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# Longitudinal predictors of word reading for children with Williams syndrome

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## Abstract

We examined the cognitive, language, and instructional predictors of early word-reading ability in a sample of children with Williams syndrome longitudinally. At Time 1, sixty-nine 6–7-year-olds (mean age = 6.53 years) completed standardized measures of phonological awareness, visual-spatial perception, vocabulary, and overall intellectual ability. Word-reading instruction type was classified as (systematic) Phonics ($n = 35$) or Other ($n = 34$). At Time 2, approximately 3 years later (mean age = 9.47 years), children completed a standardized assessment of single-word reading ability. Reading ability at Time 2 varied considerably, from inability to read any words to word-reading ability slightly above the level expected for age. The results of a multiple regression indicated that Time 1 word-reading instruction type, phonological awareness, and visual-spatial perception (as assessed by a matching letter-like forms measure) 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. Exploratory analyses suggested that the relations between these factors were complex. Considered together, these findings strongly suggest that, in line with the Cumulative Risk and Resilience Model of reading disability, word-reading (dis) ability in Williams syndrome is probabilistic in nature, resulting from the interaction of multiple individual and environmental risk and protective factors. The results also have educational implications: early word-reading instruction for children with Williams syndrome should combine systematic phonics and phonological awareness training while also incorporating letter discrimination instruction highlighting the visual-spatial differences between similar-appearing letters.

**Keywords** Williams syndrome · Word-reading ability · Word-reading instruction type · Phonological awareness · Visual-spatial perception ability · Intellectual disability

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## Introduction

Acquiring functional literacy is foundational to full inclusion 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 important for providing guidance for targeted assessments and interventions that promote the development of functional literacy.

In the present study, we examined the cognitive, language and instructional predictors of early word-reading ability in children with Williams syndrome (WS), a rare genetic disorder associated with mild to moderate ID (Kozel et al., 2021). Although phonological awareness, a key predictor of word-reading ability in alphabetic orthographies (e.g., Caravolas et al., 2005, 2013; Landerl et al., 2013; Melby-Lervåg et al., 2012), is a relative strength for individuals with WS (e.g., Mervis, 2009; Mervis et al., 2022; Miezah et al., 2021), reading difficulty is very common. Some individuals with WS are not able to read at all, and only about 30% of older teenagers and adults attain functional literacy (Brawn et al., 2018). At the same time, several research groups have documented cases of individuals with WS who read at or above age level (Brawn et al., 2018; Levy & Antebi, 2004; Mervis, 2009; Mervis et al., 2022; Pagon et al., 1987). Taken together, this evidence suggests that, similarly to what has been recently proposed for reading disability in individuals without ID, word-reading (dis)ability in individuals with WS might best be conceived as resulting from a constellation of risk and resilience factors that interact to determine an individual's reading skills (e.g., Catts & Petscher, 2022; Haft et al., 2016; van Bergen et al., 2014; Yu et al., 2018; Zuk et al., 2021). The results of recent studies suggest that, in addition to phonological awareness (Brawn et al., 2018; Laing et al., 2001; Levy & Antebi, 2004; Levy et al., 2003; Menghini et al., 2004; Mervis et al., 2022), two other factors might play a critical role in learning to read for individuals with WS, namely visual-spatial skills (e.g., Brawn et al., 2018; Dessalegn et al., 2013; Mervis et al., 2022) and type of word-reading instruction (Levy & Antebi, 2004; Mervis, 2009; Mervis et al., 2022). However, all these studies were cross-sectional, and the participants in all but one were teenagers or adults.

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 such as small sample size and problems with the analytic strategy, including use of age equivalents and raw scores rather than standard scores (SSs), and lack of control for chronological age despite a 3-year range at the start of the study. Furthermore, the study did not address the role of reading instruction type, a major contributing factor to the reading outcome of children at risk for reading disabilities (e.g., Catts & Petscher, 2022). In the present article, we report the results of a longitudinal study examining the contributions of early phonological awareness, visual-spatial perception, and type of word-reading instruction, along with more general verbal and nonverbal abilities, to later word-reading ability in a relatively large sample of similar-aged children with WS. In the remainder of the Introduction, we briefly review the theoretical framework for this study and the relevant empirical literature and then describe the design of the study.

