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**Using developmental trajectories to examine verbal and visuospatial short-term memory
development in children and adolescents with Williams and Down syndromes**

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ABSTRACT

Williams (WS) and Down (DS) syndromes have been associated with specifically compromised short-term memory (STM) subsystems. Individuals with WS have shown impairments in visuospatial STM, while individuals with DS have often shown problems with the recall of verbal material. However, studies have not usually compared the *development* of STM skills in these domains, in these populations. The present study employed a cross-sectional developmental trajectories approach, plotting verbal and visuospatial STM performance against more general cognitive and chronological development, to investigate how the domain-specific skills of individuals with WS and DS may change as development progresses, as well as whether the difference between STM skill domains increases, in either group, as development progresses. Typically developing children, of broadly similar cognitive ability to the clinical groups, were also included. Planned between- and within-group comparisons were carried out. Individuals with WS and DS both showed the domain-specific STM weaknesses in overall performance that were expected based on the respective cognitive profiles. However, skills in both groups developed, according to general cognitive development, at similar rates to those of the TD group. In addition, no significant developmental divergence between STM domains was observed in either clinical group according to mental age or chronological age, although the general pattern of findings indicated that the influence of the latter variable across STM domains, particularly in WS, might merit further investigation.

Keywords: Williams syndrome, Down syndrome, short-term memory, developmental trajectories.

INTRODUCTION

Aetiological and cognitive characteristics of Williams and Down syndromes:

Williams syndrome (WS) is a genetic condition occurring in between 1 in 7500 (Strømme, Bjørnstad, & Ramstad, 2002) and 1 in 20,000 (Morris, Demsey, Leonard, Dilts, & Blackburn, 1988) live births. It is caused by a microdeletion at the chromosomal locus 7q.11.23. This normally includes the elastin gene (e.g. Ewart et al., 1993; Lowery et al., 1995). Physically, WS is primarily characterised by distinct facial dysmorphia, and other musculoskeletal, cardiovascular, and renal abnormalities (e.g. Jones & Smith, 1975; Lenhoff, Wang, Greenberg, & Bellugi, 1997; Wang, Doherty, Rourke, & Bellugi, 1995). Psychological and behavioural markers include mild-moderate intellectual disability (ID; Udwin, Yule, & Martin, 1987), a sociable disposition (e.g. Jones et al., 2000), heightened anxiety (Dykens, 2003) and hyper-sensitivity to sound (e.g. Gallo, Klein-Tasman, Gaffrey, & Curran, 2008).

Down syndrome (DS) is the most common known genetic condition involving intellectual disabilities (Pennington, Moon, Edgin, Stedron, & Nadel, 2003), occurring in approximately 1 in every 700-1000 live births (Kittler, Krinsky-McHale, & Devenny, 2008). It is caused by a triplication on chromosome 21 (LeJeune, Gautier, & Turpin, 1959). Physical characteristics include a distinctive facial appearance, heart and gastrointestinal anomalies, immunodeficiency, hearing problems, and precocious aging (e.g. Korenberg et al., 1994; Zigman, Silverman, & Wisniewski, 1996). The most distinctive psychological features of the condition are moderate to severe ID (Pennington et al., 2003), and an increased risk of age-related cognitive decline (e.g. Rowe, Lavender, & Turk, 2006).

Both conditions have been associated with a fractionation of cognitive skills. Individuals with WS have shown relative verbal strengths alongside visuospatial impairments (e.g. Bellugi, Wang, & Jernigan, 1994; Udwin & Yule, 1991), with the latter particularly evident on tasks involving a constructive requirement (e.g. Hoffman, Landau, & Pagani, 2003). This is consistent with evidence for vulnerability of the dorsal stream (e.g. Atkinson et al., 2003; Galaburda & Bellugi, 2000), a visual cortical area involved in processing location and motion (Milner & Goodale, 1995). Although atypicality has been observed in a number of verbal sub-domains (see Brock, 2007, for a review) including pragmatic language (e.g. Reilly, Losh, Bellugi, & Wolfeck, 2004) and spatial grammar (e.g. Phillips, Jarrold, Baddeley, Grant, & Karmiloff-Smith, 2004), comparisons of the verbal and performance IQ scores of individuals with WS have generally shown a verbal superiority (e.g. Grant et al., 1997; Levy & Bechar, 2003). Individuals with DS usually display “flatter” profiles (Jarrold, Baddeley, & Phillips, 2007), but have often exhibited expressive language difficulties (e.g. Abbeduto & Chapman, 2005; Roberts, Price, & Malkin, 2007), especially with syntax (e.g. Chapman, 2003). In addition, verbal ability has been reported to be below performance, or overall, IQ levels (e.g. Vicari, Caselli, & Tonucci, 2000).

Mapping development:

Most studies examining cognitive skills in WS and DS have done so by comparing the mean task scores of these populations with those of groups matched (individually or overall) for chronological age (CA) and/or mental age (MA). This method usually collapses individual totals, age and ability levels, to give a group mean representative of overall performance level. While this approach enables the direct comparison of groups, it can be argued that it masks development, offering little indication of how skills change over time, how the clinical

group may have arrived at that level of performance, and whether this differs from the typical pattern. An alternative approach is the developmental trajectories method, which attempts formally to encapsulate change in performance over time, normally by plotting it against chronological age (CA) and/or a measure of general cognitive ability. This has been claimed to provide a picture that is descriptively richer, in terms of categorising the *types* of delay or difference shown by clinical groups, than the binary distinction between these two concepts most readily predicated by matching (see Thomas et al., 2009).

