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Developmental trajectories of spatial-sequential and spatial-simultaneous working memory in Down syndrome

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Abstract

Background Working memory (WM) is generally considered an area of weakness in the cognitive profile associated with Down syndrome (DS). The great majority of studies explored WM in this population through a comparison with typical development (TD) on the basis of mental age or developmental level. However, it is also relevant to understand how these skills develop and whether such development could be more related to chronological or developmental level. In the present study, we explored cross-sectional developmental trajectories of spatial-sequential and spatial-simultaneous WM in individuals with DS across chronological age and developmental level. Typically developing children (TD) of similar mental age were also included as a comparison group.
Methods Eighty-four individuals with DS (aged between 7 and 30 years) and 327 children with TD (aged between 4 and 8 years) were administered with tasks to assess spatial-sequential and spatial-simultaneous WM, together with tasks to assess both general verbal and spatial developmental levels.
Results and conclusion Performance in spatial-simultaneous WM task was lower compared with spatial-sequential WM task in both groups. In the case of individuals with DS, the developmental trajectories of chronological age are better described through a segmented model showing increased performance until approximately 13 years of age, followed by a rather flat progress. In the case of TD children, developmental trajectories are better described through a linear model in the spatial-simultaneous WM task when chronological age is considered; in the spatial-sequential WM, the increase in performance with age was however characterised by a discontinuity at age 6. The increase in performance followed a linear pattern in both groups (DS and TD) without substantial differences between the types of measure used (verbal vs. spatial) when the developmental level is considered.

Keywords developmental trajectories, Down syndrome, spatial-sequential working memory, spatial-simultaneous working memory

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Introduction

Down syndrome (DS) is the most common genetic cause of intellectual disability (ID) (Kittler *et al.* 2008) affecting about 1 in 1000 live births (McGrother &

Marshall 1990). The vast majority of individuals with DS has characteristic features, such as craniofacial dysmorphism, low muscle tone and cardiovascular defects. A variety of other physical and medical characteristics have also been associated with the syndrome.

Down syndrome is characterised by ID, with an average IQ of 50, although high interindividual variability, in terms of the degree of impairment, has been observed (e.g. Grieco *et al.* 2015). A number of studies reported a typical profile associated with DS characterised by speech and language impairments (e.g. Chapman & Hesketh 2000), with greater difficulties in expressive compared with receptive language (Dykens *et al.* 2000). Moreover in their review of the literature, Yang *et al.* (2014) highlighted the presence of peaks and valleys also within the domain of spatial skills, within the domain of spatial skills, with areas of challenge in some areas of visuo-spatial working memory (WM), spatial visualisation, mental rotation and wayfinding.

Working memory in Down syndrome

Processing ability is expressed by WM, a temporary storage that allows information to be maintained and manipulated over a short period of time (e.g. Baddeley 1992), and is distinguished in verbal and visuo-spatial components. Concerning the verbal domain, an impairment in relation to mental age was shown in children (Jarrold & Baddeley 1997), adolescents (Marcell & Weeks 1988; Hulme & Mackenzie 1992) and adults (Numminen *et al.* 2001; Kittler *et al.* 2004, 2008), even in comparison with individuals with other intellectual disabilities (Jarrold & Baddeley 1997; Jarrold *et al.* 2002). Several studies conducted in the 1990s directly compared verbal and visuo-spatial WM through digit and Corsi span tasks – i.e. remembering digits or sequences of positions in the same order, respectively – to assess verbal and visuo-spatial WM. Results in these studies underlined the evidence of a specific impairment in verbal WM with the relative preservation of visuo-spatial WM (e.g. Wang & Bellugi 1994; Jarrold & Baddeley 1997; Jarrold *et al.* 1999).

Given that visuo-spatial WM can be differentiated in further sub-components (Logie 1995), different studies analysed visual and spatial aspects of WM in individual with DS in depth, showing a non-homogeneous profile. Some studies suggested an impairment in the former and a relative preservation in the latter in individuals with DS (e.g. Ellis *et al.* 1989; Laws 2002). Going deeper into the spatial component of WM, several studies suggested a further distinction between spatial-simultaneous and spatial-sequential components (e.g. Denis *et al.* 1999; Cornoldi & Vecchi, 2003b) based on the presentation format of the spatial locations. These locations have shown to affect performance and can be sequential in one case and simultaneous in the other and has been shown to affect performance. Some studies confirmed this distinction both in typical development (TD; e.g. Mammarella *et al.* 2008) and in atypical development (e.g. Lanfranchi *et al.* 2015a). This distinction and difference in managing sequential and simultaneous information in WM was confirmed also by studies carried out in individuals with DS. In a first study, Lanfranchi *et al.* (2009) examined the performance of participants with DS in spatial-sequential and spatial-simultaneous WM tasks while also distinguishing between passive (requiring only storage) and active tasks (requiring both storage and manipulation of material; see Cornoldi & Vecchi 2003a, 2003b). Results showed that individuals with DS performed more poorly in spatial-simultaneous tasks than TD children with the same mental age, but not in spatial-sequential WM tasks. Additionally, the differences were more evident for active tasks than for passive ones. To better understand specific impairment in spatial-simultaneous WM, Carretti and Lanfranchi (2010) explored the impact of chunking with a matrix recall task in individuals with DS. The task involved two conditions: one in which the cells to be remembered were grouped to form a pattern (structured condition) and the other consisting in a random disposition of the cells in the matrix (no structured condition). Participants with DS performed worse than TD children with the same mental age in both conditions. However, they were able to take advantage of the structured condition, although to a lesser extent than TD children. This difficulty of taking advantage of chunking seems to be specific for spatial-simultaneous WM and does not appear in spatial-sequential WM (Carretti *et al.* 2013).

