



Speech timing and verbal short-term memory: Evidence for contrasting deficits in Down syndrome and Williams syndrome[☆]

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Abstract

This study explored the degree of verbal short-term memory deficit among individuals with Down syndrome and Williams syndrome, and the extent to which any such impairment could be accounted for by a relative slowing of rehearsal and output processes. Measures of serial recall and detailed assessments of speeded articulation for short and long words were assessed among these two populations, and controls. Both clinical groups showed an impairment in serial recall. Among individuals with Williams syndrome this deficit could be explained in terms of a general slowing in speech rate. However, although aspects of speeded articulation were delayed among individuals with Down syndrome, this could not account for the extent of impairment in their verbal short-term memory performance. The implications of these findings for the source of impaired verbal short-term memory associated with Down syndrome, and for the word length effect at different levels of development, are discussed.

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Verbal short-term memory performance, which is typically assessed by measuring the maximum number (or span) of verbal items that an individual can repeat in correct serial order, is often impaired in individuals suffering from language delay (see Hulme & Roodenrys, 1995). For example, individuals with specific language impairment and individuals with Down syndrome perform poorly on tests of verbal short-term memory (see Jarrold, Baddeley, & Phillips, 1999; Montgomery, 2003), and suffer from a relative delay in language development in comparison to non-verbal skills (e.g., Bishop, 1979; Fowler, Gelman, & Gleitman, 1994).

One explanation for this co-occurrence of deficits is that the ability to maintain verbal information in correct serial order underpins aspects of individuals' language development. Baddeley and colleagues (e.g., Baddeley, Gathercole, & Papagno, 1998) argue that vocabulary development in particular is supported by a specific verbal short-term memory system—the phonological loop—that has evolved for this purpose. They suggest that in order to create a stable, long-term representation of the phonological form of a novel word, one must represent its spoken form accurately and efficiently in short-term memory. If this is the case, then aspects of language delay associated with these developmental disorders would follow as a direct result of verbal short-term memory difficulties.

An alternative suggestion is that poor verbal short-term memory performance among such populations is simply a consequence of individuals' relatively impaired language skills (Hulme & Roodenrys, 1995; Snowling, Chiat, & Hulme, 1991; see Jarrold, 2001). There is little doubt that individuals' knowledge of the language does play a part in mediating verbal short-term memory performance. For example, in the general population memory spans are consistently higher for words than for nonwords (e.g., Hulme, Maughan, & Brown, 1991) due to benefits to recall conferred to words as a result of their familiarity. Given this, individuals with relatively poor knowledge of the language may show less benefit from this process, with a consequent reduction in their level of short-term memory performance.

One way of determining whether an observed deficit in verbal short-term memory performance truly reflects a fundamental impairment to a specific short-memory subsystem is to compare the verbal serial recall performance of groups of individuals who are equated for vocabulary knowledge. In light of the theoretical arguments for an association between language knowledge and verbal short-memory performance, it is hardly surprising that individuals' levels of receptive vocabulary have been consistently shown to relate to their scores on measures of verbal short-term memory (see Baddeley et al., 1998). Indeed, studies that have compared the strength of correlations across other indices of intellectual function have shown that it is vocabulary knowledge per se, and not general intelligence or non-verbal ability, that relates specifically to verbal short-term memory performance (Baddeley et al., 1998). Given this, if poor performance is simply a consequence of language impairment, then under conditions when vocabulary knowledge is equated, then recall levels should also be equated (see Jarrold, Baddeley, Hewes, Leeke, & Phillips, 2004, Study 1). In contrast, if a group is associated with a fundamental deficit of verbal short-term memory then this should still be apparent relative to controls of an equivalent level of vocabulary (see Jarrold et al., 2004, Study 2).

However, even when groups are equated for level of language knowledge, it is still possible that factors other than pure verbal short-term memory capacity might lead to group differences in immediate verbal serial recall. In particular, there is evidence to suggest that verbal short-term memory performance depends on the time taken by participants to articulate the items to be remembered in any task. For example, individuals show better immediate serial recall of words with fewer syllables, and consequently relatively short spoken duration, than of words with more syllables and longer spoken duration (e.g., Baddeley, Thomson, & Buchanan, 1975). Although the extent to which this effect is based on the difference in spoken duration, as opposed to complexity, of long and short words has been called into question (e.g., Caplan, Rochon, & Waters, 1992; Lovatt, Avons, & Masterson, 2000; Service, 1998), it does appear that the spoken duration of to-be-remembered stimuli constrains recall independently of item complexity (Cowan, Nugent, Elliott, & Geer, 2000).

This 'word length effect' for stimulus sets is mirrored at the level of individual differences in verbal short-term memory performance. Many studies have demonstrated strong correlations between the rate at which individuals can articulate items and the accuracy with which they can recall these items in a verbal short-term memory test (e.g., Baddeley et al., 1975; Cowan et al., 1998; Gathercole, Adams, & Hitch, 1994; Standing & Curtis, 1989). In addition, developmental studies have suggested that age-related variance in children's verbal short-term memory performance is linked to age-related change in speech rate (Cohen & Heath, 1990; Henry, 1994; Hulme, Thomson, Muir, & Lawrence, 1984; Nicolson, 1981), although this may not be the sole source of developmental variation in performance (Henry & Miller, 1991; Hitch, Halliday, & Littler, 1993).

Baddeley's (1986) model accounts for these data by suggesting that the efficiency of the phonological loop depends on the interaction between an individual's capacity to store phonological information, and the rate at which decay of this information can be offset by a process of subvocal maintenance rehearsal. Individuals' overt speech rates are assumed to provide an index of their covert articulation rates, and hence greater trace decay occurs within the loop for words of a relatively longer spoken duration and among individuals with relatively slower articulation rates. A somewhat different, though related, explanation of the association between speech rate and span was put forward by Cowan et al. (1992). Cowan and colleagues argued that longer words might suffer from relatively greater trace decay during the process of outputting items in response to a serial recall task. They showed that greater forgetting of information from short-term memory occurred for word lists that began with items of a relatively long spoken duration than for lists of the same words that were

presented so that recall began with items of a shorter spoken duration. These data suggest that the time taken to output items at recall affects the ease with which subsequent items can be remembered, and imply that individual differences in output time might mediate the relationship between speech rate and span.

