Published in final edited form as:

Res Dev Disabil. 2022 January ; 120: 104129. doi:10.1016/j.ridd.2021.104129.

Spelling Abilities of School-aged Children with Williams Syndrome

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Abstract

Aims: We examined the relation between spelling ability and word-reading ability in children with Williams syndrome (WS).

Methods: Eighty 9–17-year-olds with genetically-confirmed WS completed standardized tests of spelling, word reading, and intellectual ability; 45 also completed tests of phonological awareness and vocabulary. Reading instruction method was classified as Phonics or Other.

Results: Spelling ability varied widely. Although at the group level, spelling standard scores (SSs) were significantly lower than word-reading SSs, at the individual level, this difference was significant for fewer than half the participants. Spelling and reading SSs were highly correlated, even after controlling for intellectual ability. Students taught to read using systematic phonics instruction had significantly higher spelling SSs than those taught to read using other approaches, even after controlling for intellectual ability. Spelling ability contributed significant unique variance to word-reading ability, beyond the effects of phonological awareness, vocabulary, and reading instruction method.

Conclusions: Our findings are consistent with Ehri's Word Identity Amalgamation Theory. In combination with previous meta-analytic findings for typically developing children (Graham & Santangelo, 2014) our results suggest that children with WS are likely to benefit from the inclusion of systematic spelling instruction as part of a systematic phonics approach to teaching word reading.

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Keywords

Williams syndrome; spelling ability; word-reading ability; reading instruction method; phonics; intellectual disability

1. Introduction

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 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.

1.1. 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).

1.2. 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 ($p = .058$).

1.3. 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.

1.4. 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 & 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.

2. Method

2.1. 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.

2.2. Measures

2.2.1. 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.

2.2.2. 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 $r = .93$, $p < .001$.

2.2.3. 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.

2.2.4. 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.

2.2.4. 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.

2.2.5. 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 $d = 0.61$.

2.3. 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.

3. 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).

3.1. Relations between spelling ability and word-reading ability

Descriptive statistics for all measures investigated in this study are provided in Table 1. 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 2 for comparison.

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.

3.2. 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 3, 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 3, 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 4. 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.

3.3. 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 5. 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 3), 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.

3.4. 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 3. 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 6, 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 6, 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 6, 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 method (large effect), and Spelling SS (large effect) made significant independent contributions to the variance in Basic Reading SS. As illustrated in Table 6, 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.

4. 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.

4.1. 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.)

4.2. 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).

4.4. 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 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.

4.4. 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.

Acknowledgements

This research was funded by grants WSA 0104 and WSA 0111 from the Williams Syndrome Association, R37 HD29957 from the National Institute of Child Health and Human Development, and R01 NS35102 from the National Institute of Neurological Disorders and Stroke to C. B. M. The authors are grateful to the participants and their parents for the commitment they have shown to this research and to the members of the Neurodevelopmental Sciences Laboratory at the University of Louisville who administered and scored the assessments and maintain the research database. The authors have declared that they have no competing or potential conflicts of interest. This manuscript will be included as part of C. G. M.'s doctoral dissertation.

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Highlights

- Spelling ability of school-aged children with Williams syndrome (WS) varies widely
- Spelling and reading abilities are highly correlated, even controlling for IQ
- Spelling ability contributes significant unique variance to word-reading ability
- Systematic phonics instruction is positively associated with spelling ability in WS
- Literacy instruction in WS should incorporate systematic spelling instruction

What this paper adds?

Accurate spelling is an important component of skilled writing, an ability contributing to academic and professional success. 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. For this reason, learning to spell also contributes to the development of fluent reading. Nevertheless, despite its importance, spelling ability has been investigated much less often than reading ability. In this study, we examined the relations between spelling ability, word-reading ability, and method of reading instruction for eighty 9–17-year-olds with Williams syndrome, a genetic disorder associated with intellectual disability. Students who were taught to read using a systematic phonics approach spelled significantly better than those who were taught to read using other methods, even after controlling for intellectual ability. Similar to prior findings for typically developing children, spelling ability was very highly correlated with word-reading ability; this correlation remained strong even after controlling for intellectual ability. Finally, spelling ability strongly predicted concurrent word-reading ability independently of the effects of variations in three major predictors of reading ability: reading instruction method, phonological awareness, and vocabulary. These findings are consistent with Ehri's Word Identity Amalgamation Theory. Our results in combination with previous meta-analytic findings suggest that children with WS are likely to benefit from the inclusion of systematic spelling instruction as part of a systematic phonics approach to teaching word reading.

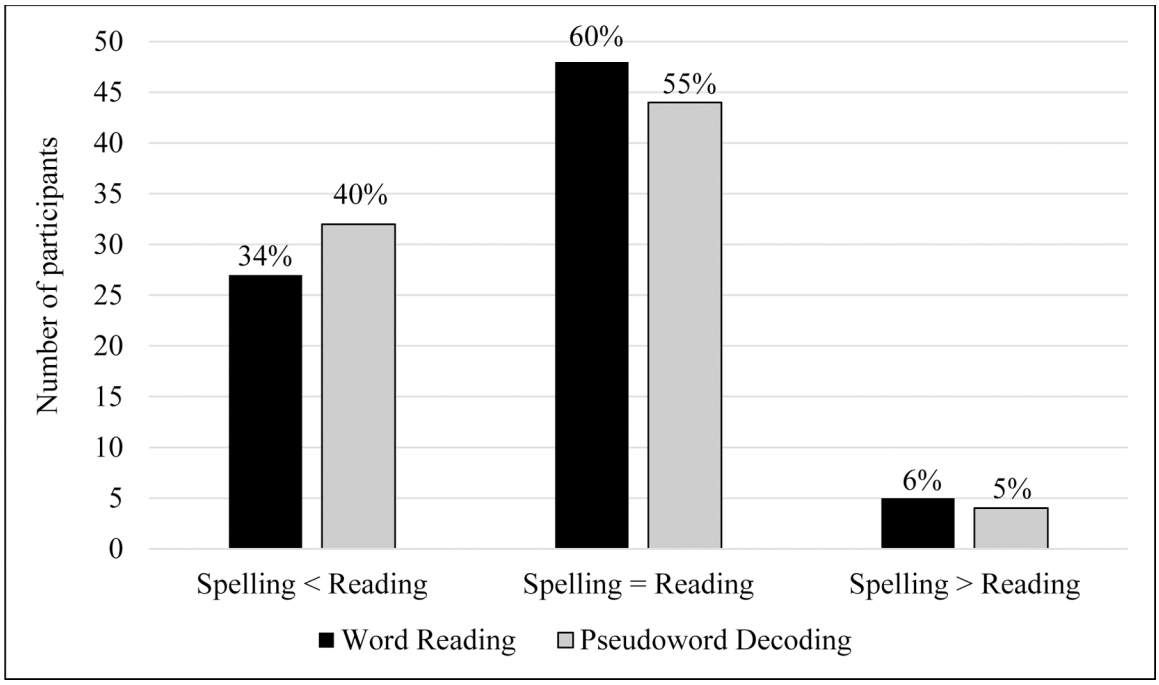

Fig. 1.

Relations between WIAT-III Spelling standard score and WIAT-III Word Reading or WIAT-III Pseudoword Decoding standard scores for individual participants. 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.

Table 1

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 2

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.

Table 3

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 4

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 5

Descriptive statistics for spelling, reading, and overall intellectual ability as a function of reading instruction method.

	Phonics (*n* = 47, 24 girls)				Other (*n* = 33, 18 girls)			
Variable	**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.

Table 6

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.