To summarise, previous studies suggested relative preservation in spatial-sequential WM in individuals with DS, as opposed to a relative impairment in

spatial-simultaneous WM (Lanfranchi *et al.* 2009; Carretti *et al.* 2013; Lanfranchi *et al.* 2015b; see also Doerr *et al.* 2019). Several causes of this impairment were proposed: from perceptual problems, such as reduced visual acuity and contrast sensitivity (e.g. Courage *et al.* 1994), to difficulties in processing more than one item at a time (e.g. Doerr *et al.* 2019 for a review), to problems in using and generalising strategies (see also Mento *et al.* 2019). Of course, it is possible that more than one of these explanations could be true at the same time.

Developmental trajectories approach

All the studies mentioned earlier were conducted with the same methodology, comparing a group of individuals with DS, more or less heterogeneous in age, with a group of TD children matched for mental age. Nonetheless, considering also high interindividual variability in DS (e.g. Karmiloff-Smith *et al.* 2012), this approach is static and does not allow variability to be examined as a function of age. The analyses of developmental trajectories allow the development of a specific skill to be described in time. Indeed, a growing body of literature demonstrated that differences between neurodevelopmental disorders and TD are not only quantitative but also qualitative (e.g. Karmiloff-Smith 2000).

Trajectories have proven to be quite beneficial for studying developmental trends in both typical and atypical populations. Thomas *et al.* (2009) reviewed different examples of the applicability of developmental trajectories in cognitive tasks, in particular with TD, DS and Williams syndrome. The authors underlined the efficacy of this approach, which focuses on changes in a specific population over time, linking variables, such as performance and chronological age in longer time frames. It is also possible to compare performance in different tasks and verify the extent to which performance could be explained by development.

The developmental trajectories approach has been already employed to explore the development of individuals with DS in several domains such as engagement behaviour (Adamson *et al.* 2009), reading (Steele *et al.* 2013), general cognitive, language, motor and socio-emotional development (Arango *et al.* 2018), as well as memory (Carney *et al.* 2013). For example, considering general developmental trajectories, Arango *et al.* (2018) showed that the changes described by developmental trajectories in cognitive, language, motor and socio-emotional skills were slower in individuals with DS compared with TD children. Moreover, the speed of development in these children seemed to be influenced by parental socio-economic status.

Focusing on memory, Carney *et al.* (2013) applied the cross-sectional developmental trajectories approach to examine verbal and spatial WM in DS. The discrepancy between verbal (assessed with the word list recall task) and spatial (assessed with the Corsi Block task) WM was confirmed, with performance in line with mental age in the spatial domain and lower performance compared with mental age in the verbal domain. This discrepancy resulted to be constant across development, considering chronological or mental age. Both verbal and spatial WM showed a linear trajectory based on chronological age, with performance increasing until approximately 20 years. A similar linear trajectory was seen also with mental age until approximately 8 years. The amount of the increase was similar in the two considered WM memory components. The study of Carney *et al.* (2013) offers interesting insight on developmental trajectories of spatial-sequential WM, because the Corsi span task requires to remember sequentially presented positions. However, there is no evidence of the spatial-simultaneous component so far. To date, no direct comparison between spatial-sequential and spatial-simultaneous WM has been carried out.

Only one study examined the latter issue considering individuals with TD (see Roberts *et al.* 2018). This study showed that performance in both spatial-sequential and spatial-simultaneous WM increased linearly until adolescence and that later profiling changed as a function of the component; the performance in spatial-simultaneous WM tended to diminish in late adolescence or early adulthood (around 18 years of age), while for spatial-sequential WM, it followed an unusual pattern with a slight decline in mid-adolescence (12–14 years of age) and an increase in early adulthood.