## Theoretical framework: Cumulative Risk and Resilience Model

According to Catts and Petscher's (2022) Cumulative Risk and Resilience Model, reading disability results from the confluence of multiple factors that interact to determine an individual's reading skills. Like other multifactorial models, the Cumulative Risk and Resilience Model emerged in response to increasing evidence against single-deficit models of reading disability (see Pennington, 2006, for a review). Take, for example, the phonological deficit hypothesis. Although there is general agreement that phonological awareness is a key predictor of word-reading ability (Caravolas et al., 2005; Ehri et al., 2001a, 2001b; Landerl et al., 2013), an isolated deficit in phonological awareness fails to account for most cases of reading disability. For example, although 73% of the participants with reading disability in Pennington et al. (2012) had a deficit in phonological awareness, most of them also had at least one other deficit. And 27% of the participants with reading disability did not have a deficit in phonological awareness. Likewise, 30% of the participants in Study 1 in O'Brien and Yeatman (2021) did not have a deficit in phonological awareness. Although phonological awareness ability significantly differentiated the group with reading disability from the group without reading disability in O'Brien and Yeatman's study, there was approximately 50% overlap in phonological awareness ability between the two groups, suggesting that some of the participants without reading disability would be misclassified as reading disabled based on their performance on the phoneme awareness measure.

Similar results have been found for other deficits frequently associated with reading disability, including deficits in processing speed, rapid naming, oral language, and visual processing (Catts & Petscher, 2022; O'Brien & Yeatman, 2021; Pennington et al., 2012), suggesting that no single deficit or combination of deficits is necessary or sufficient for the development of reading disability. Clearly, in addition to risk factors, resilience or protective factors must be taken into consideration in accounting for reading disability (Catts & Petscher, 2022).

Several potentially important risk and resilience predictors of word-reading (dis)ability were examined in the present study, namely phonological awareness, visual-spatial perception, vocabulary, overall intellectual ability, and type of word-reading instruction. Below, we review what is known about the contributions of these factors to variance in word-reading ability, first for TD children and individuals with ID other than WS and then for individuals with WS.

## Predictors of word reading for typically developing children and individuals with intellectual disabilities

### Phonological awareness

Phonological awareness is the ability to consciously attend to and manipulate speech sounds. Phonological awareness skills are central to word-level reading because children learning to read in an alphabetic writing system need to learn the arbitrary grapheme-phoneme correspondences that capture the sound sequences in the speech stream (Castles et al., 2018; Ehri, 2014; Kilpatrick, 2020). This ability 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 et al., 2001a, 2001b).

The results of cross-sectional studies have also shown that phonological awareness is strongly correlated with word reading for both children (Saunders & DeFulio, 2007) and adults (Wise et al., 2010) with ID of mixed etiology, children with Down syndrome (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 Down syndrome (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, 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 (Treiman et al., 2014), 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 strong visual-spatial perception skills performed significantly better on word reading than children with weak 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 examined the relation between visual-spatial perception ability and reading for individuals with ID.

### Vocabulary

Vocabulary knowledge also is associated with word reading ability. There are at least three reasons for this relation. First, the acquisition of phonologically-similar words contributes to the development of phonological awareness (Metsala & Walley, 1998). Second, to become a skilled reader, the child must bond meanings to spellings and pronunciations (Ehri, 2020). Third, vocabulary knowledge is essential for deciphering words whose spelling does not follow common letter-sound correspondences (Nation & Snowling, 2004; Ricketts et al., 2007; Tunmer & Chapman, 2012). Researchers have reported significant bivariate correlations between vocabulary ability and 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), and fragile X syndrome (Adlof et al., 2015).

In some of the studies of TD children, the relation between vocabulary and word reading ability remained statistically significant even after controlling for other relevant factors such as nonverbal ability and phonological skills (Nation & Snowling, 2004; Ouellette & Beers, 2010; Ricketts et al., 2007). However, other studies investigating the relation between vocabulary and word reading for TD children did not find a significant relation after accounting for initial word reading, phoneme awareness, and/or letter name/sound knowledge (Muter et al., 2004) or chronological age (Steele et al., 2013). Prior studies addressing the relation between vocabulary ability and word reading by children with ID have not controlled for these additional variables, so it is unknown whether the relation would remain statistically significant after taking these types of skills into account.

### 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).

### Type of reading instruction

Phonological awareness is crucial for cracking the alphabetic code, but this ability does not develop spontaneously (Morais et al., 1979). In natural speech, phonemes run together, making it difficult to pinpoint where one ends and the next begins. Systematic phonics instruction, which both teaches children to segment words into their constituent phonemes and teaches letter-sound relations systematically, allows children to decode by translating printed words into their spoken counterparts based on grapheme-phoneme connections. There is a large body of evidence demonstrating that systematic phonics instruction is a significantly more effective method for teaching word reading to TD children than approaches that do not teach these relations systematically (e.g., Castles et al., 2018; Ehri et al., 2001a, 2001b; Moats, 2019; Rose, 2006). 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, 2022).