The developmental trajectories approach has been employed, with regard to each group, to examine development within a number of domains. For instance, joint engagement behaviour in infants with DS has been mapped longitudinally (Adamson, Bakeman, Deckner, & Ronski, 2009), while other authors have used cross-sectional approaches to plot the development of both lexical skills (Thomas et al., 2006), and facial processing abilities (Karmiloff-Smith et al., 2004) in individuals with WS. The latter authors observed that their sample, while often equivalent to TD controls in terms of overall performance on a number of facial processing tasks, showed patterns of development that were both delayed *and* deviant in comparison. This suggests that measured between-group equivalence may not always derive from similar underlying developmental processes. In addition, other authors have indicated that the skill development of different groups may converge and diverge at different stages. Paterson and colleagues (Paterson, Brown, Gsödl, Johnson, & Karmiloff-Smith, 1999) reported similar levels of delay on a task tapping early vocabulary skills in infants with WS and DS. This parity is in marked contrast to the documented superiority of older individuals with WS, over older individuals with DS, in this domain (e.g. Paterson, 2001).

Findings such as these provide clear justification for an approach that accounts for change over time; in order to gain a more sophisticated understanding of skill profiles. This is important for an area such as facial processing, where it has been claimed that the skills of individuals with WS may proceed typically (e.g. Tager-Flusberg, Plesa-Skwerer, Faja, & Joseph, 2003).

Despite the uneven ability profiles associated with WS, as well as the usefulness of profiling skills developmentally, only a limited number of studies have used developmental trajectories to compare how verbal and non-verbal/visuospatial skills may improve with development in this population. Jarrold, Baddeley, and Hewes (1998) plotted the performance of a group of sixteen individuals with WS (aged 6-28) using both the verbal British Picture Vocabulary Scale (BPVS; Dunn, Dunn, Whetton, & Pintilie, 1982) and verbal and non-verbal subtests, such as the visuospatial measure Pattern Construction, from the Differential Abilities Scale (DAS; Elliot, 1990) against CA. Verbal performance did not develop at a typical rate across time, but was faster to improve than non-verbal performance, with difference between the two domains increasing in line with verbal ability. Although these data were cross-sectional, and as such did not enable a direct inspection of skill development within individuals, a longitudinal investigation involving testing the same group six times over approximately forty months (Jarrold, Baddeley, Hewes, & Phillips, 2001) showed that verbal skills were again faster to develop. Further, the magnitude of verbal/non-verbal differences increased significantly over time. The authors suggested that this phenomenon may explain why some studies involving younger/less able individuals with WS have not always reported the usually significant verbal/visuospatial ability differences (e.g. Pagon, Bennett, LaVeck, Stewart, & Johnson, 1987; Volterra, Capirci,

Pezzini, Sabbadini, & Vicari, 1996), as sufficient disparity between the relative scores has yet to emerge. These findings suggest that some verbal skills may develop at a faster rate than some non-verbal abilities in those with WS, and highlight the usefulness of accounting for development when examining skills across domains. However, the lack of any comparison group makes it difficult to claim, with complete confidence, that such patterns are specific to individuals with WS.

Short-term memory in individuals with WS and DS:

It is clear that to maximise the understanding of verbal and non-verbal development in populations with WS and DS, developmentally sensitive methodologies should be applied, and multiple comparison groups employed in order to identify population-specific patterns. A skill yet to be investigated in these populations in this way is short-term memory (STM), the mental storage of information over a short time period (Gathercole, 1999). STM is of developmental significance, implicated in both long-term learning (e.g. Gathercole, Willis, Emslie, & Baddeley, 1992), and positive educational outcomes (e.g. Bull, Espy, & Wiebe, 2008). Matching studies of individuals with WS and DS have suggested that they display the expected relative STM strengths and difficulties. Individuals with WS have shown relatively good verbal STM, indicated by performance equivalence with MA-matched groups on verbal STM tasks (e.g. Robinson, Mervis, & Robinson, 2003). On the other hand, they tend to show weaker visuospatial STM performance compared to MA-matched groups (e.g. O'Hearn, Courtney, Street, & Landau, 2009; Rhodes, Riby, Park, Fraser, & Campbell, 2010). By contrast, the visuospatial STM skills of individuals with DS are usually found to be equal that of MA-matched groups (e.g. Brock & Jarrold, 2005; Visu-Petra, Benga, Jîncăş, & Miclea, 2007), with corresponding deficits reported on verbal STM tasks (e.g. Vicari, Marotta, &

Carlesimo, 2004). These opposite STM findings have been reinforced by studies comparing the two populations directly (Jarrold, Baddeley, & Hewes, 1999; Kittler et al., 2008; Wang & Bellugi, 1994). Nevertheless, although the findings seem to suggest patterns of verbal and visuospatial STM development that differ both within *and* between the two groups, no study to date has attempted to plot STM performance in both domains against wider developmental indices (although see Brock & Jarrold, 2005, who examined the level of concurrence in developmental variation across the two domains). This would enable an assessment of how groups' performances may change across time, and whether developmental differences across the verbal and visuospatial domains may characterise STM. In addition, the inclusion of both groups offers an indication of the syndrome specificity of each group's performance pattern.

Overview of current study:

The present study employs the developmental trajectories approach to investigate verbal and visuospatial STM development in children and adolescents with WS and DS. As Annaz, Karmiloff-Smith, and Thomas (2008) have noted, this method allows the examination of both inter- and intra-group variability. This is useful when assessing populations such as those with WS and DS, who display uneven ability profiles, as it allows the comparison of performance across domains. Both between- and within-group analyses were conducted. The former involved comparing the trajectories for each group's performance in each STM domain, against an index of normative performance; in this case, a group of TD children. Only MA was used to compare across groups for these analyses, as overlap in CAs between the clinical groups and the TD group on this variable was minimal; such analyses would thus have necessitated a theoretically unsound level of extrapolation.

