systematic instruction in spelling not only had significantly better spelling (average weighted effect [AWE] = 0.54), but also significantly better phoneme awareness (AWE = 0.51), word reading (AWE = 0.40), reading fluency (AWE = 0.36), and reading comprehension (AWE = 0.66) skills than children who received no or unrelated (e.g., math) instruction or incidental approaches to improving spelling. Relatedly, Ouellette and Sénéchal (2017) and Treiman and colleagues (2019) have found that early spelling ability predicts later reading performance beyond the effects of well established predictors such as phoneme awareness and vocabulary.

For individuals with ID, strong correlations between word-reading ability and spelling ability also have been reported. For example, for individuals with ID of mixed etiology, correlations of .94 (Henry & Winfield, 2010) and .82 (Loveall & Conners, 2013) were found. For individuals with Down syndrome, a correlation of .85 was reported in one study (Byrne et al., 2002), and correlations of .85 (with real-word reading) and .86 (with pseudoword reading) in another (Cardoso-Martins et al., 2009).

Impact of Method of Reading Instruction on Spelling Ability

There is clear evidence that reading interventions focused on systematic phonics instruction enhance students' spelling performance. Based on a meta-analysis, Graham et al. (2018) reported that all 14 studies examining the effect of phonics instruction on spelling produced a positive effect (AWE = 0.41, 95% CI = 0.21 – 0.55), with no significant heterogeneity. These studies focused on TD children or children with learning disabilities, and participants ranged in grade from preschool to secondary school. Galuschka et al. (2020) also reported a positive effect of phonics instruction on spelling (AWE = 0.68, 95% CI = 0.15 – 1.21) in a meta-analysis of studies focusing on individuals with reading disabilities. Although there was substantial heterogeneity in effect sizes, the source(s) of the heterogeneity could not be determined. Based on a randomized clinical trial, Sermier Dessemontet et al. (2021) reported that French-speaking children with moderate ID given systematic phonics instruction made more progress in spelling than children in the control group, most of whom were exposed to unsystematic phonics instruction; the effect size was medium, and the difference was almost statistically significant ($p = .058$).

Spelling Abilities of Individuals with Williams Syndrome

WS is a rare neurodevelopmental genetic disorder caused by a hemideletion of 25 – 27 genes on chromosome 7q11.23 (Kozel et al., 2021). Individuals with WS typically have mild to moderate ID, although the full range is from severe ID to average intellectual ability (Mervis & John, 2010). Relative to their overall intellectual ability, individuals with WS typically show strengths in concrete vocabulary, nonverbal reasoning, verbal short-term memory, and phonological processing but weaknesses in spatial abilities and relational and conceptual language (Kozel et al., 2021; Mervis & Greiner de Magalhães, in press).

To the best of our knowledge, there are only seven published studies addressing the spelling abilities of individuals with WS. Pagon and colleagues (1987) described the academic achievement of nine individuals with WS (median age = 13 years, range: 10.17 – 20.67 years). Spelling GE ranged from 0.5 to 4.4 (median = 2.2) and was within one grade of reading GE for seven of the nine participants, with some participants earning higher GE for reading and others for spelling. Dessalegn and colleagues (2013) reported that two 16-year-olds with WS who were closely matched for IQ nevertheless differed considerably in their spelling abilities; one scored at the 1st grade level and the other at the 6th grade level. Laing et al. (2001) reported that the mean spelling AE for 15 individuals with WS (mean age = 15.1 years, range: 9 – 27 years) was 5.1 years (SD = 3.43), which was considerably lower than their mean word-reading AE of 6.58 years (SD = 2.61). Spelling AE was strongly correlated with IQ (r = .64). In the only study that reported SSs, Polse (2013) provided further evidence for wide variability in spelling skills. For the 10 participants (mean age = 10.86 years; SD = 1.62), the mean SS for the

Spelling Sounds subtest of the Woodcock-Johnson Tests of Cognitive Abilities-III (Woodcock et al., 2001) was 75.80, with a *SD* of 27.13, which is considerably larger than the general-population *SD* of 15 for this measure.

Udwin and colleagues conducted three studies of overlapping samples of individuals with WS that included assessment of spelling abilities. Udwin et al. (1987) reported that of forty-four 6 – 15-year-olds (mean age = 11.10 years) with WS, 58% were able to obtain a basal on the spelling measure. Their mean Spelling AE was 6.83 years (range: 5.58 – 11.33 years), which was somewhat younger than their mean word-reading AE of 7.83 years (range: 6.17 – 11.42 years). The children who were able to obtain a basal had significantly higher IQs than the children who did not meet the basal criterion.

In a longitudinal follow-up (Udwin et al., 1996) of 23 of these participants (mean age = 21.90 years, *SD* = 1.90), 19 were able to obtain a basal. The spelling AE of the 15 participants who met the basal criterion at both assessments increased from 6.62 years (*SD* = 1.15) at the first assessment to 7.57 years (*SD* = 1.89) at the second, although, as the authors noted, these AEs cannot be directly compared because they were based on different assessments. Howlin, Davies, and Udwin (1998) examined the literacy abilities of 62 adults with WS (mean age = 26.49 years, range: 19 – 39), including the 23 participants in the previous study. For the 46 participants who met the basal criterion on the spelling test, mean AE for spelling (*M* = 7.60 years, *SD* = 2.01, range 6.00 – 12.50) was significantly lower than mean AE for word-reading accuracy (*M* = 8.65 years, *SD* = 2.68, range 6.00 – 18.00). Full-scale IQ was significantly higher for the participants who met the basal criterion on the spelling test than for those who did not.

Current Study

In the present study, we addressed four research questions focused on the spelling ability of a relatively large sample of school-aged children with WS. In contrast to most of the prior studies addressing the spelling abilities of individuals with WS, our analyses were conducted using SSs. Unlike AE or GE scores, SSs provide both a standardized measure of children's ability relative to same age peers and a psychometrically sound basis for statistical comparisons. (See Brawn et al., 2018; Mervis & Robinson, 2005 for discussion of problems with interpretation of AEs or GEs.)

Our first question was: What is the relation of spelling ability to single-word reading ability for 9 – 17-year-olds with WS? This question was addressed at both the group and the individual levels. Based on prior findings (Howlin et al., 1998; Laing et al., 2001; Udwin et al., 1987) for individuals with WS suggesting that spelling ability lags behind reading ability, we predicted that at the group level, there would be a small but significant difference between spelling SS and word-reading SS favoring reading. At the individual level, based on Pagon et al.'s (1987) report that the GEs for spelling and reading ability were within one grade of each other for most participants, with some participants scoring higher on word reading and others on spelling, we predicted that the most likely pattern would be no significant difference. Based on prior findings at the group level (Udwin et al., 1987) for individuals with WS, we predicted that for cases in which there was a significant difference, the difference would favor reading SSs.

Our second question was: What are the correlations between spelling ability, word-reading ability, and overall intellectual ability for children with WS? Based on Ehri's (2020) theory and prior findings for TD children (Ehri, 2000; Ouellette &

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Sénéchal, 2017; Treiman et al., 2019) and individuals with ID (Byrne et al., 2002; Cardoso-Martins et al., 2009; Henry & Winfield, 2010; Loveall & Conners, 2013), we expected to find a strong correlation between word-reading ability and spelling ability in school-aged children with WS. Based on prior findings for individuals with WS (Laing et al., 2001), we also expected that spelling ability would be significantly correlated with IQ.

Our third question was: What is the relation between method of reading instruction and spelling ability in children with WS? Based on prior findings for TD children and children with literacy or learning difficulties (Galuschka et al., 2020; Graham et al., 2018), we expected that children with WS who were taught to read through systematic phonics instruction would evidence significantly better spelling abilities than children with WS who were taught to read using other approaches.

Finally, our fourth question was: Is spelling ability a unique concurrent predictor of word-reading ability for children with WS, even after accounting for well-established predictors of word reading for both TD children (e.g., Treiman et al., 2019) and children with WS (Mervis et al., 2021)? Based on previous findings for TD children (e.g., Treiman et al., 2019) we expected to find a unique concurrent contribution of spelling to word-reading ability, even after controlling for phonological awareness and vocabulary.

Method

Participants

Eighty children (42 females, 38 males) with genetically-confirmed classic-length deletions of the WS region participated in this study. The participants ranged in age from 9.01 to 17.98 years ($M = 12.84$ years, $Mdn = 12.13$, $SD = 3.07$). Their median grade in

school was 5th, with an interquartile range from 4th to 8th grade and a range from 2nd grade to 12th grade. Native language was English for 78 of 80 participants. All participants were fluent in English at the time of their assessment. Primary classroom placement was in a mainstream class for 47 of the 80 participants (27 with reading/language arts instruction primarily in a mainstream classroom, 20 with reading/language arts instruction primarily in a self-contained classroom) and in a special education (self-contained) class for 30 children (all with reading/language arts instruction in a special education classroom). The three remaining children were homeschooled.

Children were from 24 different U. S. states representing all U. S. census regions (31.3% Northeast, 31.3% South, 23.8% Midwest, 10.0% West) and two Canadian provinces (3.8%). The participants' racial/ethnic background was: 81.3% White non-Hispanic, 7.5% White Hispanic, 5.0% multiracial non-Hispanic, 3.8% multiracial Hispanic, 1.3% African American non-Hispanic, and 1.3% Asian non-Hispanic. Twenty of the participants' mothers (25%) did not have a bachelor degree; the remaining 60 (75%) had earned at least a bachelor degree. Participants were recruited for studies of language and cognition in children with WS. Some children were assessed multiple times (with at least 11.5 months between assessments) as part of a longitudinal study. For these children, data from the most recent assessment were used. Data collection began in March 2010 and ended in February 2020.

Measures

Spelling Ability

Spelling ability was measured by the Wechsler Individual Achievement Test-III (WIAT-III; Wechsler, 2009) Spelling subtest. This subtest requires the child to spell letter

sounds or words, arranged in order of difficulty. For the first item, the examiner asks the child to write their first name. For the next four items, the examiner asks the child to write the letter that makes a particular sound. The examiner first produces the sound in isolation, followed by a word that includes that sound (e.g., "write the letter that makes the /a/ sound in *apple"*). For the remaining 57 items, the examiner asks the child to write specific words, each of which is first provided in isolation, then within the context of a sentence, and then in isolation for a second time. The first item that is administered is based on the child's grade level. For the grade levels included in this study, the first item administered was always a word. If a score of 0 (incorrect spelling) was attained on any of the first three items, the preceding items were administered in reverse order until three consecutive scores of 1 (correct) were obtained. If the examiner could not read a letter the child had written, the child was asked what that letter was and the response scored accordingly. The task was discontinued when the child met the ceiling rule of four consecutive incorrectly spelled items. This subtest yields a SS (general population mean = 100, *SD* = 15). The average split half internal consistency for the WIAT-III norming sample was .95 for the Spelling subtest.

Word Reading Ability

Word reading was measured by the WIAT-III Basic Reading Composite SS. This measure includes two subtests, one measuring single real-word reading (Word Reading) and one measuring pseudoword decoding (Pseudoword Decoding). The standardized ceiling rule of four consecutive failed items leads to discontinuation of each subtest. For the WIAT-III norming sample, the average split half internal consistency was .97 for both

Word Reading and Pseudoword Decoding and .98 for Basic Reading Composite. For the

present participants, the correlation between the two subtest SSs was $r = .93$, $p < .001$.

Phonological Awareness

The Differential Ability Scales-II (DAS-II; Elliott, 2007) includes a supplemental

Phonological Processing subtest which assesses knowledge of the sound structure of the

English language and the ability to manipulate sounds. Four types of skills are assessed:

rhyming, blending, deletion, and phoneme identification and segmentation. The oldest

age for which this subtest is normed is 12.99 years. The 45 participants (24 females) who

were this age or younger completed this subtest. This subgroup did not differ

significantly from the remaining participants in proportion of females. The Phonological

Processing subtest yields an overall T-score (general population mean = 50, $SD = 10$).

For the DAS-II School Age version norming sample, the average IRT-based internal

consistency for 9 – 12-year-olds on the Phonological Processing subtest was .85.

Vocabulary

The Peabody Picture Vocabulary Test-4 (PPVT-4; Dunn & Dunn, 2007) is a

measure of receptive vocabulary in which participants indicate which of four colored

pictures best depicts the word said by the examiner. The Expressive Vocabulary Test-2

(EVT-2; Williams, 2007), which was co-normed with the PPVT-4, is a measure of

expressive vocabulary in which participants look at one colored picture and provide a

one-word answer to a question about the picture. There are two types of items: one where

the child is asked to name an object, action, or attribute depicted in the picture and one

where the examiner provides a label and then the child is asked to provide a synonym that

goes with the picture. Vocabulary ability was measured by a composite based on the

mean of the child's PPVT-4 and EVT-2 SSs. For the present participants, the correlation between PPVT-4 and EVT-2 SSs was $r = .86$, $p < .001$. Based on the test manuals, average split-half internal consistency for the 9 – 17-year-olds included in the norming sample was .94 for PPVT-4 Form B (the version used in this study) and .93 for EVT-2 Form B.

Intellectual Ability

The DAS-II School Age version was used to evaluate children's overall intellectual ability. The DAS-II estimates a child's General Conceptual Ability (GCA, similar to IQ) based on their performance on the subtests measuring verbal, nonverbal reasoning, and spatial abilities. Performance is reported as a SS (general population mean $= 100$, $SD = 15$). For the DAS-II School Age version norming sample, the average IRT-based internal consistency for the GCA was .96.