The current study

Considering the previous literature on the topic, the aim of the present study was to further analyse spatial

WM in children, adolescents and adults with DS using a cross-sectional developmental trajectory approach. In particular, we focused on developmental trajectories of spatial-sequential and spatial-simultaneous components. Differences in these two components of spatial WM were analysed in individuals with DS and in a large group of children with TD, aged between 3 and 8 years. Specifically, three variables – chronological age, verbal and non-verbal developmental levels – were considered. Their influence on the development of spatial-sequential and spatial-simultaneous WM was analysed in order to describe the development of such WM components in a comprehensive way. In particular, we aimed to understand whether developmental trajectories in spatial-sequential and spatial-simultaneous WM are comparable or whether differences could emerge. The trajectories were analysed considering chronological age first, then they were estimated in function of the developmental level, which was computed using verbal (vocabulary) and spatial (fluid reasoning) measures. Verbal and spatial developmental levels were considered separately, in consideration of the profile associated with DS. Keeping verbal and spatial developmental levels distinct allowed to ascertain the variations of the developmental trajectories in spatial-sequential and spatial-simultaneous WM according to which aspect (verbal vs. spatial) of developmental level is considered.

Methods

Participants

Eighty-four participants with DS (46.43% females, age range 7–30) participated in the study. Fourteen participants were under age 10, thirty-two were aged between 10 and 15, seventeen between 15 and 20, eleven between 20 and 25 and nine between 25 and 30 years. Moreover, 327 children (38.84% of females, age range 4–8) with TD were included as a comparison group. All participants had Western European origins and a medium socio-economic status according to their living area. Based on Thomas *et al.* (2009), the typically developing comparison group was identified, including participants with a chronological age range that overlapped with the mental age range of the group with DS.

All participants were selected from previously published studies. Regarding the group of individuals with DS, 29 participants took part in a series of studies on spatial cognition and wayfinding (Meneghetti *et al.* 2018; Toffalini *et al.* 2018), while 34 participants were selected from a previous study on spatial WM (Lanfranchi *et al.* 2009). Finally, 21 participants were selected from unpublished data on spatial WM. Children with TD were taken from a larger study analysing the structure of WM (Carretti *et al.* under review). Data on chronological and mental of the two-group age are reported in Table 1.

Material

Verbal developmental level

Peabody Picture Vocabulary Test-Revised. This test measures receptive vocabulary in children from 4 to 11 years of age (Dunn & Dunn 1981; Italian adaptation by Stella *et al.* 2000). It consists of a series of 175 pictorial stimuli of increasing difficulty. Each stimulus consists of four black-and-white drawings, which are shown to the child while the experimenter pronounces the word out loud. The task requires to indicate which of the four drawings best represents the meaning of the word. The task ends after six errors are made within eight consecutive responses. The reported test–retest reliability for the Italian adaptation (Stella *et al.* 2000) is good (r = 0.88). The final score is the total number of correctly identified drawings (ranging between 0 and 175), but it was transformed into an age-equivalent score for interpretive purposes linked with the goals of the present study. Note that the age-equivalent score was a linear transformation of the total raw score,[1] with an increase of approximately 9.4 correct items for every

[1]Note that age-equivalent scores were not only linearly related, but they were virtually linear transformations of the raw scores, for both PPVT-R (R^2 = 0.97) and CPM (R^2 = 0.95). In other words, the growth of the average raw score with chronological age is linear, in a nearly perfectly deterministic way, within the age range represented by the age-equivalent scores considered. This averts the psychometric problems sometimes raised using age-equivalent/mental-age scores, due to the lack of guarantee of them being on an interval scale.

Table 1 Descriptive statistics of the two groups

	Chronological age		CPM age equivalent		PPVT-R age equivalent	
Group	**DS**	**TD**	**DS**	**TD**	**DS**	**TD**
Mean	15.95	5.96	5.58	7.21	4.35	5.94
Std. deviation	6.06	1.09	1.31	1.76	2.05	1.97
Age range	7.00–30.75	4.00–8.33	3.25–7.75	3.25–10.75	2.00–8.75	3.09–11.42

CPM, Colored Progressive Matrices; DS, Down syndrome; PPVT-R, Peabody Picture Vocabulary Test-Revised; TD, typical development.

+1 year of age (starting from a score of about 60 at age 3) within the age range considered.

Visuo-spatial developmental level

Raven's Colored Progressive Matrices. This task measures fluid reasoning using items of a visuo-spatial nature (Raven *et al.* 1992; Italian adaptation by Belacchi *et al.* 2008). It consists of 36 coloured matrices of increasing complexity. Each matrix has a missing piece and the respondent is asked to choose the best fit for the missing piece, among six options. The reliability is good: the test–retest stability and convergent validity with other intelligence tests is strong in all international versions of the Colored Progressive Matrices (CPM), with r in 0.60–0.90 (Belacchi *et al.* 2008). The final score is the number of correctly completed matrices (ranging between 0 and 36). As for the Peabody Picture Vocabulary Test-Revised (PPVT-R), the latter was transformed into an age-equivalent score for interpretive purposes. Once again, the age-equivalent score was a virtually perfect linear transformation of total raw scores (see footnote 1), with an increase of approximately 2.7 correct items for every +1 year of age (starting from a score of about 9 at age 3) within the age range considered.