The within-group analyses comprised planned comparisons between the verbal and visuospatial STM developmental patterns, within each group, to: (1) investigate differences in performance levels and/or developmental rate across STM domains; (2) assess whether a faster rate of verbal than visuospatial development found for general ability levels in individuals with WS (Jarrold et al., 2001) is also observed in STM performance; and (3) establish whether a corresponding pattern, whereby the development of visuospatial STM outstrips that of verbal STM, is evident in individuals with DS. These analyses were undertaken using MA, then CA, as the developmental index variable, in order to build as comprehensive a picture as possible by examining the influence of each in turn. Plotting skill development against CA, the developmental variable employed by Jarrold et al. (2001), enabled a direct assessment of whether the verbal/visuospatial chronological divergence suggested by those authors also applies to STM.

General experimental hypotheses in accordance with previous literature were:

1. Individuals with WS would display a relative visuospatial STM weakness, as indicated by poorer visuospatial than verbal STM performance, and/or poorer visuospatial than verbal STM performance in comparison with the other two groups.
2. Individuals with DS would show a corresponding relative weakness in verbal STM, as suggested by within-group domain differences, and/or differences in between-group developmental patterns.

Two more specific hypotheses, relating to the within-group analyses, were:

3. Verbal STM would develop faster than visuospatial STM in individuals with WS.

4. Visuospatial STM would develop faster than verbal STM in individuals with DS.

METHOD

Participants:

All participant groups were composed of individuals who had undertaken *both* the IQ and the STM measures described below. The final analysis comprised 130 participants; 31 children, adolescents, and young adults with WS (15 male; age range: 8 years 2 months – 21:10), 30 children, adolescents, and young adults with DS (14 male; age range: 10:9 – 21:5), and 69 TD children (43 male; age range: 4:0 – 9:2). Although matching for cognitive ability was not the aim, older clinical individuals and younger TD participants were targeted, to ensure suitability for the tasks.

Individuals with WS were recruited through the Williams Syndrome Foundation UK.

Participants with DS were recruited via the UK branch of the Down Syndrome Association.

TD children were recruited through two primary schools in Greater London, and parenting networks local to the lead researcher. Individuals from the two clinical groups were confirmed, by parents/caregivers prior to testing, to possess formal sole diagnoses, given by appropriate health professionals according to accepted criteria. Each participant with DS displayed the full Trisomy 21 DS karyotype, the most common form of the condition (see Seung & Chapman, 2004). All TD participants were confirmed as having no diagnosis of any special educational need. Participants from all three groups had typical, or corrected-to-typical, visual and auditory abilities.

Ethical approval was given by the relevant University Research Ethics Committee. Written

consent was obtained from schools or organisations, then parents, and then the participants themselves. Table 1 summarises sample characteristics.

IQ measure:

IQ was assessed with the Stanford-Binet Abbreviated Battery (ABIQ) test; a shortened version of the full Stanford-Binet IQ test battery (Fifth Edition; Roid, 2003) which features the Non-Verbal Reasoning (NVR) and Verbal Knowledge (VKn) subtests from the full battery. As the present measures were given as part of a larger test battery, the ABIQ was deemed suitable as it takes less time to administer than the full version of the Stanford-Binet, while still giving separable non-verbal and verbal raw scores. These totals can be combined to give a composite mental age (MA) equivalent score, expressed in months.

The ABIQ is given in a fixed order, with NVR administered first. The Stanford-Binet Technical Manual (Roid, 2003) reports strong mean reliability coefficients for the ABIQ (internal, TD 5-8 year-olds: $r = .91$; test-retest, TD 2-20 year-olds: $r = .85$). Table 1 gives details of groups' ABIQ scores.

TABLE 1 ABOUT HERE

STM measures:

Verbal STM was assessed using the Word List Recall (WLR) subtest from the Working Memory Test Battery for Children (WMTB-C; Pickering & Gathercole, 2001). Participants were required to recall verbally sequences of single-syllable words, orally presented by the experimenter e.g. ("park, cod, dip"). This was a span measure, with span level (the number of words presented per trial) increasing according to performance and ranging from one to

seven. Words were presented at the rate of one per 0.75 seconds. Six trials were administered at each span level, with progress to the next span level achieved if four trials were answered correctly. Any trials unadministered as a result of this rule were automatically awarded as correct.

Two scores can be derived from this measure for each participant; overall span (0-7) and overall number of trials correct (0-42). The latter score was used in the current analysis due to claims that it is more reliable (Ferguson, Bowey, & Tilley, 2002), and less restrictive (Conway et al., 2005), than span score. WLR is a reliable measure, with the WMTB-C manual reporting strong mean internal ($r = .60$) and test-retest ($r = .80$) coefficients, derived from a sample of TD children and adolescents (age range: 4-15 years).

In order to assess visuospatial STM, the Block Recall (BR) measure, also from the WMTB-C, was employed. This required participants to recall correctly sequences of spatial locations tapped out by the experimenter, at the rate of one per second, on a plastic block board incorporating nine blocks. This was also a span test, with span level, the number of blocks tapped per trial, ranging from one to nine. Again, six trials were presented at each span level, with progress according to the same criteria as for WLR. Number of trials correct (0-54) was the score taken for each participant. The WMTB-C manual reports strong mean internal ($r = .55$) and test-retest ($r = .63$) reliability coefficients, also taken from the TD sample described above.