Reading Instruction Method

The primary approach to teaching word reading to each child was classified as Systematic Phonics (hereafter, Phonics) or Other. All available information related to the students' reading instruction was considered (e.g., reading program [if any] implemented in the primary classroom in which the child received reading instruction, Individualized Education Plan goals and progress reports, worksheets, homework assignments, conversations with parents and reading instructors). Following Mervis et al.'s (2021) procedure, reading instruction was classified as "Phonics" if the primary approach to teaching word reading was based on systematic instruction in English phonics. Reading instruction was classified as "Other" if it took a whole-language, three-cueing, or balanced literacy approach or otherwise emphasized the use of context to figure out a

word or if it focused on whole-word instruction. For older students who were no longer receiving word reading instruction, the primary approach that had been used to teach word reading was determined using the same criteria, either as documented from earlier assessments (for participants who were enrolled in a longitudinal study) or from prior IEPs or progress reports.

The primary word-reading instruction approach was Phonics for 47 (58.8%) participants (25 with reading/language arts instruction in a mainstream class, 20 with reading/language arts instruction in a self-contained class, 2 home schooled) and Other for 33 (41.2%) participants (2 with reading/language arts instruction in a mainstream class, 30 with reading/language arts instruction in a self-contained class, 1 home schooled). The Phonics group (M = 13.57 years, Mdn = 13.38, SD = 3.15, IQR: 11.03 – 17.35, range: 9.03 – 17.94) was significantly older than the Other group (M = 11.79 years, Mdn = 11.19, SD = 2.66, IQR: 9.33 – 13.55, range: 9.01 – 17.98), $t(78)$ = 2.65, p = .010, Cohen's d = 0.60. A Mann-Whitney U test indicated that the distribution of grade also was significantly higher for the Phonics group (Mdn = grade 7, IQR: 4 – 10, range: 2 – 12) than the Other group (Mdn = grade 5, IQR: 3 – 6.5, range: 2 – 12), z = -2.62, p = .009, Cohen's d = 0.61.

Procedures

The study protocol was reviewed and approved by the university's Institutional Review Board. Parents or legal guardians of all participants provided written informed consent and participants provided oral or written assent. All standardized measures were completed at the senior author's laboratory as part of a larger two-day assessment. All

measures were administered by trained doctoral students or research assistants and scored according to the standardized procedures.

Results

Data were analyzed using IBM SPSS v. 27. Cohen's *d* was used to measure effect size (0.2 = small effect, 0.5 = medium, 0.8 = large; Cohen, 1988). All assumptions of multiple linear regression analyses were met. Maternal education level was not significantly correlated with WIAT-III Spelling SS ($r = .09$, $p = .426$). Therefore, this variable was not included in the multiple regression models. For the multiple regression analyses, Cohen's f^2 was used to measure effect size (0.02 = small effect, 0.15 = medium, 0.35 = large; Cohen, 1988).

Relations Between Spelling Ability and Word Reading Ability

Descriptive statistics for all measures investigated in this study are provided in Table 6. There was considerable variability, with SSs on the spelling and word-reading measures ranging from average or above average for the general population to moderate-severe disability (including inability to spell or read any of the test items), with *SD*s at least as large as for the WIAT-III norming sample. As indicated in the Introduction, there are serious psychometric concerns regarding AE scores (e.g., Brawn et al., 2018; Mervis & Robinson, 2005). However, as AEs are the only statistical measure provided in all but one of the prior studies examining spelling abilities of individuals with WS, nonparametric descriptive statistics for the WIAT-III Spelling, Word Reading, and Pseudoword Decoding subtest AEs are provided in Table 7 for comparison.

Table 6

Descriptive Statistics for Measures Included in the Study

Variable	N	Mean	Median	SD	Range
WIAT-III Spelling SS	80	71.30	69.00	15.13	40 – 111
WIAT-III Basic Reading Composite SS	80	75.24	75.00	16.86	43 – 106
WIAT-III Word Reading SS	80	75.74	74.00	18.52	41 – 110
WIAT-III Pseudoword Decoding SS	80	76.11	76.00	15.01	50 – 107
DAS-II GCA	80	63.58	63.00	13.91	32 – 94
DAS-II Phonological Processing T	45	42.56	47.00	12.93	10 – 63
Vocabulary SS	45	77.03	77.00	15.31	33 – 109

Note. WIAT-III = Wechsler Individual Achievement Test-III; SS = standard score; DAS-II = Differential Ability Scales-II; GCA = General Conceptual Ability; T = T-score.

Table 7

Descriptive Statistics for WIAT-III Age Equivalents and Standard Scores

Variable	Median	Interquartile Range	Range
Age Equivalent			
Spelling	7.84 yrs	6.40 – 9.00 yrs	< 5.00[a] – > 19.92[b] yrs
Word Reading	7.67 yrs	6.40 – 11.25 yrs	< 6.00[a] – > 19.92[b] yrs
Pseudoword Decoding	7.17 yrs	< 6.00[a] – 9.00 yrs	< 6.00[a] – 16.00 yrs
Standard Score			
Spelling	69.00	60.00 – 82.75	40[c] – 111
Word Reading	74.00	61.25 – 91.00	41 – 110
Pseudoword Decoding	76.00	63.25 – 88.00	50 – 107
Basic Reading Composite	75.00	63.00 – 90.75	43 – 106

Note. N = 80. Age equivalents are not available for Basic Reading Composite. WIAT-III = Wechsler Individual Achievement Test-III; yrs = years.
[a] Lowest possible age equivalent.
[b] Highest possible age equivalent.
[c] Lowest possible standard score.

To determine if there was a significant difference between spelling and reading SSs for school-aged children with WS, paired sample t-tests were conducted. At the group level, Spelling SS was significantly lower than Basic Reading SS, $t(79) = -4.68$, $p < .001$, Cohen's $d = 0.23$. Spelling SS was significantly lower than both Word Reading SS, $t(79) = -4.66$, $p < .001$, Cohen's $d = 0.24$; and Pseudoword Decoding SS, $t(79) = -5.68$, $p < .001$, Cohen's $d = 0.32$. The effect sizes were small.

To investigate the individual patterns of relative strength and weakness between reading ability and spelling ability, each child's Spelling SS was compared to their Word Reading SS and Pseudoword Decoding SS, using the critical values ($\alpha = .05$) for a significant difference between SSs provided in the WIAT-III technical manual. For comparisons between Spelling SS and both Word Reading SS and Pseudoword Decoding SS, the critical values were 8 points for 9 – 11-year-olds and 7 points for 12 – 17-year-olds. As shown in Figure 1, for the majority of participants, Spelling SS did not differ significantly from reading SSs. In most cases where the differences were significant, SSs were higher for reading than for spelling.

Figure 2

Relations Between WIAT-III Spelling Standard Score and WIAT-III Word Reading or

WIAT-III Pseudoword Decoding Standard Scores for Individual Participants
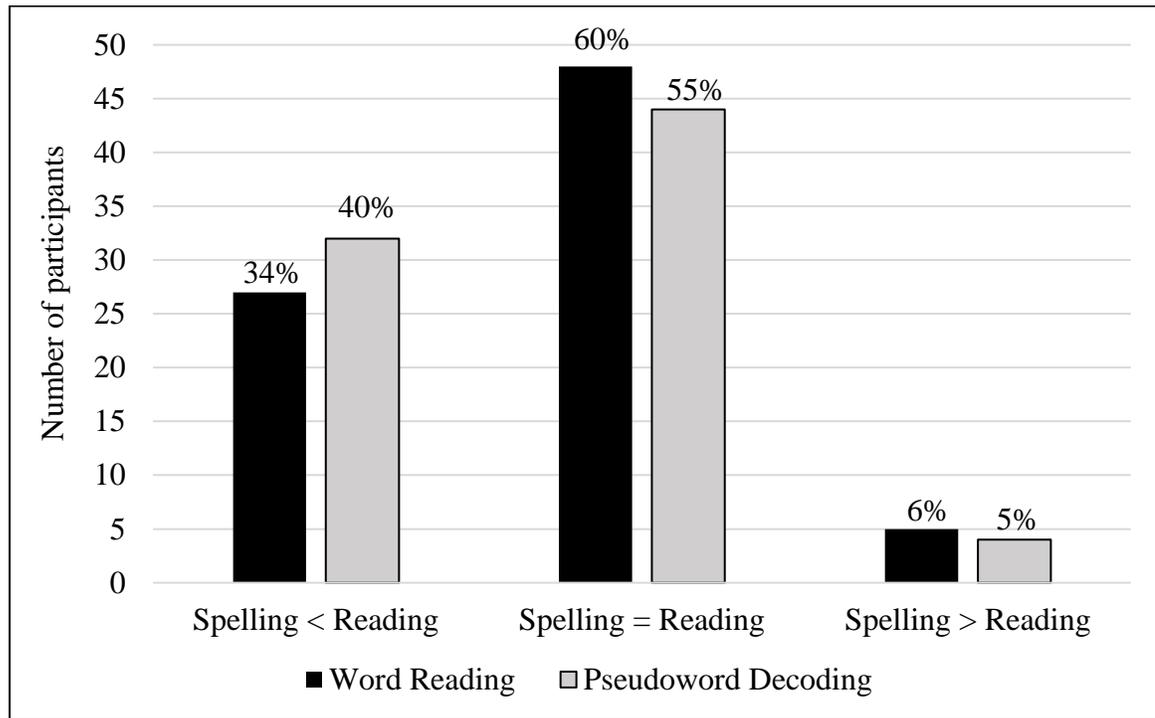
Note. Percentages indicate the percent of participants who evidenced a particular relation.

$N = 80$. < represents significantly lower; = indicates that the two standard scores do not

differ significantly; > represents significantly higher.

Correlations Between Spelling Ability, Word Reading Ability, and Overall

Intellectual Ability

Pearson correlations ($\alpha = .01$) among the students' chronological age and their

scores on the measures included in the study are displayed in Table 8, separately for the

full sample and the 45 participants who completed the DAS-II Phonological Processing

subtest. All of the correlations were statistically significant and strong, except for the

ones involving chronological age, which were weak and not statistically significant. The

lack of significant correlations between chronological age and the standardized measures

included in this study was not surprising considering that SSs and T-scores take into

account the child's chronological age.

The correlation between WIAT-III Spelling SS and WIAT-III Basic Reading SS

was very strong ($r = .90$, for both the full sample and the 45 participants who completed

all measures). As also illustrated in Table 8, children's scores on these measures also

correlated significantly and strongly with their overall intellectual ability, as measured by

the DAS-II GCA. In view of this pattern, bootstrapped partial correlations were

performed to investigate if the correlation between spelling ability and word-reading

ability remained significant after controlling for overall intellectual ability and if the

correlation between spelling ability and overall intellectual ability remained significant

after controlling for reading ability. Results are shown in Table 9. As evidenced by the

lack of overlap between the 95% confidence intervals, the partial correlation between

Spelling SS and Basic Reading SS (controlling for GCA) was significantly stronger than

the partial correlation between Spelling SS and GCA (controlling for Basic Reading SS).

Controlling for GCA resulted in very little change in the correlation between Spelling SS

and Basic Reading SS. The correlation remained significant and very strong. In contrast, controlling for Basic Reading SS changed the correlation between Spelling SS and GCA – which had been significant and moderate to strong – to very weak and no longer statistically significant. The same pattern of results was found for Word Reading SS and Pseudoword Decoding SS.

Analysis of Covariance: Relation Between Method of Reading Instruction and Spelling Ability

Descriptive statistics for spelling, reading, and overall intellectual ability as a function of reading instruction method are shown in Table 10. As indicated in the Method section, the Phonics group was significantly older than the Other group. The Phonics group also had significantly higher mean GCA than the Other group, $t(78) = 4.84$, $p < .001$, Cohen's $d = 1.10$. To compare the two groups' Spelling SSs, a between-group ANCOVA with reading instruction method as the between-group factor, controlling for GCA and chronological age, was performed. As would have been expected on the basis of the weak correlation between chronological age and Spelling SS (see Table 8), the effect of chronological age was not significant, $F(1,76) = 0.19$, $p = .663$, $\eta_p^2 = .003$, Cohen's $d = 0.10$. The effect of GCA was statistically significant, $F(1,76) = 25.22$, $p < .001$, $\eta_p^2 = .249$, Cohen's $d = 1.16$. Mean Spelling SS was significantly higher for the Phonics group than the Other group, $F(1,76) = 29.66$, $p < .001$, $\eta_p^2 = .281$, Cohen's $d = 1.25$ even after controlling for GCA and chronological age.

Table 8

Bivariate Correlations Among Chronological Age, Achievement Test Performance, Cognitive Performance, and Reading Instruction Method

Measure	1	2	3	4	5	6	7	8	9
1. Chronological age	_	-.05	-.12	-.10	-.13	-.18	-.04	-.22	_
2. WIAT-III Spelling SS	.18	_	.90***	.88***	.89***	.68***	.66***	.55***	.69***
3. WIAT-III Basic Reading SS	.14	.90***	_	_	_	.78***	.77***	.63***	.80***
4. WIAT-III Word Reading SS	.18	.89***	_	_	.93***	.76***	.74***	.62***	.76***
5. WIAT-III Pseudoword Decoding SS	.09	.87***	_	.93***	_	.74***	.73***	.59***	.83***
6. DAS-II GCA	.01	.65***	.70***	.70***	.65***	_	.84***	.83***	.56***
7. DAS-II Phonological Processing T	_	_	_	_	_	_	_	.70***	.61***
8. Vocabulary SS	.10	.53***	.61***	.63***	.55***	.80***	.70***	_	.44***
9. Reading instruction method	_	.69***	.79***	.76***	.79***	.48***	.61***	.44***	_

Note. Correlations based on *N* = 80 are reported below the diagonal; correlations based on *n* = 45 above the diagonal. WIAT-III = Wechsler Individual Achievement Test-III; SS = standard score; GCA = General Conceptual Ability; DAS-II = Differential Ability Scales II; T = T-score.
** *p* < .01; *** *p* < .001.