The literature on reading abilities of individuals with WS is limited (see Brawn et al., 2018; Mervis, 2009, 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., 2022). For example, strong correlations have been found between word-reading skills and phonological awareness (Brawn et al., 2018; Laing et al., 2001; Levy & Antebi, 2004; Levy et al., 2003; Menghini et al., 2004; Mervis et al., 2022). 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. (2022) found that phonological processing skills contributed significant unique variance to word-reading ability even after the effects of word-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., 2022). However, vocabulary did not contribute unique variance to word-reading skills after accounting for the contribution of word-reading instruction method, phonological skills, nonverbal reasoning, visual-spatial skills, verbal working memory, and rapid naming (Mervis et al., 2022).

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., 2022). 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. (2022) found that the correlation between word reading and word-reading instruction method was significantly stronger than the correlation between word reading and overall intellectual ability. In addition, the correlation between word reading and word-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 word-reading instruction method. As suggested by the results discussed previously (e.g., Dessalegn et al., 2013; Mervis et al., 2022), it is unlikely that overall IQ is the primary factor determining word-reading performance for individuals with 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 = 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. Partial correlations between Time 1 phonological awareness and Time 2 word reading controlling for Time 1 word reading or between Time 1 letter-name/sound knowledge and Time 2 word reading controlling for Time 1 word reading were not reported, so it is unknown if these partial correlations also were statistically significant. 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-same/sound 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, even though raw scores rather than SSs were used, chronological age was not controlled for, and the sample size was small.

## Current study

Reading ability varies widely among individuals with 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., 2022; 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 main research question was: What are the longitudinal predictors (measured at ages 6–7 years) of word reading abilities (measured at ages 9–10 years) for children with WS? 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 3 years later, at Time 2.[1]

The following predictors were considered: phonological awareness, visual-spatial perception, overall intellectual ability, vocabulary, and word-reading instruction type. Based on prior cross-sectional or longitudinal findings for TD children (e.g., Ehri et al., 2001a, 2001b; Greiner de Magalhães et al., 2021; Melby-Lervåg et al., 2012) or children with WS (Brawn et al., 2018; Mervis et al., 2022; Steele et al., 2013), we predicted that both phonological awareness and word-reading instruction

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

type 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). Based on prior findings for TD children (e.g., Muter et al., 2004) and children with WS (Brawn et al., 2018; Mervis et al., 2022; Steele et al., 2013), 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.

# 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. Almost all were participating in a longitudinal study of the language and cognitive development of children with WS in which they had been enrolled as toddlers or preschoolers. Parents had contacted the senior author to enroll their child after hearing presentations that she gave about language and cognitive development at national or regional meetings of the Williams Syndrome Association or after finding out about her research from other parents of children with WS or from the physician who diagnosed the child with WS. Sixty-one of the participants also were included in the cross-sectional study reported in Mervis et al. (2022). For 55 of them, the Time 2 assessment in the present study is the same assessment that was included in Mervis et al. (2022). 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% Asian 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 3 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 0.95 and 0.96, 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 0.94 and 0.90, 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 ($r = 0.87$). The IRT-based internal consistency coefficients for 6- and 7-year-olds in the norming sample were 0.77 and 0.64, 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 $r = 0.84$, $p < 0.001$. Split-half internal consistency for 6- and 7-year-olds in the norming sample ranged from 0.94 to 0.95 for the PPVT-4 and from 0.90 to 0.95 for the EVT-2.

**Word-reading instruction type** The primary approach to teaching word reading to each child was classified as Systematic Phonics (hereafter, Phonics) or Other following the procedure described in Mervis et al. (2022) and Greiner de Magalhães et al. (2022). All available information related to the students' word-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). Word-reading instruction was classified as "Phonics" if the primary approach to teaching word reading was based on systematic instruction in English phonics (either synthetic or analytic). Systematic phonics instruction included programs such as Fundations, Orton-Gillingham, Lindamood-Bell, Wilson Language Training-Phonics, Letterland, and Logic of English. Word-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 as the primary approach for figuring out an unknown word or if it focused on whole-word instruction. Examples of programs classified as Other include Edmark, Fountas and Pinnell Literacy, and Units of Study. Many of the children whose reading instruction was classified as Other were being taught to read with materials developed by their teachers rather than with formal reading programs.