Administration:

Each participant was informed, before giving their written consent prior to the commencement of testing, that they could opt out of, or interrupt, the testing sessions at any time. Clinical participants were tested at home, in one or two sittings on the same day. TD participants were mainly tested at school, across two sessions on adjacent days, although a limited number of this group completed testing within a single home visit, in one or two sittings. All participants were tested in a quiet environment, to maximise concentration.

All participants were given the ABIQ before completing the two STM subtests. The latter were counterbalanced across the whole sample, with half the participants undertaking WLR then BR, and the other half completing the measures in the opposite order.

RESULTS

For ease of interpretation, and to facilitate comparison across tasks, raw 'trials correct' scores on each of the STM measures were converted into z scores. These were calculated from the overall sample mean. Table 2 gives each group's mean trials correct score, and mean z score, along with standard deviations, for each STM task.

TABLE 2 ABOUT HERE

Trajectories were then constructed for each group, with regard to each STM task, and statistically compared using analyses of co-variance (ANCOVA). Two types of comparison were undertaken: (1) between-group differences, where the trajectories of all three groups

on each task were compared; and (2) within-group differences, where WLR and BR trajectories were compared within each group to determine whether one type of STM developed in a different manner from the other. Both MA and CA were used to plot performance for the within-group comparisons, but only MA was used for the between-group analyses, as the overlap between CA ranges of the clinical groups and that of the TD group was minimal, negating the value of such an analysis.

All assumptions with regard to ANCOVA analysis were met; the standardised residuals of each model constructed were normally distributed, and inspection of Cook's D and Leverage values generated for each analysis did not identify any cases likely to exert undue influence.

Between-group comparisons:

Trajectories for all three groups were first compared against MA for each STM task, in order to assess the performance of the two clinical groups against normative developmental performance. In these models, overall MA was re-scaled to months from the MA level of the developmentally 'youngest' participant assessed (MArs). While such an alteration does not change the analysis, it aids interpretation of regression lines by ensuring that each intercept is set at the lowest MA level measured, rather than at an "absolute" MA score of 0. These comparisons were undertaken using a 3*2 ANCOVA (IVs: Group, task type, MArS; DVs: WLR z-scores, BR z-scores), which featured MArS as the covariate.

A significant overall main effect of MArS was observed ($F[1,124]= 64.23$, $p<.001$, partial $\eta^2=.341$), with increases in this variable associated with improved STM performance overall.

The main effects of group ($F[2,124]= 1.70$, $p=.186$, partial $\eta^2= .027$), and task domain ($F[1,124]= 1.05$, $p=.306$, partial $\eta^2= .008$) were not significant. The only interaction which reached significance was that between task and group ($F[2,124]= 4.07$, $p<.05$, partial $\eta^2= .062$). Group by MArS ($F[2,124]= .09$, $p= .917$, partial $\eta^2= .001$), task by MArS ($F[1,124]= 1.16$, $p= .282$, partial $\eta^2= .009$), and task by group by MArS interactions ($F[2,124]= .44$, $p= .647$, partial $\eta^2= .007$), were not significant. Taken together, these interaction findings indicated that it was the relationships between the *overall performance levels* of the three groups - rather than the relationships between groups' respective *rates of development* - which differed across task domain.

Groups' performances were then compared for each task domain. For verbal STM (WLR), no significant differences were found between either of the clinical groups, and the TD group, with regard to either trajectory intercept (WS: $t[2,124]= -.47$, $p= .640$, partial $\eta^2= .002$; DS: $t[2,124]= 1.79$, $p= .076$, partial $\eta^2= .025$) or gradient (WS: $t[2,124]= .59$, $p= .553$, partial $\eta^2= .003$; DS: $t[2,124]= .48$, $p= .634$, partial $\eta^2= .002$). This suggested that performance levels of the two clinical groups at the lowest observed MA level were as expected on the basis of general cognitive development (although note that the lower performance of the DS group approached significance, as might have been expected based on previous literature), and did not differ – either from each other or the TD group – in the rate of subsequent development with MA. Follow-up comparisons of the WS and DS groups revealed no significant differences in intercept ($t[2,124]= -1.18$, $p= .242$, partial $\eta^2= .011$), or gradient ($t[2,124]= .05$, $p= .962$, partial $\eta^2= .000$). These trajectories are shown in Figure 1(a)².

² The unscaled MA (months) variable is used, in order to aid interpretation. For clarity, trajectories per domain are presented in separate tables.

Inspection of the analysis for the visuospatial STM task (BR) showed that the WS group were significantly less accurate at onset than the TD group ($t[2,124] = -2.25$, $p < .05$, partial $\eta^2 = .039$), who did not differ from individuals with DS ($t[2,124] = 1.02$, $p = .307$, partial $\eta^2 = .008$). Follow-up comparisons revealed that the onset performance level of the WS group was also significantly inferior to that of the DS group ($t[2,124] = 2.64$, $p < .01$, partial $\eta^2 = .053$). No differences in gradient were found between any of the groups (TD vs. WS: $t[2,124] = -.65$, $p = .514$, partial $\eta^2 = .003$; TD vs. DS: $t[2,124] = .11$, $p = .909$, partial $\eta^2 = .000$; WS vs. DS: $t[2,124] = .52$, $p = .605$, partial $\eta^2 = .002$), indicating that the *rate* of developmental improvement in visuospatial STM did not differ between groups. This meant that the lower performance of the WS group remained consistent across development. Figure 1(b) depicts these trajectories.