Table 9

Partial Correlations Between Standard Scores for Spelling Ability, Word Reading Ability, and Overall Intellectual

Ability

Variables Correlated	Bivariate corr.	Control Variable	Partial corr.	*p*	95% CI	Variables correlated	Bivariate corr.	Control Variable	Partial corr.	*p*	95% CI
Spelling and Basic Reading	.90	GCA	.81	< .001	.73 – .88	Spelling and GCA	.65	Basic Reading	.08	.488	-.11 – .27
Spelling and Word Reading	.89	GCA	.80	< .001	.71 – .88	Spelling and GCA	.65	Word Reading	.07	.570	-.12 – .28
Spelling and Pseudoword Decoding	.87	GCA	.78	< .001	.70 – .85	Spelling and GCA	.65	Pseudoword Decoding	.21	.066	.02 – .39

Note. N = 80. CI = confidence interval; corr. = correlation; GCA = General Conceptual Ability. Spelling ability and word reading ability were measured by the Wechsler Individual Achievement Test-III. Overall intellectual ability was measured by the Differential Ability Scales II GCA.

Table 10

Descriptive Statistics for Spelling, Reading, and Overall Intellectual Ability as a Function of Reading Instruction

Method

Variable	Phonics (*n* = 47, 24 girls)				Other (*n* = 33, 18 girls)			
	Mean	Median	SD	Range	Mean	Median	SD	Range
WIAT-III Spelling SS	79.94	81.00	12.84	43 – 111	59.00	60.00	7.91	40 – 77
WIAT-III Basic Reading Composite SS	86.30	88.00	11.34	55 – 106	59.48	61.00	9.04	43 – 75
WIAT-III Word Reading SS	87.40	89.00	13.73	58 – 110	59.12	60.00	9.53	41 – 78
WIAT-III Pseudoword Decoding SS	86.04	87.00	10.46	56 – 107	61.97	63.00	6.93	50 – 75
DAS-II GCA	69.15	70.00	12.62	41 – 94	55.64	53.00	11.77	32 – 79

Note. *N* = 80. WIAT-III = Wechsler Individual Achievement Test-III; SS = standard score; DAS-II = Differential Ability Scales-II; GCA = General Conceptual Ability.

Multiple Regression Analyses: Concurrent Effect of Spelling Ability on Word Reading Ability

To determine the concurrent effect of spelling ability on word-reading ability after accounting for phonological awareness, vocabulary, and reading instruction method, multiple regression analyses including the 45 participants who had completed the Phonological Processing measure were performed. Pearson correlations ($\alpha = .01$) among the variables included in the regression analyses are reported in Table 8. All correlations were statistically significant.

We began by computing a multiple regression model with Basic Reading SS as the dependent variable and Phonological Processing T-score and Vocabulary SS as predictors (Model 1). As shown in Table 11, this model explained 60% of the variance in Basic Reading SS, with significant unique variance contributed only by Phonological Processing T-score. To determine if reading instruction method contributes to word-reading ability beyond the effects of phonological awareness and vocabulary, reading instruction method was added to Model 1 (Model 2). As indicated in Table 11, Model 2 accounted for significantly more variance in Basic Reading SS than did Model 1, with large effect sizes for both Phonological Processing T-score and reading instruction method.

Finally, to determine the unique concurrent contribution of spelling ability to word-reading ability beyond the effects of phonological awareness, vocabulary, and reading instruction method, Spelling SS was added to Model 2. As shown in Table 11, Model 3 (the final model) accounted for significantly more variance in Basic Reading SS than did Model 2. Phonological Processing T-score (medium effect), reading instruction

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method (large effect), and Spelling SS (large effect) made significant independent contributions to the variance in Basic Reading SS. As illustrated in Table 11, after taking into account the contribution of vocabulary, phonological processing, and method of reading instruction, individual differences in spelling ability uniquely explained 12.3% of the variation in word reading.

Discussion

The present study is the first to describe the spelling abilities of a large sample of school- aged children with WS using SSs and to investigate the relations between spelling ability and word-reading ability at the group and individual levels. Results showed that as expected, the spelling abilities of most school-aged children with WS were more limited than those of same-aged children in the general population. However, consistent with the sparse previous literature, spelling ability ranged widely, from inability to spell any words to spelling at age level. At the group level, spelling SS was significantly lower than word-reading SS but at the individual level, for more than half of the participants, spelling SS did not differ significantly from reading SSs. Spelling SS and reading SSs were very highly correlated, even after controlling for overall intellectual ability. Children taught to read using systematic phonics instruction had significantly higher Spelling SSs than children taught to read using other approaches, even after controlling for overall intellectual ability. Spelling ability contributed significant unique variance to word-reading ability, beyond the effects of phonological awareness, vocabulary, and reading instruction method. In the remainder of the Discussion, we discuss these findings, theoretical and educational implications, limitations, and directions for future research.

Table 11

Multiple Regression Analyses Predicting WIAT-III Basic Reading Composite Standard

Score

Predictor	B	t	p-value	95% CI for B	Semi-partial r	Cohen's f^2
Model 1						
Constant	72.95	46.65	< .001	[69.80, 76.11]		
Phonological Processing T	0.80	4.73	< .001	[0.46, 1.14]	.46	0.60
Vocabulary SS	0.19	1.31	.199	[-0.10, 0.47]	.13	0.04
$R^2 = .60$, adjusted $R^2 = .59$, $F(2, 42) = 32.01$, $p < .001$						
Model 2						
Constant	65.03	36.14	< .001	[61.40, 68.67]		
Phonological Processing T	0.41	2.87	.007	[0.12, 0.70]	.21	0.36
Vocabulary SS	0.17	1.58	.122	[-0.05, 0.38]	.12	0.06
Reading instruction method	16.90	5.81	< .001	[11.02, 22.78]	.42	0.82
$R^2 = .78$, R^2 change $= .20$, adjusted $R^2 = .77$, F change $(1, 41) = 33.70$, $p < .001$						
Model 3 (Final Model)						
Constant	70.07	49.78	< .001	[67.23, 72.92]		
Phonological Processing T	0.23	2.28	.028	[0.03, 0.43]	.11	0.29
Vocabulary SS	0.08	1.10	.279	[-0.07, 0.23]	.05	0.02
Reading instruction method	9.14	4.07	< .001	[4.60, 13.69]	.20	0.40
WIAT-III Spelling SS	0.57	7.08	< .001	[0.41, 0.73]	.35	1.24
$R^2 = .90$, R^2 change $= .12$, adjusted $R^2 = .89$, F change $(1, 40) = 50.07$, $p < .001$						

Note. $n = 45$. All continuous independent variables were centered on the sample mean. WIAT-III = Wechsler Individual Achievement Test-III; CI = confidence interval; T = T-score; SS = standard score.

Relations Between Spelling Ability and Reading Ability

As hypothesized based on the previous literature for individuals with WS (Howlin et al., 1998; Udwin et al., 1987), at the group level, the single-word reading abilities (as measured by SSs) of school-aged children with WS are a strength relative to their spelling ability (SS), but the effect size is small. The parents of many participants spontaneously reported that their child had never been assigned spelling words, suggesting that spelling either was not being taught or was not being taught systematically. If this pattern characterized a large proportion of participants, the lack of spelling instruction likely would have contributed to the difference in reading and spelling SSs. Consistent with the possibility that a large proportion of the participants had not received systematic spelling instruction, based on a national survey, Cutler and Graham (2008) found that only about one-third of primary grade teachers reported using a systematic spelling program for TD children; and Pan et al. (2021) reported that in recent years, a growing number of schools in the USA have deemphasized or eliminated traditional methods of explicit spelling instruction. Similarly, an observational study of the reading instruction provided to children with ID in self-contained classrooms found that most of the teachers did not spend any time teaching spelling; on average, only 0.1% of the observed time for literacy instruction was spent on spelling instruction (Lindström & Lemons, 2021).

Although the difference between spelling ability and reading ability was significant at the group level, word-reading SSs were on average only 4 SS-points higher than spelling SSs. This is consistent with the result that at the individual level, neither Word Reading SS nor Pseudoword Decoding SS differed significantly from Spelling SS

for more than half of the participants. As hypothesized based on Udwin et al.'s (1987) findings, most of the remaining participants obtained significantly higher Reading SSs than Spelling SS.

Similar to TD children (e.g., Ehri, 2000; Ouellette & Sénéchal, 2017; Treiman et al., 2019) and children with ID (e.g., Cardoso-Martins et al., 2009; Henry & Winfield, 2010), there was a very strong correlation between reading ability and spelling ability for children with WS. This correlation remained very strong even after controlling for intellectual ability. In contrast, once word-reading ability was controlled, the correlation between spelling ability and IQ became close to 0. The very strong correlation between spelling and word reading suggests that spelling ability and word-reading ability rely on the same learning mechanisms. In line with Ehri's (2020) Word Identity Amalgamation Theory, our finding of a strong correlation between systematic phonics instruction and both spelling and word reading suggests that a major learning mechanism consists of mapping between the graphemes in written words and the phonemes in spoken words.

Similar to previous longitudinal findings for TD children (e.g., Ouellette & Sénéchal, 2017; Treiman et al., 2019), our cross-sectional finding of a unique concurrent contribution of spelling ability to word-reading ability above and beyond the effects of phonological awareness, vocabulary, and reading instruction method provides further evidence for the strong and unique relation between spelling ability and word-reading ability. This finding, in combination with prior meta-analytic findings for TD children (Graham & Santangelo, 2014), also suggests that school-aged children with WS are likely to benefit from systematic instruction in spelling to support learning to read. Given that learning to spell strengthens knowledge of the alphabetic system (Ehri, 2020; Graham et

al., 2018), systematic instruction in spelling should enhance the bond between spellings

and pronunciations in memory, resulting in higher lexical quality and stronger

orthographic representations (Ehri, 2020; Perfetti & Hart, 2002), which are key to both

skilled reading and accurate spelling (Galuschka et al., 2020; Graham & Santangelo,

2014).

With the advent of spell checkers, some teachers have suggested that spelling

instruction is unnecessary (Moats, 2005; Pan et al., 2021). However, for the spell checker

to work, spelling abilities need to be reasonably developed. For example, spell checkers

correctly identified the target word from the misspellings of students with learning

disabilities only 53% of the time (Montgomery et al., 2001; see Pan et al., 2021 for

further discussion). Furthermore, as highlighted by Moats (2005, 2019) and evidenced by

Graham and Santangelo's (2014) meta-analysis, direct, systematic instruction in spelling

is important for the development not only of spelling but also of word reading, phoneme

awareness, and reading comprehension, and for understanding of the writing system

(Treiman, 2018). We therefore suggest that direct and systematic spelling instruction be

incorporated into literacy practices for children with WS. (See Moats, 2005, 2019, for

detailed strategies for spelling instruction.)

Relation Between Method of Reading Instruction and Spelling and Reading

Abilities

As hypothesized based on prior findings for TD children and children with

learning disabilities (Graham et al., 2018), school-aged children with WS being taught to

read using a systematic phonics approach also spell significantly better than children

taught to read using other methods. This effect remained significant even after controlling

for chronological age and IQ. These findings provide further evidence of the value of systematic phonics instruction for children with WS. The finding of a unique concurrent contribution of reading instruction method to word-reading ability, above and beyond the effects of phonological awareness, vocabulary, and spelling, extends Mervis et al.'s (2021) finding of a significant positive effect of systematic phonics instruction on the word-reading abilities of 9-year-olds with WS to a considerably broader age range. Given the importance of word reading for reading comprehension (e.g., Castles et al., 2018), this benefit would be expected to extend to reading comprehension. As Moats (2005, 2019) has noted, the teaching of spelling would be enhanced by an approach that includes teaching about the structure of the language, phonological awareness, phoneme-grapheme correspondence, orthographic patterns, morphology, and etymology, in addition to systematic instruction in phonics (see also Galuschka et al., 2020).

Limitations and Future Directions

The results of the present study should be interpreted in the context of certain limitations. Despite efforts to enroll a diverse sample and the strength of including participants who resided across a very wide geographical area, most of the participants were White non-Hispanic and the majority of the participants' mothers had completed at least a bachelor's degree. Future research with more diverse samples would be valuable.

The cross-sectional nature and correlational design of the present study do not allow us to draw conclusions about causality. Longitudinal studies in which spelling abilities at one age are predicted from hypothesized predictor abilities measured at a younger age would be valuable to address the hypothesis that spelling ability and reading

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ability rely on similar processes. This type of study is crucial for beginning to address causal relations in the development of spelling abilities in individuals with WS.

