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Learning to Read in Williams Syndrome: Looking Beneath the Surface of Atypical Reading Development

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In this paper, we make a fundamental distinction between literacy attainment scores and the actual process of learning to read, and examine these two aspects of reading in atypical development. Reading skills in a group of children and adults with the genetic disorder Williams syndrome (WS) were compared to a group of typically developing children matched for reading age and receptive vocabulary scores. Study 1 focused on the product of reading and explored the relationship between reading, general cognition, and phonological skills. Phonological skills were shown to be related to individual differences in reading attainment in both groups, although more weakly in the WS group. Experiment 2 examined the process of learning to read. The two groups were taught to associate abbreviated spellings (cues) with spoken words. The cues differed in their phonetic closeness to the target words, whereas the target words differed on the semantic variable of imageability. Compared to controls, the WS group showed slower learning, less sensitivity to the phonetic quality of the cue, and reduced influence from the imageability of words. The results support the hypothesis that although reading levels in WS depend on phonological skills, the full development of their reading is compromised by weak semantics. The studies highlight the importance in atypical populations of examining both reading levels and the actual process of learning to read.

Keywords: Williams syndrome, reading scores, new word learning, imageability, phonological skills.

Abbreviations: BAS: British Ability Scales; BPVS: British Picture Vocabulary Scales; WOLD: Wechsler Objective Language Dimensions; WORD: Wechsler Objective Reading Dimensions; WS: Williams syndrome.

Williams syndrome (WS) is a rare genetic disorder affecting 1 in 20,000 live births (Beuren, 1972; Greenberg, 1990). It is caused by a hemizygous submicroscopic deletion of several contiguous genes on chromosome 7q11.23 (Ewart et al., 1993; Tassabehji et al., 1996). WS presents with an IQ range between 50 and 65 and an uneven cognitive profile. Many nonlinguistic functions, such as spatial cognition, number, planning, and problem solving are seriously impaired (Arnold, Yule, & Martin, 1985; Bellugi & Bihrle, 1993), whereas language is often surprisingly good in comparison (Bellugi, Bihrle, Jernigan, Trauner, & Doherty, 1990; Clahsen & Almazan, 1998). However, within this domain of relative strength, weaknesses in both morphosyntax and semantics have been shown to occur (Karmiloff-Smith et al., 1997, 1998; Mervis & Bertrand, 1997; Stevens & Karmiloff-Smith, 1997; Thomas et al., 2001; Volterra, Capirci, Pezzini, Sabbadini, & Vicari, 1996).

A number of studies have investigated the scholastic achievements of individuals with WS. Pagon, Bennett, LaVeck, Stewart, and Johnson (1987) examined the skills of nine individuals with WS between the ages of 10 and 20, using the Peabody Individual Achievement Test. They found that reading was the strongest area of school performance and that standard scores on the reading test exceeded Full Scale IQ in all nine cases. However, the relationship between Verbal IQ and reading was not examined separately. Given the uneven cognitive profile in WS, it is possible that while the reading scores exceeded Full Scale IQ, they were actually at a similar level to Verbal IQ. Nonetheless, the fact that even the participants who fell into the "severely retarded" category obtained first-grade level reading led the authors to suggest that reading ability was "surprisingly strong".

Others have not interpreted their findings so positively. Udwin, Davies, and Howlin (1996) reported a longitudinal follow-up of 23 young adults with WS. Because different reading measures were used on the two occasions, the results are not directly comparable. However, they did report a small, but nonsignificant, increase in reading age between time 1 and time 2 (by 1 year 1 month), together with a slight decrease in reading comprehension scores.

In a subsequent study, Howlin, Davies, and Udwin (1998) examined the cognitive skills of 67 adults with WS, with IQ scores in the 50–69 range. The adults were all given the Wechsler Objective Reading Dimensions

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(WORD) test (Rust, Golombok, & Trickey, 1993). The authors found that 47 of the participants were able to score above basal level on the tests of single word recognition and reading comprehension, and 46 were able to score above basal level on the spelling test. The mean basic reading age obtained was 8 years, 8 months (range 72–216 months), the reading comprehension age equivalent was 7 years, 2 months (range 72–132 months) and the mean spelling age was 7 years, 7 months (range 75–150 months). Reading comprehension was found to be significantly worse than single word reading. Verbal, Performance, and Full Scale IQ scores were significantly higher for the readers than the nonreaders, and higher for the spellers than the nonspellers. In contrast to Pagon et al. (1987), the authors of this study concluded that far from being an area of strength, reading attainments were "disappointingly low" (p. 187).

Studies such as these provide important information about the level of reading skill obtained in the phenotypic outcome by individuals with WS. However, they do not address the question of why some learn to read while others do not. Despite the fact that spoken language has been shown to play an important role in normal reading development, this relationship has not been examined in previous studies of WS. Yet children with WS come to the task of learning to read with a different and uneven pattern of language skills compared to those of typically developing children. They often have higher vocabulary scores than would be predicted by their level of general intelligence (Bellugi et al., 1993; Levine, 1993), but morphosyntactic levels have been shown to be closer to their nonverbal mental age than to their vocabulary age (Karmiloff-Smith et al., 1997; Mervis & Bertrand, 1997; Volterra et al., 1996; but see Clahsen & Almazan, 1998, for a different view of WS grammar). Others have argued that, despite high vocabulary scores, people with WS process semantic information differently, use more unusual words, and are less sensitive to word frequency than controls (Reilly, Klima, & Bellugi, 1990; Rossen, Bihrle, Klima, Bellugi, & Jones, 1996; Stevens & Karmiloff-Smith, 1997; Vicari, Brizzolara, Carlesimo, Pezzini, & Volterra, 1996). Some studies have suggested that individuals with WS are relatively over-dependent on phonological encoding and may not process semantic information in the normal way (Grant et al., 1997; Johnson & Carey, 1998; Thomas et al., 2001; Vicari, Carlesimo, Brizzolara, & Pezzini, 1996). For example, children and adults with WS show normal similarity and length effects in memory tasks, but a reduced word frequency effect (Vicari, Brizzolara et al., 1996). They also perform better in phonological fluency tasks than in semantic fluency tasks (Volterra et al., 1996). Finally, Grant et al. (1997) demonstrated a persistant relationship between nonword repetition and vocabulary at all ages in WS, suggesting that this clinical group continues to rely more heavily on phonological short-term memory to support word learning than is usual in normal development beyond 5 years.

