A case of exceptional reading accuracy in a child with Down syndrome: Underlying skills and the relation to reading comprehension

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We report on a case of a girl with Down syndrome (DS), K.S., whose reading accuracy is exceptional. This ability is associated with robust phonological skills and relative strengths in visual and verbal short-term memory, articulation, and speech fluency. Although her reading comprehension is age appropriate when it comes to the retention of literal information, K.S. has some difficulties in using knowledge-based inferences in reading comprehension. Reading comprehension in that sense is at a level commensurate with her oral language skills. Her reading performance parallels that of children with reading comprehension difficulties who do not have DS. This reading profile is in contrast with claims that individuals with DS mainly use sight-word strategies in reading and shows that the phonological pathway can be highly proficient in a child with DS. However, even in a case such as K.S. where reading accuracy is good, functional literacy is constrained by limited comprehension skills.

Reading is the process of retrieving and comprehending information from print. It comprises the ability to recognize printed words on one hand (reading accuracy) and to understand what those words mean on the other (reading comprehension). In this paper, we report a case of exceptional word recognition ability in a child with Down syndrome (DS), explore the skills underlying this ability, and assess whether exceptional word recognition is accompanied by good reading comprehension.

It is only in the last 20 years that acquiring some level of reading ability has become an attainable goal for individuals with DS. It is not straightforward to get an idea of the level of reading ability generally achieved by such individuals. This is due to changing expectations of achievement, differences in early intervention, school placement, and type of reading instruction received, and the variety of standardized and nonstandardized tests used and scores reported in assessments. Table 1 summarizes the mean reading age achieved by individuals with DS taken from 10 studies in which single-word reading ability was reported and shows an average word recognition level equivalent to that

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Reference	Word recognition test		Mean CA ^a	Mean RAª
K.S.	TOWRE Sight Word Efficiency		98	102
Pieterse and Treloar (1981) ^b	Schonell Graded Reading Test	8	95	85
A. Byrne et al. (1995)	K-ABC Reading Decoding	24	98	75
Cupples and Iacono (2000)	Woodcock Word Identification	22	101	79
A. Byrne et al. (2002)	K-ABC Reading Decoding	24	109	78
A. Byrne et al. (2002)	K-ABC Reading Decoding	24	121	80
Fletcher and Buckley (2002)	BAS Word Reading Test	17	137	86
Evans (1994)	Schonell Graded Reading Test	4	147	78
Snowling et al. (2002)	BAS Word Reading Test	23	156	75
Laws and Gunn (2002)	K-ABC Reading Decoding	30	198	81
Bochner et al. (2001)	Waddington Diagnostic Reading Test	24	252	99

Table 1. Chronological and reading age of K.S. and individuals with Down syndrome in studies of word recognition ability that used standardized tests

Note: CA = chronological age. RA = reading age. TOWRE = Test of Word Reading Efficiency. K-ABC = Kaufmann Assessment Battery for Children. BAS = British Ability Scales.

^aAge in months. ^bCited in Buckley (1985).

of 6- to 8-year-old typically developing children. Although one has to be careful in comparing ageequivalent scores on different reading tests because they can be highly misleading, it was the only score that was reported in many of these studies. Interestingly, the reading level achieved appears to be independent of age. It should be noted though that, within study groups, wide variability in word recognition skills has been reported (e.g., Bochner, Outhred, & Pieterse, 2001; Laws & Gunn, 2002).

An important question is whether reading development in DS is limited by specific cognitive impairments associated with this syndrome. Children with DS were initially thought to rely heavily on sight-word strategies in learning to read (Buckley, 1985; Cossu, Rossini, & Marshall, 1993; Evans, 1994). The extent to which individuals with DS can make use of phonological decoding strategies, as reflected in the ability to read pronounceable nonwords, is again difficult to ascertain for many of the same reasons discussed earlier for word recognition. However, four studies that report scores on a nonword reading test (the Word Attack subtest of the Woodcock Reading Mastery Tests; e.g., Woodcock, 1998) suggest that some decoding skills are present, but that they are reduced compared to word recognition skills (Boudreau, 2002; Cupples & Iacono, 2000; Fowler, Doherty, & Boynton,

1995; Kay-Raining Bird, Cleave, & McConnell, 2000). An exception to this is the performance of 5 young adults with DS, who had the highest level of proficiency in reading in the group of individuals assessed by Fowler and colleagues (1995). Their age-equivalent score on the nonword reading test was ahead of their word reading score. This raises the question of whether such individuals have unusual cognitive strengths compared to other people with DS.

Research with typically developing children has documented a variety of underlying cognitive– linguistic processes in addition to phonological awareness that support successful word recognition and decoding, including general cognitive ability (Boudreau, 2002; Ellis & Large, 1988), visual and verbal short-term memory skills (Brady, Mann, & Schmidt, 1987; Brady, Shankweiler, & Mann, 1983; Ellis & Large, 1988), and oral language skills, in particular receptive grammar skills (e.g., Muter & Snowling, 1998).

In individuals with DS a similar picture has emerged, although factors such as hearing status and reading instruction received have not always been taken into account. Readers with DS have been found to perform better than nonreaders with DS on measures of nonverbal cognitive ability, even after differences in hearing were controlled (Laws & Gunn, 2002). However, nonverbal cognitive ability was not a significant predictor for word recognition in a study carried out by Boudreau (2002) that included readers and nonreaders with DS. In that study, nonverbal cognitive ability did predict decoding (i.e., nonword reading) skills (Boudreau, 2002). Visual and verbal short-term memory spans have consistently been reported as predictive for word recognition abilities in individuals with DS (Fidler, Most, & Guiberson, 2005; Fowler et al., 1995; Kay-Raining Bird et al., 2000; Laws, 1998; Laws, Buckley, Bird, MacDonald, & Broadley, 1995; Laws & Gunn, 2002). In addition, Kay-Raining Bird et al. (2000) found that decoding ability is predicted by verbal short-term memory span after chronological age, mental age, and phoneme segmentation are controlled for. Fowler et al. (1995) even concluded that a verbal shortterm memory span of four was a necessary, but not sufficient, prerequisite for acquiring decoding skills. Finally, differences in oral language skills have also been associated with individual differences in word recognition and decoding in DS in such a way that individuals with stronger oral language skills also showed stronger word recognition and decoding skills (Fowler et al., 1995; Kay-Raining Bird et al., 2000; Laws et al., 1995; Laws & Gunn, 2002).

Although some anecdotal descriptions of highly proficient readers with DS exist (e.g., Buckley, 1985), no extensive quantitative report of their skills has been published as far as we are aware. From the literature on underlying individual differences associated with reading success in DS it appears that we might expect advances in general cognitive ability, vocabulary, grammar, and visual and verbal short-term memory in a proficient reader with DS.

We report a case (K.S.) of exceptional word recognition and decoding ability in a girl with DS. In the first study, we compare K.S.'s performance with that of other readers with DS to establish how unusual her word recognition and decoding abilities are. We also include a range of language and memory tests in an attempt to ascertain the levels of different aspects of cognitive-linguistic functioning associated with K.S.'s word recognition and decoding abilities. In the second study, we address the question of whether K.S.'s exceptional word recognition and decoding abilities are accompanied by good phonological skills. In the third study, we investigate her reading comprehension ability. We ask whether it matches her word recognition and decoding skills and compare K.S.'s performance on a listening comprehension task with that of children with reading comprehension problems who do not have DS. Finally, we compare her level of reading comprehension to the level of her oral language abilities.

Participant: Case K.S.

K.S., a girl with Trisomy 21, is a monolingual speaker of English and was assessed over a period of several days when she was 8;2 and 8;6 (years;months). She is the second child in a family with three children, and both parents completed higher education degrees. The pregnancy and birth of K.S. were unproblematic. A report from a qualified audiologist states that K.S., at age 8, had a mild conductive loss in both ears at the lower frequencies and suffers from glue ear. Grommets were inserted into her right ear at age 7, but were no longer in place by age 8. K.S. has always been integrated into mainstream primary school and at the time of testing she was in Year 3, receiving 20 hours of support. K.S. was supported by the Portage programme¹ and was taught Makaton² from the age of 3 months and became a skilled user, but does not currently use signs. Her parents started to teach her reading whole words at the age of 2;6. At 3;9 she started to attend the Early Development Group at the Sarah Duffen Centre in Portsmouth. By the age of 4;6 K.S. was reading simple books. At that

¹ A home visiting educational service for preschool children with additional support needs and their families.

² A communication programme using gestures derived from British Sign Language.

stage, her parents started to teach her letter sounds and blending letter sounds into words.