Conclusions

The spelling ability of school-aged children with WS is characterized by considerable variability, ranging from inability to spell any words to spelling at age level. Spelling ability and reading ability are very highly correlated and remain very highly correlated even after controlling for IQ. Furthermore, the advantage of systematic phonics instruction for facilitating word-reading development and, by extension, reading comprehension previously found for a large sample of 9-year-olds with WS (Mervis et al., 2021) extends to spelling skills for 9 – 17-year-olds with this syndrome. Our findings have a clear educational implication: Literacy instruction for children with WS should include a systematic phonics component that incorporates systematic spelling instruction.

CHAPTER IV

GENERAL DISCUSSION

The purpose of the present dissertation was to examine and characterize the word-reading and spelling abilities of children with WS and to identify their cognitive, language, and instructional correlates. I took two different approaches to characterize these abilities. In Study 1, individual differences in phonological awareness, visual spatial perception, vocabulary, overall intellectual abilities, and reading instruction approach (systematic phonics vs. other approaches) for sixty-nine 6 – 7-year-olds (most of whom were in kindergarten) were used to predict word-reading abilities three years later. In Study 2, relations between spelling, word reading, and vocabulary abilities and method of reading instruction were investigated for eighty 9 – 17-year-olds. Findings from both Study 1 and Study 2 provide further evidence that the word-reading ability and spelling ability of individuals with WS vary broadly, ranging from inability to read or spell any of the words on the standardized assessment to word-reading or spelling ability at age level. The magnitude of the individual differences among children with WS was similar to the magnitude of individual differences found in the general population, as evidenced by the fact that the SDs for the WS sample on the standardized measures used in Study 1 were almost as large as for the general population and in Study 2 were as large as for the general population. At the same time, the means for the academic achievement measures for the samples included in this dissertation are almost

two SDs below the mean for the WIAT-III norming sample, indicating that, on average, the word-reading and spelling abilities of children and adolescents with WS are well below the average level of their chronological age peers from the general population.

The results reported in my dissertation extend previous findings regarding the literacy abilities of individuals with WS in at least six ways. First, Study 1 was the first longitudinal study to use standardized scores (SSs or T-scores) for both predictors and word-reading measures. The only previous longitudinal study of the reading abilities of children with WS (Steele et al., 2013) used raw scores (despite the 3-year range in age among the participants) and age equivalents. Second, Study 1 was the first to demonstrate, for children with WS, the indirect effect of vocabulary on later word reading through its effect on phonological awareness for children with WS. Third, Study 1 was also the first to document the positive longitudinal relation between systematic phonics instruction and later word-reading skills. Fourth, Study 2 characterized the spelling skills of children with WS using SSs for a considerably larger sample than the only previous study that reported SSs (Polse, 2013). Fifth, Study 2 was the first to systematically investigate the relations between word-reading and spelling skills for children and adolescents with WS. Finally, Study 2 was the first to demonstrate the relevance of systematic phonics instruction for individuals with WS not only for word reading but also for spelling skills.

As indicated in Chapter 1, I had two overarching goals for my dissertation research: 1) evaluate the applicability of theories of reading and spelling acquisition developed based on TD children for children with WS; and 2) provide results that will inform the development of targeted assessments and interventions. Regarding my first

overarching goal, the findings from the two studies reported on children with WS provide

support for the universality of theoretical models of reading development developed

based on TD children. Regarding my second overarching goal, findings from my

dissertation suggest that educational approaches known to be effective for children who

are having difficulty learning to read are likely to be appropriate for children with WS. In

the remainder of the Discussion, I consider the findings from both studies, highlighting

the similarities between the early literacy of children with WS and TD children.

Theoretical and educational implications and directions for future research are addressed.

Early Literacy of Children with Williams syndrome is Similar to Typically

Developing Children

Broadly, the findings from Study 1 and Study 2 indicate that the correlates of the

word-reading and spelling abilities of children with WS are similar to those for TD

children. This was demonstrated in at least four ways. First, I demonstrated a positive

relation between systematic phonics instruction and both word-reading ability (Study 1)

and spelling ability (Study 2) for children with WS. Systematic phonics has been shown

to be more effective than less-systematic phonics or no phonics for teaching word reading

to TD children as demonstrated by a large body of research (e.g., Castles et al., 2018;

Ehri, 2020; NRP, 2000). Second, similar to what has been demonstrated for TD children

(Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002) and children at

familial risk for reading difficulties (Hulme et al., 2015), the contribution of vocabulary

to word reading of children with WS, rather than being direct, was indirect through

phonological awareness. Third, in Study 2 spelling ability and reading ability were highly

correlated, which is consistent with findings for TD children (e.g., Ehri, 2000; Pan et al.,

2021; Peterson et al., 2021). Fourth, as would be expected given these findings, the results reported for children with WS are consistent with theories that were developed for children from the general population.

Theoretical Implications

The findings for children with WS presented in this dissertation fit with the previous results that were used to develop theoretical models of reading development (described in the previous chapters) for TD children or for children with reading difficulties. These models include: Phase Theory (Ehri, 1987, 2005, 2020), Word Identity Amalgamation Theory (Ehri, 2020), the lexical quality hypothesis (Perfetti & Hart, 2002), the triangle model (Seidenberg & McClelland, 1989), and the self-teaching hypothesis (Share, 1995). In addition, findings from Study 1 fit with the Lexical Restructuring Model (Metsala & Walley, 1998), a theoretical model of phonological awareness development. Consequently, my findings for children with WS provide support for the universality of these models, as described in the remainder of this section.

According to Ehri's Phase Theory (Ehri, 1987, 2005, 2020; see Chapter 1), phonological awareness is a central foundational skill for learning to read. In addition, although I was not able to investigate letter name/sound knowledge in my dissertation, this skill has been shown repeatedly to be essential for beginning to read (Clayton et al., 2020; Ehri, 2020; Ehri, Nunes, Stahl, et al., 2001; Ehri, Nunes, Willows, et al., 2001). To move from the pre-alphabetic phase to the partial and full alphabetic phases, the beginning reader needs to form grapheme-phoneme connections. To be successful in this task, young children need to be able to recognize and discriminate letter shapes and be aware of the sounds in speech. The Word Identity Amalgamation Theory (Ehri, 2020)

explains the last phase of Ehri's Phase Theory (consolidated alphabetic phase), stating

that to become a fluent reader, the child must bond the various aspects of a word together

as a lexical unit. These aspects include orthography (spelling), phonology, morphology,

syntax, and semantics (vocabulary). In oral language, most of these aspects have already

been bonded together. However, to complete the bonding process and allow for fluent

reading, the child must bond spellings to pronunciations. Knowledge of the alphabetic

system is the glue that bonds orthography to phonology. Decoding has a crucial role in

this process. (See also Share's [1995] self-teaching hypothesis.) As a result of decoding,

the spellings of words are stored in memory by forming connections between letter-sound

subunits within individual words. My findings for Study 1 and 2 are consistent with

Ehri's theories.

In addition to Ehri's (2020) theories, the lexical quality hypothesis (Perfetti &

Hart, 2002) and the triangle model (Seidenberg & McClelland, 1989) both emphasize the

importance of forming connections between orthography, phonology, and semantics in

order to read words automatically. All three theories also predict the high correlation

observed between spelling and reading ability for children with WS in Study 2. These

theories have in common that learning to spell allows for better understanding of the

alphabetic principle (i.e., the understanding that graphemes represent phonemes) and

facilitates learning letter-sound regularities, which are necessary for children to be able to

read words that they have not encountered before.

Phonological Awareness

Consistent with all the above-mentioned theories, the findings from both Study 1

and Study 2 demonstrated the key role of phonological awareness in word reading for

children with WS. In Study 1 (see Table 3), early phonological awareness was strongly correlated with word-reading ability measured three years later ($r = .62$, $p < .001$) and continued to explain significant unique variance in word reading even after accounting for visual-spatial perception, overall intellectual ability, and reading instruction method. In Study 2 (see Table 8), phonological awareness was strongly correlated with both word reading ($r = .77$, $p < .001$) and spelling ($r = .66$, $p < .001$) and continued to explain significant unique variance in word reading even after accounting for vocabulary, reading instruction method, and spelling performance. Theoretically, these findings suggest that phonological awareness has a central role in learning to read for children with WS, providing support for the universality of the central role of phonological awareness in learning to read. These results are consistent with previous studies of TD children (e.g., Caravolas et al., 2013; Clayton et al., 2020; see Melby-Lervåg et al., 2012 for a meta-analysis) and individuals with WS (Brawn et al., 2018; Laing et al., 2001; Levy et al., 2003; Levy & Antebi, 2004; Menghini et al., 2004; Mervis et al., 2021; Pezzino et al., 2021).

Visual-spatial Perception

As stated previously, to transition from the pre-alphabetic phase to the partial alphabetic phase (Phase Theory; Ehri, 1987, 2005, 2020), recognizing and differentiating letter shapes is key for the beginning reader to be able to start forming grapheme-phoneme connections. Consistent with Ehri's Phase Theory, in Study 1, the visual-spatial perception task – in which children had to match identical letter-like forms by discriminating among forms that differed only by spatial rotation – was strongly correlated with word reading ($r = .55$, $p < .001$) and continued to explain significant

unique variance in word reading even after accounting for phonological awareness, overall intellectual ability, and reading instruction method. These findings are consistent with Dessalegn et al.'s (2013) conclusions based on the two cases they reported. The two 16-year-olds with WS in their study differed considerably in their visual-spatial perception skills, which the authors suggest could be driving the striking difference in their word-reading skills. In sum, our results suggest that visual-spatial perception skills are particularly important in the early stages of learning to read, allowing children to differentiate letters that differ only in spatial orientation (e.g., *b* and *d*). Telling letters apart is difficult for very young TD children (e.g., Kaufman, 1980; Lachmann & Geyer, 2003) and considering the hallmark weakness in visual-spatial skills in individuals with WS (Mervis et al., 1999), differentiating letters might be particularly challenging and will probably be mastered more slowly.

Vocabulary

The simple correlation between vocabulary and word-reading skills was strong in both Study 1 ($r = .51$, $p < .001$) and Study 2 ($r = .61$, $p < .001$). However, consistent with findings from studies of TD children (e.g., Hjetland et al., 2019; Hulme et al., 2015; Storch & Whitehurst, 2002), the direct effect of vocabulary was not statistically significant after accounting for other relevant variables (phonological awareness, visual-spatial perception, intellectual ability, and reading instruction method in Study 1; phonological awareness, reading instruction method, and spelling in Study 2). On the other hand, the results of the mediation analysis presented in Study 1 demonstrated that vocabulary had an indirect effect on word-reading ability through phonological awareness. This finding is consistent with the Lexical Restructuring Model (Metsala &

Walley, 1998) that was developed based on TD children and also with previous

longitudinal studies of TD children (e.g., Hjetland et al., 2019; Hulme et al., 2015; Storch

& Whitehurst, 2002). According to the Lexical Restructuring Model, vocabulary

development contributes to phonemic segmentation. As vocabulary size increases,

children's phonological representations need to become more segmented to allow for the

differentiation of words that are phonologically similar. According to this model,

phoneme awareness emerges as a consequence of vocabulary development (Walley et al.,

2003). Based on Ehri's theories, the lexical quality hypothesis (Perfetti & Hart, 2002), the

triangle model (Seidenberg & McClelland, 1989), and findings from previous studies, I

expect that vocabulary will play a central role not only in word-reading, but also in

spelling development for children with WS and therefore should also be emphasized in

their literacy instruction. Consistently, van Rijthoven et al. (2021) demonstrated that

children with reading disability who have better semantic skills (including vocabulary)

not only performed better in spelling but were also able to make more progress during a

phonics intervention that incorporated direct spelling instruction than their peers with

reading disability with weaker semantic representations.

Although reading comprehension was not investigated in this dissertation, I have

previously demonstrated in a cross-sectional study (Mervis et al., 2021) that the Simple

View of Reading (Gough & Tunmer, 1986) applies to children with WS. According to

the Simple View of Reading, reading comprehension is the product of decoding and

listening comprehension. Therefore, both are necessary for successful reading

comprehension. In Mervis et al.'s (2021) study, 79% of the variance in reading

comprehension of 9-year-olds with WS was explained by the combination of word

reading and listening comprehension. Considering that comprehension is the final goal of reading, this finding adds to the theoretical implications of this dissertation.

Overall Intellectual Ability

Overall intellectual ability was strongly correlated with word reading in both Study 1 ($r = .54$, $p < .001$) and Study 2 ($r = .70$, $p < .001$) and with spelling in Study 2 ($r = .65$, $p < .001$). Despite these strong bivariate correlations, overall intellectual ability did not explain a statistically significant amount of unique variance in word-reading skills in Study 1, above and beyond what was explained by phonological awareness and visual-spatial perception (see Table 4). Findings from both Study 1 and Study 2 demonstrated that variations in overall intellectual ability did not play a central role in accounting for individual differences in word reading and spelling for the groups of children with WS who participated in these studies, whose intellectual ability ranged from severe intellectual disability to average intellectual ability. The partial correlations reported in Study 2 (see Table 9) further demonstrate that word reading and spelling are strongly correlated above and beyond overall intellectual ability. Together, these findings are consistent with Pezzino et al.'s (2021) finding that many individuals with ID learn to read. My results support this same conclusion for spelling. These findings are also consistent with the reading models previously described given that these models do not consider the role played by overall intellectual ability.