The suggestion that individuals with WS are relatively over-dependent on phonological encoding will have important implications for the way in which children with WS learn to read. It is possible, for example, that while they may learn letter-sound correspondences fairly easily and use phonological processing in reading, their understanding of what they read will be impaired.

Studies of reading in typically developing children have demonstrated a close relationship, independent of IQ, between individual differences in phonological awareness and the development of reading skills (see Rack, Hulme, & Snowling, 1993; and Wagner & Torgeson, 1987, for reviews). The mechanisms underlying this association are, however, not well understood. It is clear that phonological awareness tasks measure more than just phonological processing skills. Such tasks require, in addition, a degree of metacognitive skill. Whereas some argue that this metacognitive component is important, others maintain that phonological awareness tasks merely tap the quality of the underlying representations that are used in learning to read (Fowler, 1991; Hulme & Snowling, 1992; Swan & Goswami, 1997).

The role of metacognitive skill in underpinning the relationship between phonological awareness and learning to read is clearly important in investigating the reading skills of clinical populations. It is likely that such groups will find the cognitive demands of many phonological awareness tasks too great. This in itself may lead to an apparent lack of relationship between phonological awareness and reading. It is thus essential to measure phonological skills not only with tasks that involve explicit awareness (phonological awareness tasks) but also with phonological processing tasks (such as rapid naming and memory tasks) that do not require explicit awareness.

The present studies investigate both the end-state of reading development, as well as the process of learning to read. Study 1 considers the phonological processing and explicit phonological awareness skills of a group of children and adults with Williams syndrome. Study 2 investigates this group's ability to learn to read a series of novel words.

Study 1

Study 1 examines levels of reading attainment and relationships between reading, language, and phonological skills in a group of participants with WS and a younger typically developing group matched for receptive vocabulary and single word reading. A strong relationship between phonological skills and reading has already been demonstrated in normal development (Goswami & Bryant, 1990; Rack, Hulme, Snowling, & Wightman, 1994; Wagner & Torgeson, 1987). The relationship has also been shown to hold for people with Down's syndrome, provided phonological awareness tasks tap implicit rather than explicit skills (Buckley & Bird, 1993; Cardoso-Martin & Frith, 1997; Mercer, 1997). However, it is unknown whether the same will obtain for individuals with WS. An important question is whether general intelligence is the primary predictor of reading skill or whether, as in normal development, individual differences in phonological processing skill in individuals with Williams syndrome will also play an important role.

Method

Participants. Fifteen individuals with Williams syndrome with a mean age of 15 years, 1 month (range 9 years to 27 years, 7 months) took part in this study. The participants were all recruited through the Williams Syndrome Foundation, the U.K. support group for parents and were from middle- to lower-middle-class socioeconomic backgrounds. The participants with WS had a mean General Cognitive Ability score on the British Ability Scales (BAS) of 43.8 (*SD* 5.12, range 39 to 54). The mean verbal standard score was 56.3 (*SD* 6.02, range 49 to

74), the mean nonverbal standard score was 50.08 (*SD* 3.4, range 41 to 61), and the mean spatial standard score was 46.9 (*SD* 3.4, range 40 to 52).

The participants with WS were individually matched to a group of typically developing children on the basis of their reading age and verbal mental age. Reading age was calculated from the BAS II test of single word reading (Elliot, Smith, & McCulloch, 1996). The children in the control group all came from a single primary school in East Sussex, and were of similar socioeconomic status to the group with WS. The mean chronological age of the children in the control group was 6 years 9 months (range 5 years to 9 years 2 months). They were all within the normal range for reading ability, with a mean standard score on the BAS Reading Test of 103.27 (*SD* 9.81, range 83 to 118). Time constraints meant that it was not possible to obtain full BAS scores for the children in the control group. As a result, a measure of nonverbal reasoning was obtained by administering the matrices subtest from the BAS and of verbal skills from the British Picture Vocabulary Scale (BPVS).

Design and materials. We administered a battery of phonological awareness and phonological processing tasks, specifically chosen because they have been shown to correlate with individual differences in reading skill in normally developing young children. We also assessed single word and nonword reading skills as well as reading comprehension skills, in order to gain a broader picture of WS reading skills.