Working Memory Matrices – spatial-sequential and spatial-simultaneous. This task consists of a series of matrices presented on a sheet of paper (Lanfranchi *et al.* 2004). Each matrix has a 3 × 4 to 4 × 5 layout, which increases with the increased level of the task (see Carretti *et al.* 2013). Each cell of the matrix is a square with a side measuring 3 cm. Two trials are presented for each level of difficulty (memory span). Levels range from 1 to 8, corresponding to the number of elements that have to be recalled. In the *spatial-sequential* condition, a pathway is presented with positions pointed one after the other. In the *spatial-simultaneous* condition, all target cells are presented simultaneously, with different colours. In both conditions, the child has to respond immediately after the presentation of the trial by indicating the pointed or coloured squares on a blank matrix. One point is given only if the child indicated correctly all the pointed or coloured squares. The task ends when the child fails both trials at the same level of difficulty. The final score for each condition is the number of correctly performed trials.

Results

Data analysis

Statistical analyses were carried out with R software, version 4.0.3 (R Core Team 2019). Descriptive statistics were reported with mean and standard deviations. WM performance (spatial-sequential and spatial-simultaneous) was evaluated in both DS and TD groups as a function of chronological age through null, linear models and segmented regression models. The latter, also known as piecewise regression, represents linear models with breakpoints so that the independent variable is partitioned into intervals with different regression coefficients. The 'segmented' library (Muggeo 2008) of R was used to fit those models. For any purpose of model selection, the best fitting model was chosen according to the Akaike information criterion (AIC, lower is better; Akaike 1973).

Subsequently, we analysed WM performance as a function of age-equivalent scores of PPVT-R and CPM tests (representing measures of verbal and

spatial development), comparing both groups through linear models.

The 'compute.es' R package (Del Re 2013) was used to calculate effect sizes (Cohen's d; Cohen 1988) related to age-equivalent and WM scores.

Spatial-sequential and spatial-simultaneous working memory

Descriptive statistics and paired samples *t*-tests for spatial-sequential and spatial-simultaneous WM tasks were calculated (Table 2). A higher mean score was found in spatial-sequential WM compared with spatial-simultaneous in both groups of individuals with DS, $t(83) = 5.36$, $P < 0.001$, Cohen's $d = 0.46$, and TD group, $t(326) = 5.57$, $P < 0.001$, Cohen's $d = 0.36$.

Spatial-sequential working memory, spatial-simultaneous working memory and chronological age across groups

Performance in spatial-sequential and spatial-simultaneous WM related to chronological age was examined by means of regression models with null, linear and segmented relationships. A series of regressions were calculated, considering DS and TD groups separately because the age range in each group was not comparable. The final estimated effects are depicted, along with the scatter plots of all observations, in Fig. 1.

In the DS group, spatial-sequential WM was best explained by the segmented model (AIC = 372.99), which fitted better than the null model (AIC = 380.08), $F_{3,80} = 4.49$, $P = 0.006$, and better than the linear model (AIC = 380.40), $F_{2,80} = 5.82$, $P = 0.004$. Therefore, the segmented model was ultimately kept. The estimated breakpoint was at age 13.67 [95% confidence interval (CI): 10.99, 16.34]. The overall segmented model had $R^2 = 0.14$. The estimated relationship before the breakpoint was $B = 0.50$, SE = 0.16, $P = 0.004$, $R^2 = 0.21$. The estimated relationship after the breakpoint was $B = -0.07$, SE = 0.06, $P = 0.277$, $R^2 = 0.03$ (Fig. 1a). Also, in spatial-simultaneous WM, in the DS group, the segmented model (AIC = 389.10) fitted better than the null model (AIC = 391.87), $F_{3,80} = 2.94$, $P = 0.038$, and better than the linear model (AIC = 392.89), $F_{2,80} = 3.89$, $P = 0.024$. Therefore, the segmented model was ultimately kept. The estimated breakpoint was at age 13.50 [95% CI: 10.15, 16.85]. The overall segmented model had $R^2 = 0.10$. The estimated relationship before the breakpoint was $B = 0.31$, SE = 0.19, $P = 0.103$, $R^2 = 0.09$. The estimated relationship after the breakpoint was $B = -0.09$, SE = 0.07, $P = 0.203$, $R^2 = 0.03$ (Fig. 1b).