Along these lines, for the Study 1 sample, as demonstrated in the scatterplot in Figure 3, despite having a GCA below 40, one child with WS who was receiving systematic phonics instruction attained a word-reading SS of 76. Similarly, two children with GCAs below 60 who were receiving systematic phonics instruction attained word-

reading SSs of at least 85. For Study's 2 sample, one child whose GCA was below 50 attained a spelling SS above 85 (See Figure 4). This child was also receiving systematic phonics instruction. Finally, three children in the phonics instruction group whose GCA was below 60 attained a spelling SS above 70.

Method of Reading Instruction

My findings of a major contribution of systematic phonics instruction to both word reading in Study 1 (see Tables 4 and 5) and spelling in Study 2 (see Table 10) for children with WS are consistent with previous findings for TD children (e.g., Castles et al., 2018; National Reading Panel, 2000) and for individuals with ID (Reichow et al., 2019). These findings are also in alignment with Ehri's theories (Ehri, 1987, 2005; 2020) and provide further support for the use of systematic phonics instruction, integrated with systematic spelling instruction (Graham & Santangelo, 2014) for children with WS. These findings along with the findings for overall intellectual ability, provide hope for families of children with WS that if provided with adequate systematic phonics instruction integrated with systematic spelling instruction, their child will be able to attain their full potential in word reading, which in turn will likely have a positive impact on their community living skills (Brawn et al., 2018; Greiner de Magalhães & Mervis, 2021).

Figure 3

Association Between Overall Intellectual Ability at Time 1 and WIAT-III Basic Reading

Standard Score at Time 2 as a function of Reading Instruction Method: Study 1
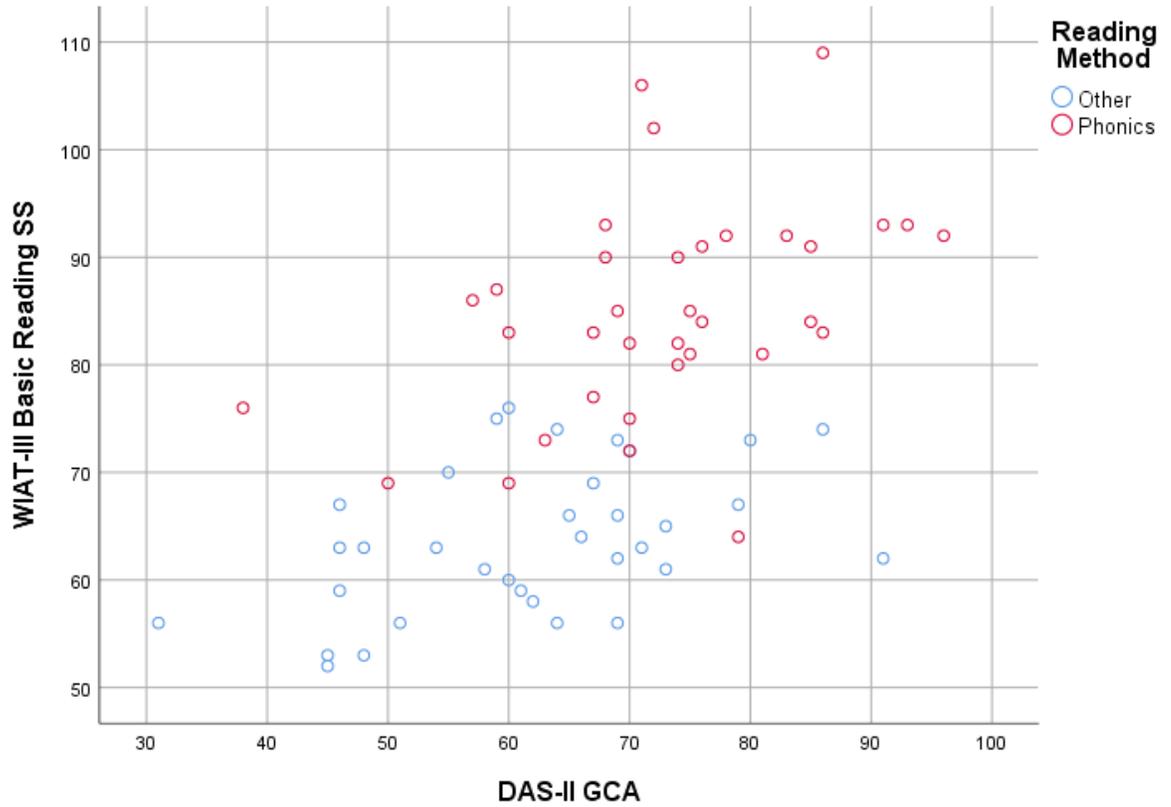
Note. Each dot represents an individual participant. Children in the Phonics instruction group are shown in red and children in the Other instruction group are shown in blue. *N* = 69.

Figure 4

Association Between Overall Intellectual Ability and Concurrent WIAT-III Spelling

Standard Score as a function of Reading Instruction Method: Study 2
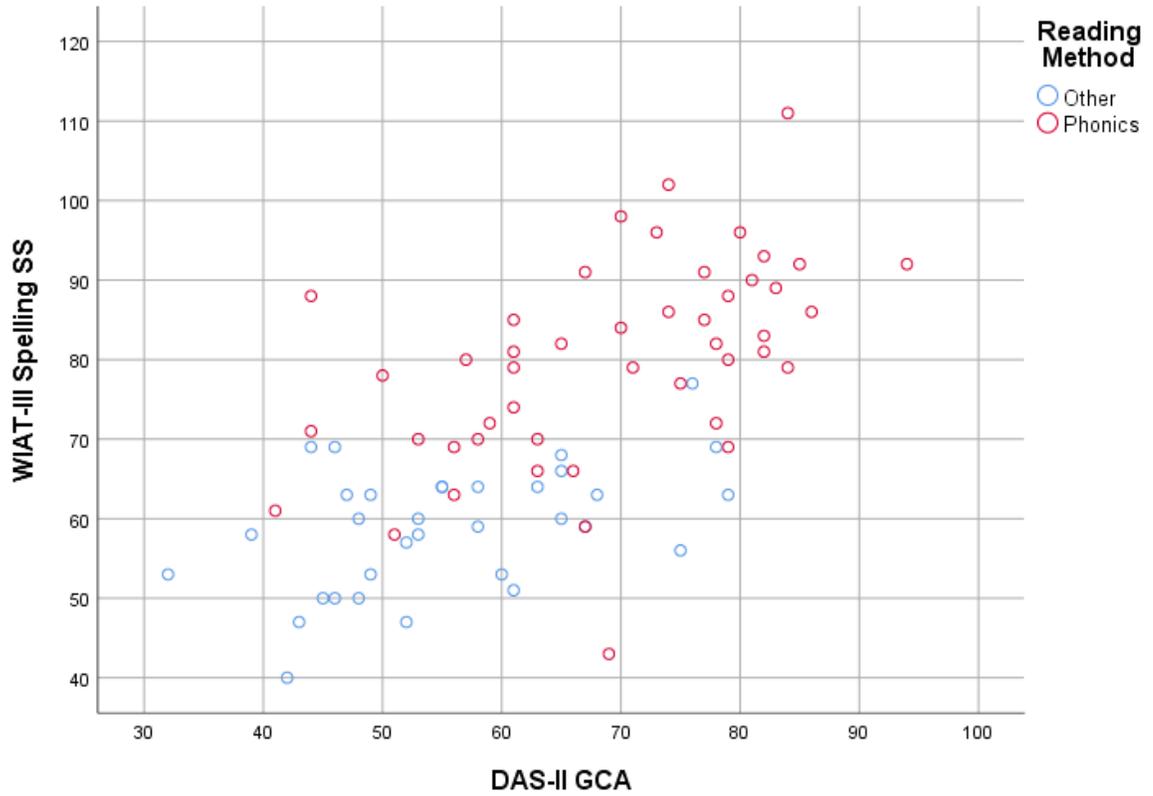

Note. Each dot represents an individual participant. Children in the Phonics group are

shown in red and children in the Other instruction group are shown in blue. *N* = 80.

Educational Implications

The first important point to make relative to practical educational implications is the presence of very large individual differences for both the children included in Study 1 and those included in Study 2. Individual differences were present not only for the academic achievement measures but also for all the correlates considered. Given this variability, it is important that well-designed individual assessments be performed using standardized assessments to address both the child's performance relative to the general population (as measured by SSs) and also the child's pattern of relative strengths and weaknesses among skills shown to be important for the development of literacy. A carefully designed assessment will inform the design of an intervention that is best suited for each child with WS. However, the general guidelines presented in this dissertation should be suitable for most children with WS and may be used as a basis for the intervention, keeping the unique characteristics of each individual child in mind. In sum, my findings suggest that a targeted word reading and spelling assessment for children with WS should include (at least) word reading and spelling, phonological awareness, letter name and letter sound knowledge, vocabulary, and visual-spatial perception (whether the child is able to differentiate the orientation of letter-like forms).

Regarding literacy intervention, the fact that my findings for children with WS fit with prior findings for TD children suggests that educational approaches that have been shown to be effective for TD children who are having difficulty learning to read are likely to be appropriate for children with WS. Therefore, literacy intervention should focus on explicitly teaching grapheme-phoneme relations for young children, which is best accomplished through systematic phonics instruction that integrates systematic

spelling instruction. In addition, phonological awareness training should be a key part of instruction, in addition to vocabulary and broad language skills. As emphasized by Ehri (2020), orthography (spelling), phonology, semantics (vocabulary), syntax, and morphology are all relevant to learning to read. In particular, studying morphology (e.g., '*ology*' means 'the study of') can lead to improvements in children's vocabulary and help with both reading and spelling skills (Joshi, 2019; Moats, 2019). Along these lines, Moats (2019) provided a detailed guide on how to integrate systematic phonics instruction with instruction that focuses on the structure of the language. As suggested by Mervis et al. (2021), such instruction is likely to be effective for children with WS.

Translating Findings from Research to Practice

The biggest challenge that the science of reading faces is translating the findings from research to practice (e.g., Seidenberg et al., 2020). It is my hope that the findings of this dissertation can be used by educational and clinical psychologists, principals, and teachers to address the needs of children with WS so they can attain their full potential in the area of literacy, which is likely to have a positive impact on other aspects of their lives (Brawn et al., 2018; Greiner de Magalhães & Mervis, 2021).

Although addressing the issue of translation is not easy, it is possible. For example, Ehri and Flugman (2018) demonstrated that a very intensive (135 hours) training for kindergarten – third grade teachers in urban, low SES schools, composed of a 45-hour course on how to teach phonics explicitly and systematically followed by a 90-hour year-long in-school mentoring, led to statistically significant improvements in teachers' ability to provide systematic phonics instruction to their students. In the instruction used in Ehri and Flugman (2018)'s study, teachers were taught to deliver

instruction that integrated the teaching of reading, spelling, and handwriting. In order to investigate if the gains in performance made from Fall to Spring by the students who participated in this intervention study were significantly larger than the typical gain expected, the gains made by this sample were compared to those in previous studies and to normative samples representative of the U.S. population. Results indicated that participants in Ehri and Flugman (2018)'s study had significantly greater growth in reading than might be expected if students had not received this intervention. These results suggest that the intensive training teachers received transferred to children's reading and spelling skills. For children with WS, elevating the knowledge of special education teachers on both *what* (academic content knowledge), and *how* (knowledge of instructional methods, or pedagogical knowledge) to teach their students is particularly important (Porter et al., 2021). Adequate implementation of teacher instruction is likely to lead to improvement in the word-reading and spelling performance of children with WS.

Protective Factors

According to Catts and Petscher's (2021) Cumulative Risk and Resilience Model, learning to read includes both risk and resilience factors that interact in complex ways to influence the reading development of children who are having difficulty learning to read. Their model is rooted in models that propose that no single deficit is necessary or sufficient to cause reading disability and that multiple risk and protective factors interact to cause difficulty in learning to read (e.g., Multiple Deficit Model; Pennington, 2006). The Cumulative Risk and Resilience Model is consistent with the proposal that developmental disorders are best explained by a constellation of strengths and

weaknesses, all of which must be taken into account in designing effective interventions (Astle & Fletcher-Watson, 2020). A similar model that applied the concept of resilience to understanding protective factors for children at high risk of developing a reading disability, who sometimes thrive in learning to read and comprehend despite their high risk, was proposed by Haft et al. (2016). In that model, cognitive protective factors included, among others, oral language skills, vocabulary, morphological awareness, and executive functioning. In addition, Haft et al. (2016) emphasized that low levels of task-focused behavior and high levels of task avoidance differentiated children who later developed reading difficulties from those who did not. Finally, in Haft et al. (2016)'s model, socio-emotional protective factors included, among others, positive peer relationships and teacher and parental support.

The Componential Model of Reading (Joshi, 2019) similarly emphasizes three domains that are important for reading development: cognitive, psychological, and ecological. The cognitive domain consists of the above-mentioned components of the Simple View of Reading (i.e., decoding and oral language). The psychological domain includes factors such as motivation and teacher knowledge, and the ecological domain includes factors such as home environment and socioeconomic status. Thus, beyond the cognitive and instructional factors investigated in this dissertation, psychological and ecological factors are also important for reading development.

The results of both Study 1 and 2 are consistent with these models, considering the interaction of factors that are considered a relative strength (phonological awareness) and a relative weakness (visual-spatial skills) in WS, and the central role of reading instruction in both studies. The results of this dissertation suggest that focusing on the

resilience factors proposed by Catts and Petscher (2021), such as instruction, growth

mindset, task-focused behavior, adaptive coping and strategies, and family and peer

support might be particularly important so individuals with WS can better face and

overcome challenges in attaining satisfactory literacy skills, which in turn would likely

have a positive impact on their adaptive skills (Brawn et al., 2018; Greiner de Magalhães

& Mervis, 2021).