The primary word-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, $t(67) = 0.18$, $p = 0.855$, Cohen's $d = 0.04$; or Time 2, $t(67) = 0.16$, $p = 0.876$, Cohen's $d = 0.02$. A Mann–Whitney U test indicated that the distribution of grade did not differ significantly as a function of Word-reading Instruction type group either at Time 1 ($Mdn$ = Kindergarten, $IQR$ = Kindergarten–Kindergarten for each group), $z = 0.32$, $p = 0.752$, Cohen's $d = 0.08$, or at Time 2 ($Mdn$ = 3rd grade, $IQR$ = 2nd grade–3rd grade for each group), $z = 0.20$, $p = 0.844$, 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 $r = 0.90$, $p < 0.001$. According to the WIAT-III technical manual, split half internal consistency for both Word Reading and Pseudoword Decoding was 0.98 for the 9-year-olds in the norming sample and 0.97 for the 10-year-olds. Split half internal consistency for Basic Reading Composite was 0.99 for the 9-year-olds in the norming sample and 0.98 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 2-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. Funds were available to offset travel expenses for families for whom these expenses would have been a financial hardship.

## Data analysis

Data were analyzed using IBM SPSS v. 27. 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. 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).

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 word-reading instruction type. 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 word-reading instruction type. Given the very high correlation between GCA and Vocabulary SS ($r = 0.83$), GCA was not included in the second regression analysis.

**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 |

$N = 69$. *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 years | 6.00–7.80 years | < 6.00[a]–13.00 years |
| Pseudoword Decoding | 6.40 years | < 6.00[a]–7.40 years | < 6.00[a]–12.40 years |
| *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 |

$N = 69$. Age equivalents are not available for Basic Reading Composite

*WIAT-III* Wechsler Individual Achievement Test-III

[a]Lowest possible age equivalent

## Results

### 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 moderate-severe disability to average or above average for the general population. 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 subtest 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, $t(68) = 1.00$, $p = 0.320$, Cohen's $d = 0.05$.

### Multiple regression analyses: longitudinal predictors of word reading abilities

Maternal Education level was not significantly correlated with Basic Reading SS ($r = 0.05$, $p = 0.657$). Therefore, Maternal Education was not included in the multiple regression models. Pearson correlations ($\alpha = 0.01$) among the variables included in the regression analyses are reported in Table 3. All correlations were statistically significant.

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

**Table 3** Bivariate correlations among the measures included in the regression analyses

| Measure | 2 | 3 | 4 | 5 | 6 | 7 | 8 |
|---|---|---|---|---|---|---|---|
| 1. Word-reading instruction type | 0.40* | 0.37* | 0.41* | 0.34* | 0.78** | 0.75** | 0.77** |
| 2. Time 1 Phonological Processing T | | 0.52** | 0.72** | 0.72** | 0.62** | 0.61** | 0.60** |
| 3. Time 1 Matching Letter-like Forms T | | | 0.70** | 0.65** | 0.55** | 0.53** | 0.55** |
| 4. Time 1 General Conceptual Ability | | | | 0.83** | 0.56** | 0.54** | 0.56** |
| 5. Time 1 Vocabulary SS | | | | | 0.53** | 0.51** | 0.54** |
| 6. Time 2 Basic Reading Composite SS | | | | | | – | – |
| 7. Time 2 Word Reading SS | | | | | | | 0.90** |
| 8. Time 2 Pseudoword Decoding SS | | | | | | | |

$N = 69$. *T* T-score, *SS* standard score

*$p < 0.01$, **$p < 0.001$

**Table 4** First multiple regression analysis predicting Time 2 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 | < 0.001 | [63.61, 68.77] | | |
| Phonological Processing T | 0.30 | 3.19 | 0.002 | [0.11, 0.48] | 0.20 | 0.16 |
| Matching Letter-like Forms T | 0.21 | 2.33 | 0.023 | [0.03, 0.38] | 0.15 | 0.09 |
| General Conceptual Ability | −0.04 | −0.38 | 0.707 | [−0.26, 0.18] | −0.02 | < 0.01 |
| Word-reading instruction type | 16.42 | 8.59 | < 0.001 | [12.60, 20.25] | 0.54 | 1.15 |
| | $R^2 = 0.75$, adjusted $R^2 = 0.73$, $F$ (4, 64) = 47.19, $p < 0.001$ | | | | | |