Although these two accounts differ in where they place the precise source of forgetting in verbal short-term memory, they have in common the assumption that verbal short-term memory performance is not simply constrained by phonological storage capacity, but also depends on the speed with which individuals can articulate to-be-remembered items. Indeed, both Baddeley and Cowan have argued that both rehearsal and output effects might operate and contribute to phenomena such as the word length effect (Baddeley et al., 1998; Cowan et al., 1998). Consequently, there are at least two reasons why individuals with a developmental disorder might show impaired verbal short-term memory relative to controls of an equivalent level of vocabulary; members of a group may suffer from a fundamental deficit in phonological storage capacity, or they may have reduced rehearsal or articulation rates.

The purpose of the current study was to explore in detail the extent to which verbal short-term memory deficits among individuals with developmental disorders reflect these two possibilities. Individuals with two different developmental disorders—Down syndrome and Williams syndrome—were assessed in this study. Both conditions have a genetic basis, Down syndrome being caused by triplication of chromosome 21 (trisomy 21), and Williams syndrome occurring as a result of a micro-deletion of genes from the long arm of chromosome 7. While Down syndrome is the most common form of genetically determined learning disability, occurring in approximately 5 in 10,000 live births (Steele & Stratford, 1995), Williams syndrome is much rarer and has an estimated incidence of approximately 1 in 20,000 live births (Morris & Mervis, 1999). In addition, these two conditions are associated with quite different psychological profiles. Individuals with Down syndrome tend to suffer from a degree of generalised intellectual delay, although the extent of learning difficulties do vary considerably among individuals. Furthermore, and as noted above, there is reasonable evidence to suggest that the language skills of individuals with Down syndrome are relatively more impaired than their non-verbal abilities (e.g., Chapman, 1995; Fowler, 1990; Fowler et al., 1994). In contrast, while individuals with Williams syndrome suffer from marked difficulties in non-verbal areas, their verbal skills are a relative strength. Indeed, early accounts of the condition suggested that language abilities might be ‘spared’ in Williams syndrome (Bellugi, Sabo, & Vaid, 1988; Bellugi, Wang, & Jernigan, 1994). Although subsequent studies have shown that this is typically not the case (e.g., Karmiloff-Smith et al.,

1997), it is fair to say that language skills in Williams syndrome tend to be in advance of non-verbal abilities (Jarrold, Baddeley, & Hewes, 1998; Jarrold, Baddeley, Hewes, & Phillips, 2001).

These two conditions also appear to give rise to contrasting short-term memory skills that, perhaps unsurprisingly, match these different profiles of verbal and non-verbal abilities. Wang and Bellugi (1994) compared verbal and visuo-spatial short-term memory abilities in these two conditions by presenting individuals with both digit span and Corsi span tasks; the latter being an analogue to digit span in which participants respond by selecting a series of visuo-spatial locations in the order demonstrated to them by the experimenter (Corsi, cited in Milner, 1971). Wang and Bellugi found that individuals with Down syndrome showed significantly poorer digit recall, but significantly better Corsi performance than individuals with Williams syndrome (see also Jarrold, Baddeley, & Hewes, 1999). Other studies that have examined the verbal short-term memory performance in Down syndrome have confirmed that individuals with the condition perform significantly less well on digit and word span tasks than would be expected given their level of receptive vocabulary (e.g., Brock & Jarrold, in press-a; Jarrold & Baddeley, 1997; Jarrold, Baddeley, & Hewes, 2000; Jarrold, Baddeley, & Phillips, 2002; Laws, 2002; Marcell, Harvey, & Cothran, 1988; Marcell & Weeks, 1988).

The extent of any impairment, or relative sparing, of verbal short-term memory performance in Williams syndrome is less clear. Vicari and colleagues reported unimpaired verbal short-term memory in Williams syndrome (Vicari, Brizzolara, Carlesimo, Pezzini, & Volterra, 1996; Vicari, Carlesimo, Brizzolara, & Pezzini, 1996), but this was relative to controls matched for level of non-verbal rather than verbal ability (see also Crisco, Dobbs, & Mulhern, 1988). Grant et al. (1997) showed that the performance of individuals with Williams syndrome on a nonword repetition task—a possible index of verbal short-term memory capacity—was poorer than expected for their level of receptive vocabulary (see also Barisnikov, Van der Linden, & Poncelet, 1996; Grant, Karmiloff-Smith, Berthoud, & Christophe, 1996). However, Majerus, Barisnikov, Vuillemin, Poncelet, and Van der Linden (2003) found no evidence for impaired nonword repetition relative to vocabulary matched controls, albeit among a relatively small sample of four individuals with Williams syndrome. Studies employing digit or word span tasks have provided similarly inconsistent findings to date. Although there is a general tendency for individuals with Williams syndrome to perform less well than controls of an equivalent level of vocabulary, this deficit has been significant in some studies (Brock, 2002; Laing, Hulme, Grant, & Karmiloff-Smith, 2001) but not in others (Laing et al., in press; Robinson, Mervis, & Robinson, 2003).

One aim of the current study, therefore, was to verify the extent of any verbal short-term memory difficulties in Williams syndrome relative to vocabulary-matched controls. A further objective was to determine whether any observed deficit in verbal short-term memory performance in this group, as well as that expected among individuals with Down syndrome, are related to group differences in speech rates. Hulme and Mackenzie (1992) found that individuals with Down syndrome and with severe learning difficulties both showed impaired word spans and reduced speech rates relative to typically developing individuals of an equivalent vocabulary level. However, these two clinical groups did not show the typical relationship between mean speech rate and mean span for word sets of different lengths, and as a result, their verbal short-term memory deficit could not be readily explained in terms of a reduction in speech rate affecting rehearsal or output efficiency. Similar results have been reported in other studies that have examined this relationship at the level of individual differences, and which have failed to demonstrate a reliable relationship between speech rate and verbal short-term memory performance among individuals with Down syndrome (Jarrold et al., 2000; Seung & Chapman, 2000; Vicari, Marotta, & Carlesimo, 2004). To our knowledge, only one study has previously examined this relationship in Williams syndrome. Laing et al. (in press) found that their participants with Williams syndrome had slower speech rates than controls, suggesting that speech rates may be relatively impaired in the condition, but they did not report the extent to which articulation rates correlated with recall performance.