(1) *Reading and spelling tests.*
(a) *Single word reading.* Reading ability was assessed using a graded test of single word naming, the BAS II Word Reading test (Elliot et al., 1996), and the single word reading test from the WORD (Rust et al., 1993).
(b) *Reading comprehension.* The reading comprehension test from the WORD was administered (Rust et al., 1993). In this test the participant is required to read a short story and then answer questions on it.
(c) *Nonword reading.* To assess participants' ability to use letter-sound rules for decoding novel words, the Graded Nonword Reading Test (Snowling, Stothard, & McLean, 1996) was used. This test consists of 10 one-syllable and 10 two-syllable nonwords, graded in difficulty.
(d) *Letter knowledge.* Letter knowledge was assessed by asking the participants to give the name or sound of the letters of the alphabet, presented in random order on printed cards.
(e) *Spelling test.* The WORD spelling test (Rust et al., 1993) was administered. The participant is required to spell a series of words graded in difficulty, presented both in isolation and in a sentence context.
(2) *Phonological awareness tests.* Two standardised tests of phonological awareness were used, the Phonological Abilities Test (PAT; Muter, Hulme, & Snowling, 1997), and the Phonological Awareness Battery (PHAB; Frederickson, Frith, & Reason, 1997)
(a) *Phonological Abilities Test.*
(i) *Rhyme detection.* All the items in this test are accompanied by pictures. The participant is required to indicate which of three words rhyme with a stimulus word.
(ii) *Rhyme production.* The participant is given 30 seconds to supply words or nonwords that rhyme with each of the two stimulus words, DAY and BELL.
(iii) *Phoneme deletion.* The participant is required to remove the initial phoneme from a single-syllable word. The correct response is also a word. For example, meat without the /m/ is … eat. The test is then repeated with another set of single-syllable words and the participant is now asked to delete the final phoneme.
(iv) *Syllable and phoneme identification.* Two sets of items are presented with pictures. The first set requires participants to supply the final phoneme of a single-syllable word. In the second set the examiner supplies the first part of the word, which the participant is then required to finish; for example, "here is a ra … (rabbit)" and the participant is expected to say /bit/.
(b) *Phonological Awareness Battery.*
(i) *Rhyme.* The participant listens to three single-syllable words and is required to say which two ended with the same sound. Unlike the rhyme detection test described above, the words in this test are not accompanied by pictures.
(ii) *Spoonerisms.* This test assesses the ability to segment single syllable words and then to synthesise the segments to provide new words. In part 1 of the test of the participant is required to replace the first sound in a word with a new one (e.g. *cot* with a /g/ gives *got*). The answers are always words. In part 2 the individual is required to swap the initial sounds of two words (e.g. "*sad cat*" gives "*cad sat*"). The answers in this part are sometimes nonwords.
(iii) *Naming speed.* Part 1 of the test is a rapid picture naming task (drawings of five everyday objects). Part 2 is a rapid digit naming (the printed numbers 1–9). Participants are given a practice trial for each test to ensure that they are able to name the pictures and digits. The participant is then shown a visual display of randomly presented items and asked to name them in sequence as quickly as possible.
(3) *Memory tests.*
(a) *Word span.* Word span was measured using a test devised by McDougall, Hulme, Ellis, and Monk (1994). Participants are presented with lists of one-syllable words to repeat in order, matched for frequency and conceptual class. There are four lists at each list length, starting with lists of two words. Testing is discontinued when participants fail to repeat all lists of a given length correctly.
(b) *Digit span.* The digit span subtest from the BAS was administered according to the manual.
(c) *Nonword repetition.* The Children's Nonword Repetition (CNRep) task (Gathercole & Baddeley, 1996) was administered.
(d) *Listening comprehension.* The listening comprehension test from the Wechsler Objective Language Dimensions (WOLD) (Rust, 1996) was administered.
(4) *Background measures.*
(a) *Vocabulary.* The British Picture Vocabulary Scales (Dunn, Dunn, Whetton, & Pinitilie, 1982) were administered.

Procedure. The participants with WS were seen individually at the Neurocognitive Development Unit at the Institute of Child Health in London. Testing lasted approximately 2 hours and breaks were given as needed. The tests were administered in random order. The control group was seen at school and tested individually in a quiet area outside their classroom. Due to the practical difficulties of seeing school children for such long sessions, testing was broken up into a number of shorter sessions, depending on the age of the child.

Results

Comparing the Williams syndrome and control groups. The reading, verbal and nonverbal scores of the WS and control groups are shown in Table 1. It can be seen from this table that the groups are well matched for single word reading ability, and for nonverbal (matrices) and verbal (BPVS) mental age. As is often the case in studies of rare disorders, the range of age and reading scores in the WS group and, as a consequence, in the reading-age-matched control group, is rather broad. The groups were therefore split into subgroups on the basis of their BAS reading scores, using median scores as the cutoff point. The BAS reading score means, *SD*s, and ranges were: Low scorers: WS ($N = 8$): mean = 12.50, *SD* 8.47, range 0–22; Control

Table 1
Mean Scores (and SDs) of the Williams Syndrome and Control Groups on the Measures of Reading, Nonverbal, and Verbal Ability and Digit Span

Measure	Groups: WS	Control	*F*-value
BAS reading[a]	29.3 (21.4)	31.0 (22.0)	0.04
Matrices[a]	6.8 (3.4)	7.5 (4.5)	0.21
Digit span[a]	14.7 (3.7)	18.4 (4.5)	6.80**
BPVS test age[b]	87.2 (22.9)	79.3 (22.6)	1.46

[a]Raw scores.
[b]In months.
$**p \leqslant .01$.

Table 2
Performance of Participants with Williams Syndrome on Reading, Spelling, and Language Tests

Measure	Test age in months (*SD*)
WORD word recognition	82.2 (31.3)
WORD reading comprehension	58.2 (44.0)
WORD spelling	61.2 (41.1)
BAS single word reading	76.1 (34.4)
WOLD listening comprehension	82.0 (11.9)
BAS Verbal Mental Age	86.2 (24.7)
BPVS	87.7 (22.9)

Average chronological age of sample = 181 months.

($N = 8$): mean = 13.88, *SD* 8.66, range 0–23; High scorers: WS ($N = 7$): mean = 48.57, *SD* 13.40, range 32–65; Control ($N = 7$): mean = 50.57, *SD* 14.48, range 33–65. Where relevant in the analyses that follow, results for the subgroups are reported in addition to the overall group results, with low scorers referred to as the "low ability subgroups" and high scorers as the "high ability subgroups".

*The reading skills of individuals with Williams syndrome.*The performance of the Williams syndrome group on the standardised reading and spelling tests is shown in Table 2. The mean reading age obtained, according to the two tests of single word reading, was around 6 years 5 months, for a group whose mean chronological age was 15 years 1 month. The spelling ages and reading comprehension ages of this group were even lower.