Another important resilience factor highlighted by the Cumulative Risk and

Resilience Model (Catts & Petscher, 2021) and the Componential Model of Reading

(Joshi, 2019) and that was the focus of this dissertation is literacy instruction. As

demonstrated in both Study 1 and Study 2, having adequate literacy instruction via

systematic phonics instruction is a major predictor of word reading and is also strongly

related to spelling ability for children with WS. As demonstrated by Graham and

Santangelo's (2014) meta-analysis, for TD children, explicit and systematic instruction in

spelling has positive effects not only on spelling but also on reading skills. Considering

these meta-analytic findings, similar to what is recommended for TD children (Graham &

Santangelo, 2014), systematic phonics instruction for children with WS should

incorporate systematic spelling instruction. However, as described in Chapter 1, a

substantial proportion of early childhood teachers do not themselves know phonics

(Brady et al., 2009; Campbell et al., 2014; Cunningham et al., 2004, 2009; Nelson &

Machek, 2007; Porter et al., 2021), making translation of these research findings into

practice even harder. Along these lines, as part of the psychological domain, intensive

teacher training in systematic phonics can improve teachers' knowledge, which would be

expected to lead to improved instruction which in turn can improve students' academic performance (Joshi, 2019; see also Ehri & Flugman, 2018).

Considering the above-described models, strengthening protective factors may help children to thrive despite their risk for having reading difficulties. In this context, improving children's grit and resilience might be particularly important. In a 3-year longitudinal cohort study, Hossain et al. (2021) demonstrated that improved grit and resilience was associated with improved academic performance in children with reading difficulties. Despite their high levels of social motivation, children with WS have low levels of mastery motivation, "or the willingness to persevere on a moderately difficult task [that is not social in nature] and the expression of pleasure upon mastery the task" (Mervis & John, 2010, p. 241). Considering the importance of high levels of task-focused behavior and low levels of task avoidance for the successful development of reading skills (Eklund et al., 2013), interventions addressing the low levels of mastery motivation that are typical of children with WS (Rowe, 2007) may lead to increased persistence on task-related behavior, which in turn would be likely to have a positive impact on their academic performance as well as on their levels of independent living and employment success (Rowe, 2007).

Prevention and Early Intervention

Considering that the median age of diagnosis for children with WS in the United States is about 12 months (Kozel et al., 2021), opportunities to develop pre-literacy skills are present from an early age, which gives the child a much better chance to successfully learn to read. Focusing on prevention strategies (e.g., Catts & Hogan, 2021) such as implementing a validated curriculum like Letterland (Manson & Wendon, 2003;

Wendon, 1992) in preschool is particularly important. In most U.S. states, the diagnosis of WS makes any child with this syndrome eligible for free public preschool, and families and schools can use this opportunity to focus on the early protective factors that will help children with WS develop successful reading and spelling skills later. These protective factors include providing the child with a literacy-rich environment that fosters oral language and curiosity to learn. The findings from this dissertation suggest that the early literacy strategies shown to be effective for TD children (e.g., Ehri, 2020; National Reading Panel, 2000) are likely to be well suited for children with WS. As described by Dennis and Horn (2011), these include instruction that focuses on oral language, vocabulary, word combinations (grammar, syntax, morphology), phonological awareness, print awareness, and letter name/sound knowledge. Finally, a strategy that is likely to work for improving oral language is careful selection of books that are age and developmentally appropriate for each child and use of dialogic reading to engage young children with WS in book reading (Dennis & Horn, 2011).

Embedded letter mnemonics. Considering the crucial importance of explicitly teaching children letter-sound correspondences at a young age, strategies that support this process might be particularly helpful. For instance, the commercially available Letterland curriculum (Manson & Wendon, 2003; Wendon, 1992; see https://us.letterland.com/), a systematic phonics and foundational literacy program, provides a guide for teaching letter-sound correspondences to young children using embedded letter mnemonics. These letter shapes are embedded in a familiar object, action, or character whose name begins with that sound. For example, the letter S is drawn as the body of "Sammy Snake." This curriculum not only efficiently teaches letter-sound relations, which is the *sine qua non*

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skill for learning to read, but it also has positive effects on motivation and engagement of preschoolers during instruction (Roberts, 2021). The value of this curriculum has been supported by research with TD preschoolers (Roberts, 2021; Shmidman & Ehri, 2010), by the National Reading Panel (Ehri, Nunes, Stahl, et al., 2001), and by Ehri (2020). Mervis (2009) previously suggested that this curriculum might be especially useful for children with WS. In particular, considering the relation between visual-spatial perception and word reading found in Study 1, for young children with WS, embedding hard-to-differentiate letters (such as *d* and *b*) in objects, actions or characters might help to reinforce the visual-spatial differences between similar-appearing letters representing different sounds. Another advantage of this curriculum is its positive effect on motivation for learning to read (Roberts, 2021) which might help to address the low levels of mastery motivation of children with WS (Rowe, 2007), providing a fun and enjoyable way to learn letter-sound correspondences.

Future Directions

Future directions that are specific to each of the studies are described in chapters 2 and 3. In this section, general future directions are described.

There has been no research regarding the impact that difficulty learning to read has on the mental health of children with WS. However, it is known that having WS is associated with increased mental health problems, in particular anxiety (Kozel et al., 2021; Leyfer et al., 2006) and that reading ability is positively associated with community living skills in individuals with WS (Brawn et al., 2018; Greiner de Magalhães & Mervis, 2021). Considering the results of Haft et al.'s (2019) study that peer mentoring has a positive effect on socio-emotional and health outcomes of children with

learning disabilities, an intervention designed to train TD peers to support the learning of children with WS may be valuable. Further research is needed in this area, including the effects of this type of support on social-emotional and health outcomes of children with WS. The focus should be on the strengths of the individuals with WS, such as high social motivation and a relative strength in phonological awareness, and how these can be used to address the challenges in acquiring literacy that these children might encounter. In addition, peer mentors should be provided with ongoing training and support (Lansey et al., 2021). A program like this could also be suitable to address the mastery motivation problems children with WS face (Rowe, 2007) and might provide valuable support for individuals with WS in the transition between high school to post-secondary education) (college or vocational training) and/or meaningful employment.

Intervention studies that are adapted to the individual and general needs of children with WS should be performed to identify best practices in teaching children with WS letter-sound relations, word reading, spelling, and reading comprehension. Given the low level of mastery motivation characteristic of children with WS, which presumably has a negative impact on their academic skills, adaptive skills, and employability (Rowe, 2007), studies addressing whether interventions focusing on improving grit and resilience would have a positive impact on their academic performance, as Hossain et al. (2021) found for children with learning disabilities, would be valuable.

In Study 2, to describe the characteristics of the spelling skills of children and adolescents with WS relative to the general population, I scored the spellings of the children dichotomously – as correct or incorrect – following standardized procedures. However, there is more information available in children's spellings that could be further

studied. Future studies could investigate the types of spelling errors committed by children with WS using scoring procedures that take into consideration the plausibility of misspellings (Treiman et al., 2016), allowing researchers to address the hypothesis that, similar to TD children, children with WS base their spellings on English orthographic rules. Consistent with this hypothesis, in a pilot study conducted in my mentor's laboratory, Williamson et al. (2021) demonstrated that most school-aged children and adolescents with WS have some knowledge of English orthographic spelling rules.

Conclusions

The results presented in this dissertation demonstrate that the reading and spelling skills of children and adolescents with WS are on average considerably more limited than those of same-aged children in the general population. However, there is considerable variability among children with WS, with some not able to read or spell any words and others able to read and/or spell at age level. Several variables that account for much of this variability were investigated in my dissertation, including cognitive, language, and instructional factors. Cognitive factors investigated in this dissertation that are relevant to intervention in preschool and kindergarten years include vocabulary, phonological awareness, and visual-spatial perception. Systematic phonics instruction emerged as a key factor that positively contributed to better word-reading and spelling outcomes. Spelling is an important skill that reinforces the child's knowledge of the alphabetic principle and strengthens the connections between the various components involved in learning to read. Therefore, spelling should be explicitly taught as part of literacy instruction.

The findings for children with WS presented in this dissertation fit with previous results for children in the general population that were used to develop theoretical models of reading development for TD children or children with reading difficulties. This fact suggests that educational approaches shown to be effective for children who are having difficulty learning to read are likely to be appropriate for children with WS. Adaptations should be made to take into account the patterns of relative strengths and weaknesses in each child's abilities. Other factors that were discussed in this chapter but were not investigated in this dissertation – including mastery motivation, task-focused behavior, family/school and peer support, and intensive teacher instruction in evidence-based literacy practices – should also be considered so that children with WS have the opportunity to attain their full potential.

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APPENDIX

Supplemental Materials – Study 1

Regression Analyses for Wechsler Individual Achievement Test-III Word Reading and Pseudoword Decoding Subtests

Two additional multiple regression analyses with Time 1 Phonological Processing T-score, Matching Letter-like Forms T-score, General Conceptual Ability standard score (SS), and reading instruction method as the independent variables were conducted, one for each of the subtests included in the Wechsler Individual Achievement Test-III (WIAT-III; Wechsler, 2009) Basic Reading Composite. The independent variables were measured by performance on the Differential Ability Scales-II (DAS-II; Elliott, 2007). For the regression with Time 2 WIAT-III Word Reading SS as the dependent variable, R^2 = .70, adjusted R^2 = .68, $F(4,64) = 37.14$, $p < .001$. The effects of Phonological Processing T-score ($p = .002$, semi-partial $r = .22$), Matching Letter-like Forms T-score ($p = .041$, semi-partial $r = .14$), and reading instruction method ($p < .001$, semi-partial $r = .52$) were significant. For the regression with Time 2 WIAT-III Pseudoword Decoding SS as the dependent variable, R^2 = .72, adjusted R^2 = .71, $F(4,64) = 41.62$, $p < .001$. The effects of Phonological Processing T-score ($p = .008$, semi-partial $r = .18$), Matching Letter-like Forms T-score ($p = .029$, semi-partial $r = .15$), and reading instruction method ($p < .001$, semi-partial $r = .54$) were significant.

Regression Analyses Excluding Participants with Raw Scores of 0 on the WIAT-III Word Reading and/or Pseudoword Decoding Subtests

In some of the previous studies addressing the single-word reading ability of individuals with Williams syndrome (WS), participants who were not able to read any of

the real words on the assessment were excluded from the analyses. In other studies, participants who were not able to read any of the pseudowords were excluded. To provide parallel analyses, two additional multiple regressions including Time 1 Phonological Processing T-score, Matching Letter-like Forms T-score, General Conceptual Ability SS, and reading instruction method as the independent variables and Time 2 WIAT-III Basic Reading Composite SS as the dependent variable were conducted (Final model). In the present sample, three of the 69 participants (4.35%) did not read any of the real words correctly and 11 (15.94%) – including the three who did not read any of the real words correctly – did not read any of the pseudowords correctly. All 11 children were in the Other group. When the three children who did not read any real words correctly were excluded, $R^2 = .72$, adjusted $R^2 = .70$, $F(4, 61) = 39.09$, $p < .001$. There were significant effects of Phonological Processing T-score ($p = .006$, semi-partial $r = .19$), Matching Letter-like Forms T-score ($p = .024$, semi-partial $r = .16$), and reading instruction method ($p < .001$, semi-partial $r = .57$). When the 11 children who did not read any pseudowords correctly were excluded, $R^2 = .66$, adjusted $R^2 = .64$, $F(4, 53) = 26.04$, $p < .001$. There were significant effects of Phonological Processing T-score ($p = .007$, semi-partial $r = .22$), Matching Letter-like Forms T-score ($p = .040$, semi-partial $r = .17$), and reading instruction method ($p < .001$, semi-partial $r = .57$).

Table S1

Model Coefficients for the Simple Mediation Model Displayed in Figure 1

Antecedent		Consequent						
		Time 1 Phonological Processing T				Time 2 WIAT-III Basic Reading SS		
		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
Time 1 Vocabulary SS	a	0.77	0.09	< .001	c'	0.21	0.15	.169
Time 1 DAS-II Phonological Processing T		--	--	--	b	0.49	0.14	< .001
Constant	i_M	-24.98	7.96	.003	i_Y	36.41	9.73	< .001
		$R^2 = .51$				$R^2 = .40$		
		$F(1, 67) = 70.73, p < .001$				$F(2, 66) = 21.77, p < .001$		

Note. Coeff. = unstandardized coefficient; DAS-II = Differential Ability Scales-II; EVT-2 = Expressive Vocabulary Test-2; PPVT-4 = Peabody Picture Vocabulary Test-4; *SE* = standard error; SS = standard score; T = T-score; TRC = Test of Relational Concepts.