FIGURE 1 ABOUT HERE.

Within-group comparisons: MA

Trajectories plotting performance in each STM task domain against MA were then constructed and analysed for each group, using 2*2 ANCOVAs (IVs: MA, task type; DVs: WLR z scores, BR z scores), which included MA as the covariate. Analysis of the TD group's performance showed that the effect of task type was not significant ($F[1,68] = .00$, $p = .964$, partial $\eta^2 = .000$): this indicated that the performance levels of the TD group did not differ across STM domains. A significant overall effect of MA was, however, observed ($F[1,67] = 79.45$, $p < .001$, partial $\eta^2 = .542$), with increases in this variable significantly related to

improvements in performance across both tasks. The interaction between the MA and task type variables did not reach significance ($F[1,67] = .11$, $p = .743$, partial $\eta^2 = .002$), indicating that the positive relationship between MA and performance was consistent across both STM domains. Trajectories constructed for the TD group are given in Figure 2(a).

Analysis of the performance of individuals with WS showed a significant effect of task type ($F[1,30] = 21.74$, $p < .001$, partial $\eta^2 = .420$), with overall performance level on the verbal STM measure significantly superior to performance on the visuospatial STM measure. This group also showed a significant positive effect of MA ($F[1,29] = 29.42$, $p < .001$, partial $\eta^2 = .504$), indicating that STM performance increased with MA. Finally, there was no MA by task interaction ($F[1,29] = 1.71$, $p = .201$, partial $\eta^2 = .056$), meaning that the effect of MA did not differ across STM domains. In sum, performance levels on verbal STM were consistently higher than on visuospatial STM, but scores in both domains increased with MA to the same extent. Trajectories from individuals with WS can be seen in Figure 2(b).

Individuals with DS also displayed a significant effect of task type ($F[1,29] = 16.56$, $p < .001$; partial $\eta^2 = .364$), with overall accuracy on the visuospatial STM task higher than shown on the verbal STM task. As with the other two groups, a significant main effect of MA ($F[1,28] = 9.17$, $p < .01$, partial $\eta^2 = .247$) was found, indicating that STM performance increased with MA. Finally, there was no significant interaction between MA and task type ($F[1,28] = .13$, $p = .721$, partial $\eta^2 = .005$), indicating that the effect of MA did not differ across STM domains. In sum, performance levels on visuospatial STM were consistently higher than on verbal STM, but scores in both domains increased with MA to the same extent. Figure 2(c) shows the trajectories of the DS group.

FIGURE 2 ABOUT HERE.

Within-group comparisons: CA

Each group's performance was then plotted against CA, and analysed using 2*2 ANCOVAs featuring this variable as the covariate. Effects of task type were identical to those given above. The effect of CA on STM abilities was significant for the TD group ($F[1,67]= 75.44$, $p<.001$, partial $\eta^2= .530$), with performance improving as CA increased. The CA by task interaction was not significant ($F[1,67]= .03$, $p= .865$, partial $\eta^2= .000$), indicating that this pattern was consistent across STM domains. These trajectories are depicted in Figure 3(a).

CA did not significantly affect the overall performance of individuals with WS ($F[1,29]= 2.56$, $p= .120$, partial $\eta^2= .081$), indicating that performance did not vary with CA. Further, the CA by task interaction missed significance for this group ($F[1,29]= 2.56$, $p= .099$, partial $\eta^2= .091$), suggesting that changes in performance on both STM variables with CA were not significantly different. Figure 3(b) gives these trajectories.

Finally, a significant positive relationship between CA and STM performance was observed in the DS group ($F[1,28]= 12.95$, $p<.01$, partial $\eta^2= .316$). This improvement in STM scores with CA was true for both STM domains, as indicated by a non-significant CA by task interaction ($F[1,28]= 1.18$, $p= .287$, partial $\eta^2= .040$). These trajectories are shown in Figure 3(c).

FIGURE 3 ABOUT HERE

The next part of the analysis was conducted in order to establish formally whether any of the groups showed an increase in verbal/visuospatial *disparity* as a function of MA and/or CA. A new variable, difference in z-scores, was computed by subtracting each participant's BR z-score from their WLR z-score. Two simple linear regression analyses were then conducted for each group, using this measure as the dependent variable, and MA and CA, respectively, as predictors. These showed no significant relationships, in any group, between between-domain z-score disparity and either MA (TD: $t[1,67] = .33$, $p = .743$; WS: $t[1,29] = 1.31$, $p = .201$; DS: $t[1,28] = .36$, $p = .721$), or CA (TD: $t[1,67] = -.17$, $p = .865$; WS: $t[1,29] = 1.70$, $p = .099$; DS: $t[1,28] = -1.08$, $p = .287$).

DISCUSSION

The present study involved the administration of both verbal and visuospatial STM measures to groups of children, adolescents and young adults with WS and DS. Performance was plotted against MA and CA in turn, to give developmental trajectories for each of these variables. Both groups were compared with a group of TD children of broadly similar MAs. Both between- and within-group comparisons were undertaken. Results from each of these will be discussed in turn.

Between-group comparisons:

The between-group analyses enabled a direct inspection of whether the clinical groups' performance deficits constituted *genuine* impairments against the comparison of typical

performance (mental age level). The three groups did not differ on the verbal STM task, either in terms of performance level at onset, or the rate at which skills improved in conjunction with general cognitive development. This goes somewhat against the literature (although note the tendency for individuals with DS to get lower scores – see below) where studies of matched groups in individuals with DS and typical development often report poorer performance in DS groups. Findings were more in line with previous literature with respect to visuospatial STM: individuals with WS displayed significantly lower visuospatial STM scores than both the TD and DS groups; although no differences were found between any of the groups in terms of the rate of developmental improvement in performance with MA.