The difference between performance on the single word reading and reading comprehension tests was examined. Twelve of the 15 WS participants scored above floor on the WORD test of single word reading. The scores of these 12 participants on the single word reading and comprehension tests were compared. The standardised scores on the single word reading task were significantly higher than those on the reading comprehension test, $F(1, 11) = 6.04$, $p < .05$. In contrast to this poor performance on the reading comprehension test, listening comprehension skills were considerably stronger.

Relationship between measures of general cognitive ability and reading skill in the WS group. Comparisons were made between age-equivalent scores on the tests of reading and the tests of general ability. Reading ages (as measured by the BAS) of the WS group were not significantly different from their vocabulary age (BPVS), $F(1, 14) = 2.65$, $p > .05$, or from their verbal mental age (BAS), $F(1, 14) = 0.87$, $p > .05$. The subgroup analyses also yielded no significant differences between these variables.

As a large number of participants were at floor on one or more of the subtests, it was not possible to calculate accurate mental ages. It was therefore impossible to compare reading age to overall mental age or to nonverbal mental age. In order to investigate the relationship between reading and cognitive ability, correlations between measures were calculated. Because of the problem of floor effects, raw scores were used in the correlational analysis. Composite verbal, nonverbal, and spatial scores were created by summing z-scores. To create the verbal measure, for example, the z-scores for the definitions and verbal similarities subtests were summed. For the nonverbal measure, the z-scores for the quantitative reasoning and matrices subtests were summed, and for the spatial measure the z-scores for the pattern construction and recall of designs task were summed. These measures were then correlated with single word reading as measured by the BAS and the WORD as well as with the measures of spelling and reading comprehension from the WORD. The raw scores from the BPVS were also correlated with these measures of reading ability.

These correlations are shown in Table 3.The measures of verbal ability and receptive vocabulary (BPVS) are moderately correlated with the measures of literacy attainment in the WS group. In contrast, the measures of spatial and nonverbal ability correlate more strongly with the measures of literacy attainment. Given the large number of correlations reported, the adoption of a conservative alpha level is appropriate (.05/20 = .003). Correlations that remain significant are shown in italic in Table 3. It is clear from these data that cognitive ability (particularly nonverbal ability) is highly predictive of individual differences in literacy attainment in the WS group.

Although numbers in the subgroups are necessarily small, the above findings were confirmed for the high ability group. Scores on one or both single word reading

Table 3
Correlations between Measures of Reading and Spelling and General Cognitive Ability in the WS Group

	Verbal	Spatial	Nonverbal	Full Scale	BPVS
BAS read	.51*	*.82****	.66**	*.76****	.56*
WORD read	.48	*.75****	*.72***	*.75****	.52*
WORD spell	.35	.64**	.67**	.64**	.44
Reading comprehension	.53*	.68**	*.76****	*.76****	.56*

Correlations that remain significant at $\alpha = .003$ (see text) are shown in italic.
$*p \leqslant .05$; $**p \leqslant .01$; $***p \leqslant .001$.

Table 4
Correlations between Measures of Reading and Listening Comprehension in the WS Group

	Comprehension	
	Reading	Listening
Reading comprehension	1.00	.62**
WORD single word	*.92****	.41
Listening comprehension	.62**	1.00
BPVS	.53*	.58*
Word span	.30	.46
Digit span	.43	*.73***
CNRep	.26	.68**
Verbal IQ	.53*	.66**
Nonverbal IQ	*.76****	.63**
Spatial IQ	.68**	.47

Correlations that remain significant at $\alpha = .003$ (see text) are shown in italic.
*$p \leqslant .05$; **$p \leqslant .01$; ***$p \leqslant .001$.

tests and spelling were significantly related to spatial and nonverbal ability. Reading comprehension was significantly related to general cognitive ability (alpha level = .05). However, none of the correlations were significant for the low ability group.

Relationships between measures of reading and listening comprehension. Table 4 shows the correlations between the measures of reading and listening comprehension, and memory and cognitive abilities, in the WS group. Given the large number of correlations reported, the adoption of a conservative alpha level is again appropriate (.05/20 = .003). There was a fairly strong correlation between the measures of reading and listening comprehension in this group, as is found in studies of typically developing children (Townsend, Carrithers, & Bever, 1987). In addition, as would be expected, reading comprehension is highly correlated with single word reading ability. Listening comprehension, but not reading comprehension, is highly correlated with digit span. Finally, there are moderate correlations between listening comprehension and verbal and nonverbal ability (as there are for reading comprehension, see Table 3).

We again examined the pattern of these correlations in the high and low ability groups separately. The strong relationship between single word reading and reading comprehension was confirmed in the low ability subgroup, but not in the high ability subgroup. Digit span and Verbal IQ were significantly related to listening comprehension in the low ability group and two measures of phonological short-term memory (word span and nonword repetition) were significantly related to listening comprehension in the high ability subgroup (alpha = .05).

Phonological skills in individuals with WS. Table 5 shows the performance of both groups on the measures of phonological awareness and phonological processing ability. A one-way MANOVA on these data indicated that there was no reliable effect of Group, Wilks Lambda, $F(11, 18) = 1.86$; n.s. This null result clearly needs to be interpreted with caution given the small sample size. Univariate tests indicated that the difference between groups on the phoneme deletion measure was reliable, $F(1, 28) = 9.49$; $p < .005$, but none of the other differences were. In summary, in comparing the group of participants with Williams syndrome to a younger group of normally developing children (matched for vocabulary age using BPVS and single word reading ability) we see that the groups are approximately equivalent in terms of their phonological skills.

Relationships between reading and phonological skills. The relationships between reading skills and performance on the phonological awareness tasks was examined for both the WS group and the control group. In order to simplify the analysis, composite measures of phonological ability were created by summing z-scores. These composite measures were a phoneme measure (derived from the phoneme deletion task and spoonerism task), a rhyme measure (derived from the three rhyme tasks) and a speeded naming measure (derived from the digit and picture naming tasks).