$N = 69$. All continuous independent variables were measured at Time 1 and centered on the sample mean

*WIAT-III* Wechsler Individual Achievement Test-III, *CI* confidence interval, *T* T-score, *SS* standard score. Word-reading instruction type was classified as Phonics (coded as 1) or Other (coded as 0)

**Table 5** Second multiple regression analysis predicting Time 2 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 | < 0.001 | [44.37, 82.95] | | |
| Phonological Processing T | 0.26 | 2.78 | 0.007 | [0.07, 0.45] | 0.18 | 0.12 |
| Matching Letter-like Forms T | 0.18 | 2.15 | 0.035 | [0.01, 0.34] | 0.14 | 0.07 |
| Vocabulary SS | 2.56 | 0.27 | 0.789 | [− 16.53, 21.65] | 0.02 | < 0.01 |
| Word-reading instruction type | 16.37 | 8.58 | < 0.001 | [12.56, 20.18] | 0.54 | 1.15 |
| | $R^2 = 0.75$, adjusted $R^2 = 0.73$, $F$ (4, 64) = 47.12, $p < 0.001$ | | | | | |

$N = 69$. All continuous independent variables were measured at Time 1 and centered on the sample mean

*WIAT-III* Wechsler Individual Achievement Test-III, *CI* confidence interval, *T* T-score, *SS* standard score. Word-reading instruction type was classified as Phonics (coded as 1) or Other (coded as 0)

other predictors. After controlling for the effects of the remaining independent variables, a 1-point increase in Phonological Processing T-score resulted in a 0.30-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 0.21-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 word-reading instruction type (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 effects 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 Supplemental Materials). In addition, as reported in the Supplemental Materials, 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.

## Exploratory analyses of more complex relations between phonological awareness, visual-spatial perception, and word-reading instruction type

To begin to address the possibility of more complex relations between phonological awareness ability, visual-spatial perception ability, and word-reading instruction type in accounting for the variation in single-word reading ability of children with WS, we dichotomized phonological awareness ability and visual-spatial perception ability into deficit (< 10th percentile for the general population; T < 37) and low average + (≥ 10th percentile; T ≥ 37). We then crossed these two variables with Word-reading Instruction type to form eight groups. In Table 6, we report

**Table 6** Performance as a function of group (Word-reading Instruction Type X Phonological Processing Skill Level X Visual-Spatial Skill Level)

| Measure | Group | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 |
| N | 13 | 3 | 8 | 10 | 4 | 3 | 9 | 19 |
| Word-reading instruction type | Other | Other | Other | Other | Phonics | Phonics | Phonics | Phonics |
| Phonological Processing T (classification) | Deficit | Deficit | Low Av+ | Low Av+ | Deficit | Deficit | Low Av+ | Low Av+ |
| Matching Letter-like Forms T (classification) | Deficit | Low Av+ | Deficit | Low Av+ | Deficit | Low Av+ | Deficit | Low Av+ |
| Basic Reading Composite SS ≥ 81 (≥ 10th percentile) | 0% | 0% | 0% | 0% | 50% | 33% | 67% | 89% |
| Basic Reading Composite SS—Mean | 60.00 | 68.00 | 62.25 | 68.50 | 77.50 | 74.00 | 79.78 | 90.79 |
| Basic Reading Composite SS—SD | 6.71 | 7.00 | 4.37 | 5.76 | 6.46 | 8.08 | 6.30 | 8.29 |
| Basic Reading Composite SS—range | 52–75 | 63–76 | 56–70 | 58–74 | 69–83 | 64–85 | 69–87 | 77–109 |
| Phonological Processing T—Mean | 24.31 | 30.67 | 43.63 | 46.10 | 23.25 | 29.00 | 48.11 | 53.58 |
| Phonological Processing T—SD | 9.66 | 5.77 | 3.85 | 6.12 | 12.58 | 7.00 | 3.95 | 5.47 |
| Matching Letter-like Forms T—Mean | 20.69 | 39.67 | 28.25 | 44.40 | 25.75 | 53.00 | 29.33 | 48.32 |
| Matching Letter-like Forms T—SD | 8.62 | 0.58 | 3.28 | 10.38 | 10.53 | 10.39 | 4.53 | 9.80 |
| GCA—Mean | 52.46 | 59.67 | 62.50 | 73.90 | 62.00 | 70.33 | 65.33 | 78.89 |
| GCA—SD | 11.00 | 13.50 | 7.25 | 9.64 | 21.91 | 8.08 | 6.40 | 8.67 |
| GCA—Range | 31–69 | 46–73 | 51–73 | 62–91 | 38–86 | 63–79 | 57–75 | 67–96 |
| Vocabulary SS—Mean | 73.35 | 88.00 | 85.56 | 89.50 | 75.88 | 84.83 | 85.72 | 97.18 |
| Vocabulary SS—SD | 11.01 | 8.35 | 7.31 | 13.85 | 15.50 | 5.25 | 4.01 | 8.06 |