The visuospatial STM performance patterns shown by the WS group demonstrated that they were significantly less accurate, at the youngest MA measured (i.e. 55 months), but their subsequent rate of skill development did not reliably differ from that shown by the other groups. The observed reduction in accuracy in the WS group is, therefore, present throughout the MA range measured. This is in keeping with what may be expected, given the claim that visuospatial STM skills are relatively weak in this population (e.g. Wang & Bellugi, 1994). It is also potentially encouraging, as it indicates that visuospatial STM in this population, across the developmental range assessed here, may not only improve at a “typical” speed in relation to MA, but also be positively linked to general cognitive development. Future studies should examine whether this may be the case, and assess whether specific learning interventions selected on the basis of overall MA can promote enhanced improvement across time, and also prevent deficits from enduring as cognitive development proceeds.

The within-group pattern of weaker verbal than visuospatial STM performance displayed by individuals with DS (see below for separate discussion of this) did not translate into a significant verbal deficit in direct comparison with the other groups. It is, however, worth noting that the lower intercept performance of this group, in comparison to that of TD individuals, was marginally significant ($p = .076$). Furthermore, the lack of differences between the gradients of groups' trajectories demonstrated that this reduction in accuracy, while not statistically significant, persisted throughout development. Although this "typical" rate of development is encouraging, it is difficult to claim from these findings that verbal STM skills in children and adolescents with DS are unimpaired, especially given previous studies showing localised deficits in this area (e.g. Jarrold et al., 1999). The present findings may thus indicate that the verbal skills of the individuals with DS in the current study were relatively strong, in comparison with those of the population as a whole. Alternatively, the present sample, while sufficient to detect a within-group difference in performance across domains, may not have encompassed a range of MAs – and/or CAs - wide enough to allow for the emergence of a statistically reliable deficit in comparison to the TD group. Both these possibilities would be minimised in the future by the measurement of a larger number of individuals with the condition.

Indeed, the lack of significant between-group differences in the gradient of STM improvement, although potentially encouraging, merits further examination. It may be that the MA ranges of the groups tested did not cover the developmental period in which reliable differences between groups exist. Improvement in one or more of the clinical groups may be slower at an earlier point in general cognitive development, before "righting"

itself; similarly, development may tail off in those with MAs higher than the present range. Recruitment of samples with wider MA ranges than those tested here would enable these possibilities to be examined.

Finally, the recruitment of groups with overlapping CA ranges – something which was not achieved in the present study – would enable between-group comparisons of the developmental influence of CA, with regard to each STM domain. This would not only complement MA-based between-group trajectory comparisons, but also extend the within-group findings with regard to the influence of CA, discussed above.

Within-group comparisons:

The within-group analyses complemented the between-group analyses by enabling a comparison of verbal versus visuospatial STM performance patterns within each of the three groups. This is useful when assessing populations with WS and DS, as both have been associated with selective areas of STM difficulty. Comparisons plotting STM performance against MA within each group showed that both clinical groups displayed a relative impairment in STM which would be expected based on their proposed cognitive profiles. Hence, individuals with WS were less accurate on the visuospatial STM measure, and individuals with DS displayed a relative difficulty with the verbal STM task. These relationships remained consistent as cognitive development proceeded (i.e. when plotted against MA), as rates of subsequent STM development with MA did *not* significantly differ across task type (verbal, visuospatial) for either group. Further, all three groups showed clear increases in STM as MA increased, regardless of domain. This implies that STM abilities

do improve with level of cognitive development in individuals with DS and WS.

Analyses plotting STM performance in each domain against CA demonstrated a similar significant relationship between older ages and increased overall accuracy in the DS and TD groups, but this relationship between STM performance and CA was not found for individuals with WS. Non-significant interactions between CA and STM task type in all three groups further suggested that these patterns did not reliably differ across domains in either group. Therefore, for TD children, both MA and CA were, unsurprisingly, significant positive predictors of performance in each STM domain, and performance levels on both tasks were highly similar. For individuals with DS, performance on the verbal STM task was consistently lower than performance on the visuospatial STM task across development, whether assessed by MA or CA, but consistent developmental improvements were shown for both tasks. Individuals with WS differed slightly. They showed the expected poorer performance on visuospatial STM compared to verbal STM tasks, and developmental improvements in STM associated with increases in MA; but developmental increases in STM performance in relation to CA were not found.

These findings suggest that each clinical group's relative STM difficulty is manifest in a lower level of performance which remains consistent throughout development, with skills showing broadly the same relationship to both MA and CA across task domain. With regard to WS, this indicates that Jarrold and colleagues' observation of faster verbal than non-verbal/visuospatial development may not apply to STM when performance is plotted against MA or CA. In addition, it suggests that no corresponding pattern – namely, an increase in visuospatial superiority in conjunction with MA or CA - is observable for

individuals with DS. These findings were confirmed by more formal analyses, which found no significant developmental increases, in either group, in differences between z scores across domains (verbal/visuospatial STM score ‘disparities’ did not reliably increase in conjunction with either MA or CA, in any of the groups).