Methodology

In single-case studies, one seeks to demonstrate that a participant is behaving differently from a small sample of controls, either healthy participants or a group of other patients. A range of methods has been employed to substantiate such claims statistically (for a review, see Mycroft, Mitchell, & Kay, 2002). Two very common methods are descriptive statistics (e.g., the participant's score does not fall within two standard deviations of the mean for controls) and statistics based on z. The problem with using descriptive statistics alone is that they are based on the observation of the participant's performance in relation to the mean and distribution of the control scores, but do not offer any formal test of the likelihood that the participant's data are drawn from the control population (Crawford, Howell, & Garthwaite, 1998; Mycroft et al., 2002). The use of z statistics is inappropriate because it treats control sample statistics as population parameters, rather than as sample statistics. With the modest sample sizes typically used in single-case reports, this leads to an increase in the Type I error rate, and hence the abnormality of the participant's score will be exaggerated (for a detailed description of this argument, see Crawford et al., 1998). In this paper, we follow the statistical methods based on modified t tests suggested by Crawford and colleagues (Crawford & Garthwaite, 2002, 2005; Crawford, Garthwaite, Howell, & Gray, 2004; Crawford et al., 1998) that treat data from the control sample as sample statistics rather than as population parameters. Specifically, we use two different statistical tests that both test whether one can reject the null hypothesis that the case's scores are observations from the control population (i.e., they serve as significance tests) but simultaneously provide a point estimate

of the abnormality of the cases's scores, as well as a confidence interval (CI) that quantifies the uncertainty associated with this estimate. First, to compare K.S.'s score on a (sub)test with the score of the control sample the programme SINGLIMS.EXE (Crawford & Garthwaite, 2002) was used. Second, to test whether the difference between scores on two (sub)tests observed for K.S. is significantly greater than the differences observed for the control sample, the revised standardized difference test was used (RSDT.EXE; Crawford & Garthwaite, 2005).³ Two-tailed tests were performed on all occasions unless explicitly stated otherwise.

STUDY 1: WORD RECOGNITION, DECODING, AND COGNITIVE– LINGUISTIC FUNCTIONING IN K.S. AND OTHER READERS WITH DS

We compare K.S.'s performance with that of other readers with DS to establish how unusual her word recognition and decoding abilities are. A wide range of hearing, language, and memory tests was administered in an attempt to shed some light on the cognitive-linguistic skills associated with her word recognition and decoding abilities. The few existing studies that have looked at individual differences in reading accuracy in DS suggest that we might expect advances in vocabulary, grammar, verbal short-term memory, and visual processing skills in a proficient reader with DS. Because of these a priori hypotheses, we performed one-sided significance tests for the comparisons of K.S. with the DS control group on the language and memory tests.

Participants and methods

K.S.'s word recognition, decoding, language, and memory abilities are compared with the performance of 13 readers with DS, of similar nonverbal

³ Both methods have been shown to be robust to departures from normality in terms of control of Type I and Type II error rates (Crawford & Garthwaite, 2006; Crawford, Garthwaite, Azzalini, Howell, & Laws, 2006; Garthwaite & Crawford, 2004).

cognitive ability, t(12) = -0.636, p = .536, chronological age, t(12) = -1.003, p = .336, and hearing ability, t(9) = -0.471, p = .649. See Table 2 for information on the sample. All the children were supported by Portage programmes from an early age onwards and were taught Makaton. Several of them also attended early development programmes for children with DS. All attended mainstream primary schools in England at the time of testing, from which it follows that they would have been exposed to the National Literacy Strategy. It should be noted that an additional 6 children with DS were tested, but were not included because they did not obtain a score on the reading measure. Either they failed to read at least one of the practice items successfully (3 children), or the test was judged to be too difficult for them by a parent or a teacher (3 children). As a consequence, the control group of children with DS is not representative of the population, but rather constitutes a more able proportion of it.

Because standard scores were either heavily skewed or not informative, as they did not differentiate between individuals, we report raw scores on all the tests. The only exceptions are the performance on the reading measure and the Children's Communication Checklist-2, where we report standard scores. Standard scores on these measures showed reasonable variability among the individuals and were not heavily skewed.

 Table 2. Comparison of K.S. with other readers with Down
 syndrome on background and reading measures

	DS readers ^a	K.S.
Age ^b	111.2 (12.68)	98
Hearing level ^c	24 (8.10)	20
Nonverbal cognitive ability ^d	4.69 (2.56)	3
TOWRE Words ^e	78.69 (11.61)	106
TOWRE Nonwords ^e	80.62 (9.89)	122

Note: DS = Down syndrome. TOWRE = Test of Word Reading Efficiency. Mean values are shown, with standard deviations in parentheses.

Hearing level

After an otological inspection of the ear by a qualified audiologist, an air pure-tone audiogram for 1,000 Hz in the right ear was carried out. Three children were unable to comply with the procedure. All audiological assessments followed procedures as recommended by the British Society of Audiology. The score reported refers to the threshold in dB HL.

Word recognition and decoding ability

Word recognition and decoding ability were assessed at the single-word level using the two subtests of the Test of Word Reading Efficiency (TOWRE; Torgesen, Wagner, & Rashotte, 1999): the "Sight Word Efficiency" or Words subtest and the "Phonemic Decoding Efficiency" or Nonwords subtest, respectively. In this American test the child is asked to read as many words or nonwords as possible in 45 seconds of a list that increases in difficulty. The test words are preceded by eight practice words. To proceed with the test items, the child has to read at least one practice item successfully. As the variation of standard scores for this test is reasonable, and their distribution does not appear to violate assumptions of normality, standard scores are reported.

General cognitive ability

Nonverbal cognitive ability was assessed using the Matrix Reasoning subtest of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999). The tasks involved in completing the matrices are pattern completion, classification, analogy, and serial reasoning. Verbal cognitive ability was assessed using the Similarities subtest of the WASI (Wechsler, 1999). Similarities is a measure of verbal concept formation and abstract verbal reasoning ability. Reported scores for both subtests are raw scores, reflecting the number of items completed successfully. To facilitate the comparison of K.S.'s nonverbal cognitive ability with her other skills, her percentile rank on the WASI Matrix Reasoning is also reported.

^a*n* = 13 (8 girls). ^bIn months. ^cIn dB HL; *n* = 10 (7 girls). ^dRaw score. ^cStandard score (*M* = 100, *SD* = 15).

Children's Communication Checklist-2

To obtain an estimate of the child's use of language in everyday situations and to assess aspects of communication that are not easily evaluated using more traditional language tests, parents completed the Children's Communication Checklist-2nd edition (CCC-2; Bishop, 2003a). This questionnaire consists of 70 multiple-choice items, divided into 10 scales. The first 4 scales (speech, syntax, semantics, and coherence) assess aspects of language structure, vocabulary, and discourse. The next 4 (inappropriate scales initiation, stereotyped language, use of context, and nonverbal communication) cover pragmatic aspects of communication. The last 2 scales (social relations and interests) assess behaviours that are usually impaired in cases of autistic disorder.

Articulation and Oromotor skills

The Goldman-Fristoe Test of Articulation (Goldman & Fristoe, 1986) was used to test the children's ability to produce the consonant sounds of English in isolated words. Scores reported are raw scores, reflecting the percentage of correctly pronounced consonants of the total number of elicited consonants.

Articulatory ability was further assessed by using the Oromotor Sequences subtest of the NEPSY (Korkman, Kirk, & Kemp, 1998). This test assesses the child's articulatory fluency and the ability to organize and produce rhythmic oral sequences. The child is asked to produce five repetitions of a range of articulatory sequences such as tick tock, split splat, and scoobelly doobelly, followed by a series of increasingly difficult tonguetwisters. The test phase is preceded by a practice item on which corrective feedback is given. In order to minimize effects of articulation difficulties and memory problems, only the results for singleonset, one-syllable items (i.e., tick tock, mish mash, and puh tuh kuh) are included in this paper. The scores reported are raw scores, reflecting the number of correct responses for these items. Errors included interruptions longer than the time of one sequence and omissions, distortions, and substitutions of sounds. Stable misarticulations were not penalized.

Grammar

Understanding of a range of grammatical constructions was assessed using the Test for Reception of Grammar–Version 2 (TROG-2; Bishop, 2003b). In the TROG-2, the child is presented with four trials for a range of grammatical constructions, and all four have to be answered successfully in order to pass the block. The child is asked to point to the picture (out of four choices) that best represents the sentence spoken by the examiner. The test is discontinued after five consecutive blocks have been failed. Scores reported are raw scores, reflecting the number of blocks completed successfully.