The current study explored this issue in more detail among individuals with Down syndrome and Williams syndrome by examining the relationship between short-term memory performance and two particular aspects of articulation rate. As noted above, Cowan and colleagues have argued that the rate at which individuals can output information in response to a verbal short-term memory task constrains performance, and accounts, in part at least, for the relationship between articulation rate and span. However, Cowan (1992) suggested that in addition to the rate at which individuals can output the to-be-remembered words in response to a short-term memory task, the rate at which individuals are able to 'reactivate' decaying memory traces in the pauses between word articulation might account for separate variance in recall performance. In support of this view, Cowan et al. (1994) conducted a detailed analysis of the duration of spoken words and of the pauses between words in children's output to a verbal short-term memory task. They found that while word length unsurprisingly affected the length of spoken words, it did not affect pause durations during output, which by contrast were sensitive to age differences (see also Cowan, 1999; Hulme, Newton, Cowan, Stuart, & Brown, 1999). More directly, Cowan et al. (1998) showed that a traditional

index of speech rate and a measure of the duration of individuals' pauses during output from a verbal short-term memory task accounted for separate variance in recall performance. Jarrold et al. (2000) extended this work by showing that measures of the time taken to articulate words and of the pauses between spoken words from within a speeded articulation rate test, though inter-correlated, predicted separate variance in children's immediate serial recall of words. These data support Cowan's suggestion that two potentially separable speech rates may underpin verbal short-term memory performance, with time taken to produce words providing an index of the extent of information loss due to time-based forgetting during rehearsal or output, and with length of pauses between words reflecting the efficiency with which individuals can reactivate these decaying traces.

Given this possible dissociation between different speech timing measures and their relation to verbal short-term memory performance, previously reported speech rate data from individuals with Down syndrome (Jarrold et al., 2000) were reanalysed for the purposes of this current study. The duration of both words and pauses within speeded articulation data from these individuals, and measures of their verbal short-term memory performance were compared to those seen among a newly recruited control group of typically developing individuals. In addition, a similar set of data were collected for this study from a sample of individuals with Williams syndrome, who were also compared to a separate group of typically developing individuals. Each typically developing control group was matched to their respective clinical group for level of receptive vocabulary. This single matching measure was employed for two reasons. First, the discrepancy between verbal and non-verbal abilities associated with Williams syndrome, and to a lesser extent also Down syndrome, makes it practically impossible to match a control group to these clinical populations for both verbal and non-verbal abilities (cf. Klein & Mervis, 1999). Indeed, given the variability in different aspects of language functioning in these two conditions (e.g., Bellugi, Lichtenberger, Jones, Lai, & St. George, 2000; Chapman, 1995; Karmiloff-Smith et al., 1997), it is likely that multiple matching groups would be needed to control for aspects of language in addition to receptive vocabulary. While it would, of course, be possible to include such groups, a second point is that the theoretical arguments and empirical data reviewed above highlight the fact that it is vocabulary knowledge, and not general level of intelligence, that is particularly related to verbal short-term memory performance, and is the crucial measure to control for in a study of this kind. The measure used to equate groups for receptive vocabulary was the long form of the British Picture Vocabulary Scale (BPVS, Dunn, Dunn, Whetton, & Pintilie, 1982), which has a published reliability of between .70 and .95 (median

.91) as assessed in groups of typically developing children aged between 3 and 17 years.

Method

Participants

Four groups of participants took part in this study, individuals with Down syndrome and individuals with the Williams syndrome phenotype of a similar average age, as well as two samples of typically developing children. The group of individuals with Down syndrome consisted of 14 children and teenagers, six male and eight female, aged between 9 years 8 months and 18 years 8 months. All individuals had confirmed trisomy 21 without mosaicism, and were recruited from local schools for children with special educational needs, or in the case of individuals in mainstream education via the local Down Syndrome Association. Speech rate and short-term memory data from this group of individuals were previously reported in Jarrold et al. (2000, Experiment 1), although further analysis of these data were conducted for the purposes of the current study.

The group of individuals with the Williams syndrome phenotype consisted of 16 children, teenagers and young adults, aged between 6 years 5 months and 28 years 0 months; six of the group were male and nine female. All individuals were recruited via the United Kingdom Williams Syndrome Foundation, and had received a formal clinical diagnosis of Williams syndrome (eight individuals) or Infantile Hypercalcaemia (seven individuals). The latter diagnostic label is a term that was commonly applied to the condition within the UK until recently, and was consequently more common among the older members of the sample. Four individuals in this group had received a 'fluorescence in situ hybridisation' (FISH) test to assess deletion of the elastin gene on chromosome 7. Elastin is deleted in over 90% of individuals with Williams syndrome (Lowery et al., 1995; Nickerson, Greenberg, Keating, McCaskill, & Schaffer, 1995), and hence a positive test provides relatively strong confirmation of a diagnosis. All four tests were positive confirming elastin deletion. No individual in the sample had received a

FISH test with a negative result, but the majority of individuals had been diagnosed prior to the development and common usage of the FISH test for elastin deletion, and had not subsequently received it.

In addition, 30 typically developing were selected to form two separate control groups. These individuals were recruited from local mainstream schools that were associated with average indicators of parental socio-economic status and level of student achievement. These individuals were matched, at the group level, to the two clinical groups for receptive vocabulary mental age as assessed by the long form of the BPVS. Controls for the 16 individuals with Williams syndrome were taken from an existing data set from a previous study of speech timing and short-term memory measures in typically developing 8-year-olds (Jarrold, Hewes, & Baddeley, 2000; Experiment 1). These individuals were aged 8 years 1 month to 8 years 10 months, and there were 10 boys and 6 girls in this group. These particular individuals were selected to ensure that the mean vocabulary mental age of this subsample matched that of the group of individuals with Williams syndrome; and no consideration was given to these participants' performance on speech times or memory measures at this selection stage. A further 14 typically developing 4- and 5-year olds (age range 4 years 6 months to 5 years 0 months) were recruited for this study to provide controls for the individuals with Down syndrome. This group consisted of 5 boys and 9 girls. These individuals were selected from a larger sample of children who were screened for vocabulary mental age, prior to the administration of any other tasks, to ensure that the mean vocabulary mental age of this control group matched that of the group of individuals with Down syndrome.