In the TD group, spatial-sequential WM was best explained by the segmented model (AIC = 1107.26), which fitted better than the null model (AIC = 1232.79), $F_{3,323} = 53.31$, $P < 0.001$, and better than the linear model (AIC = 1115.11), $F_{2,323} = 5.96$, $P = 0.003$. Therefore, the segmented model was ultimately kept. The estimated breakpoint was at age 6.33 [95% CI: 5.67, 6.99]. The overall segmented model had $R^2 = 0.33$. The estimated relationship before the breakpoint was $B = 1.16$, SE = 0.16, $P < 0.001$, $R^2 = 0.19$. The estimated relationship after the breakpoint was $B = 0.24$, SE = 0.18, $P = 0.173$, $R^2 = 0.02$ (Fig. 1c). However, we can consider this segmentation as an effect of the fact that, after the age of 6.30, most of the children have a performance near to the ceiling. Instead, spatial-simultaneous WM was best explained by a linear model (AIC = 1367.96), which fitted better than the null model (AIC = 1453.65), $F_{1,325} = 99.96$, $P < 0.001$. At the

Table 2 Working memory performance across groups

	Spatial-sequential WM		Spatial-simultaneous WM	
	DS	**TD**	**DS**	**TD**
Mean	5.06	5.79	3.98	5.09
SD	2.28	1.59	2.45	2.22

DS, Down syndrome; SD, standard deviation; TD, typical development.

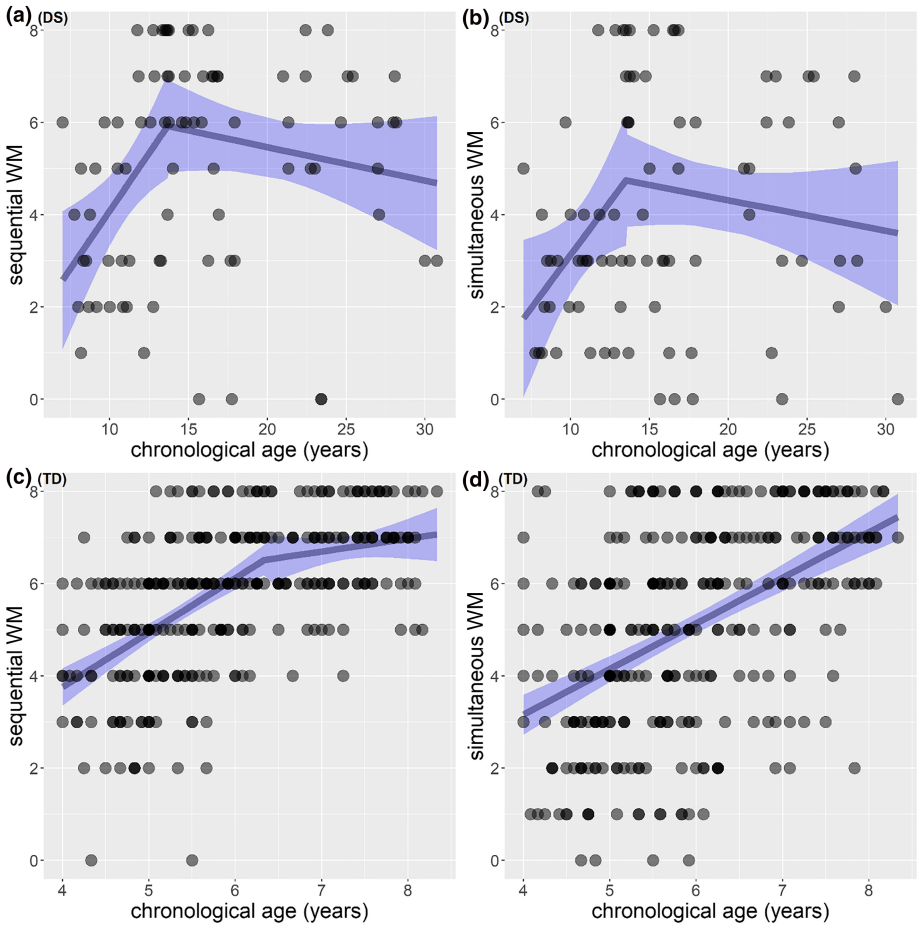

Figure 1. Regression models explaining progress in sequential and simultaneous WM in both groups separately. (a) Segmented model, (b) segmented model, (c) segmented model and (d) linear model. DS, Down syndrome; TD, typical development; WM, working memory. [Colour figure can be viewed at wileyonlinelibrary.com]

same time, the segmented model did not fit better than the linear model (AIC = 1367.12), $F_{2,323} = 2.41$, $P = 0.091$. Therefore, the linear model was kept to avoid overfitting of data. The estimated relationship was $B = 0.99$, SE = 0.10, $P < 0.001$, $R^2 = 0.23$ (Fig. 1d).