Mediation Analysis with Vocabulary Measured by a Composite Based on the Mean

of Each Child's Peabody Picture Vocabulary Test-4, Expressive Vocabulary Test,

and Test of Relational Concepts SSs

The kindergarten Oral Language measure used by Storch and Whitehurst (2002) in their path analysis included a measure of relational language in addition to measures of single-word receptive and expressive vocabulary. To provide a more direct comparison to Storch and Whitehurst (2002)'s findings, we created another composite measure based on the mean of each child's SSs on the Peabody Picture Vocabulary Test-4 (PPVT-4; Dunn & Dunn, 2007), the Expressive Vocabulary Test-2 (EVT-2; Williams, 2007), and the Test of Relational Concepts (TRC; Edmonston & Litchfield Thane, 1988) and conducted a second simple mediation analysis using this composite as the Vocabulary measure. The TRC was used because it was the closest available measure to the Clinical Evaluation of Language Fundamentals—Preschool (Wiig et al., 1992) Basic Concepts subtest, which Storch and Whitehurst (2002) included along with the Peabody Picture Vocabulary Test—Revised (PPVT–R; Dunn & Dunn, 1981), and the One Word Picture Vocabulary Test (Gardner, 1990 – a test of expressive vocabulary) in their kindergarten measure of Oral Language. The TRC measures conceptual /relational language. Five types of relational concepts are included: temporal (e.g., before/after), quantitative (e.g., many/few), dimensional (e.g., tall/short), spatial (e.g., under/over), and other (e.g., same/different). The TRC is normed for ages 3.00 – 7.99 years and yields a T-score ($M = 50$, $SD = 10$). T-scores were converted to SSs. For the present participants, the correlations between TRC SS and the other vocabulary measures was $r = .70$, $p < .001$ for

PPVT-4 SS; $r = .64$, $p < .001$ for EVT-2 SS; and $r = .69$, $p < .001$ for the PPVT-EVT composite.

Findings from a simple mediation analysis using the new Vocabulary composite measure, conducted using ordinary least squares path analysis (Hayes, 2018), once again indicated that Time 1 Vocabulary SS indirectly influenced Time 2 WIAT-III Basic Reading SS through its effect on Time 1 Phonological Processing T. As can be seen in Figure S1 and Table S2, children who had higher Time 1 Vocabulary SSs had higher Time 1 Phonological Processing Ts ($a = 0.68$), and children who had higher Time 1 Phonological Processing Ts also had higher Time 2 WIAT-III Basic Reading SSs even after controlling for Time 1 Vocabulary SSs ($b = 0.46$). A percentile bootstrap 95% confidence interval for the indirect effect ($ab = 0.31$) based on 5,000 bootstrap samples with seed number 25071990 was entirely above zero (0.12 to 0.51). There was no evidence that Time 1 Vocabulary SS influenced Time 2 WIAT-III Basic Reading SS independent of its effect on Time 1 Phonological Processing T ($c' = 0.23$, $p = .081$).

Table S2

Model Coefficients for the Simple Mediation Model Displayed in Figure S1

Antecedent		Consequent						
		Time 1 Phonological Processing T				Time 2 WIAT-III Basic Reading SS		
		Coeff.	*SE*	*p*		Coeff.	*SE*	*p*
Time 1 Vocabulary SS (PPVT-4, EVT-2, TRC)	*a*	0.68	0.08	< .001	*c'*	0.23	0.13	.081
Time 1 DAS-II Phonological Processing T		--	--	--	*b*	0.46	0.14	.001
Constant	i_M	-11.73	6.61	.081	i_Y	37.51	7.53	< .001
		$R^2 = .50$				$R^2 = .40$		
		$F(1, 67) = 66.35, p < .001$				$F(2, 66) = 22.74, p < .001$		

Note. Coeff. = unstandardized coefficient; DAS-II = Differential Ability Scales-II; EVT-2 = Expressive Vocabulary Test-2; PPVT-4 = Peabody Picture Vocabulary Test-4; *SE* = standard error; SS = standard score; T = T-score; TRC = Test of Relational Concepts.

Figure S1

Simple Meditation Model Predicting Time 2 WIAT-III Basic Reading Standard Score
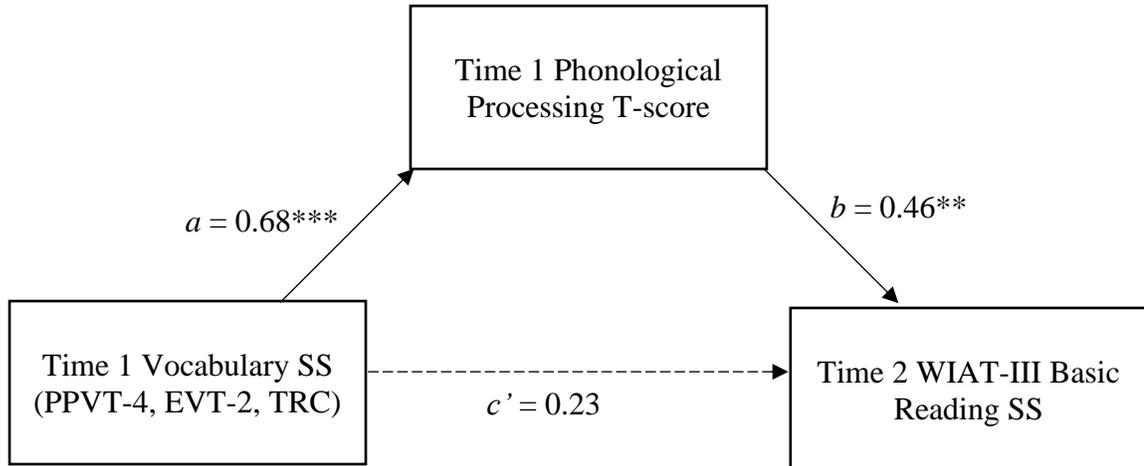
Note. This simple mediation model represents the indirect effect of Time 1 vocabulary

standard score (as measured by a composite based on the mean of each child's PPVT-4,

EVT-2 and TRC SSs) on Time 2 WIAT-III Basic Reading standard score through Time 1

Phonological Processing T-score. Statistics are unstandardized regression coefficients.

Solid lines indicate statistically significant relations and broken lines indicate statistically

nonsignificant relations. a = effect of X on the mediator; b = effect of the mediator on Y

partialling out the effect of X; c' = direct effect of X on Y; EVT-2 = Expressive

Vocabulary Test-2; PPVT-4 = Peabody Picture Vocabulary Test-4; SS = standard score;

TRC = Test of Relational Concepts; WIAT-III = Wechsler Individual Achievement Test-

III.

$**p < .01. ***p < .001.$

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1. University Fellowship, University of Louisville (2017 – 2018; 2020 – 2021).
2. Experimental Psychology Ph.D. Program Award for Excellence in Teaching. Department of Psychological & Brain Sciences, University of Louisville (Spring 2020).
3. 2019 Dr. M. Celeste Nichols Professional Development Award given by the University of Louisville Women's Center in conjunction with the George J. Howe Student Leadership Fund, the University of Louisville Women's Center, and a private donor (November 2019).
4. NIH Student Travel Award for the 40th Annual Symposium on Research in Child Language Disorders (SRCLD), University of Wisconsin-Madison (June 2019).
5. Experimental Psychology Ph.D. Program Award for Excellence in Research–Junior Researcher. Department of Psychological & Brain Sciences, University of Louisville (Spring 2018).
6. Graduate Network in Arts and Sciences Travel Funding. College of Arts & Sciences, University of Louisville (Fall 2017, Spring 2019, Fall 2019, Spring 2020).
7. Graduate Student Council Travel Award. School of Interdisciplinary and Graduate Studies, University of Louisville (Fall 2017, Spring 2018, Fall 2018, Fall 2019, Spring 2020).
8. Graduate Student Fellowship (January 2016 – December 2017). Department of Psychology. Universidade Federal de Minas Gerais, Belo Horizonte, Brazil.
9. Second place, Luiz Marcellino de Oliveira Prize in the Masters category at the 45th Annual Meeting Brazilian Society of Psychology (SBP). Universidade Federal de Minas Gerais, Belo Horizonte, Brazil (October, 2015).
10. Best poster, II World Dyslexia Forum. Universidade Federal de Minas Gerais, Belo Horizonte, Brazil (August, 2014).
11. Scientific Initiation Stipend (fellowship) (August 2011 – December 2014). Department of Psychology. Universidade Federal de Minas Gerais, Belo Horizonte, Brazil.
12. Scientific Initiation Stipend (fellowship) (August 2010 – August 2011). Department of Medicine. Universidade Federal de Minas Gerais, Belo Horizonte, Brazil.

RESEARCH

Peer-reviewed Publications

1. **Greiner de Magalhães, C.**, Cardoso-Martins, C., & Mervis, C. B. (in press). Spelling abilities of school-aged children with Williams syndrome. *Research in Developmental Disabilities.*

2. Mervis, C. B., **Greiner de Magalhães, C.**, & Cardoso-Martins, C. (2021). Concurrent predictors of word- reading and reading comprehension for 9-year-olds with Williams syndrome. *Reading and Writing: An Interdisciplinary Journal.* https://doi.org/10.1007/s11145-021-10163-4

3. Greiner de Magalhães, C., Pitts, C. H., & Mervis, C. B. (2021). Executive function as measured by the Behavior Rating Inventory of Executive Function-2: Children and adolescents with Williams syndrome. *Journal of Intellectual Disability Research* (special issue on Executive Function). https://doi.org/10.1111/jir.12858

4. Greiner de Magalhães, C., Mervis, C.B. & Cardoso-Martins, C. (2021). Cognitive predictors of arithmetic, reading, and spelling in Brazilian Portuguese-speaking children. *Reading and Writing: An Interdisciplinary Journal, 34*, 174-198. https://doi.org/10.1007/s11145-020-10062-0

5. Greiner de Magalhães, C., O'Brien, L. M. & Mervis, C. B. (2020). Sleep characteristics and problems of 2-year-olds with Williams syndrome: Relations with language and behavior. *Journal of Neurodevelopmental Disorders, 12*(32), 1-16. https://doi.org/10.1186/s11689-020-09336-z

6. Mervis, C. B. & **Greiner de Magalhães, C.** (in press). Williams syndrome. In M. Beauchamp, R. Peterson, M. D. Ris, H. G. Taylor, & K. O. Yeates (Eds.). *Pediatric neuropsychology: Research, theory, and practice*, 3rd ed. New York: Guilford.

7. Abreu, A., Ohno, P. M., **de Magalhães, C. G.**, & Barreto, I. S. (2016). Treinamento de pais e autismo: uma revisão de literatura [Parent training and autism: a literature review]. *Ciências & Cognição, 21*, 7-22.

8. Cardoso-Martins, C., Gonçalves, D. T., **de Magalhães, C. G.**, & da Silva, J. R. (2015). Word reading and spelling ability in school-age children and adolescents with autism spectrum disorders: evidence from Brazilian Portuguese. *Psychology & Neuroscience, 8*, 479-487. https://doi.org/10.1037/pne0000029

9. Cardoso-Martins, C., Gonçalves, D. T., & **de Magalhães, C. G.** (2013). What are the mechanisms behind exceptional word reading ability in hyperlexia?: Evidence from a 4-year-old hyperlexic boy's invented spellings. *Journal of Autism and Developmental Disorders, 43*, 3001-3003. https://doi.org/10.1007/s10803-013-1857-0

10. Barreto, I. S., **de Magalhães, C. G.**, Gonçalves, D. T., & Abreu, A. (2013). Processos de intervenção para crianças e adolescentes com Síndrome de Asperger: uma revisão de literatura [Intervention processes for children and adolescents with Asperger syndrome: A literature review]. *Contextos Clínicos, 6*, 132. http://dx.doi.org/10.4013/ctc.2013.62.06

11. de Magalhães, C. G., Barbosa, P. G., Abreu, C. S. D., & Cardoso-Martins, C. (2012). Early joint attention deficits in autism: evidence from a retrospective case study. *Revista Brasileira de Psiquiatria, 34*, 509-510. https://doi.org/10.1016/j.rbp.2012.04.004

Manuscripts Under Review

1. Haft, S. L., **Greiner de Magalhães, C**., & Hoeft, F. A meta-analytic review of the consequence of stigma and stereotype threat for individuals with specific learning disabilities. *Journal of Learning Disabilities.*

Conference Presentations (peer-reviewed)

1. Greiner de Magalhães, C., Cardoso-Martins, C., & Mervis, C. B. (2021, July). Spelling abilities of children and adolescents with Williams syndrome. Oral presentation to be presented at the Society for the Scientific Study of Reading Conference 2021 (online).
2. Greiner de Magalhães, C., O'Brien, L. M. & Mervis, C. B. (2021, July). Nonverbal reasoning ability and nighttime sleep duration predict expressive and receptive language ability in 2-year-olds with Williams syndrome. Poster to be presented at the International Association for the Study of Child Language, Philadelphia, PA. Originally accepted for presentation at the 2020 conference, which was canceled.
3. Greiner de Magalhães, C., Cardoso-Martins, C., & Mervis, C. B. (2021, June). Longitudinal predictors of word reading for children with Williams syndrome. Poster to be presented at the 41st annual, virtual Symposium on Research in Child Language Disorders.
4. Greiner de Magalhães, C. & Mervis, C. B. (2021, April). Emotion regulation and word reading ability as concurrent predictors of community living skills for children and adolescents with Williams syndrome. Poster presented at the Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities (online).
5. Greiner de Magalhães, C., Pitts, C. H., & Mervis, C. B. (2021, April). Executive functions of children and adolescents with Williams syndrome. Poster presented at the Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities (online). Originally accepted for presentation at the 2020 conference, which was canceled.
6. Greiner de Magalhães, C., Cardoso-Martins, C., & Mervis, C. B. (2020, July). Reading and spelling abilities of school-aged children with Williams syndrome. Poster accepted for presentation at the meeting of the Society for the Scientific Study of Reading, which was canceled.
7. Greiner de Magalhães, C., Mervis, C. B., & Cardoso-Martins, C. (2019, October). Cognitive predictors of reading, spelling, and arithmetic in Brazilian Portuguese-speaking children. Poster presented at the Cognitive Development Society Biennial Conference, Louisville, KY.
8. Greiner de Magalhães, C., Pitts, C. H., & Mervis, C. B. (2019, June). Phonological processing profiles of 9-year-old children with Williams syndrome and relations to word reading ability. Oral presentation at the Symposium on Research in Child Language Disorders, University of Wisconsin-Madison, WI.
9. Greiner de Magalhães, C. & Mervis, C. B. (2019, April). Mathematical abilities of 9-year-old children with Williams syndrome. Poster presented at the Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities, San Antonio, TX.
10. Gonçalves, D. T., Greiner de Magalhães, C., Mervis, C. B., & Cardoso-Martins, C. (2018, July). The development of orthographic knowledge in children with reading disabilities in Portuguese. Poster presented at the Society for the Scientific Study of Reading, Brighton, UK.
11. Greiner de Magalhães, C., Becerra, A. M., Pitts, C. H., O'Brien, L. M. & Mervis, C. B. (2018, July). Sleep disordered breathing is associated with increased

developmental delay and behavioral difficulties in Williams syndrome. Poster presented at the International Congress of Infant Studies, Philadelphia, PA.