Table 1 presents summary statistics for age and vocabulary mental age for each of these four groups. A series of one factor analyses of variance confirmed that the group of individuals with Williams syndrome was matched to their typically developing controls for verbal mental age, $F(1, 30) = 0.20, p = .66, MSE = 1246.57$, partial $\eta^2 < .01$, although the individuals with Williams syndrome were significantly older than these controls, $F(1, 30) = 15.08, p < .01, MSE = 3935.06$, and partial $\eta^2 = .33$. Similarly, the group of individuals with Down syndrome was

Table 1

Summary statistics for age and verbal mental age by group (TD-WS, typically developing controls for individuals with Williams syndrome; TD-DS, typically developing controls for individuals with Down syndrome)

Group	Chronological age (months)		Verbal mental age (months)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
WS	186.63	88.67	105.31	45.90
TD-WS	100.50	2.63	99.75	19.66
DS	166.43	37.69	54.93	13.38
TD-DS	56.86	2.03	56.93	9.47

matched to their typically developing controls for verbal mental age, $F(1,26)=0.21$, $p=.65$, $MSE=134.38$, and partial $\eta^2<.01$, but was again reliably older than these individuals, $F(1,26)=117.96$, $p<.01$, $MSE=712.43$, and partial $\eta^2=.82$. The individuals with Williams syndrome and individuals with Down syndrome did not differ significantly in chronological age, $F(1,28)=0.63$, $p=.44$, $MSE=4872.04$, and partial $\eta^2=.02$. However, the individuals with Williams syndrome had significantly higher vocabulary mental ages than the individuals with Down syndrome, $F(1,28)=15.64$, $p<.01$, $MSE=1211.80$, and partial $\eta^2=.36$, confirming the need to include separate control groups for each of these two clinical samples. Both sets of typically developing individuals had verbal mental ages that were consistent with their chronological ages, $F(1,15)=0.02$, $p=.88$, $MSE=198.83$, and partial $\eta^2<.01$ controls for individuals with Down syndrome, $F(1,13)<0.01$, $p=.98$, $MSE=43.19$, and partial $\eta^2<.01$ controls for individuals with Williams syndrome.

Procedure

All individuals received the British Picture Vocabulary Scale to determine their receptive vocabulary mental age in an initial testing session. In a subsequent assessment participants were given measures of spoken articulation time, and short-term memory. Both of these employed the same two sets of stimuli. These were a pool of nine 'short words' of relatively brief spoken duration (*bee, bone, cup, gate, lamp, nail, pig, shoe, and tent*) and a pool of nine 'long words' of longer spoken duration (*bananas, bicycle, caterpillar, elephant, grandmother, newspaper, radio, telephone, and policeman*). These sets were matched for approximate familiarity for children using Carrol, Davies, and Richman (1971) Grade 3 frequency counts, $t(16)=0.18$, $p=.86$, unpaired t test.

Verbal short-term memory was assessed in two conditions, one for each word set, that were counterbalanced for order of presentation across participants. These were presented via a Macintosh G3 laptop computer using pre-recorded stimuli presented in a male voice. The spoken duration of short words as presented in this task was between 586 and 845ms; the durations of long words were between 876 and 906ms. However, on trials when more than one word was presented, words were played at the rate of 1 per second to ensure that total presentation time for any given list length was comparable across conditions. Both conditions began with a familiarisation phase in which participants heard all the nine words in the appropriate set. Testing proper then began, and participants were instructed that they would hear lists of these words being spoken, and that their task was to repeat the words they had heard, in the correct order, once the computer had finished 'speaking.' A span procedure was employed, with four trials at

each successive list length, starting at list length one. All individuals received the same set of pre-determined lists, which ensured approximately equal distribution of all stimuli within each list length. On trials at list lengths greater than one, words were never repeated within a particular list. If participants were correct on three or four trials at any given list length, they moved on to four trials at the subsequent list length; correct recall required repetition of all items in their correct serial order. If participants were correct on only 2 or fewer trials at any length, that condition was ended once the four trials at that length had been presented. Rather than taking span as the dependent measure for short and long word recall, performance was scored in terms of the more sensitive measure (cf. Oberauer & Süß, 2000) of the total number of trials correctly recalled in each condition.

Articulation times for short and long spoken duration words were assessed in two conditions in which participants were requested to repeat pairs of words as rapidly as possible. The nine words in each stimulus set were divided into five word pairs, with one word included in two pairs (short word pairs: *bee-tent, cup-gate, lamp-bone, pig-tent, and shoe-nail*; long word pairs: *bananas-policeman, bicycle-telephone, caterpillar-radio, elephant-bananas, and grandmother-newspaper*). Participants were required to repeat each pair five times, and were practiced on word pairs not included in either stimulus set. The order of presentation of short and long word conditions was counterbalanced across participants. Responses were recorded and subsequently digitised for analysis using Sound Edit for the Macintosh. Two measures were derived from this analysis. Spoken word durations were calculated by first averaging the time taken to say the 10 words included in five repetitions of each word pair. Similarly, pause durations were estimated by averaging the nine silent intervals between words in the repetitions of each pair.

Results

Preliminary analyses

The reliability of the short-term memory and articulation time measures was assessed in the whole sample of 60 individuals, prior to further separate analyses of each clinical group and their associated controls. Split-half reliability was calculated for recall of both short and long words by comparing the number of trials correct calculated across the first two and the last two trials at each span length. Spearman–Brown reliability estimates for short word and long word recall trials were .86 in each case. However, it should be noted that these estimates may potentially be inflated by the lack of independence between the first two and last two trials at any list length, given that the task ended when participants were

correct on 2 or fewer trials at any length. Reliability of articulation time measures was determined separately for word and pause durations and for each word length. This was done by determining Cronbach's α for the five estimates of word or pause duration obtained from the repetition of the five different word pairs in each word length set. Cronbach's α values for word and pause durations from the five short word pairings were .88 and .85 respectively. The α values for word and pause durations from long word pairings were .52 and .83. Subsequent item analysis showed that the reliability of spoken durations of long words was reduced by the inclusion of data from the repetition of the *grandmother-newspaper* pairing. Removing data from this item increased α values to .71 and .84, for word and pause durations, respectively, and consequently all long word articulation data in all subsequent analyses were derived from averaging across the four remaining word pairs in this stimulus set.

Individuals with Williams syndrome and their controls

The performance of individuals with Williams syndrome and their associated controls on the short and long word conditions of the short-term memory and articulation tasks are shown in Table 2. The short-term memory data were subjected to a two factor analysis of covariance, with group and word length as independent and repeated measures, respectively, and with verbal mental age covaried from the group effect to account for any residual individual variation in this measure not controlled for by group matching (cf. Evans & Anastasio, 1968). This revealed a significant main effect of group, $F(1, 29) = 6.60$, $p = .02$, $MSE = 4.58$, partial $\eta^2 = .19$, reflecting poorer overall performance among individuals with Williams syndrome. The main effect of word length was significant, $F(1, 30) = 19.64$, $p < .01$, $MSE = 1.83$, and partial $\eta^2 = .40$, due to poorer recall of long as opposed to short words. The interaction between group and word length was non-significant, $F(1, 30) = 0.55$, $p = .47$, $MSE = 1.83$, and partial $\eta^2 = .02$.