An estimate of expressive grammatical complexity of the child's speech in the form of a "mean length of utterance" was derived from the speech sample collected as part of the administration of the Expression, Reception, and Recall of Narrative Instrument (ERRNI; Bishop, 2004). The ERRNI tests a child's ability to relate a story, comprehend it, and remember it after a delay. A pictorial context is used to elicit a narrative, which is recorded and transcribed. The test phase is preceded by a "warm-up", where the child is asked to tell the examiner as much as he or she can about a picture of a swimming pool. The goal of the warm-up picture is to encourage the child to talk about pictorial material. This is followed by a preview of the story book. In this paper, only the mean length of utterance in words (MLUw) based on the initial story telling part of the ERRNI is reported. In this part, the child is asked to tell the story of what happens in the pictures. Prompting during the test phase was restricted to general encouragement and relatively nonspecific prompts such as "What happened next?" in order to elicit full utterances as responses. The normal procedure for the computation of the MLUw from the ERRNI involves combining the results from the initial story telling and the story recall phase to get a more reliable estimate. However, the manual mentions that it is acceptable to compute MLUw on the basis of just one story as the resulting standard score correlates .94 with the MLUw based on both initial story

telling and story recall. The ERRNI has two parallel forms; we used the Fish story.

Vocabulary

Vocabulary was assessed using the Expressive and Receptive One-Word Picture Vocabulary Tests (EOWPVT, ROWPVT; Brownell, 2000a, 2000b). The EOWPVT is a measure of an individual's expressive vocabulary, which is assessed by asking the child to name objects, actions, and concepts pictured in illustrations. The ROWPVT is a measure of receptive vocabulary, which is assessed by asking the child to identify, from four choices, the illustration that depicts the meaning of a word presented orally by the experimenter. As these are American tests, some of the items had to be adapted to reflect British English vocabulary more closely (for instance the word "lorry" for "truck"). Scores reported are raw scores, reflecting the number of items completed successfully.

Short-term memory

The Digit Span task of the Wechsler Intelligence Scale for Children-Third Edition UK (WISC-III^{UK}; Wechsler, 1992), and the Sentence Repetition and Repetition of Nonsense Words subtests of the NEPSY (Korkman et al., 1998) were used to assess verbal short-term memory, while a Corsi Span measure probed visuo-spatial short-term memory. In the Digit Span task, the child was asked to repeat increasingly long sequences of digits, presented orally at a rate of one per second. Two items were presented at each list length, starting at a list length of two, and at least one had to be completed successfully in order to continue with the next list length. A practice item on which corrective feedback was given preceded the test phase. The same procedure was followed in the Corsi Span task, except that the presentation of the items and the response of the child consisted of pointing at increasingly long sequences of blocks in a fixed array of nine randomly arranged blocks. Only the forward span parts of the tasks were carried out. Scores reported are raw scores and reflect the mean of the two longest memory spans repeated successfully by the child.

In the Sentence Repetition subtest of the NEPSY the child is asked to repeat sentences of increasing complexity and length after a single oral presentation. Two points are awarded for each correct verbatim response, and one point per item if the child makes one or two errors. Omitting, changing, or adding a word or changing word order counts as an error. The test is discontinued after four consecutive scores of zero. Scores reported are raw scores, which reflect the sum of the points awarded on each item.

The Repetition of Nonsense Words subtest of the NEPSY assesses phonological decoding of sound patterns as well as encoding and articulating complex and unfamiliar words. The child listens to recorded nonsense words increasing in length and complexity and repeats each word after it is presented. Errors include distortions and omissions of syllables. Stable misarticulations and stressing the wrong syllable are not counted as errors. Scores reported are raw scores, reflecting the number of syllables pronounced correctly.

Results

How does K.S.'s word recognition and decoding ability compare to the performance of other readers with DS?

Compared with other readers with DS, K.S. scores significantly higher on the Words subtest, t(12) = 2.267, p = .043. The estimated percentage of the DS population in the age range examined that would obtain a score higher than K.S. is 2.13%, and the 95% CI on this percentage is from 0.03% to 10.32%. On the Nonwords subtest, K.S. again scores significantly higher than the DS control group, t(12) = 4.033, p = .002. The estimated percentage of the DS population in the age range examined that would obtain a score higher than K.S. is 0.08%, and the 95% CI on this percentage is from 0% to 0.74%. See Table 2 for means and standard deviations.

How unusual is K.S.'s discrepancy between word recognition and decoding ability compared to other readers with DS?

Although on average the score on the Nonwords subtest of the TOWRE seems to be slightly

ahead of the score on the Words subtest in the DS control group, this difference is not statistically significant, t(12) = -0.711, p = .491. When looking at individuals within this group, three children show a difference big enough to reach statistical significance according to the data reported in the test manual. Two of these children scored higher on the Nonwords subtest than on the Words subtest. For the remaining child, the difference was in the opposite direction. In none of these children was the difference large enough to be clinically significant, defined in the TOWRE test manual as a difference that occurred in less than 5% of the children of the normative sample. K.S. showed a difference of 16 points between her standard scores on the Nonwords subtest and the Words subtest. Although statistically significant, a difference of this magnitude is not that unusual to be of clinical significance, according to the TOWRE test manual. In order to compare K.S.'s difference score to the difference scores obtained by the DS control sample directly, the revised standardized difference test was used (Crawford & Garthwaite, 2005), and a trend towards statistical significance was found, t(12) =1.825, p = .093. Only an estimated 4.64% of the DS population would exhibit a difference in favour of nonwords that is more extreme than K.S.

How is K.S. different from other readers with DS in her cognitive-linguistic functioning?

K.S.'s score on the WASI Matrix Reasoning subtest falls in the 3rd percentile, which indicates substantially lower nonverbal cognitive skills than those of typically developing children her age but, as stated earlier, similar to those of the DS control group. Tables 3 and 4 summarize the results of the comparison of K.S. with the other readers with DS on the language and memory tests. K.S. scores significantly better on Digit Span than does the DS control group, and there are also strong trends for K.S. to score higher on two other short-term memory measures (Corsi Span and Sentence Repetition) and on the speech subscale of the CCC-2. No significant differences were found on any of the other measures. Closer examination of the results does

provide a clear profile of her strengths in some areas of language and memory compared to other readers with DS. On the CCC-2, K.S.'s score falls in the top 10% on the speech subscale and in the top 20% on the pragmatic subscale related to the initiation of conversation. K.S.'s score also falls in the top 10% on the visuo-spatial shortterm memory test (Corsi Span) and two of the verbal short-term memory tests (Digit Span and Sentence Repetition). On the articulation measures (Goldman-Fristoe and Oromotor Sequences), the nonword repetition test, and the estimate of the grammatical complexity of her expressive language as reflected in the MLUw, K.S.'s score falls in the top 20%. It is worth mentioning that although her utterances were relatively long (MLUw = 8.61), she made grammatical errors in about 20% of them. The two most common errors were omission of the subject and subject-verb agreement problems. The ERRNI does not provide norms for such errors as they were extremely rare in children of 5 years or above in the normative sample.

Discussion

With a standard score of 106 on the Words subtest, K.S.'s word recognition ability at the single-word level is age appropriate. Although anecdotal reports of children with DS whose word recognition abilities are age appropriate or ahead of those expected for typically developing children of their age exist (Buckley, 1985), to our knowledge no systematic studies have been published. As mentioned earlier and as illustrated in Table 1, on average, individuals with DS seem to achieve a word recognition level equivalent to that of 6- to 8-year-old typically developing children, and this appears to be independent of age. Even for children in K.S.'s age range, aged 7;11-8;5 (years; months), word recognition ability lags behind their chronological age (A. Byrne, Buckley, MacDonald, & Bird, 1995; Cupples & Iacono, 2000; Pieterse & Treloar, 1981, cited in Buckley, 1985). In this sense, K.S.'s performance is unusual. This picture from the literature is confirmed by the comparison to

Subscale	DS readers ^b	<i>K.S.</i>	Percent. below ^c	t	p^{d}
Speech	1.30 (1.50)	4	93.99 (77.54-99.75)	1.723	.059
Syntax	2.90 (4.12)	3	50.90 (27.55-74.01)	0.023	.491
Semantics	4.50 (2.76)	2	20.51 (5.12-44.26)	-0.864	.205
Coherence	4.00 (1.63)	3	28.65 (10.04-53.20)	-0.584	.287
Inappropriate initiation	4.60 (1.08)	6	87.61 (66.35-98.36)	1.242	.123
Stereotyped language	5.50 (2.51)	5	42.68 (20.60-66.73)	-0.190	.427
Use of context	2.50 (0.71)	2	25.94 (8.27-50.35)	-0.674	.259
Nonverbal communication	7.20 (2.66)	6	33.86 (13.72-58.43)	-0.430	.338
Social relations	5.90 (2.33)	4	22.84 (6.41-46.94)	-0.777	.228
Interests	6.40 (2.17)	7	60.10 (35.82-81.64)	0.264	.399

Table 3. Scaled scores^a for K.S. and the control group of readers with Down syndrome on the CCC-2

Note: Percent. below = estimated percentage of DS population below K.S.'s score. DS = Down syndrome. CCC-2 = Children's Communication Checklist-2nd edition.