Spatial-sequential working memory, spatial-simultaneous working memory and verbal developmental level

Two linear models were run to examine the relationship between WM and developmental verbal (PPVT-R) age-equivalent scores, comparing DS and TD groups (Fig. 2). Specifically, in this case, group (DS vs. TD) was inserted as a factor in the model; the interaction between group type and developmental level on WM scores was examined as well.

In sequential WM, the linear model (AIC = 1572.46) outperformed the null model (AIC = 1630.56), $F_{2,407} = 33.19$, $P < 0.001$, but the segmented model outperformed the linear model (AIC = 1569.12), $F_{2,405} = 3.65$, $P = 0.027$. Nonetheless, the linear model was kept because the segmented model failed to identify a breakpoint within the range of scores of both groups. The estimated coefficient for age was $B = 0.28$ [95% CI: 0.11, 0.45], SE = 0.09, $P = 0.001$. The estimated

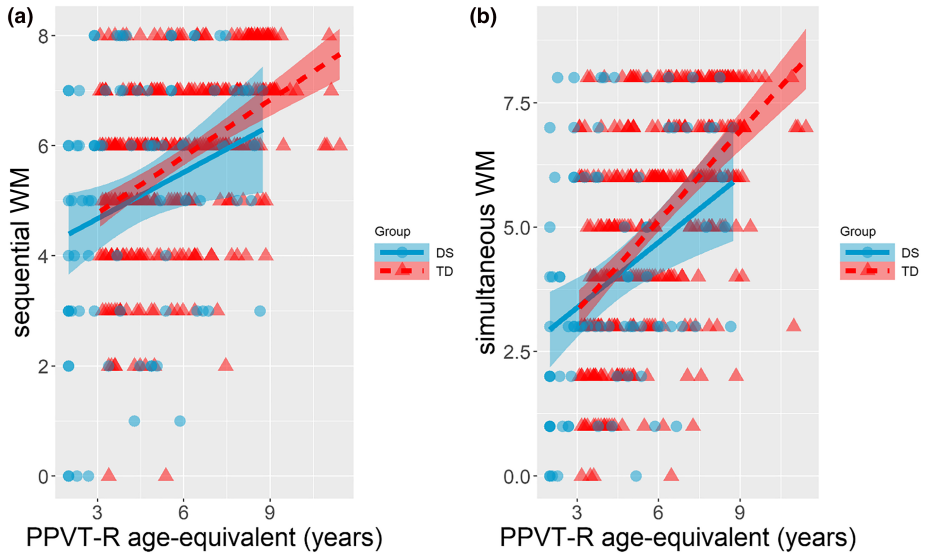

Figure 2. Linear regressions for WM and verbal developmental level, according to each group. DS, Down syndrome; PPVT-R, Peabody Picture Vocabulary Test-Revised; TD, typical development; WM, working memory. [Colour figure can be viewed at wileyonlinelibrary.com]

coefficient for the group factor was $B = -0.08$ [95% CI: −1.08, 0.91], SE = 0.51, $P = 0.867$, suggesting no between-group difference at the intercept. The estimated coefficient for the interaction between age and group was $B = 0.06$ [95% CI: −0.13, 0.25], SE = 0.10, $P = 0.532$. Therefore, the model suggested a similar increase of sequential WM with verbal developmental level in both groups. The model had $R^2 = 0.16$ (Fig. 2a).

In simultaneous WM, the linear model (AIC = 1732.18) fitted better than the null model (AIC = 1844.70), $F_{2,407} = 66.70$, $P < 0.001$. At the same time, the segmented model did not fit better than the linear model (AIC = 1730.58), $F_{2,405} = 2.78$, $P = 0.063$. Therefore, the linear model was kept to avoid overfitting of data. The estimated coefficient for age was $B = 0.44$ [95% CI: 0.23, 0.65], SE = 0.10, $P < 0.001$. The estimated coefficient for group was $B = -0.53$ [95% CI: −1.74, 0.68], SE = 0.62, $P = 0.388$. The interaction coefficient of age with group was $B = 0.16$ [95% CI: −0.07, 0.39], SE = 0.12, $P = 0.182$, again suggesting no relevant between-group difference at the intercept. Also in this case, the model showed a similar increase of sequential WM with verbal developmental level in both groups. The model had $R^2 = 0.27$ (Fig. 2b).

Spatial-sequential working memory, spatial-simultaneous working memory and spatial developmental level

Two regression models were run to examine the relationship between WM and developmental spatial (CPM) age-equivalent scores, comparing DS and TD participants (Fig. 3).