The articulation time data summarised in Table 2 were subjected to a doubly multivariate analysis of variance, performed on word and pause indices, with group as an independent measure and condition (short or long word sets) as a multivariate repeated measure. This revealed a main effect of group (test of levels) that was close to significant, multivariate $F(2, 29) = 2.98$, $p = .07$, and partial $\eta^2 = .17$, due to somewhat slower articulation among individuals with Williams syndrome. The main effect of condition (deviation from flatness) was significant, multivariate $F(2, 29) = 320.90$, $p < .01$, and partial $\eta^2 = .96$. Further stepdown analysis confirmed a length effect for both pause duration, $F(1, 30) = 19.01$, $p < .01$, and $MSE = 2056.65$, and for word duration, $F(1, 28) = 633.95$, $p < .01$, and $MSE = 2487.05$. The condition effect for words reflected longer durations for long than short duration words, but the condition effect for pauses reflected shorter pauses for long than for short duration word sets. The interaction between group and condition (deviation from parallelism) was non-significant, multivariate $F(2, 29) = 0.53$, $p = .60$, and partial $\eta^2 < .01$.

The above analyses show that individuals with Williams syndrome have poorer verbal short-term memory than their controls given their level of receptive vocabulary. They also showed a tendency to take longer to articulate word pairs drawn from the same stimuli sets. To determine whether these two group effects were related, correlations between average word recall and average word and pause duration were examined for each group separately. Among individuals with Williams syndrome average word recall was reliably related to average word duration, $r = -.50$, $df = 15$, $p = .05$, and pause duration, $r = -.59$, $df = 15$, $p = .02$. Among controls the corresponding relationships between average word recall and both word duration, $r = -.38$, $df = 15$, $p = .15$, and pause duration, $r = -.42$, $df = 15$, and $p = .11$, were moderate in size but not significant. To determine whether an association between articulation time and recall might mediate the group difference in verbal short-term memory performance, analysis of covariance was conducted

Table 2
Summary statistics for short-term memory performance and articulation rates among individuals with Williams syndrome and associated controls

Measure	Word length	WS		TD-WS	
		<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Short-term memory score	Short	12.31	2.75	14.31	2.75
	Long	11.06	3.09	12.56	1.79
Spoken word duration (ms)	Short	304	35	253	69
	Long	629	98	556	98
Silent pause duration (ms)	Short	151	56	127	71
	Long	96	66	83	62

on average recall score with group as an independent measure and average word and pause durations as covariates. A preliminary analysis revealed no violation of the assumption of homogeneity of regression for either covariate ($p = .29$, mean word duration; $p = .21$, mean pause duration). A further regression analysis among individuals from both groups with recall score as the dependent measure and the two covariates as independent variables accounted for a reliable 39% of the variance in recall score, $F(2, 29) = 9.08$, $p < .01$, and revealed no potential outliers (standardised residuals < 1.8 , Tabachnick & Fidell, 2001, p. 67). Subsequent analysis of covariance on average word recall scores employing both covariates revealed a main effect of group that was non-significant, $F(1, 28) = 0.93$, $p = .34$, $MSE = 4.38$, and partial $\eta^2 = .03$. Fig. 1 presents the scatterplot that best corresponds to this analysis by plotting

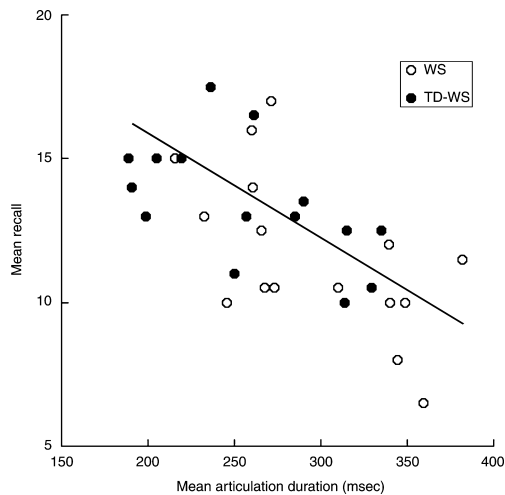


Fig. 1. Scatterplot of the relationship between articulation duration and word recall for individuals with Williams syndrome and their controls. Single line of best fit shown as analysis of covariance revealed no significant group difference in mean recall (see text).

mean recall score against mean articulation time (words and pauses combined).

Individuals with Down syndrome and their controls

Table 3 summarises the performance of the individuals with Down syndrome and their associated controls on the short and long word conditions of the short-term memory and spoken articulation tasks. These data were analysed as above. Analysis of covariance performed on recall scores for short and long words, with group and word length as independent and repeated measures, respectively, and with verbal mental age as a covariate on the group effect, revealed a main effect of group, $F(1, 25) = 12.12$, $p < .01$, $MSE = 2.20$, and partial $\eta^2 = .33$, due to poorer performance among individuals with Down syndrome than among controls. The main effect of word length was significant, $F(1, 26) = 7.95$, $p < .01$, $MSE = 2.91$, and partial $\eta^2 = .23$, reflecting poorer recall of words of long as opposed to short spoken duration. The group by word length interaction was non-significant, $F(1, 26) = 0.39$, $p = .54$, $MSE = 2.91$, and partial $\eta^2 < .01$.

The speech timing measures reported in Table 3 were subjected to a doubly multivariate analysis of variance, with word and pause indices as dependent variables, and with group as an independent measure and condition (short or long word sets) as a multivariate repeated measure. This revealed a significant main effect of group (test of levels), multivariate $F(2, 25) = 4.12$, $p = .03$, and partial $\eta^2 = .25$. Subsequent Roy–Bargman stepdown analysis showed that the groups did not differ reliably in duration of silent pauses, $F(1, 26) = 0.95$, $p = .34$, and $MSE = 46,255.06$, but did differ significantly in duration of spoken words, $F(1, 25) = 7.08$, $p = .01$, and $MSE = 17041.67$, due to individuals with Down syndrome taking less time to articulate words than their controls (see Table 3). The main effect of condition (deviation from flatness) was significant, multivariate $F(2, 25) = 104.90$, $p < .01$, and partial $\eta^2 = .89$. Stepdown analysis revealed a non-significant condition effect on pause durations, $F(1, 26) = 0.01$, $p = .92$, and