Deficit: < 10th percentile (T-score < 37); Low Av+: low average range or above (≥ 10th percentile; T-score ≥ 37)

*T* T-score, *SS* standard score, *GCA* General Conceptual Ability (similar to IQ) basic descriptive statistics for performance on the dependent variable and each of the continuous independent variables included in the regression analyses for each of the eight groups. We also dichotomized Basic Reading Composite into deficit (below the 10th percentile; SS < 81) and low average + (≥ 10th percentile; SS ≥ 81) and report the percentage of participants in each group in the low average + classification. The < 10th percentile criterion for "deficit" was based on Pennington et al. (2012).

As indicated in Table 6, 38% of the participants (74% of those being taught word reading using a systematic phonics approach and 0% of those being taught word reading with other types of approaches) scored at least in the low average range for the norming sample on the composite word reading measure. Two-thirds of the participants (67%) scored at least in the low average range on our measure of phonological awareness, and more than half (51%) scored at least low average on our measure of visual-spatial perception. Of the children with word reading composites at or above the low average range, all were being taught word reading with a systematic phonics approach. Although 88% had phonological awareness scores at least in the low average range (that is, were in Groups 7 or 8) and 69% had visual-spatial perception scores at least in the low average range (that is, were in Groups 6 or 8), 8% scored below the 10th percentile on both measures (that is, were in Group 5). At the same time, 50% of the participants who scored at least in the low average range on phonological awareness scored below the 10th percentile on word reading (100% of Groups 3 and 4, 33% of Group 7, and 11% of Group 8), as did 49% of the children who scored at least in the low average range on visual-spatial perception (100% of Groups 2 and 4, 67% of Group 6, and 11% of Group 8).

## Discussion

The present study examined longitudinal predictors of word-reading ability in a relatively large sample of children with WS. In line with the results of previous studies of individuals with WS (e.g., Brawn et al., 2018; Levy & Antebi, 2004; Mervis et al., 2022; Pagon et al., 1987), word-reading ability varied considerably, from inability to read any words to word-reading ability above chronological age. Consistent with our predictions, word-reading instruction type, phonological awareness, and visual-spatial perception at ages 6–7 years (Time 1) each 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 either overall intellectual ability or vocabulary was taken into account. In what follows, we discuss the theoretical and educational implications of these findings.

### Longitudinal predictors of word-reading ability in Williams syndrome: theoretical implications

As predicted, phonological awareness at age 6–7 years was strongly correlated with single-word reading ability at age 9–10 years ($r = 0.62$). Although a significant relation between phonological processing and word reading has been reported previously in several cross-sectional studies of individuals with WS (Brawn et al., 2018; Laing et al., 2001; Levy & Antebi, 2004; Levy et al., 2003; Menghini et al., 2004; Mervis et al., 2022) as well as in studies of individuals with Down syndrome (Cardoso-Martins & Frith, 2001; see Lemons & Fuchs, 2010 for a review and Næss, 2016 for a meta-analysis), children with reading disability and TD children (see Melby-Lervåg et al., 2012 for a meta-analysis), the present findings provide the first evidence that early phonological awareness ability is related to word reading ability several years later for children with WS. This longitudinal finding is consistent with previous longitudinal findings for TD children (e.g., Caravolas et al., 2013; Clayton et al., 2020) and children with reading disability (e.g., Snowling et al., 2019).

At the same time, once the effects of overall intellectual ability, visual-spatial perception, and word-reading instruction approach were taken into account, the effect size for the unique contribution of early phonological awareness to later word-reading ability was medium rather than large, accounting for only 4% of the variance in the word-reading measure. Furthermore, although 67% of the participants in the present study performed at least in the low average range on the phonological awareness measure, only 38% (88% of whom had phonological processing abilities at or above the 10th percentile) scored in the low average range or higher on the single-word reading composite. Clearly, other factors must also contribute to variance in word-reading ability in children with WS.