 ${}^{a}M = 10$, SD = 3. ${}^{b}n = 10$ (6 girls); mean values, with standard deviations in parentheses. Mean values, with 95% confidence interval (CI) in parentheses. d One-sided.

the DS control group of similar nonverbal cognitive ability and chronological age. K.S.'s score on the TOWRE Words subtest is significantly higher than that of the DS control group, with an estimated percentage of less than 3% of the DS population aged 7–11 years achieving a higher score. With regard to decoding ability as indexed by her performance on the TOWRE Nonwords subtest, K.S. stands out even more. With a standard score of 122, she performs above average compared to typically developing children 8 years of age. Her score is again significantly higher than that of the DS control group, with an estimated percentage of less than 1% of the DS population aged 7–11 years achieving a higher score. K.S.'s decoding ability is ahead of her word recognition ability, as it was for 5 young adults with DS, who had the highest level of proficiency in reading in the group of individuals assessed by Fowler and colleagues (1995).

Test		DS readers ^a	K.S.	Percent. below ^b	t	₽°
Articulation	Goldman-Fristoe	73.33 (15.01)	92.42	87.81 (69.75-97.72)	1.226	.122
	Oromotor sequences	9.23 (3.75)	14	87.83 (69.79-97.73)	1.227	.122
Grammar	ERRNI MLŪw ^d	5.33 (2.53)	8.61	87.86 (67.92-98.19)	1.241	.121
	TROG-2	3.54 (2.82)	5	68.70 (47.15-86.23)	0.500	.313
Vocabulary	EOWPVT	53.62 (16.59)	55	53.13 (32.16-73.44)	0.080	.469
,	ROWPVT	58.77 (17.55)	67	67.03 (45.46-84.96)	0.452	.330
Short-term memory	Corsi Span	3.12 (0.85)	4.5	92.56 (77.43-99.23)	1.579	.070
	Digit Span	2.96 (0.52)	4	95.50 (88.37-99.77)	1.928	.039
	Sentence Repetition	8.54 (2.57)	13	93.99 (80.15-99.53)	1.673	.060
	Nonword Repetition	17.91 (7.23)	25	81.50 (59.38-95.47)	0.939	.185
Verbal cognitive ability	WASI Similarities ^e	11.30 (6.82)	14	64.28 (39.77-84.89)	0.378	.357

Table 4. Raw scores for K.S. and the control group of readers with Down syndrome on the language and memory tests

Note: Percent. below = estimated percentage of DS population below K.S.'s score. DS = Down syndrome. ERRNI MLUw = Expression, Reception and Recall Narrative Instrument Mean Length of Utterance in Words. TROG-2 = Test for Reception of Grammar–Version 2. EOWPVT = Expressive One-Word Picture Vocabulary Test. ROWPVT = Receptive One-Word Picture Vocabulary Test. WASI = Wechsler Abbreviated Scale of Intelligence.

 ${}^{a}n = 13$ (8 girls); mean values, with standard deviations in parentheses. ^bMean values, with 95% confidence interval (CI) in parentheses. ^cOne-sided. ${}^{d}n = 11$ (9 girls). ${}^{e}n = 10$ (6 girls).

In the DS control group 7 children showed a difference score in the same direction, but only two of these scores were big enough to reach statistical significance as defined by the test manual. On average there was a trend for K.S.'s difference score to be significantly more extreme than the difference scores found in the DS control group, with less than 5% of the DS population age 7–11 years exhibiting a difference more extreme than K.S. From this we conclude that K.S.'s word recognition and decoding ability is exceptional for a child with DS of her age and that she is particularly proficient in decoding. This proficiency has developed in the presence of limited nonverbal cognitive abilities.

K.S. scores significantly higher than the other readers with DS on the Digit Span task. In addition, there were strong trends for her word recognition and decoding abilities to be accompanied by relatively high capacities in two other tests from the short-term memory domain and tests from the articulation and speech fluency domain. Her scores on these tests fall in the top 10% and top 20%, respectively. In addition, her MLUw is relatively high, again falling in the top 20%. In contrast, K.S. does not stand out in this way when it comes to grammar comprehension, vocabulary, or cognitive ability. These differences do not appear to be explained by differences in hearing level as K.S. did not differ from the other readers with DS on this.

Her strengths in the short-term memory domain fit in well with the reports of associations between both visual and verbal short-term memory and word recognition and decoding in individuals with DS (Fidler et al., 2005; Fowler et al., 1995; Kay-Raining Bird et al., 2000; Laws, 1998; Laws et al., 1995; Laws & Gunn, 2002). Also, strong expressive grammar skills as estimated by MLUw have been found to be associated with individual differences in word recognition in DS in earlier studies (Laws et al., 1995; Laws & Gunn, 2002). In contrast, strengths in articulation and speech fluency have not previously been specifically reported in individuals with DS with high levels of proficiency in word recognition and decoding. Other studies have reported

associations of word recognition and decoding with receptive vocabulary and grammar and cognitive ability, but the individuals in these studies were mostly teenagers and young adults (Fowler et al., 1995; Kay-Raining Bird et al., 2000; Laws et al., 1995; Laws & Gunn, 2002). It is possible that through their longer reading experience, older readers might have had more opportunity to develop stronger skills in these areas.

STUDY 2: FUNCTIONING OF THE PHONOLOGICAL PATHWAY

From K.S.'s performance on the Words and Nonwords subtests of the TOWRE it appears that she has a well-developed orthographic system and makes use of grapheme-phoneme correspondences. To examine functioning of the phonological pathway in K.S. further, her ability to read nonwords was examined in more detail, and her phonological skills were assessed. Specifically, people can use analogies with words rather than their knowledge of grapheme-phoneme correspondences to read nonwords (Treiman, Goswami, & Bruck, 1990). In a study with French individuals with DS, Gombert found a strong trend for them to do worse than controls when reading nonwords without orthographic neighbours, suggesting that they relied more heavily on analogies between the nonwords and words in reading them (Gombert, 2002). We investigated how heavily K.S. relied on these analogies in reading nonwords.

While the ability to read nonwords shows implicit knowledge of grapheme-phoneme correspondences, phonological awareness can be defined as the ability to focus explicitly on the sound structure of language. Phonological awareness can be conceptualized as consisting of awareness of larger phonological units, such as rhymes on one hand and the smallest units, phonemes, on the other. Rhyming skills may or may not be necessary in acquiring the alphabetic principle, but they generally precede phonemic awareness skills in typically developing children (Stanovich, Cunningham, & Cramer, 1984; Yopp, 1988).

With regard to phonemic awareness, two abilities seem to be important in acquiring alphabetic decoding skills: phoneme identity and phoneme manipulation (B. Byrne, 1998; Murray, 1998). Phoneme identity (also referred to as "phoneme invariance") involves detecting that two phonemes are in some respects the same across differing phonetic contexts, which is typically assessed in phoneme matching tasks. Phoneme manipulation skills refer to the ability to break down spoken words into discrete phonemes ("segmentation") and to smoothly assemble an ordered phoneme sequence to identify a spoken word ("blending"). This is typically assessed in tasks in which phonemes have to be counted, tapped, segmented, inverted (e.g., spoonerisms), or blended.

As mentioned earlier, readers with DS were initially thought to rely heavily on sight-word strategies and to lack phonological awareness skills (Buckley, 1985; Cossu et al., 1993; Evans, 1994). Cossu et al. (1993) even concluded that the absence of phonological awareness skills in a group of children with DS who could read shows that these skills are not necessary in learning to read. However, recently it has become clear that readers with DS can show measurable levels of phonological awareness and that these skills are associated with word recognition and decoding ability (Cardoso-Martins & Frith, 2001; Cupples & Iacono, 2000; Fletcher & Buckley, 2002, Fowler 1995, Gombert, et al., 2002; Kay-Raining Bird et al., 2000; Kennedy & Flynn, 2003a; Laws & Gunn, 2002; Snowling, Hulme, & Mercer, 2002). Furthermore, two small-scale intervention studies indicate that children with DS can be taught phonological skills (Kennedy & Flynn, 2003b) and that phonological reading instruction improves the ability to read unfamiliar words in children with DS (Cupples & Iacono, 2002). Although these last two studies should be interpreted with caution because of their extremely small sample sizes, they do suggest that this area merits further investigation. One explanation for earlier findings of a lack of phonological awareness skills in readers with DS is of a methodological nature. It has been suggested that the children with DS in Cossu

et al.'s (1993) study performed poorly on the phonological awareness tasks as a result of poor general cognitive ability, rather than poor phonological awareness (Bertelson, 1993; B. Byrne, 1993; Cupples & Iacono, 2000; Morton & Frith, 1993). As Cupples and Iacono (2000) noted, the tasks used by Cossu et al. (1993) have a high memory load. Specifically, although the majority of children with DS had digit spans of 3 or lower, they were asked to blend 4 or 6 individual phonemes together in the phoneme blending task. The same criticism applies to the study carried out by Evans (1994). In some subsequent studies, memory load has been minimized by supplementing auditory presentation with pictorial presentation of the items (e.g., Cardoso-Martins & Frith, 2001; Cupples & Iacono, 2000).