Table 3

Summary statistics for short-term memory performance and articulation rates among individuals with Down syndrome and associated controls

Measure	Word length	DS		TD-DS	
		<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Short-term memory score	Short	9.64	1.91	11.36	2.10
	Long	8.07	1.38	10.36	2.13
Spoken word duration (ms)	Short	274	66	365	124
	Long	586	104	652	149
Silent pause duration (ms)	Short	264	162	297	128
	Long	356	257	211	81

$MSE=11083.60$, but a significant effect of condition on word durations, $F(1,25)=209.70$, $p<.01$, and $MSE=5,973.70$. The latter effect was unsurprisingly due to longer word durations in the long as opposed to short word condition. The interaction between group and condition (deviation from parallelism) was also significant, multivariate $F(2,25)=4.97$, $p=.02$, and partial $\eta^2=.29$. Stepdown analysis showed that the group by length interaction was significant for pause durations, $F(1,26)=10.02$, $p<.01$, and $MSE=11,083.60$, but not for word durations, $F(1,25)=0.22$, $p=.64$, and $MSE=5973.70$. Fig. 2 plots mean pause durations for the two groups across short and long word set conditions (and also shows the corresponding data for individuals with Williams syndrome and their controls for comparison). Simple effects analysis confirmed that the effect of group was not significant for pause durations between short words, $F(1,26)=0.36$, $p=.55$, $MSE=21,341.82$, and partial $\eta^2=.01$, but was close to significant for pause durations between long words, $F(1,26)=4.09$, $p=.054$, $MSE=35,966.84$, and partial $\eta^2=.14$; individuals with Down syndrome tended to pause for longer between long words than did their controls. Both groups showed a significant effect of word set condition on pause duration. Among individuals with Down syndrome pauses were longer between long words than short words, $F(1,26)=5.34$, $p=.03$, $MSE=11,083.60$, and partial $\eta^2=.17$, but the opposite effect was observed among controls, $F(1,26)=4.69$, $p=.04$, $MSE=11,083.60$, and partial $\eta^2=.15$.

A correlational analysis was performed to explore the possibility that group differences in short-term

memory performance might be mediated by differences in speech timing measures. However, in both groups neither average word duration nor average pause duration were reliably related to average word recall score, r values from .11 to $-.13$. Further analysis exploring these relationships at the level of short and long words separately also showed no reliable correlation between timing measures and recall in either group for short word stimuli, r values from .02 to .24, or between word duration and word recall for long words, $r=.04$ individuals with Down syndrome, $r=-.10$ controls. There was a slight trend for long word pause duration to relate, in the expected direction, to long word recall among individuals with Down syndrome, $r=-.42$, $df=13$, and $p=.13$, but this trend was less marked among controls, $r=-.33$, $df=13$, and $p=.25$, and the relationship between long word pause duration and average word recall score was clearly non-significant among individuals with Down syndrome, $r=.05$, $df=13$, and $p=.85$. Consequently, and in contrast to the data from individuals with Williams syndrome, there was no clear indication of a causal relationship between speech timing measures and recall for individuals with Down syndrome. Indeed, a regression of the form conducted above among individuals with Williams syndrome and their controls to detect outliers in the relationship between average recall and speech timing showed that average word duration and average pause duration together accounted for a non-significant 7% of the variance in the average recall performance of individuals with Down syndrome and associated controls, $F(2,25)=0.88$, $p=.43$.

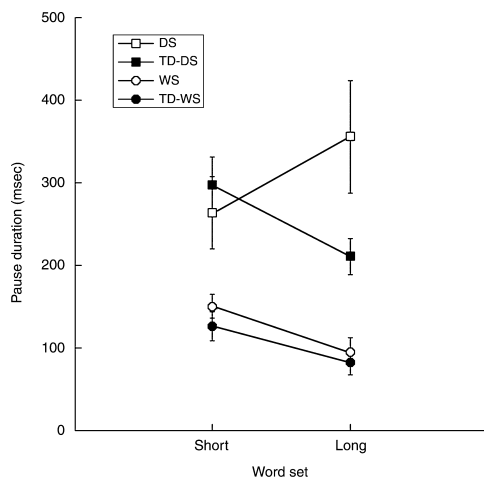


Fig. 2. Mean plot of the group by condition (word length set) interaction for pause duration data from individuals with Down syndrome and their controls. Corresponding data from individuals with Williams syndrome and their controls also shown. (Error bars are ± 1 SE.)

Discussion

The main aim of this study was to explore the impact of speech production skills on short-term memory performance among individuals of a comparable age, but with two different developmental disorders, Williams syndrome and Down syndrome. Although data from these two populations were not analysed together in the above analyses, because of the difficulties of equating these groups on any meaningful matching variable (cf. Klein & Mervis, 1999), the fact that both groups were themselves matched to similar control groups allows for a comparison across these conditions. In particular, the two analyses of verbal short-term memory performance in which verbal mental age was covaried showed that both individuals with Williams syndrome and with Down syndrome were significantly impaired relative to controls given their level of receptive vocabulary, although the magnitude of this impairment was less marked among individuals with Williams syndrome than with Down syndrome (effect sizes of .19 and .33, respectively).

However, although verbal short-term memory impairments were seen among the individuals with Williams syndrome and Down syndrome assessed here, a key implication of the current data is that these deficits appear to be driven by different underlying mechanisms. Among individuals with Williams syndrome there was a relationship between speech timing measures and recall. These individuals also tended to take longer to articulate word pairs than their controls. The fact that a broadly similar relationship between speech timing measures and recall was seen among the associated control participants (see Fig. 1) allowed for analysis of covariance that examined group differences in recall performance once differences in speech timing had been accounted for. This analysis showed that the group effect on recall was reduced to a non-significant level, with a corresponding reduction of effect size from .19 to .03 when both word and pause duration were covaried.

In contrast, among individuals with Down syndrome there was no comparable relation between average estimates of immediate serial word recall and either speech timing measure (cf. Vicari et al., 2004). Indeed, a similar absence of reliable associations was observed between average measures of recall and speech timing among the controls for this group, and together spoken word duration and pause duration only accounted for 7% of the variation in recall performance of individuals with Down syndrome and their controls (in comparison to 39% of the variance accounted for among individuals with Williams syndrome and their controls). Consequently, this difference in overall memory performance observed between individuals with Down syndrome and their matched controls appears not to be easily explicable in terms of general differences in speech timing measures. Indeed, there was no suggestion in the data that individuals with Down syndrome produced *generally* longer responses in the speeded articulation task than their controls. This replicates findings from Jarrold et al. (2000), who reported no reliable difference in overall total articulation time between these individuals with Down syndrome and individuals with moderate learning difficulties matched for verbal mental age. Kanno and Ikeda (2002) also found that individuals with Down syndrome performed less well than controls on a verbal serial recall task, despite the fact that the two groups had comparable articulation rates. Similarly, although Seung and Chapman (2000) found that individuals with Down syndrome had significantly lower digit spans than mental age matched controls, these groups showed comparable rates of responding at recall.