Our results suggest that another important child factor is visual-spatial perception ability. In keeping with the findings of previous cross-sectional studies of individuals with WS (Brawn et al., 2018; Dessalegn et al., 2013), we found that individual differences in visual-spatial perception at 6–7 years were strongly related to word reading ability at age 9–10 years (bivariate $r = 0.55$). Furthermore, we found that these individual differences in visual-spatial perception ability significantly and uniquely contributed to the variance in word reading ability at 9–10 years. However, the effect size for the unique contribution of visual-spatial perception was small, accounting uniquely for only about 2% of the variance in single-word reading ability once we controlled for the effects of phonological awareness, word-reading instruction type, and either overall intellectual ability or vocabulary. As discussed further below, it is possible that the effects of phonological awareness and visual-spatial perception are enhanced in certain instructional contexts.

Unquestionably, the primary predictor of word reading ability was type of reading instruction, with a systematic phonics approach associated with considerably better word reading ability than other word-reading instruction approaches. This finding is consistent with Catts and Petscher's (2022) statement that the primary factor that is likely to have a positive impact on reading development for children at risk of reading disability is "explicit instruction on how to decode and read printed words" (p. 176). This result both extends prior cross-sectional findings for individuals with WS (Mervis, 2009; Mervis et al., 2022) and provides longitudinal evidence for the importance of early systematic phonics instruction to later word reading ability for children with WS. In addition, it is consistent with the results of intervention studies both for children without ID (e.g., Ehri et al., 2001a, 2001b) and children with ID of mixed or unknown etiology (Allor et al., 2014; Browder et al., 2012; Hunt et al., 2020).

Although word reading ability at 9–10 years was as strongly correlated with overall intellectual ability at 6–7 years as with phonological awareness and visual-spatial perception ability at that age, overall intellectual ability did not contribute significant unique variance to later word reading ability after the effects of phonological awareness, visual-spatial perception, and word-reading instruction approach were taken into account. It is thus not surprising that, as indicated in Table 6, there was considerable overlap in the participants' overall intellectual ability across most of the range of word reading ability. In conjunction with the results of previous cross-sectional (Mervis et al., 2022) and intervention (Pezzino et al., 2019; Stuebing et al., 2009) studies, the present findings suggest that limited overall intellectual ability, including IQs in the moderate-severe disability range, does not preclude word reading acquisition.

Individual differences in vocabulary at 6–7 years also did not add unique variance to word-reading ability at 9–10 years beyond that accounted for by variations in phonological awareness, visual-spatial perception, and type of word-reading instruction at 6–7 years. It is possible that vocabulary influences word reading ability indirectly, via its impact on the development of phonological awareness (see, e.g., Metsala & Walley, 1998). The strong bivariate correlation between vocabulary ability and phonological awareness ability at age 6–7 years ($r = 0.72$) is consistent with this hypothesis.

The results of the regression analyses, in combination with the exploratory analyses reported in Table 6, are consistent with multifactorial models of reading (dis)ability (e.g., Catts & Petscher, 2022; Pennington, 2006). In particular, as indicated by the results of the regression analyses, multiple factors—both child (phonological awareness ability, visual-spatial perception ability) and instructional (word-reading instruction type)—measured at 6–7 years contributed significant unique variance to word reading ability 3 years later, together accounting for 75% of the variance in the word reading composite at 9–10 years. At the same time, although broadly consistent with the regression results, the findings from the exploratory analyses suggest that the three factors identified by the regression analyses interacted in complex, likely nonlinear ways. For example, no child whose word-reading instruction used an approach other than systematic phonics evidenced word reading ability at the low average level or higher regardless of how strong their phonological processing and visual-spatial perception skills were and how high their overall intellectual ability (GCA) was. For children being taught with a systematic phonics approach, 43% with deficits in phonological awareness were able to read words at the low average level, whether or not they also had a deficit in visual-spatial perception, as were 62% with a deficit in visual-spatial perception, regardless of whether they also had a deficit in phonological awareness. If both phonological awareness and visual-spatial perception were at least in the low average range, it was very likely that word reading performance also would be at least in the low average range. For the children who were being taught to read with an approach other than systematics phonics, better word-reading performance (although still below the low average range) was more likely for children with low average level or higher visual-spatial perception ability, whether or not they had a deficit in phonological awareness.