Although recent studies found that individuals with DS can succeed on phonological awareness tasks, they also suggest that in some respects, the development of reading and phonological awareness skills is qualitatively different in children with DS. For instance, while the ability to analyse rime has been shown to precede phonemic awareness skills in typically developing children (Stanovich et al., 1984; Yopp, 1988), readers with DS show a deficit in this area (Boudreau, 2002; Cardoso-Martins, Michalick, & Pollo, 2002; Gombert, 2002; Snowling et al., 2002). Not only did they perform significantly more poorly on rhyming tasks than did controls matched on nonverbal mental age and/or on reading ability (Boudreau, 2002; Gombert, 2002; Snowling et al., 2002), they also found a rime-matching task more difficult than a middle-phoneme-matching task, whereas this was reversed for typically developing children (Cardoso-Martins et al., 2002). Their lack of rhyming skills does not seem to be due to decreased familiarity with nursery rhymes, but it has been proposed that children with DS might be selectively more sensitive to sounds at the beginnings than at the ends of words (Snowling et al., 2002).

With regard to phonemic awareness skills, in general the performance of individuals with DS on phoneme manipulation tasks seems poorer than their performance on phoneme identity tasks, although findings are complicated by

differences in task design. Specifically, in a number of studies, their performance on phonemematching tasks requiring judgements as to whether a target word began with the same sound as one of the test words was not significantly different from the performance of typically developing children, matched on nonverbal mental age or reading ability (Cardoso-Martins & Frith, 2001; Snowling et al., 2002). However, in two studies where the phoneme-matching tasks employed an odd-one-out procedure they performed significantly poorer than the control group (Boudreau, 2002; Gombert, 2002). Individuals with DS were significantly poorer on phoneme manipulation tasks involving tapping and segmentation (Gombert, 2002). Concerning the opposite manipulation of phoneme blending, children and adolescents with DS performed as well as nonverbal-mental-age-matched controls in the study by Boudreau (2002), but worse than reading-age-matched controls in the study by Gombert (2002). Although these findings initially seem contradictory, three possible explanations exist. First, the tasks differ in the level at which sounds had to be blended. Boudreau's task included items at the word (e.g., birth-day), syllable (e.g., pen-cil), onset-rime (e.g., f-un), and phoneme (e.g., f-i-s-h) level, whereas Gombert's task only looked at the phoneme level. Second, it could be that both groups performed at floor level on the task in Boudreau's experiment, which, given the age of the control group (3;6-5;3 years;months) is not unlikely. Third, it could be that individuals with DS are able to achieve a certain reading level in the absence of the development of additional skills that are necessary to complete the phoneme blending task, and these skills are reflected in measures of nonverbal mental ability. Finally, individuals with DS were significantly poorer on phoneme deletion tasks than were controls matched on reading ability (Cardoso-Martins & Frith, 2001; Gombert, 2002). In a way, this is not surprising as typically developing children find phoneme deletion the most difficult phonological awareness task. It should be noted though that in many segmentation, tapping, blending, and deletion tasks, oral

presentation was not supplemented by pictures, placing a high demand on memory. To see whether good word recognition is accompanied by appropriate phonological awareness skills, we assessed K.S.'s rhyming, phoneme identity, and phoneme manipulation skills.

In addition to phonological awareness, it has been proposed that phonological production skills, as assessed in rapid automatic naming (RAN) tasks and as reflected in speech rate, contribute to the reading process (Bowers & Ishaik, 2003; McDougall, Hulme, Ellis, & Monk, 1994; Muter & Snowling, 1998; Wolf, 1991). Both measures can be seen as providing an index of the speed and efficiency with which phonological codes of words long-term memory can be activated in (McDougall et al., 1994; Muter & Snowling, 1998; Wagner & Torgesen, 1987). In a RAN task the participant is asked to name digits or a limited set of pictures as fast as possible. Bowers and colleagues have found that performance on these tasks predicts word recognition level independently of variance explained by phonological awareness skills, and individuals with word recognition deficits, such as dyslexics, show deficits on RAN tasks (Sunseth & Bowers, 2002; Wolf & Bowers, 1999). As far as we are aware, performance on a RAN task has not been investigated in individuals with Down syndrome, but since it is an important predictor of word recognition in typically developing children, we assessed K.S.'s phonological production skills using a RAN task.

Finally, given the significant predictive value of speech rate for verbal short-term memory and reading accuracy in typically developing children (McDougall et al., 1994; Muter & Snowling, 1998), we decided to include this measure in the examination of K.S.'s phonological production skills. Although a close association between verbal short-term memory span and its development with age and speech rate has been reported for typically developing children and other atypical groups such as Williams syndrome, this appears to be less straightforward in individuals with DS (Hulme & Roodenrys, 1995; Jarrold, Cowan, Hewes, & Riby, 2004). Although individuals with DS have typically been found to have impaired verbal short-term memory spans (e.g., Fowler et al., 1995; Hulme & Mackenzie, 1992; Jarrold & Baddeley, 1997) and reduced speech rates relative to typically developing individuals of an equivalent vocabulary level in one study (Hulme & Mackenzie, 1992), but not in other studies (Jarrold et al., 2004; Seung & Chapman, 2000), a reliable relationship between speech rate and verbal short-term memory performance among individuals with DS has not been demonstrated so far (Jarrold, Baddeley, & Hewes, 2000; Jarrold et al., 2004; Seung & Chapman, 2000; Vicari, Marotta, & Carlesimo, 2004). To our knowledge, no study has investigated the relationship between speech rate and reading accuracy in individuals with DS.

Participants and methods

K.S.'s nonword reading performance is compared with that of a group of 15 typically developing children matched on chronological age (M =100.73 months, SD = 2.94), t(14) = 0.418, p =.682, taken from the larger control group described in Griffiths and Snowling (2002).

The control group was not given the phonological awareness tasks as ceiling level performance would have been expected. For those tasks that are standardized measures, it is possible to compare K.S.'s performance with that of the normative sample. The same holds for the tasks assessing K.S.'s phonological production skills.

Nonword reading

Two measures of nonword reading were used: the Graded Nonword Reading Test (GNWRT; Snowling, Stothard, & McLean, 1996) and a list of nonwords that vary in their similarity to words, compiled by Griffiths and Snowling (2002). The GNWRT is a standardized test that contains 20 nonwords (10 monosyllabic and 10 two-syllable nonwords) varying in phonological complexity. The test trials are preceded by 5 practice nonwords. The list compiled by Griffiths and Snowling (2002) consists of 24 monosyllabic and 8 two-syllable words. In this paper, we only report on the performance on the monosyllabic words.

The 24 one-syllable nonwords varied in the frequency of their orthographic rime unit; 8 items contained high-frequency rimes (e.g., yoal, vag), 8 items contained low-frequency rimes (e.g., choub, vep), and 8 nonwords had no close orthographic neighbours (e.g., phuve, glaje).

Phonological skills

Phonological skills were assessed using a variety of tests. Rime analysis skills and phoneme identity were probed in the two tests as used by Bird, Bishop, and Freeman (1995). In the rimematching test, the child has to select, from an array of four pictures, the picture whose name rhymes with the name of the puppet. For example, the child would be shown "Dan", told that he "liked things that sounded like his name", and asked to find what he would like from an array of house, boat, car, and van. Two practice trials with corrective feedback were followed by nine test trials. The same procedure was used in the phoneme-matching test, except that the task was to pick the picture whose name "began with the same sound" as the puppet's name. In addition, phoneme identity skills were assessed in the Alliteration with Pictures subtest of the Phonological Assessment Battery (PhAB; Frederickson, Frith, & Reason, 1997). This test was included because it is a frequently used test of phonological awareness skills in the UK. The supplementary Alliteration with Pictures was used instead of the Alliteration test because K.S. was unable to comply with the procedure of the latter task, probably due to its high memory load. In the Alliteration with Pictures subtest the examiner says three words, while the child is presented simultaneously with three pictorial representations of these words. The child is asked to say the words or point to the pictures of the words that start with the same sound. There are 10 trials in total: 5 with single onsets and 5 with cluster onsets. Each section is preceded by 2 practice trials on which corrective feedback is given.