Despite this, both the WS and DS groups showed developmental patterns that were visibly, if not statistically, more divergent as CA increased. This particularly applied to the WS group, whose visuospatial STM by CA line-of-best-fit was “flatter” than that for verbal STM by CA, with regression analyses showing that CA missed being a significant positive predictor of greater disparity between domains in this group ($p = .099$). One explanation for this is that chronological age divergence in STM skill domains may exist, but statistically significant CA by task interactions are only found in groups with considerably wider age ranges than those in the current study. The age ranges of the present clinical groups (WS: 13 years, DS: 11 years) were markedly more restricted than that of Jarrold et al.’s sample of individuals with WS (22 years), in whom a statistically divergent pattern was observed. Further testing of STM skill separation, preferably employing a longitudinal approach rather than a cross-sectional one, is thus required before its presence in the two populations can be conclusively ruled out.

Concluding remarks:

The present study presented a comprehensive cross-sectional developmental analysis of verbal and visuospatial STM skills in individuals with WS and DS. Within- and between-group comparisons indicated that individuals with WS displayed a relative weakness in visuospatial

STM. This was in comparison to both their own verbal STM performance and the visuospatial STM performance of the other two groups. Hypothesis 1 was supported for this reason. Secondly, individuals with DS displayed a difficulty on the verbal STM measure, but only in comparison with their own visuospatial performance and not in comparison to either of the other groups. Hence, hypothesis 2 was tentatively supported. Finally, the notion of developmental divergence in verbal/visuospatial STM skills was not supported with regard to either clinical group, as disparity between the two domains did not significantly increase across development as measured by mental or chronological age. Although these means that hypotheses 3 and 4 are not formally supported, a visual inspection of the groups' performance trajectories according to CA suggests that the chronological divergence of STM skills in these populations may need further examination.

In general, the current findings indicate that, while the STM profiles of individuals with WS and DS show domain-specific weaknesses, in terms of overall performance level, which may be expected, the development of skills in both groups, across verbal and visuospatial domains, proceeds at a "typical" rate according to overall cognitive development. Secondly, the relationship between CA and STM performance in the clinical groups - particularly individuals with WS - merits further investigation.

In methodological terms, the validity of using the developmental trajectories approach to link task performance with cognitive and chronological development is further emphasised. Such a methodology gives a more nuanced picture of performance; one which – by treating development itself as a meaningful factor – enables a description of change over time which goes beyond the dichotomous *delay vs. deviance* and *intact vs. impaired* descriptive axes

underpinned by matching methods (e.g. Annaz, et al., 2008; Thomas et al., 2009). For this reason, it is recommended that studies of cognitive skills in diagnostic populations, particularly those with purportedly uneven skill profiles, should formally incorporate a measure of intellectual and/or chronological development. Procedural enhancements which the authors of any future work of this nature may wish to consider include the collection of longitudinal rather than cross-sectional data, the measurement of individuals across a greater developmental and/or chronological period, and the equation of gender ratios across groups.

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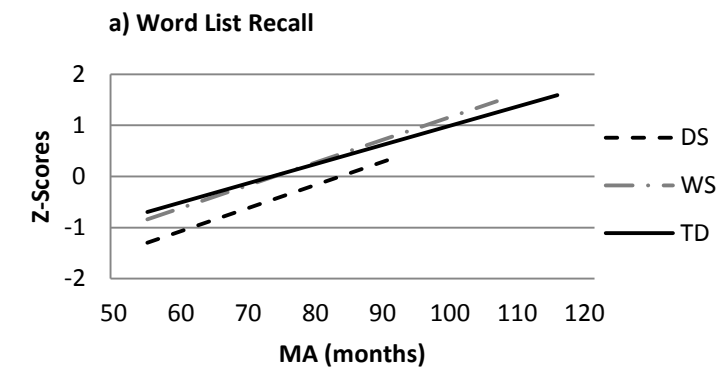
Table 1: Means, standard deviations and ranges for chronological age in months (CA), raw non-verbal ABIQ score (NVR), raw verbal IQ score (VR), and overall mental age equivalent in months (MA) by group (n=130).

	WS (n=31)			DS (n=30)			TD (n=69)		
	M (SD)	Min	Max	M (SD)	Min	Max	M (SD)	Min	Max
CA	178.32 (38.22)	98	262	174.07 (33.07)	129	257	75.07 (16.14)	48	110
NVR	14.39 (4.24)	8	24	15.30 (3.41)	10	26	15.96 (5.13)	8	25
	(max: 36)								
VR	25.71 (4.01)	20	38	22.50 (2.82)	16	29	24.80 (3.80)	18	35
	(max: 74)								
MA	76.71 (14.23)	55	107	72.03 (9.55)	55	92	78.16 (16.30)	55	116

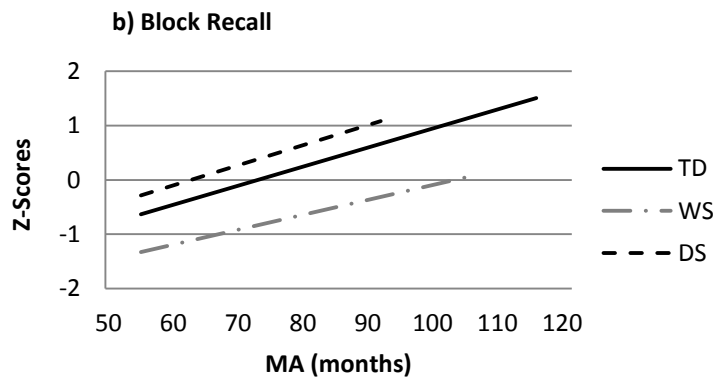
Table 2: Means, standard deviations and ranges, per group, for Word List Recall (WLR) and Block Recall (BR) raw trials correct and z scores (calculated from overall sample mean).