In fact, the analysis of speech timing measures in these groups showed that individuals with Down syndrome actually produced words more rapidly than their controls. However, this was offset, in part, by a tendency to produce longer pause durations when reproducing

pairs of long words (see Fig. 2).¹ Indeed, among individuals with Down syndrome, pauses between long words were significantly longer than those between short words. This effect was the opposite of that seen among the associated controls, as well as among the individuals with Williams syndrome and their controls. It also contrasts with the effect of word length on pause duration reported among 60 typically developing 5–10-year-olds by Jarrold et al. (2000, Experiment 2), where pauses were again shorter between long as opposed to short words (see also Hulme et al., 1984, Experiment 2). Jarrold et al. (2000) argued that this typical shortening of pauses between long words reflected processes of coarticulation whereby participants plan their articulation of the subsequent words while producing the current word. Words of a relatively long spoken duration would provide relatively more opportunity to plan this subsequent articulation, with a consequent shortening of pauses between words. If so, then the increase in pauses between long words seen among individuals with Down syndrome could well reflect speech planning problems, which are known to be relatively common in the condition (e.g., Dodd, 1975; Gibson, 1978; Gunn & Crombie, 1996). Indeed, Seung and Chapman (2000) found that while individuals with Down syndrome produced digits at the same rate as controls, they took longer to initiate their responses, a finding which is consistent with specific speech planning problems rather than a generalised slowing of articulation. Furthermore, one would expect difficulties in planning to particularly affect long words, given that longer words invariably contain more phonemes and are associated with more complex articulatory demands (Waters, Rochon, & Caplan, 1992).

These data therefore provide evidence of deficits in verbal short-term memory performance in Williams syndrome and Down syndrome that are different in nature. Among individuals with Williams syndrome relatively poor verbal short-term memory performance appears to be driven by a generalised relative slowing of speech rates; although clearly the above analyses do not indicate the direction of causal association between these measures. The results are, however, consistent with two possible explanations of the apparent verbal short-term memory deficit, which themselves are not necessarily mutually exclusive. First, it may be that longer articulation times among individuals with Williams syndrome, relative to controls, leads to relatively greater time-based forgetting of information. This might follow as a result of relatively slower subvocal rehearsal (cf.

¹ An indication of this effect is also seen in the Jarrold et al. (2000) articulation time data, where the interaction between group (individuals with Down syndrome or moderate learning difficulties) and word length on overall articulation time approached significance ($p = .09$).

Baddeley et al., 1975), or relatively longer spoken output times when responding (cf. Cowan et al., 1992), or both. Second, it may be that individuals with Williams syndrome have relatively slower memory search times, and so take longer to re-activate memory items for subsequent recall, with a consequent increase in forgetting of these items. The current data do not conclusively favour either suggestion. Certainly, a general slowing of speed of processing might well be expected if these individuals with Williams syndrome were of a lower developmental level than their controls. This is certainly possible given the matching procedure employed here. As noted above, verbal abilities are a relative strength in Williams syndrome (Bellugi et al., 2000; Jarrold et al., 2001; Morris & Mervis, 1999), and one might therefore expect these individuals' general speed of processing to be somewhat delayed relative to their vocabulary level (cf. Laing et al., *in press*). Consequently, although the individuals with Williams syndrome assessed here did show some evidence of impaired verbal short-term memory relative to their controls, there is a sense in which one would not want to explain these data in terms of a fundamental verbal short-term memory deficit (cf. Majerus et al., 2003). Instead, it seems more likely that a more general slowing of speech rates relative to controls of a similar level of vocabulary, which could reflect a difference in general level of development, is the cause of this apparent problem. This would account for the relatively small group effect on recall performance observed here, and the fact that other studies have not consistently shown reliable verbal short-term memory deficits among individuals with Williams syndrome (Brock, 2002; Laing et al., 2001, *in press*; Robinson et al., 2003).

In contrast, the deficit in verbal short-term memory performance seen in Down syndrome is not explicable in the same terms. Although the individuals with Down syndrome assessed here do appear to have speech production problems these do not reflect a general slowing relative to controls, but instead appear to result from more specific difficulties in speech planning and articulation. In addition, this particular difficulty is not obviously related to the deficit in verbal short-term memory performance (cf. Caplan et al., 1992, Experiment 3). This is consistent with other evidence that suggests that verbal short-term memory problems persist in Down syndrome even on tasks that reduce or remove the need to respond by articulating the list of to-be-remembered items. These include tests of probed recall (Jarrold et al., 2000), tasks that require non-verbal responses (Brock & Jarrold, *in press-a*; Marcell & Weeks, 1988), and tests of individuals' ability to recognise changes in the ordering of items in memory lists (Brock & Jarrold, *in press-b*; Jarrold et al., 2002). Taken together, these data suggest that slowed responding at the output stage of a verbal serial recall task cannot be the prime cause of poor verbal short-term memory performance in Down syndrome.

The data also count against the possibility that slowed sub vocal rehearsal gives rise to verbal short-term memory problems in Down syndrome (Kay-Raining Bird & Chapman, 1994). In that sense they are, once again, consistent with previous data. Jarrold et al. (2000, Experiment 2) found that individuals with Down syndrome performed less well than controls on a verbal short-term task that involved probed recall, but that neither group showed evidence of a word length effect under these conditions. The absence of a word length effect in this previous study suggested that neither group were engaging in rehearsal, and that, consequently, a rehearsal deficit could not explain the impaired recall that was still observed among individuals with Down syndrome in this case. Gathercole, Henry, and others have argued that typically developing individuals do not engage in subvocal rehearsal until around 7 years of age (e.g., Flavell, Beach, & Chinsky, 1966; Gathercole et al., 1994; Henry, 1991). The absence of a clear relationship between speech timing measures and recall in both individuals with Down syndrome and their controls in the current data is consistent with this suggestion (cf. Cowan et al., 1994; Gathercole & Adams, 1993; Gathercole et al., 1994), as individuals in both groups are either aged less than 7 or functioning below that level of development (see Table 1).