In sequential WM, the linear model (AIC = 1538.82) fitted better than the null model (AIC = 1630.56), $F_{2,407} = 53.38$, $P < 0.001$. At the same time, the segmented model did not fit better than the linear model (AIC = 1540.49), $F_{2,405} = 1.15$, $P = 0.318$. Therefore, the linear model was kept to avoid overfitting of data. The estimated coefficient for age was $B = 0.60$ [95% CI: 0.35, 0.86], SE = 0.13, $P < 0.001$. The estimated coefficient for group was $B = 0.85$ [95% CI: −0.79, 2.49], SE = 0.83, $P = 0.311$. The interaction coefficient of age with group was $B = -015$ [95% CI: −0.43, 0.12], SE = 0.14, $P = 0.275$. Therefore, the model showed a similar increase of sequential WM with spatial developmental level in both groups. The model had $R^2 = 0.23$ (Fig. 3a).

In simultaneous WM, the linear model (AIC = 1775.18) fitted better than the null model

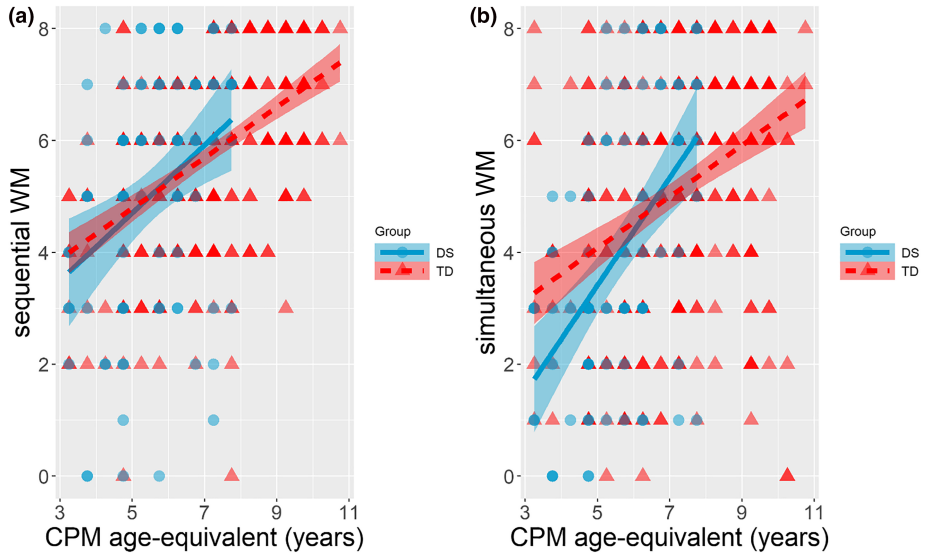

Figure 3. Linear regressions for WM and spatial developmental level, according to each group. CPM, Colored Progressive Matrices; DS, Down syndrome; TD, typical development; WM, working memory. [Colour figure can be viewed at wileyonlinelibrary.com]

(AIC = 1844.70), $F_{2,407} = 39.87$, $P < 0.001$. At the same time, the segmented model did not fit better than the linear model (AIC = 1775.51), $F_{2,405} = 1.81$, $P = 0.164$. Therefore, the linear model was kept to avoid overfitting of data. The estimated coefficient for age was $B = 0.96$ [95% CI: 0.62, 1.30], SE = 0.17, $P < 0.001$. The estimated coefficient for group was $B = 3.17$ [95% CI: 0.98, 5.36], SE = 1.11, $P = 0.005$, suggesting a lower intercept for the DS as compared with the TD group. The age by group interaction coefficient, however, was $B = -0.50$ [95% CI: −0.87, −0.13], SE = 0.19, $P = 0.007$, suggesting the possible presence of an effect that reduced the regression coefficient in the TD group. The latter may be due to a higher intercept combined with a partly ceiling effect in the TD group in this variable. The model had $R^2 = 0.19$ (Fig. 3b).

Discussion

The aim of the present study was to explore spatial WM in individuals with DS more in depth, considering the distinction between the spatial-simultaneous and spatial-sequential components, in relation with age (chronological or developmental level).

In particular, the developmental trajectories of these two components were analysed in a range of participants with DS aged between 7 and 30 years and compared with those of TD children with similar mental age, aged between 4 and 8 years. The design was cross-sectional; therefore, different participants were tested based on the different ages. To our knowledge, this is one of the few studies analysing developmental trajectories, in particular those of WM. As previously mentioned in the introduction, Carney *et al.* (2013) considered the developmental trajectories of verbal and spatial WM, reporting a linear increase with chronological age, with performance increasing until approximately 20 years, and mental age, until approximately 8 years. Carney *et al.* (2013) did not find differences in the slope of the increase between the two WM components. Nonetheless, it is noteworthy that only the linear term was tested in both chronological and mental age, whereas some studies in the literature on TD suggested the presence of non-linear trajectories (see Roberts *et al.* 2018).