Table 6 shows the correlations between reading, as assessed by the BAS single word reading test, and the measures of phonological skill. Partial correlations controlling for differences in age and general ability (measured by summing z-scores for BPVS and matrices raw scores) are also shown. Once again, given the number (8) of correlations reported for each group, it is appropriate to adopt a conservative alpha level (.05/8 = .006).

The pattern of correlations shows some interesting similarities and differences between the groups. As expected, nonword reading ability correlates very strongly, and letter knowledge correlates moderately strongly, with reading ability in both groups. Our measures of

Table 5
Mean (and SDs) Raw Scores of the Williams Syndrome and Control Groups on the Phonological Awareness and Memory Tests

	Groups		
	WS	Control	*F*-value
Rhyme detection	6.80 (2.96)	8.60 (1.76)	4.09
Rhyme production	5.93 (4.17)	7.40 (2.64)	1.32
Spoonerisms	5.00 (3.95)	7.33 (7.27)	1.19
PHAB rhyme task	8.27 (6.32)	11.33 (5.95)	1.87
Nonword reading	4.87 (5.53)	9.73 (7.70)	3.95
Phoneme deletion	6.73 (6.22)	12.73 (4.23)	9.49**
Identification	12.67 (2.82)	14.07 (2.40)	2.14
Digit naming	98.73 (39.30)	96.73 (46.18)	0.02
Picture naming	125.27 (33.68)	130.73 (41.97)	0.15
Word span	3.47 (0.48)	3.75 (0.54)	2.29
CNrep	29.80 (7.13)	30.53 (3.76)	0.12

**$p \leqslant .01$.

Table 6
Correlations and Partial Correlations (Controlling for Age and General Ability) between Single Word Reading and Phonological Awareness for the WS and Control Groups

	Group			
	WS		Control	
	r	partial r	r	partial r
Nonword reading	*.88****	*.79***	*.91****	.66*
Rhyme awareness	.59*	.05	*.88****	.54
Phoneme awareness	.66**	.19	*.86****	.53
Letter knowledge	.65**	.47	.55*	.54
Speeded naming	*−.77****	−.56*	*−.79****	−.66**
Nonword rep.	−.01	−.42	.59*	.22
Digit span	.19	−.19	.35	.02
Word span	−.02	−.62*	.34	.04

Correlations that remain significant at $\alpha = .006$ (see text) are shown in italic.
*$p \leqslant .05$; **$p \leqslant .01$; ***$p \leqslant .001$.

phonological awareness (rhyme and phoneme awareness) show moderate to strong correlations with reading ability in both groups, but these relationships in the WS group are not independent of age and general ability. It is very interesting to note that naming speed shows strong correlations with reading ability in both groups, and this relationship is only slightly weakened when the effects of age and general ability are partialled out. Finally, there is a moderate correlation between reading ability and nonword repetition in the control group, but not in the WS group.

In summary, it appears that phonological skills are related to individual differences in single word reading ability in both the WS and control groups, though arguably these relationships are stronger (and less closely related to age and general ability) in the control group.

Discussion

The results of Study 1 show that although some individuals with WS can learn to read, the level of reading ability obtained is low (although roughly at a level that would be predicted by their verbal mental age). Howlin et al.'s (1998) finding of a discrepancy between single word reading and reading comprehension skills was replicated in the present study. On average, individuals with WS have better decoding than reading comprehension skills. This pattern has also been found in other clinical populations such as Down's syndrome (Fowler, 1995). There was a moderate correlation between reading comprehension and listening comprehension in the WS group, as is found in typically developing children.

The WS and control groups were matched on verbal skills (as measured by the BPVS) and nonverbal skills (as measured by the matrices task). Despite this, there is a tendency for the WS group to perform more poorly on the phonological tasks, with a significant impairment on the phoneme deletion measure.

The correlations suggest that there is a stronger relationship between phonological abilities and reading in the control group than in the WS group (although any conclusions must be tempered by the relatively small sample sizes studied). Although for the participants with WS there were moderately strong relationships between phonological abilities and reading, these relationships became much weaker once individual differences in general ability were controlled for. By contrast, for the control group the relationships between the phonological measures and reading ability appeared more independent of variations in age and general ability, as would be expected from many earlier studies of normal reading development (for reviews see Rack, Hulme, & Snowling, 1993; Goswami & Bryant, 1990).

The results of this study show that general ability and phonological processing skills are both related to the variations in reading ability found in the WS population. The results extend previous findings that have only examined the relationship between reading and overall IQ in this population. However, although our results suggest that phonological skills play a role in reading in WS, they cannot tell us how these individuals use phonology in learning to read. For this reason, it is crucial to examine the actual process of learning to read in WS rather than simply looking at the product of such learning reflected by performance on standardised reading tests.

Study 2

In the second study we assessed more directly the importance of phonological and semantic processes in learning to read words, using a paradigm developed by Ehri and Wilce (1985) and subsequently modified by Rack, Hulme, Snowling, and Wightman (1994) and by Laing and Hulme (1999). This learning paradigm taps phonological processes at an implicit level and therefore provides a good way of examining the role of phonology in learning to read in clinical populations who may have problems with the metacognitive aspects of phonological awareness tasks.

Using the method developed by Laing and Hulme (1999), WS and control participants were taught to associate three-letter cues with spoken words, differing in their phonetic similarity to the target word. If phonological processing is important in learning to read words in the WS population, as it has been shown to be in typically developing children, then we would expect the WS group to learn to read the phonetic cues more easily than the phonetically distant control cues. (This is called the phonetic cue effect.) If, however, the WS group use a different strategy to learn to associate the cues with the spoken words, they may not show a phonetic cue advantage and instead learn both types of cue with equal ease or difficulty.