12. Greiner de Magalhães, C., Becerra, A. M., Eovino, J., Speak, J. M., Thurman, A. J., Pitts, C. H., & Mervis, C. B. (2018, April). Lexical and grammatical abilities of 3- and 4-year-old children with Down syndrome or Williams syndrome. Poster presented at the Gatlinburg Conference on Research and Theory in Intellectual and Developmental Disabilities, San Diego, CA.

13. Piuzana-Barbosa, D. C. B., Greiner de Magalhães, C., & Abreu, A. (2016). Relato de um caso de autismo na 3ª infância [Report of a case of autism in early childhood]. Oral presentation at the XXV Encontro Brazileiro de Psicologia e Medicina Comportamental, Foz do Iguaçu, PR, Brazil.

14. Greiner de Magalhães, C., Gonçalves, D. T., & Cardoso-Martins, C. (2015). As crianças com dificuldades de matemática apresentam déficits no processamento fonológico?: Resultados preliminares [Do children with difficulties in mathematics have deficits in phonological processing?: Preliminary results]. Poster presented at the 45ª Reunião Anual da Sociedade Brazileira de Psicologia, Belo Horizonte, MG, Brazil.

15. Gonçalves, D. T., Greiner de Magalhães, C., Michalick-Triginell, M. F., & Cardoso-Martins, C. (2015). 'Febriu' ou 'febril'? O desenvolvimento do conhecimento ortográfico em crianças com dislexia do desenvolvimento ["Febriu" or "febril"? The development of orthographic knowledge in children with developmental dyslexia]. Poster presented at the 45ª Reunião Anual da Sociedade Brazileira de Psicologia, Belo Horizonte, MG, Brazil.

16. Greiner de Magalhães, C., Gonçalves, D. T., & Abreu, A. (2015). Atendimento da criança com TEA [Intervention with the child with Autism Spectrum Disorder]. Oral presentation at the IV Congresso Brazileiro de Terapia Cognitiva da Infância e Adolescência, Belo Horizonte, MG, Brazil.

17. Maranhoto, A. C. S., Greiner de Magalhães, C., Gonçalves, D. T., & Cardoso-Martins, C. (2014). Déficits de atenção em crianças com dificuldades na aprendizagem da leitura: um estudo transversal [Attention deficits in children with learning disabilities: A cross-sectional study]. Poster presented at the XXII Semana de Iniciação Científica da UFMG, Belo Horizonte, MG, Brazil.

18. Greiner de Magalhães, C., Gonçalves, D. T., & Cardoso-Martins, C. (2014). Dificuldades persistentes de leitura em português e sua relação com outros transtornos do desenvolvimento: o papel do processamento fonológico, da velocidade de processamento e da inteligência verbal e não-verbal [Persistent reading difficulties in Portuguese and their relation with other developmental disorders: The roles of phonological processing, processing speed, and verbal and nonverbal intelligence]. Poster presented at the XXII Semana de Iniciação Científica da UFMG, Belo Horizonte, MG, Brazil.

19. Gonçalves, D. T., Greiner de Magalhães, C., & Cardoso-Martins, C. (2014). Does speed of processing contribute to variance in reading ability? Evidence from Brazilian Portuguese. Poster presented at the II World Dyslexia Forum, Belo Horizonte, MG, Brazil.

20. Gonçalves, D. T., Greiner de Magalhães, C., & Cardoso-Martins, C. (2013). A relação entre o processamento fonológico e as habilidades acadêmicas: um estudo

longitudinal [The relation between phonological processing and academic abilities: A longitudinal study]. Oral paper presented at the XXII Congresso Brazileiro e II Congresso Internacional da Associação Brazileira de Neurologia, Psiquiatria Infantil e Profissões Afins, ABENEPI, Belo Horizonte, MG, Brazil.

21. Gonçalves, D. T., Greiner de Magalhães, C., & Abreu, A. (2013). Intervenção comportamental em uma criança autista: habilidades sociais [Behavioral intervention in a child with autism: Social skills]. Poster presented at the XXII Congresso Brazileiro e II Congresso Internacional da Associação Brazileira de Neurologia, Psiquiatria Infantil e Profissões Afins, ABENEPI, Belo Horizonte, MG, Brazil.

22. Greiner de Magalhães, C., Gonçalves, D. T., & Abreu, A. (2013). Intervenção no comportamento verbal de uma criança autista: um estudo de caso [Intervention in the verbal behavior of a child with autism: A case study]. Poster presented at the XXII Congresso Brazileiro e II Congresso Internacional da Associação Brazileira de Neurologia, Psiquiatria Infantil e Profissões Afins, ABENEPI, Belo Horizonte, MG, Brazil.

23. Greiner de Magalhães, C., Gonçalves, D. T., Barreto, I. S., Mendonça, L. M., Ribeiro, A. A., de Paula, L., & Cardoso-Martins, C. (2012). Dificuldades persistentes de leitura e escrita em português: resultados preliminares [Persistent difficulties of reading and spelling in Portuguese: Preliminary results]. Poster presented at the XXI Semana de Iniciação Científica da UFMG, Belo Horizonte, MG, Brazil.

24. Gonçalves, D. T., Greiner de Magalhães, C., Barreto, I. S., Mendonça, L. M., de Paula, L. D., Ribeiro, A. A., & Cardoso-Martins, C. (2012). Preditores precoces das habilidades de leitura e escrita no português [Early predictors of reading and spelling skills in Portuguese]. Poster presented at the XXI Semana de Iniciação Científica da UFMG, Belo Horizonte, MG, Brazil.

25. Greiner de Magalhães, C., Barreto, I. S., Gonçalves, D. T., & Cardoso-Martins, C. (2012). Há vantagens posteriores em adquirir a habilidade de leitura precocemente? [Are there any advantages in learning to read early?]. Poster presented at the VI Simpósio Internacional de Neurociências: Percepção, Belo Horizonte, MG, Brazil.

26. Gonçalves, D. T., Greiner de Magalhães, C., Barreto, I. S., & Cardoso-Martins, C. (2012). Habilidade de escrita na hiperlexia: evidência de um estudo de caso [Spelling ability in hyperlexia: Evidence from a case study]. Poster presented at the VI Simpósio Internacional de Neurociências: Percepção, Belo Horizonte, MG, Brazil.

27. Gonçalves, D. T., Greiner de Magalhães, C., Silva, J., R., & Cardoso-Martins, C. (2012). Estratégias de leitura em indivíduos com Transtornos do Espectro Autista. [Reading strategies used by individuals with autism spectrum disorders]. Poster presented at the I Congresso Mineiro de Neuropsicologia, Belo Horizonte, MG, Brazil.

28. Greiner de Magalhães, C., Barbosa, P. G., Abreu, C. S., Jesus, J. C., & Cardoso-Martins, C. (2012). O desenvolvimento da atenção compartilhada no autismo: um estudo de caso [The development of joint attention in autism: A case study]. Poster presented at the I Congresso Mineiro de Neuropsicologia, Belo Horizonte, MG, Brazil.

29. Gonçalves, D. T., Greiner de Magalhães, C., & Cardoso-Martins, C. (2012). O processamento fonológico prediz a habilidade inicial de matemática?: Evidência de um estudo longitudinal [Does phonological processing predict initial math skills ?: Evidence from a longitudinal study]. Poster presented at the I Congresso Mineiro de Neuropsicologia, Belo Horizonte, MG, Brazil.

30. Greiner de Magalhães, C., Gonçalves, D. T., Barreto, I. S., & Abreu, A. (2012). Processos de intervenção para crianças e adolescentes com síndrome de Asperger: uma revisão sistemática [Intervention for children and adolescents with Asperger syndrome: A systematic review]. Poster presented at the I Congresso de Terapias Cognitivas – a clínica em foco, Belo Horizonte, MG, Brazil.

31. Greiner de Magalhães, C., & Camargos, V. P. (2011). Fatores associados à satisfação com a vida em adolescentes de 11 a 17 anos em dois distritos de Belo Horizonte [Factors associated with life satisfaction in adolescents aged 11 to 17 years in two districts of Belo Horizonte]. Poster presented at the XX Semana de Iniciação Científica da UFMG, Belo Horizonte, MG, Brazil.

ADDITIONAL RESEARCH TRAINING

The Basics of Peer Review. APS free webinar with Amy Drew, Robert L. Goldstone, Erin B. Tone, and Becca White (August 2020).

Early Start Denver Model: Introductory Course. Taught by Thiago Lopes, with certificate from Sally Rogers, Ph.D., UC Davis, MIND Institute. Belo Horizonte, Minas Gerais, Brazil (2016).

Factorial and Principal Component Analysis (15 hours). Department of Psychology, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil (2015).

Course in Linear Regression (15 hours). Department of Psychology, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil (2015).

Introduction to R and Data Analysis (15 hours). Department of Psychology, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil (2012).

TEACHING AND MENTORING EXPERIENCE

University of Louisville, Louisville, KY, Department of Psychological and Brain Sciences

Online Education Technical Consultant (Spring 2021). Psychology 306 – Life Span Developmental Psychology: A Cultural Perspective and Psychology 201 – Introduction to Psychology.

Mentor, Psychology 492 Culminating Undergraduate Experience (CUE) Undergraduate Research (Fall 2020 – Spring 2021).

Graduate Teaching Assistant (Fall 2020). Psychology 609 – Language Development. (Graduate course, 10 students, 100% synchronous remote via Microsoft Teams).

Graduate Teaching Assistant (Fall 2020). Psychology 306 – Life Span Developmental Psychology: A Cultural Perspective. (Undergraduate course, 105 students, hybrid course via synchronous Blackboard Collaborate and Microsoft Teams).

Graduate Teaching Assistant (Summer 2020). Psychology 420 – History of Psychology. (Undergraduate course, 24 students, 100% remote via Blackboard Collaborate).

Graduate Teaching Assistant (Spring 2020). Psychology 306 – Life Span Developmental Psychology: A Cultural Perspective. (Undergraduate course, 153 students, in-person with transition to hybrid course via synchronous Blackboard Collaborate due to the pandemic).

Graduate Teaching Assistant (Fall 2019). Psychology 301 – Quantitative Methods in Psychology. (Undergraduate course, 99 students, in-person, I was responsible for two lab sections, one with 26 students and one with 23 students).

Universidade Federal de Minas Gerais, Belo Horizonte, Brazil, Department of Psychology

Graduate Teaching Assistant (Spring 2016, Fall 2016). Transtornos do Neurodesenvolvimento

[Neurodevelopmental Disorders]. (Undergraduate course, around 30 students each, in person).

ADDITIONAL TRAINING IN TEACHING

Introduction to Teaching in Higher Education. Graduate School, University of Louisville (Fall 2020). University of New Hampshire virtual conference on "Empower Students for Academic Success: Teaching Students Study Skills Informed by the Science of Learning" organized by Dr. Catherine Overson and Dr. Victor Benassi (November 6, 2020).

Remote Teaching: The Perusall Platform with Dr. Eric Mazur (August 14, 2020).

Blackboard Collaborate training session with Linda A. Leake, Instructional Technology Consultant Senior. Delphi Center for Teaching and Learning, University of Louisville (August 12, 2020).

iClicker Cloud Demo – Virtual session with Linda A. Leake, Instructional Technology Consultant Senior. Delphi Center for Teaching and Learning, University of Louisville (August 05, 2020).

DEPARTMENT/ UNIVERSITY SERVICE

University of Louisville, Louisville, KY, Department of Psychological and Brain Sciences

Planning Committee, reading group on diversity: So You Want to Talk about Race, by Ijeoma Oluo (Fall 2020 – Spring 2021).

Planning Committee, reading group on diversity: White Fragility, by Robin Diangelo (Summer 2020 – Fall 2020).

Brief survey evaluating the Experimental Psychology Peer Mentor Program (Summer 2020). Judge, Undergraduate Arts & Sciences Research Showcase, University of Louisville (April 2020). Judge, Junior Science & Humanities Symposium, Kentucky (February 2020).

Student Representative, Experimental Psychology Ph.D. Program (Fall 2018 – Spring 2020).

Planning Committee, Statistics Series Group. Department of Psychological & Brain Sciences (Spring 2019 – Fall 2019).