Phoneme manipulation skills, and in particular phoneme segmentation, were assessed in the Word Completion (syllables and phonemes) subtest of the Phonological Abilities Test

(PAT; Muter, Hulme, & Snowling, 1997). In this test the examiner supplies the first "part" of the word which the child "finishes off". There are 16 trials in total: 8 that require the child to supply the last syllable of a two-syllable word and 8 that require the child to supply the final phoneme of a one-syllable word. Each section is preceded by 2 practice trials on which corrective feedback is given, and all the trials are accompanied by pictures. In addition, the Phoneme Deletion (beginning and end sounds) subtest of the PAT was administered. In this test the child has to remove a phoneme of a one-syllable word. There are 16 trials in total, 8 in which the initial phoneme has to be removed. In these trials the correct response is also a word (e.g., meat-eat). In the remaining 8 trials, the child has to remove the final phoneme. The resulting correct response is not usually a word (e.g., bag-ba). Each section is preceded by 4 practice trials on which corrective feedback is given, and all the trials are accompanied by pictures.

The RAN task administered consisted of the naming speed sections (Picture Naming Speed and Digit Naming Speed) from the PhAB (Frederickson et al., 1997). In these tests the child is asked to name a series of pictures or digits as fast as possible. Each test consists of two trials and is preceded by a practice trial. Phonological production skills were further assessed by measuring speech rate using the Speech Rate subtest of the PAT (Muter et al., 1997). This subtest requires the child to repeat the word "buttercup" 10 times, as quickly as possible while the examiner records the time taken. The three test trials are preceded by a practice trial where the child is asked to repeat his or her name 10 times, as quickly as possible. The raw score reflects speech rate in words per second.

Results

Can K.S. read nonwords that have low or no similarity to words?

On the GNWRT, K.S. read 17 out of 20 nonwords correctly, falling in the 75th percentile for typically developing children of her age.

On the Griffiths and Snowling list (Griffiths & Snowling, 2002) as a whole, K.S.'s performance is not significantly different from that of the typically developing 8-year-olds, t(14) =0.409, p = .689. The estimated percentage of the control population falling below K.S.'s score is 65.55%, and the 95% CI on this percentage is from 45.47% to 82.76%. In addition, we looked at K.S.'s performance profile on the three subtests: items containing high-frequency rimes, items containing low-frequency rimes, and items that had no close orthographic neighbours. As illustrated in Table 5, no significant differences were found between K.S. and the control group on the items containing high, t(14) = -0.503, p = .623, or low, t(14) =-0.527, p = .607, frequency rimes. On the other hand, there was a strong trend for her performance on items that had no close orthographic neighbours to be significantly better than that of the control group, t(14) = 2.108, p = .054. To quantify the abnormality of the difference between K.S.'s ability to read nonwords with and without close orthographic neighbours we averaged the scores on the nonwords with high- and low-frequency rimes and compared this combined score (M = 5.80, SD = 1.28, for the control group) to the performance on the nonwords without close

Table 5. Performance of K.S. and the typically developing controls and K.S. on the list of nonwords that varied in the frequency of their rime unit

	<i>Controls</i> ^a	K.S.	Percent. below ^b
Overall	14.40 (3.79)	16	65.55 (45.47-82.76)
High-frequency rime	6.67 (1.29)	6	31.14 (14.64–51.17)
Low-frequency rime	4.93 (1.71)	4	30.34 (14.02-50.34)
No close orthographic neighbours	2.80 (1.47)	6	97.32 (88.90–99.91)

Note: Percent. below = estimated percentage of control population below K.S.'s score.

^an = 15; mean values, with standard deviations in parentheses.
^bMean values, with 95% confidence interval (CI) in parentheses.

orthographic neighbours by using the revised standardized differences test. K.S.'s z-scores for her performance on the nonwords with and without close orthographic neighbours were -0.625 and 2.177, respectively. The difference between K.S.'s performance on the nonwords with close orthographic neighbours and the performance on nonwords without orthographic neighbours was significantly bigger than that observed in the control group, t(14) = 3.549, p = .003. It is estimated that only 0.16% of the control population would exhibit a difference in favour of nonwords without close orthographic neighbours that is more extreme than that observed in K.S.

Does K.S. have good phonological skills?

On both the rime-matching and the phonemematching tests, K.S. performed well above chance with 89% and 100% correct, respectively. the other phoneme identity On test-Alliteration with Pictures subtest of the PhAB-K.S. achieved a standard score of 84. Initially, this appears to contradict the result of the phoneme-matching test. However, it should be noted that K.S. scored 100% correct on the first part of the test (involving single onsets) and 60% correct on the second part (involving cluster onsets). It is reported in the PhAB manual (Frederickson et al., 1997) that children in the normative sample typically reached ceiling on this test by the age of 7. They therefore recommend using this supplementary test only for children who score less than 3 on the Alliteration test (without pictures). The mean performance on the Alliteration with Pictures test of 8-year-old children in the normative sample who fall into that category (n = 2) is 7.7, which is similar to K.S.'s raw score of 8.

With regard to K.S.'s phoneme segmentation skills, she performed at ceiling (100% correct) on both sections of the Word Completion subtest of the PAT. She also performed at ceiling on both sections of the Phoneme Deletion test of the PAT (100% correct on initial phoneme deletion, 87.5% correct on final phoneme deletion). The PAT does not have norms for 8-year-olds, but from about age 7, children seem to perform at ceiling level.

K.S.'s standard scores on the Picture Naming Speed and Digit Naming Speed subtests of the PhAB were 97 and 96, respectively, indicating age-appropriate performance. K.S.'s speech rate, as measured by the speech rate subtest of the PAT, is 1.16 words per second. With this score, her performance falls between the 25th and 50th percentiles for typically developing children aged 7;6–7;11 (the oldest age group for which norms are provided). A score of 1.2 words per second is average (50th percentile) for typically developing children aged 6;6–7;5, which is ahead of her mental age as indicated by her score on the receptive vocabulary measure (ROWPVT ageequivalent score = 6;0).

Discussion

K.S.'s ability to read nonwords to a high standard, as indicated by her achievement on the GNWRT and her age-appropriate performance on the Griffiths and Snowling list as a whole, indicates that she is not "just" using a whole-word strategy to read, but uses knowledge of graphemephoneme correspondences as well. In contrast to the finding reported by Gombert (2002) that the individuals with DS in her sample relied more heavily on analogies between nonwords and words in reading the nonwords, this was not found in K.S. Instead, there is a strong trend for her to outperform typically developing children of her age in reading nonwords that have no close orthographic neighbours. The discrepancy observed between her ability to read nonwords with and without orthographic neighbours suggests that she might rely on the use of grapheme-phoneme correspondences more heavily than would the control group. Possibly, this is the result of the intensive practice in this reading strategy provided by her parents. It is worth noting that this superior performance in reading nonwords without orthographic neighbours occurred in the face of very poor nonverbal cognitive ability (3rd percentile on the WASI Matrix Reasoning subtest).

In her performance on the rime analysis, phoneme identity, phoneme segmentation, and phoneme deletion tasks K.S. displays a robust level of phonological awareness skills. Her high performance on the rime-matching task is in sharp contrast with the reports in the literature of a deficit in rime analysis skills in individuals with DS. Although none of the studies looking at rime analysis skills in individuals with DS used the task used here, rime-matching tasks very similar to it were used by Cardoso-Martins et al. (2002), Gombert (2002), and Snowling et al. (2002). These rime-matching tasks generally involved matching of a target word with one out of three test words. In all these tasks, the verbal presentation of the words was accompanied by pictures. Therefore, it seems unlikely that K.S.'s high performance on the rime-matching task can be explained by differences in task design. It should also be noted that a few children with DS did perform above chance in the rime analysis tasks in other studies, varying from 1 out of 29 to 5 out of 30 children with DS in studies by Snowling et al. (2002), for instance.

Also, the speed with which K.S. can activate phonological representations of words in longterm memory is more or less age appropriate as both her performance on the RAN task and her speech rate indicate.

These results confirm earlier reports that readers with DS can show measurable levels of phonological awareness (Cardoso-Martins & Frith, 2001; Cupples & Iacono, 2000; Fletcher & Buckley, 2002; Fowler et al., 1995; Gombert, 2002; Kay-Raining Bird et al., 2000; Kennedy & Flynn, 2003a; Laws & Gunn, 2002; Snowling et al., 2002) and add that the phonological pathway can be highly proficient in a child with DS.