Following the method of Laing and Hulme (1999), the influence of imageability was also considered in the present experiment. Laing and Hulme showed that normally developing young children learned to associate three- and four-letter cues more easily with high-imageability than with low-imageability words. This finding therefore provides evidence that learning to recognise printed words may be mediated by semantic as well as by phonological processes in typically developing young children. Previous research on language acquisition and short-term memory (Grant et al., 1997; Thomas et al., 2001; Vicari, Brizzolara, et al., 1996; Vicari, Carlesimo, et al., 1996) has suggested that individuals with WS rely less on semantic processing than typically developing children. This led to the prediction that individuals with WS would be relatively insensitive to the effects of imageability in the visual cue learning task.

As well as considering the phonological and semantic processing of the WS group, the present experiment had two further aims: (1) to consider the rate of overall learning of the cues of the WS and control groups; (2) to examine the relationship between learning to read the cues and performance on phonological tasks. Typically developing children's phonological awareness skills have been shown to be strongly related to their ability to learn to associate cues with target words (Laing & Hulme, 1999).

Method

Participants. Fourteen of the 15 WS participants, and all 15 normally developing children from Study 1, took part in this study. The mean age of the WS participants in Study 2 was 15 years 6 months (range 10 years 9 months to 27 years 7 months) and their mean reading age was 6 years 1 month (range < 5 years to 9 years 9 months). The removal of one participant from the WS group did not significantly alter the mean scores on any of the phonological awareness, language, or memory tasks.

Design and materials. Participants were taught to associate three-letter abbreviations (cues) with spoken target words. Sixteen target-cue pairings were taught in two separate sessions. The target words were all bisyllabic words with a CVCVC or CVCV phonemic structure. The cues contained two letters whose corresponding phonemes occurred in the target word and a single medial letter whose corresponding phoneme did not occur in the target word. In the phonetic cue condition the middle phoneme was similar to the corresponding phoneme in the target word in terms of place and manner of articulation but different in voicing. Five voice/voiceless pairs were used to create the phonetic cues: t/d, p/b, s/z, f/v, and k/g. In the control cue condition, the central phoneme in the cue always differed from that in the target word in place of articulation (and sometimes also voicing). So, for example, for the target word *ladder* the phonetic cue was *ltr* and the control cue *lkr*.

As well as the cues differing in their phonetic closeness to the target words, the target words themselves differed in imageability. Half of the target words were of high imageability and half were of low imageability. The imageability of the words was rated by 15 individuals, all of whom were professionals working with children. The raters were asked to rate the extent to which each word could evoke an image or clear picture in the mind of young children on a scale of 1 (low) to 7 (high). The familiarity of words was also rated. Raters were asked to rate words on a scale from 1 to 5 for the extent to which an average 5-year-old would have heard a word and would know its meaning. Finally, age of acquisition was also rated using a 1–7 scale, with each point on the scale representing the average age at which a word is considered to be acquired by a child. The reliability of these ratings was high (for imageability, $\alpha = .98$, for familiarity, $\alpha = .89$, and for age of acquisition, $\alpha = .89$).

There were four learning conditions in the experiment: high-imageability phonetic cues, high-imageability control cues, low-imageability phonetic cues, and low-imageability control cues. Two forms of the experiment were constructed so that each target word appeared in each form once, in either the phonetic cue or control cue conditions. Each form of the experiment contained 16 words: 4 in each condition. The cues were grouped into two blocks of eight words that were taught in two separate sessions. The order of presentation of blocks was counter-balanced. Each of the blocks of cues contained items that shared common first letters. This might be expected to encourage children to attend more to the medial letters in the cues in order to learn to read them effectively. The cues are listed in Appendix A, and the properties of the word sets in Appendix B.

Procedure. The WS participants in this study did the word learning task during the same session as the tasks in Study 1. The two word learning sessions were administered as far apart as possible such that each block was usually given at the very start and the very end of the testing session. This meant, on average, a gap of around 2 hours between the administration of the two blocks. Where possible, blocks were administered to the control participants in the same way. A number of the control participants were, however, tested on two consecutive days for the two blocks.

All participants were seen individually in a quiet area. In each session they were taught to associate the 16 printed cues with their pronunciations. They were told that they were going to learn some "funny made-up spellings". Each cue was presented to the participant and the experimenter said the target word twice, with the participant repeating the word once. On the first presentation the card remained in view for approximately 5 seconds. The cards were then shuffled and presented again to the participant, who was asked to say the spoken word that had been paired with each cue. Refusals and errors were corrected. Testing was discontinued after eight test runs. The participant scored 1 for a correct response and 0 for an incorrect response, giving a maximum possible score of 8 for each cue word; with eight words in each condition, the maximum possible score for each condition was 64.

Results

Word learning task. The results of the word learning task are shown in Fig. 1. The data were subjected to an analysis of variance in which the factors were Group, Phonetic Cue Type (phonetic or control), and Imageability (high or low). The main effect of Group was significant: by subjects, $F(1, 27) = 4.11$, $p = .05$, and by items, $F(1, 14) = 37.5$, $p < .05$, showing that the control group learned better than the WS group. The main effect of Imageability was not significant: by subjects, $F(1, 27) = 3.46$, $p > .05$, or by items, $F(1, 14) = 1.34$, $p > .05$, but critically the Group x Imageability interaction was significant: by subjects, $F(1, 27) = 9.47$, $p < .05$, and by items, $F(1, 14) = 16.48$, $p < .05$. Tests of simple effects confirmed that imageability had a significant effect on learning in the control group, by subjects $F(1, 27) = 0.62$, $p < .001$, by items $F(1, 14) = 6.66$, $p < .05$, but not in the WS group, by subjects, $F(1, 27) = 0.72$, n.s., by items, $F(1, 14) = 0.14$, n.s.