First of all, our data confirmed better performance in spatial-sequential compared with spatial-simultaneous WM in individuals with DS; a similar pattern of results has been found also in TD. These results are in line with those reported in previous studies (e.g. Lanfranchi *et al.* 2009; Carretti & Lanfranchi 2010), although in the present work we

did not directly compare the performance of the two groups due to differences in numerosity and mean developmental level.

Secondly, our study examined cross-sectional developmental trajectories of spatial WM in individuals with DS in comparison with TD children. Considering chronological age, the segmented regression model fitted the data better for both spatial-sequential and spatial-simultaneous WM in individuals with DS. In particular, a similar trajectory emerged with chronological age for both spatial WM components with an increase until approximately 13 years of age: after such period, the curve tended to flatten. Therefore, after this age, spatial-simultaneous and spatial-sequential WM tend to be stable and no further improvements can be seen. In the case of children with TD, it emerged that spatial-simultaneous WM performance is linearly associated with age, whereas in the spatial-sequential task, there was a discontinuity after age 6, probably due to a partial ceiling effect in this task in older children. However, the age range considered in the current study was more restricted compared with previous studies (e.g. Roberts *et al.* 2018 or Pickering *et al.* 2001); therefore, it was unable to make direct comparisons, except for the fact that the performance increased in both WM tasks.

Comparing developmental trajectories of individuals with DS with TD literature, a pattern of results with similarities and differences emerged. In fact, similarly to what was found by Roberts *et al.* (2018) in TD, we found a linear increase in the group of individuals with DS in both spatial components until adolescence. Moreover, the age at which an increase in performance was observed was similar to TD for the sequential component (approximately 13 years), while it was precocious for the simultaneous component, considering that the development stops at approximately age 18 in TD. No further increase was observed in individuals with DS in the spatial-sequential component in early adulthood as in TD.

To sum up, developmental trajectories in DS are similar to TD until early adolescence, while they are different in late adolescence and adult age. The earlier developmental stop seen in DS is coherent with data suggesting an early decline of cognitive development (Dykens *et al.* 2000). However, on the basis of this literature, we would have expected a more marked decline of WM performance in adolescence and adulthood. This could be explained by the fact that our adolescent and adult participants were recruited in daily centres for individuals with ID, where participants were regularly involved in several activities and autonomies that could have contributed to contrasting the decline of spatial WM. Of course, more studies with larger samples are needed in order to better clarify this aspect. Indeed, the lack of clear decline can also be influenced by the restricted number of participants in the older age range.

When developmental trajectories were analysed in function of developmental level, the results showed that linear regression models fit better, both for sequential and simultaneous components, in both individuals with DS and TD children; from a descriptive point of view, the trajectories of the two groups overlapped. Interestingly, our results showed that spatial-sequential and spatial-simultaneous WM increase in individuals with DS as the developmental level increases, both considering the verbal, and especially the visuo-spatial, domain, similarly to what happens in TD. In our hypothesis, we expected a greater relationship of both WM components with the spatial developmental level, considering the fact that all these measures tap the spatial domain and that such domain represents an area of relative strength in the DS cognitive profile. However, it is interesting to note that the verbal developmental level is also related to spatial WM.

Similar results were found by Carney *et al.* (2013) who analysed the discrepancy between verbal and spatial (sequential) memory: their data suggest the hypothesis that the developmental trajectory of all WM components might be similar in DS, although a more comprehensive study considering all WM components would be suitable in order to directly compare developmental trajectories.

From a practical point of view, our data suggest that both TD and individuals with DS could benefit from the sequential presentation of simple visual material to memorise. Moreover, our data suggest that it is important to work not only on WM specifically but also to contextually support and foster verbal and visuo-spatial cognition, in order to increase visuo-spatial performance in individuals with DS (e.g. Carney *et al.* 2013).

Despite these results being new and interesting, some limitations must be mentioned. The main limitation of the study consists in the fact that the reported data are cross-sectional and not longitudinal, which prevents a more detailed account of the precise development of spatial WM. Future studies assessing spatial WM longitudinally should be conducted in order to better describe developmental trajectories in DS. This is important both from a theoretical point of view and also from an applied perspective, in order to understand which aspects should be targeted through intervention.

To conclude, our study suggests that the developmental trajectories of spatial WM follow different pathways depending on the performance being plotted against chronological age or developmental level. In the former case, the development is better described through a segmented trajectory with a peak followed by a flat course, whereas in the latter case (developmental level), a linear increase clearly emerges. Interestingly, the linear increase is similar when both verbal and visuo-spatial general abilities are used.

These results offer new insight on developmental changes in WM in individuals with DS and on the factors that are involved in these changes.

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Data availability statement

The data that support the findings of this study are openly available in figshare at https://doi.org/10.6084/m9.figshare.17032628.v1 reference number: 17032628.

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Conflict of interests

The authors declare no conflict of interest.

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