This pattern of findings is consistent with Catts and Petscher's (2022) Cumulative Risk and Resilience Model, with deficits in phonological awareness and visual-spatial perception as risk factors and systematic phonics instruction as a resilience factor and the three factors interacting in nonlinear ways. These findings provide the first evidence that this model, developed for children in the general population who have difficulty learning to read, also is applicable to children with a genetic syndrome associated with ID. Our findings also are consistent with Astle and Fletcher-Watson's (2020) 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.

## Educational implications

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, as has been found for young TD children (e.g., Ehri et al., 2001a, 2001b; Roberts, 2021; Shmidman & Ehri, 2010), children with WS are likely to benefit from preschool and early primary school programs that incorporate embedded letter mnemonics to teach letter-sound correspondences (see also Mervis, 2009). Embedding hard-to-differentiate letters (such as *d* and *b*) in objects, actions or characters should help to reinforce the visual-spatial differences between similar-appearing letters representing different sounds. Together with prior evidence that broader language skills, including vocabulary, contribute to the development of phonological awareness and letter-sound knowledge both for children at risk of reading disability (Hulme et al., 2015; Storch & Whitehurst, 2002) and children at no known risk (Hjetland et al., 2019), our finding of a strong concurrent relation between vocabulary and phonological awareness suggests that activities that promote the development of oral language also are likely to support the development of word reading for children with WS. Thus, preschool programs that provide systematic phonological and phonemic awareness training incorporating embedded-letter mnemonics and also include opportunities for the development of broader language skills provide children with WS a solid foundation for learning to read once they enter primary school. In turn, systematic phonics instruction, beginning in kindergarten, that is coordinated with continuing systematic phonological and phonemic awareness instruction and broad language activities would offer children with WS the opportunity to optimize their early word reading abilities, at the same time as supporting listening comprehension and reading comprehension.

## Limitations

The results of the present study should be interpreted in the context of certain limitations. Most importantly, as single-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 previous reports of a strong concurrent correlation between phonological awareness (which we did measure at Time 1) and letter-sound knowledge (e.g., Clayton et al., 2020).

Our models explained 75% of the variance in the word reading composite, leaving 25% unaccounted for. While some of the unexplained variance is due to measurement error, some may be attributable to individual differences in child psychological factors such as motivation or ecological factors such as family support or socioeconomic status (see Joshi, 2019; Catts & Petscher, 2022) which were not measured in the present study. Future larger-N studies of children with WS that also measure these types of variables would allow for more nuanced evaluations of complex multifactorial models of the development of word reading.

Another limitation is that the classification of reading instruction type was not based on direct observation. Given the rarity of WS and therefore the need to include participants who resided across a very wide geographical area to obtain a substantial number of same-aged children, this limitation was inevitable. To address this limitation, the senior author used all available information, including direct conversations with parents and/or reading instructors, to classify the primary approach to teaching reading in the classroom in which the children received all or most of their reading instruction as Systematic Phonics or Other. 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.

## Conclusions

The present longitudinal results indicate that Catts and Petscher's (2022) Cumulative Risk and Resilience Model of reading (dis)ability can be successfully extended to children with WS. As discussed above, although phonological awareness and visual-spatial perception each predicted unique variance in later word reading ability, their effects were modulated by type of word reading instruction. In particular, systematic phonics instruction emerged as a strong resilience factor for children with WS. These results have clear educational implications. Early word reading instruction for children with WS should combine systematic phonics instruction and phonological awareness training while also incorporating letter discrimination instruction highlighting the visual-spatial differences between similar-appearing letters.

**Supplementary Information** The online version contains supplementary material available at https://doi.org/10.1007/s11145-022-10370-7.

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**Author contributions** CGR: Conceptualization, methodology, formal analysis, investigation, writing original draft. CC-M: Conceptualization, writing—review and editing. CBM: Conceptualization, methodology, formal analysis, data curation, writing—review and editing, supervision, funding acquisition.

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**Availability of data and material** The data that support the findings of this study are available from the corresponding author upon reasonable request.

**Code availability** Not applicable.

## Declarations

**Conflicts of interest** The authors have declared that they have no competing or potential conflicts of interest.

**Ethics approval** The study protocol was reviewed and approved by the University of Louisville's Institutional Review Board.

**Related Reports** An earlier version of this manuscript was included as part of the doctoral dissertation completed by Caroline Greiner de Magalhães (now Caroline G. Richter; 2021). As described in the Participants section of the present manuscript, many of the children included in the present longitudinal study also participated in the cross-sectional study reported in Mervis et al. (2022).



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