STUDY 3: READING COMPREHENSION

Although K.S. is highly proficient in word recognition and decoding, the main goal of reading is extracting meaning from print. Abilities associated with successful reading comprehension include broader language skills (Nation & Snowling, 2004) and working memory (Carretti, Cornoldi, De Beni, & Romano, 2005; Nation, Adams, Bowyer-Crane, & Snowling, 1999; Oakhill, Cain, & Bryant, 2003). Specifically, deficits in vocabulary (Nation & Snowling, 2004), grammar comprehension (Nation, Clarke, Marshall, & Durand, 2004), and semantic knowledge (Nation & Snowling, 1998, 1999) have been reported in individuals with poor reading comprehension. Oakhill and colleagues have focused on explanations for poor comprehension at the text level. They found deficits in the ability to derive structure from text and to integrate information in a text, and in inferential skills in children with poor reading comprehension. In addition, children with deficits in reading comprehension were worse at metacognitive skills such as monitoring their own comprehension (e.g., Oakhill, 1994, for a review). Comprehension problems are not restricted to the written domain as several studies have reported deficits in listening comprehension in these children (Nation & Snowling, 2004; Oakhill, 1982, 1983), even when memory load is controlled.

Few studies have investigated reading comprehension in individuals with DS. However, from the studies that do report reading comprehension levels (see Table 6), one can conclude that it almost always lags behind word recognition levels (A. Byrne et al., 1995; A. Byrne, MacDonald, & Buckley, 2002; Fowler et al., 1995; Laws & Gunn, 2002; Moni & Jobling, 2001). Also, in longitudinal studies progress in word recognition appears greater than progress in reading comprehension (A. Byrne et al., 2002; Moni & Jobling, 2001). In terms of the abilities underlying reading comprehension, working memory and grammar are two areas of particular difficulty for individuals with DS (e.g., Chapman, 1997; Jarrold, Baddeley, & Phillips, 2002). In contrast, semantic skills in the form of vocabulary knowledge have often been reported as an area of relative strength (e.g., Chapman, 1997). Given the reports of the predictive value of oral language skill in reading comprehension

	Test		Mean age ^a			
Reference	Word recognition	Comprehension	N	СА	<i>RAacc</i>	RAcomp
K.S.	NARA-II	NARA-II		102	95	82
Pieterse and Treloar (1981) ^b	NARA	NARA	8	95	91	81
A. Byrne et al. (1995)	K-ABC	K-ABC	24	98	75	72
A. Byrne et al. (2002)	K-ABC	WORD	24	109	78	72
A. Byrne et al. (2002)	K-ABC	WORD	24	121	80	72
Laws and Gunn (2002)	K-ABC	K-ABC	30	198	81	81
Moni and Jobling (2001)	NARA	NARA	9	249	114	108

Table 6. Chronological and reading age of K.S. and individuals with Down syndrome in studies of reading comprehension ability that used standardized tests

Note: CA = chronological age. RAacc = reading age for accuracy, RAcomp = reading age for comprehension, NARA = Neale Analysis of Reading Ability. NARA-II = Neale Analysis of Reading Ability-2nd Edition. K-ABC = Kaufmann Assessment Battery for Children. WORD = Wechsler Objective Reading Dimensions.

^aIn months. ^bCited in Buckley (1985).

in children without DS and the reduced language abilities in individuals with DS, deficits in reading comprehension are not unexpected in this population. It seems likely that reading comprehension might function at the level of their oral language skills.

In this section, we look at K.S.'s ability to comprehend what she has read. After having established her level of reading comprehension, we compare it to the performance of a group of poor comprehenders without DS. We also ask whether her reading comprehension functions at the same level as her oral language skills as has been found in previous studies of poor comprehenders who do not have DS.

Participants and methods

K.S.'s performance is compared with the performance of a group of typically developing children and a group of poor comprehenders (Table 7). K.S. is matched on decoding ability as assessed on the GNWRT (see Study 2) with both the average readers, t(16) = -0.399, p = .695, and the poor comprehenders, t(16) = -0.135, p =.894. The groups also did not differ from K.S. in chronological age, t(16) = 0.000, p = 1.000; t(16) = 0.146, p = .886. Nation and Snowling have reported on these groups of children in earlier publications (Nation & Snowling, 1997, 1998).

Because of the problems in reading comprehension and skills underlying it in individuals with DS reported in the literature, we expect K.S. to do worse on the reading comprehension tests than the typically developing children with average reading comprehension ability. We therefore performed a one-tailed test for this particular comparison and the comparison of K.S. with the average comprehenders on the discrepancy scores

Table 7. Raw scores for K.S., the average readers, and the poor comprehenders, and K.S. on decoding, reading accuracy, reading comprehension, and listening comprehension

	Reading comprehension		
	Average ^a	Poor ^b	K.S.
Age ^c	102 (7.71)	100.9 (7.05)	102
GNWRT	17.65 (1.58)	17.29 (2.08)	17
Reading accuracy	66.35 (16.57)	52.00 (11.73)	40
Reading comprehension	22.88 (4.29)	11.75 (2.14)	9
Listening comprehension	9.08 (2.36)	3.63 (2.13)	2

Note: GNWRT = Graded Nonword Reading Test. Mean values are given, with standard deviations in parentheses. ^an = 17 (9 girls). ^bn = 17 (12 girls). ^cIn months. between reading accuracy and comprehension on the Neale Analysis of Reading Ability-Revised 2nd edition (NARA-II). As we had no a priori reason to expect such a difference between K.S. and the group of poor comprehenders, we used two-tailed tests in these comparisons. In addition, K.S.'s oral language skills were assessed using a large battery of standardized tests, including verbal cognitive ability, vocabulary, semantic skills, and grammar. K.S.'s standard score on the reading comprehension test is compared with standard scores on these language tests.

Word recognition at text level

Word recognition at text level was assessed using the NARA-II (accuracy score; Neale, 1997), in which the child reads aloud a series of short, graded passages. The examiner corrects any words that the child reads incorrectly or has difficulty reading, so that comprehension of the text is maintained.

Reading comprehension

Reading comprehension was assessed using the NARA-II (Comprehension score; Neale, 1997, Form I) and the Reading Comprehension subtest of the Wechsler Objective Reading Dimensions (WORD; Wechsler, 1993). As the NARA-II, the WORD is also composed of a series of graded printed passages. After the child has read the passage, he or she is asked one (in the WORD) or several (in the NARA-II) comprehension questions about the passage. The child may refer back to the text to answer the questions. Passages in the WORD start at a simpler level than in the NARA-II, and only the first eight passages are accompanied by illustrations, while all the passages in the NARA-II are illustrated. The most important difference between the two tests is that in order to be able to answer the comprehension questions on the NARA-II the participant has to make knowledge-based inferences, while the WORD is more biased towards the retention of literal information (Bowyer-Crane & Snowling, 2005).

Listening comprehension

Listening comprehension was included as a measure of oral language comprehension and was assessed by using the Form 2 of the NARA-II, following Nation and Snowling (1997). Instead of the child reading the story, the examiner read the story, while the child could see the picture.

Verbal cognitive ability, vocabulary, and semantic skills

Verbal cognitive ability was assessed using the verbal subtests of the WASI (Wechsler, 1999). In the Vocabulary subtest of the WASI, the child is asked to define a series of words. It is a measure of the individual's expressive vocabulary, verbal knowledge, and fund of information. For details on the Similarities subtest of the WASI, see Study 1.

Vocabulary was assessed using the Expressive and Receptive One-Word Picture Vocabulary Tests (EOWPVT, ROWPVT; Brownell, 2000a, 2000b). See Study 1 for details.

Semantic skills were assessed using the Word Opposites (Level 1) and Synonyms (Level 1) subtests of the Test of Word Knowledge (TOWK; Wiig & Secord, 1992). The Word Opposites and Synonyms subtests of the TOWK are two receptive measures of semantic knowledge. In the Word Opposites task the child is asked to choose the word with a meaning opposite to that of the target word from three choices. In the Synonyms task the child is asked to select the synonym of the target word from three choices. In both tasks, the words are presented orally as well as in print, and each task includes two practice items.

Grammar

Receptive grammar skills were assessed using the TROG-2 (Bishop, 2003b), and two measures of expressive grammar were used: MLUw as derived from the narrative sample as part of the administration of the ERRNI (Bishop, 2004), and the Word Structure subtest of the Clinical Evaluation of Language Fundamentals–Third Edition UK (CELF-3^{UK}; Semel, Wiig, & Secord, 2000). For details on the TROG-2 and

the ERRNI, see Study 1. In the Word Structure subtest of the CELF-3^{UK}, a range of morphological constructions (e.g., possessives, plurals, tense markers) are elicited, supported by pictures. The child is asked to finish sentences such as "Here is one bus and here are two ...?" (correct answer: buses). Each class of items is preceded by a demonstration, and all items are administered irrespective of the achievement of the child.

Results

Is K.S.'s reading comprehension ability at a level similar to her word recognition and decoding abilities?