	WLR						BR					
	Raw			Z-Score			Raw			Z Score		
	M (SD)	Min	Max	M (SD)	Min	Max	M (SD)	Min	Max	M (SD)	Min	Max
TD	16.20 (3.38)	7	23	.17 (.96)	-2.44	2.10	20.51 (4.49)	9	31	.18 (.93)	-2.21	2.36
WS	16.03 (3.35)	8	22	.12 (.95)	-2.16	1.82	16.13 (3.89)	6	23	-.73 (.81)	-2.83	.70
DS	13.73 (3.47)	9	22	-.53 (.99)	-1.87	1.82	21.30 (4.72)	9	27	.34 (.98)	-2.21	1.53

Figure 1: Developmental trajectories for performance on a) Word List Recall and b) Block Recall, plotting the z scores of all three groups against mental age (MA) in months.



$$TD: y = 0.0375x - 2.7591, R^2 = 0.406; WS: y = 0.0444x - 3.2806, R^2 = 0.440; DS: y = 0.0453x - 3.7886, R^2 = 0.192.$$



$$TD: y = 0.0351x - 2.5607, R^2 = 0.375; WS: y = 0.0274x - 2.836, R^2 = 0.233; DS: BR: y = 0.0369x - 2.3151, R^2 = 0.129.$$

Figure 2: Developmental trajectories, per group, for both Word List Recall (WLR) and Block Recall (BR) performance, plotting z scores against mental age (MA) in months.

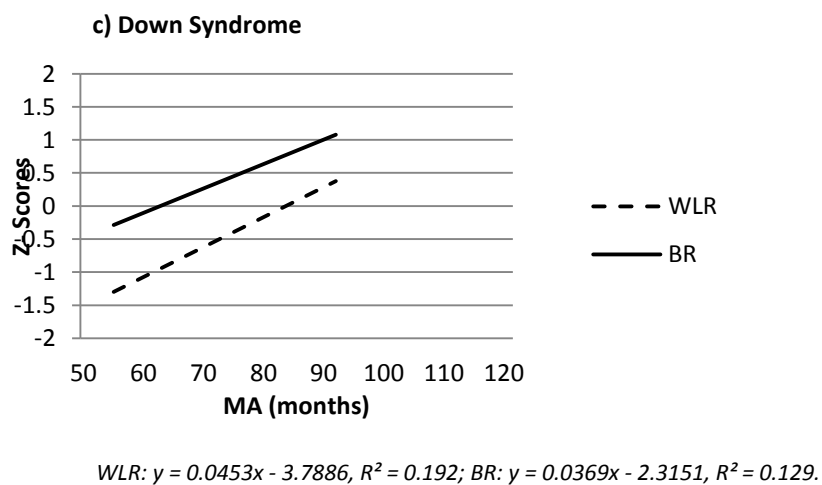
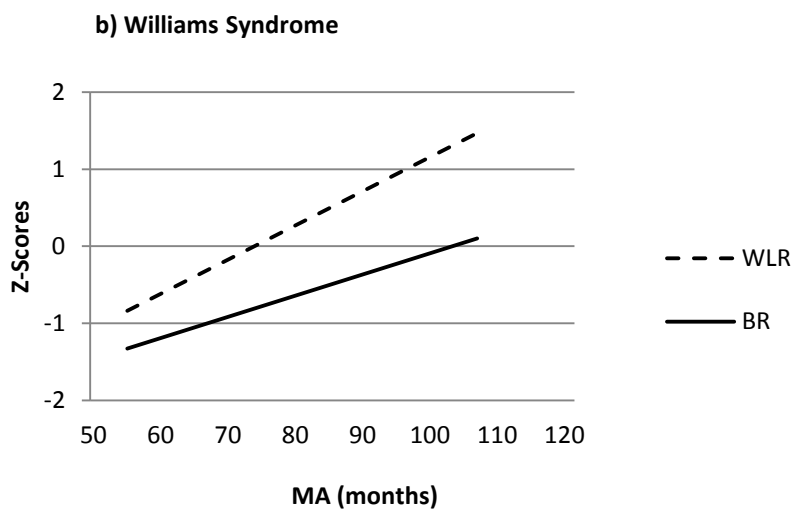
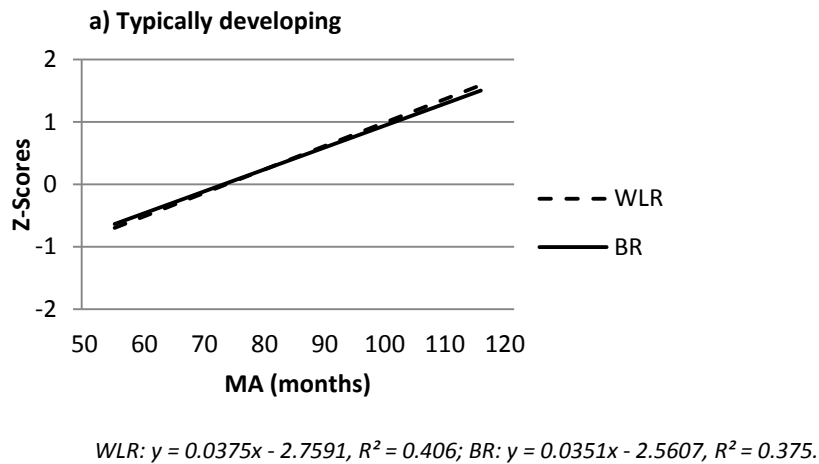


Figure 3: Developmental trajectories, per group, for both Word List Recall (WLR) and Block Recall (BR) performance, plotting z scores against chronological age (CA) in months.