There was a significant main effect of Phonetic Cue Type: by subjects, $F(1, 27) = 12.15, p < .05$, and by items, $F(1, 14) = 9.06$, $p < .05$, but no significant interaction of Group x Cue Type: by subjects, $F(1, 27) = 0.45, p > .05$, or by items, $F(1, 14) = 2.98$, $p < .05$. No other effects in this analysis approached significance.

Subgroup analyses (considering the high and low ability groups separately) were carried out by subjects only and the results were similar to those for the whole group. Although there was no significant effect of Group

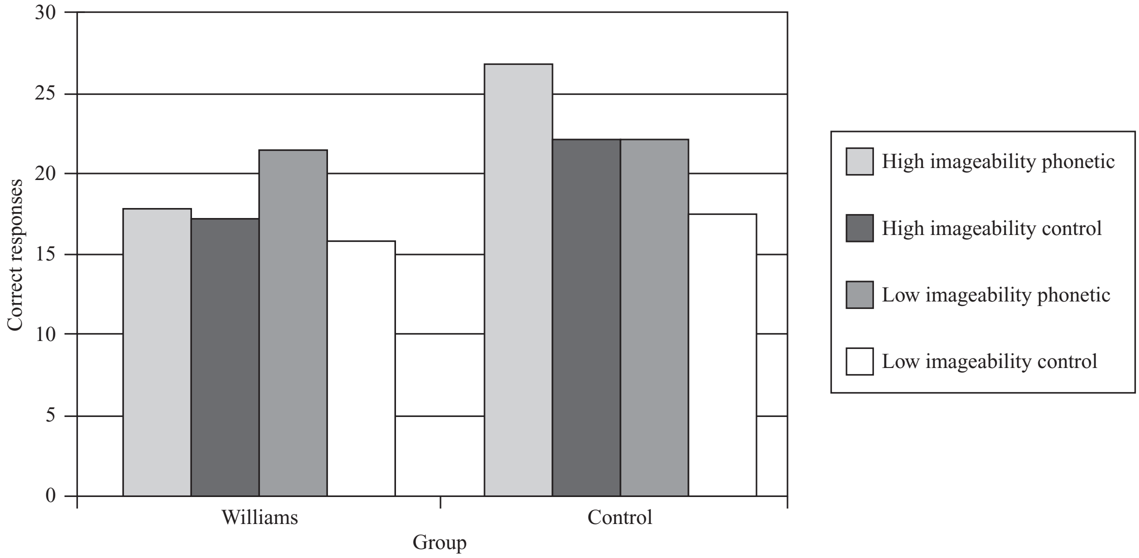

Figure 1. Number of correct responses to the cues for the WS and control groups in Experiment 2.

Table 7
Correlations and Partial Correlations (Controlling for Age and General Ability) between Cue-word Learning and Measures of Phonological Skill in the WS and Control Groups in Experiment 2

	Group			
	WS		Control	
	r	partial *r*	*r*	partial *r*
BAS reading	.85***	.66*	.78***	.62*
Nonword reading	.60*	.22	.65**	.23
Letter knowledge	.69**	.53	.88***	.89***
Phoneme awareness	.64**	.07	.82***	.73**
Rhyme awareness	.63*	.03	.79***	.62*
Segmentation	.52	−.03	.21	−.18
Speeded naming	−.73**	−.46	−.91***	−.85***
Nonword repetition	−.18	−.62*	.65**	.43
Memory span	.12	−.39	.27	.15
Digit span	.37	−.12	.46	.36

$^{*}p \leqslant .05$; $^{**}p \leqslant .01$; $^{***}p \leqslant .001$.

or of Imageability for either subgroup: for Group, low ability $F(1, 13) = 3.44$, $p > .05$, high ability $F(1, 12) = 3.11$, $p > .05$, there was a significant interaction of Group x Imageability in the low ability subgroup, $F(1, 13) = 5.83$, $p < .05$, and the interaction in the high ability subgroup was close to being significant, $F(1, 13) = 4.09$, $p = .07$. As in the whole group, the effect of Phonetic Cue type was significant, whereas the Cue x Group interaction was nonsignificant: for Phonetic Cue Type, low ability $F(1, 13) = 5.81$, $p < .05$, high ability $F(1, 12) = 5.52$, $p < .05$; Phonetic Cue x Group, low ability $F(1, 13) = 0.45$, $p > .05$, high ability $F(1, 12) = 0.08$, $p > .05$.

Relationships between cue-word learning and phonological skills. We examined the relationship between overall performance on the word learning task and measures of phonological skill. The total score (the sum of the correct responses to the phonetic and control cues) gives an overall measure of learning in the task. The correlations between total cue learning and the measures of phonological skills are presented in Table 7 for both groups. In view of the small sample sizes, these correlations clearly need to be interpreted with caution.

It can be seen from Table 7 that for the typically developing control group there are moderate to strong relationships between overall cue learning and measures of speeded naming, rhyme detection, phoneme awareness, and letter knowledge, even when age and general ability (BPVS and matrices) are controlled for. These findings are consistent with those of Laing and Hulme (1999) and demonstrate that children's ability to learn to read words is directly related to their level of phonological processing skill. Even adopting a conservative alpha level (.005), cue learning is significantly related to letter knowledge and speeded naming in the control group.

A similar pattern can be seen for the WS group in that phonological skills are related to cue learning ability. However, when differences in general ability and age are taken into account none of the correlations is significant at the .005 level. This pattern of results reflects the fact that the correlation between general cognitive ability and reading is much stronger for the WS group than for the control group. These data, along with those from Study 1, suggest that the relationship between phonological skills and reading is weaker for the WS group. This is confirmed by the subgroup analysis for the low ability readers. Although only the relationship between cue-word learning and letter knowledge was significant in the WS group, once age and general cognitive ability were controlled for, in the control group cue-word learning was significantly related to letter knowledge, phoneme awareness, and speeded naming (as well as to BAS reading scores). In the high ability subgroups, no significant relationships emerged.