Table 7 summarizes the raw scores for K.S., the group of average readers, and the group of poor comprehenders on decoding, reading accuracy and reading comprehension. K.S. achieved standard scores of 94 and 83 for accuracy and comprehension, respectively, on the NARA-II. Although K.S.'s decoding (i.e., nonword reading) ability is at a similar level as that of the average readers, K.S.'s discrepancy between performance on reading accuracy and comprehension is significantly greater than that observed in this group, t(16) =1.839, one-tailed p = .042. Only an estimated 4.23 % of the population of average readers would exhibit a difference between accuracy and comprehension score more extreme than that of K.S. The discrepancy between reading accuracy and comprehension observed in the group of poor comprehenders is not significantly different from the one observed in K.S., t(16) = 0.101, p = .460. On the WORD, K.S. reached a standard score of 94 for comprehension.

Does K.S.'s reading comprehension ability parallel her oral language ability?

K.S.'s performance on the listening comprehension task (Table 7) differed significantly from the performance of the group of average readers, t(16) = -2.915, p = .010, but not from the performance of the group of poor comprehenders, t(16) = -0.744, p = .468.

As illustrated in Table 8, K.S.'s scores on the majority of the standardized language tests are in

the same range as her reading comprehension score on the NARA-II. Specifically, her scores on the verbal cognitive ability, receptive vocabulary, and receptive semantic knowledge measures all fall around the 10th percentile, as does her reading comprehension score on the NARA-II. She scores slightly lower on the expressive vocabulary measure, the receptive grammar measure, and one of the expressive grammar measures (Word Structure). Her MLUw on the other hand is age appropriate. Although the results for the two expressive grammar tests seem contradictory at first sight, this merely reflects the different aspects of grammar ability assessed by the tests. Whereas MLUw indicates the complexity of a child's expressive language, it does not pick up grammatical morphology errors that are

Table 8. K.S.'s performance on the reading and language tests

Test	Percentile rank
Word recognition	
NARA-II Accuracy	34 (14-60)
Reading comprehension	
NARA-II Comprehension	13 (3-37)
WORD Comprehension	34 (16-58)
Verbal cognitive ability	
WASI Verbal IQ	16 (8-32)
Vocabulary	
EOWPVT	4 (1-10)
ROWPVT	11 (4-23)
Semantics	
TOWK Word Opposites	9 (2-25)
TOWK Synonyms	9 (5-16)
Grammar	
ERRNI MLUw	55
CELF-3 ^{UK} Word Structure	1 (1-22)
TROG-2	<1

Note: Percentile ranks are given, with 95% confidence interval in parentheses. NARA-II = Neale Analysis of Reading Ability-2nd Edition. WORD = Wechsler Objective Reading Dimensions. WASI = Wechsler Abbreviated Intelligence Scale. EOWPVT = Expressive One-Word Picture Vocabulary Test. ROWPVT = Receptive One-Word Picture Vocabulary Test. TOWK = Test of Word Knowledge. ERRNI MLUw = Expression Reception and Recall Narrative Instrument, mean length of utterance in words. CELF-3^{UK} = Clinical Evaluation of Language Fundamentals-3rd Edition UK. TROG-2 = Test for Reception of Grammar-Version 2. pinpointed in the Word Structure test of the CELF-3^{UK}. As mentioned in Study 1, although K.S.'s MLUw is relatively long, about 20% of her utterances contained grammatical errors (mainly subject omissions and subject–verb agreement errors). Such errors are rarely seen in typically developing children above the age of 5, which is reflected in her low score on the Word Structure test of the CELF-3^{UK}.

Discussion

K.S.'s reading comprehension ability is not at the same level as her word recognition ability. With a standard score of 83 for comprehension on the NARA-II, K.S.'s performance falls just outside one standard deviation of the mean for typically developing children of her age. The discrepancy between her word recognition skills at text level and her reading comprehension, as assessed in the NARA-II, is very similar to the discrepancy observed in the group of children with reading comprehension problems who do not have DS. It is also similar to the discrepancy reported by Pieterse and Treloar (1981, cited in Buckley, 1985) for children with DS of similar chronological age to K.S., with similar word recognition abilities as K.S. and using an earlier version of the NARA-II. They describe a mean difference in reading ages of 10 months between reading accuracy and comprehension; for K.S. this is 13 months.

To shed some light on the nature of her comprehension difficulties, we administered the WORD Reading Comprehension test in addition to the NARA-II. Bowyer-Crane and Snowling (2005) found that the type of inferences that the child needs to make in order to answer the comprehension questions on the NARA-II and the WORD differs. While the WORD is more biased towards the retention of literal information, the NARA-II requires more knowledge-based inferences and inferences that rely on linguistic cues. K.S.'s standard score of 94 for comprehension on the WORD falls within the age-appropriate range. In their study, Bowyer-Crane and Snowling (2005) found a mean difference of 10.7 points between the standard scores on the NARA-II and the WORD in "less skilled" comprehenders. This is very similar to K.S.'s difference of 11 points. It suggests that K.S. has difficulties in making use of real-word knowledge to generate inferences during reading.

Previous research has found that oral language skills are good predictors of reading comprehension (Nation & Snowling, 2004). We compared K.S.'s performance on one oral language task (the listening comprehension task) with the performance of children who do not have Down syndrome. All had the same standard of decoding skills, but one group had average reading comprehension skills, while the second group was poor at reading comprehension. In parallel to the poor comprehenders, K.S. performed significantly worse on the listening comprehension task than did the group with average reading comprehension skills. This suggests that reading comprehension difficulties in children with and without Down syndrome can be associated with similar difficulties in linguistic comprehension. However, the listening comprehension task inevitably placed high demands on verbal shortterm memory. Given the known deficits in this area in individuals with Down syndrome (e.g., Jarrold et al., 2002), this might well have contributed to K.S.'s poorer performance on the listening comprehension task. We looked at K.S.'s oral language skills more closely by administering a large battery of standardized language tests. K.S.'s profile of standard scores on this test battery indicates that her reading comprehension ability beyond the retention of literal information as assessed by the NARA-II functions at a level similar to her oral language skills and her verbal cognitive ability. All scores were in the range of her reading comprehension score, except expressive and receptive grammar and expressive vocabulary on which she scored lower. This is in agreement with the general findings on language abilities in individuals with DS that grammar skills in general (and specifically grammatical morphology) and expressive vocabulary are particular weaknesses (e.g., Chapman, 1997).

In conclusion, her reading comprehension is age appropriate when it comes to the retention of literal information. It therefore seems sufficient for reading to be a useful tool in her everyday activities. In addition to the enjoyment of simple books, she can also use lists to remind her of things to do or to buy, and she can read signs, recipes, menus, and so on. When it comes to reading comprehension beyond the retention of literal information, K.S. has some difficulties. This implies that she would benefit from support in the use of real-world knowledge to generate inferences during reading.

GENERAL DISCUSSION

K.S. has exceptional word recognition and decoding ability for a child with DS of her age, and she is particularly proficient in decoding. There is some evidence that K.S.'s word recognition and decoding abilities are associated with strengths in visual and verbal short-term memory and articulation and speech fluency. Although K.S. only scored significantly higher on the Digit Span task, her scores on other tests in these domains did fall in the top 10-20%. Her high performance on verbal short-term memory tasks is in agreement with the literature; however, the strength in articulation and speech fluency has not been reported before. A fuller understanding of the individual differences underlying reading success in children with DS would be beneficial for developing reading instruction programmes, and we hope that these results can be used as pointers to guide future research in this direction.

K.S.'s exceptional word recognition and decoding skills are accompanied by robust phonological awareness skills, which confirms reports that children with DS can show measurable levels of phonological awareness and do not rely solely on sight-word strategies as thought initially. In contrast, K.S.'s nonword reading abilities suggest that she might rely more heavily on the use of grapheme-phoneme correspondences than do typically developing children her age. This shows that the phonological pathway can be highly proficient in a child with DS and emphasizes the value of conducting a full assessment of each individual's strengths and weaknesses to identify the most appropriate reading instruction strategies.

A key aspect of her good performance on the phonological awareness tasks seems to be the modification of the tasks, minimizing memory load. This implies that it is of key importance to carefully evaluate the memory demands of phonological awareness tasks and adjust them appropriately to prevent poor memory capacity obscuring phonological awareness skills in children with DS.

With regard to her reading comprehension skills, they are of a sufficient standard for reading to be a useful tool in her everyday activities, but when it comes to reading comprehension beyond the retention of literal information, K.S. has some difficulties. This result also highlights that reading comprehension performance can be dramatically influenced by the structure of the task. K.S.'s deficits in the area of comprehension using knowledge-based inferences parallel those found in children with poor reading comprehension who do not have Down syndrome. Therefore, it follows that similar strategies used to improve their understanding should be explored in an attempt to improve K.S.'s reading comprehension ability. This includes stimulating the use of background knowledge, training inferential skills and question generation.

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