Of course word length effects are seen for both individuals with Down syndrome and their controls in the current data (cf. Kanno & Ikeda, 2002; Vicari et al., 2004), a finding that at first sight might appear problematic for this account. However, many other studies have shown word length effects among individuals younger than 7 years of age, or functioning below the seven-year level (e.g., Hitch, Halliday, Dodd, & Littler, 1989; Hulme et al., 1984; Johnston, Johnson, & Gray, 1987). The fact that these word length effects are removed by probing for recall (Allick & Siegel, 1976; Balthazar, 2003; Henry, 1991; Jarrold et al., 2000; Turner, Henry, & Smith, 2000), thereby removing the need to produce a full spoken repetition of the item list, has led to the claim that word length effects at this level are due to output effects, with poorer recall of longer words reflecting the greater time to reproduce rather than rehearse these items (cf. Cowan et al., 1992).

One of the broader implications of the current findings, therefore, is that they raise questions about the nature of the word length effect in young children (cf. Vicari et al., 2004). Among individuals with Down syndrome and their controls a clear effect of word length on recall was observed, but there was no clear and consistent relationship between individuals' recall performance and their rate of articulation. Obviously, care should be exercised in interpreting these null results, particularly given the small sample sizes involved in these correlational analyses, and the slight suggestion of a relationship between duration of pauses in speeded

articulation of long words and long word recall among individuals with Down syndrome. However, the reliability of the measures employed here was good, and correcting for attenuation would not raise the correlations observed in these groups between average indices of recall and speech timing to significant levels.² Consequently, these data do suggest that the word length effect among these participants may not have been caused by speech rate effects at either rehearsal or output. Instead, the findings are more consistent with the view that this effect reflects differences in the complexity, rather than duration per se, of short and long words (Brown & Hulme, 1995; Caplan et al., 1992; Lovatt et al., 2000; Neath & Nairne, 1995; Service, 1998). Indeed, the suggestive evidence of a relationship between pause duration and recall for long words only might support this view, given the arguments made above for the relatively greater complexity of words of a long as opposed to short spoken duration and for the fact that pause durations might index planning processes affected by complexity.

Of course, this raises the possibility that the word length effect seen among individuals with Williams syndrome and their controls arose for the same reasons, and not because of any link between speech timing measures and extent of forgetting as the above analysis appears to suggest. There are two related reasons for thinking this unlikely. First, there is some evidence that the magnitude of this effect is larger among individuals with Williams syndrome and controls than among individuals with Down syndrome and their controls. A formal comparison of the size of the word length effect across each group was not carried out, and would arguably not be particularly meaningful given the differences in absolute recall level of the groups (cf. Logie, Della Sala, Laiaccona, Chambers, & Wynn, 1996). Nevertheless, the effect size for the word length effect on recall was .40 for individuals with Williams syndrome and controls, compared to .23 for individuals with Down syndrome and their controls. Second, as Fig. 2 indicates, pause durations were considerably shorter among individuals with Williams syndrome and controls than among the other two groups. If pause duration does provide an index of speech planning or memory search processes that are affected by complexity (Cowan et al., 2000; Jarrold et al., 2000), then these data suggest that these factors are likely to be less of a constraint on performance in these two groups. Indeed, given the more advanced developmental level of individuals with Williams syndrome

and controls, relative to the individuals with Down syndrome and their controls (see Table 1), one plausible suggestion is that specific complexity constraints on children's verbal short-term memory are likely to be more dominant at early developmental levels, with general speed of articulation becoming relatively more important later on. Again, this is consistent with the suggestion that rehearsal in verbal short-term memory develops at around the seven-year level.

The current study therefore has important implications for the understanding of the word length effect in general, and among children of different levels of development in particular. The other key implication of this work is that verbal short-term memory impairments can arise for different reasons. Although individuals with Williams syndrome did perform less well than their controls on the verbal serial recall tasks, this difference in performance is attributable to differences in rate of articulation, which in turn may reflect the matching procedure employed. Indeed, the correlation between individual differences in speech rate and recall among individuals with Williams syndrome and their controls is problematic for the view that word length effects are not in any way time-dependent (cf. Lovatt & Avons, 2001; see Cowan et al., 2000). Although the fact that covarying speech time differences removes recall differences in these two groups certainly does not prove that articulation rate constrains verbal short-term memory performance, it does suggest that temporal factors need to be considered when evaluating such short-term memory impairments (cf. Avons & Hanna, 1995; Bosshardt, 1993; Raine, Hulme, Chadderton, & Bailey, 1991; Swanson & Ashbaker, 2000; White, Craft, Hale, & Park, 1994).

However, among individuals with Down syndrome the observed deficit in verbal short-term memory performance, relative to vocabulary matched controls, is not obviously mediated by speech timing effects. This finding shows that temporal factors are not always causally related to verbal short-term memory performance, nor the sole source of variation in verbal immediate serial recall. Indeed, it implies that Down syndrome is associated with a more fundamental verbal short-term memory deficit than is Williams syndrome. It is, of course, possible that individuals with Down syndrome perform poorly on tests of verbal short-term memory because of hearing or speech perception difficulties. These are known to be relatively common in Down syndrome (Dahle & McCollister, 1986; Limongi, Carvallo, & Souza, 2000; Marcell & Cohen, 1992; Welsh & Elliot, 2001), and might well be expected to have a detrimental effect on recall, given that verbal short-term memory tasks are typically presented auditorially. In fact, however, what evidence there is relevant to this issue suggests that such difficulties do not have a dramatic impact on performance, and cannot account for the extent of the difficul-

² At the level of correlations between average recall and timing measures (across short and long words combined), correcting for attenuation due to lack of reliability would raise correlation coefficients among individuals with Down syndrome and their controls to, at best, $-.14$.

ty on verbal short-term memory tasks seen in Down syndrome (Brock & Jarrold, in press-b; Jarrold & Baddeley, 1997; Jarrold et al., 2002; Marcell & Cohen, 1992). Instead it seems that the condition is associated with a more fundamental verbal short-term memory deficit, that may be associated with the maintenance of verbal material in correct serial order (Brock & Jarrold, in press-a; Jarrold et al., 2002). Given that intact verbal short-term memory may be crucially important for aspects of typical language development, and vocabulary learning in particular (e.g., Baddeley et al., 1998), this claim has clear implications for the development of these individuals' language skills. Consequently, future work could usefully test and map out the potential association between impaired verbal short-term memory and language acquisition in Down syndrome.

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