Discussion

The learning of individuals with WS in a task that mimics the acquisition of a sight vocabulary reveals both similarities to, and differences from, reading-age-matched controls. Learning in the WS group was less efficient overall and they were not influenced by a semantic property (imageability) of the words in the same way as the control group. However, both groups benefited from the phonetic relationship between the printed cue and the target word's pronunciation to a similar degree. Thus, a parsimonious summary of these findings is that for the typically developing children, learning a sight vocabulary is mediated by creating mappings between the orthographic form of a word and both its phonological and semantic representations (see Laing & Hulme, 1999). By contrast, for the WS group, learning appears to involve the creation of mappings between orthography and phonology only. Their slower rate of learning may be related to their failure to create mappings between orthography and semantics.

The fact that the WS group did not show the imageability effect in the word learning task suggests that they were not processing semantic information to support word learning in the same way as the control group. This lack of an imageability effect fits with other findings that suggest that individuals with WS draw less on the semantic system and rely more on phonological encoding in language processing tasks (Grant et al., 1997; Thomas et al., 2001; Vicari, Brizzolara, et al., 1996; Vicari, Carlesimo, et al., 1996; Volterra et al., 1996). Even when phonological processing was constrained, as in the control cue condition, the WS group did not show an imageability effect.

Although the control group learned more cues overall, the difference between groups was not as large as might have been expected. However, this experiment did not measure long-term retention of cues, and this factor may well turn out to differ in the two groups. Indeed, in another study of typically developing children, Laing (1998) demonstrated that typically developing children are able to recall many of the newly learnt words a week later, suggesting that durable learning had taken place. It is possible that, although the WS group learnt many cues in the present experiment, their long-term retention of the cues would be impaired.

The findings of Study 2 highlight the importance of considering, in addition to levels of reading, the process of learning to read. Despite the groups in this study being well matched in terms of their levels of reading skill, it is clear that they approached the task of learning to read new words differently.

General Discussion

Had we simply measured reading scores in our WS population our studies would have revealed relatively few differences between the WS and control groups beyond developmental delay.

The experiments reported in this paper have explored, for the first time, some of the factors that underlie the (limited) ability of people with WS to learn to read. As predicted, reading skills in this group were poor, although there was considerable variability within the group that was associated with individual differences in intelligence and phonological skills. Reading ages of the WS group were lower in this study than has previously been reported (Howlin et al., 1998), but the discrepancy between single word decoding and reading comprehension was replicated. The lack of semantic activation in single word learning demonstrated in Study 2, together with evidence that people with WS rely less on semantics than phonology in their decoding skills (Grant et al., 1997; Thomas et al., 2001; Vicari, Brizzolara, et al., 1996; Vicari, Carlesimo, et al., 1996; Volterra et al., 1996), is likely to contribute to these difficulties with reading comprehension.

Study 1 also demonstrated, for the first time in this atypical group, that phonological skills are related to reading skills. As mentioned, the use of phonological awareness tasks with clinical groups can be problematic because of the metacognitive demands of such tasks. This is why we also used measures of phonological processing that do not require explicit awareness. In WS reading was found to be related to some of these measures of phonological processing skill, particularly speeded naming. Overall, however, the relationship between learning and performance on the phonological tasks appeared to be stronger for the control group than for the WS group.

Even when matched to participants with a similar level of reading skill, children and adults with WS approach the task of learning to read novel words differently. In learning to associate the cues with spoken words, the WS group did show a phonetic cue effect, which suggests that the relationships found between phonological skills and levels of reading attainment in Study 1 are of importance, because learning to read words in the WS group was affected by phonological processes. However, the WS group was not influenced by the imageability of the words, whereas the control group displayed a large imageability effect. In other words, semantics plays a much weaker role in learning to read in Williams syndrome than it does in typical development. The fact that the WS group learnt significantly fewer cues overall suggests that their learning strategy is not only different from that seen in typically developing children but also less effective.

Our study started out by taking a rare genetic disorder, Williams syndrome, as a model for studying both the process and product of learning to read. We have shown that the differential weighting of phonology and semantics plays an important role in how individuals with WS learn to read. Such discoveries help us to go beyond reading scores and explain their reading impairments. It then becomes clear that when a person with a developmental disorder attains reading scores at the level of a 6-year-old, for example, this cannot be taken to imply that the process of learning to read is the same as that of a typically developing 6-year-old. The different strategies involved in learning to read play a vital role. Whether the learning style we have identified in children and adults with WS is specific to this group or also holds for other clinical populations, is an important question for future studies, with crucial syndrome-specific implications for reading remediation.

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Appendix A

Target Words and Cues Used in Experiment 2

High imageability			Low imageability		
Target word	Phonetic cue	Control cue	Target word	Phonetic cue	Control cue
Ladder	ltr	lkr	Middle	mtl	mkl
Jacket	jgt	jpt	Heavy	hfi	hzi
Robber	rpr	rgr	Listen	lzn	lfn
Ticket	tgt	tpt	Hidden	htn	hgn
Coffee	kvi	ksi	Ready	rti	rki
Wizard	wsd	wfd	Because	bgs	bps
Bottle	bdl	bpl	Noisy	nzi	nfi
Rabbit	rpt	rgt	Better	bdr	bpr

Appendix B

Properties of the High- and Low-imageability Word Sets in Experiment 2

	Imageability		
Property	High	Low	t value
Imageability	5.8	2.6	7.90**
Familiarity	3.8	3.8	0.00
Age of acquisition	2.6	2.7	−0.17
Number of letters	6.0	5.8	1.00
Number of phonemes	4.5	4.4	0.48

$^{**}p < .001$.