

Towards an integrative model accounting for typical and atypical development of visuospatial short-term memory

Short title: Development of visuospatial STM

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Abstract

The origin of visuospatial short-term memory (STM) impairment is poorly investigated and is generally considered to be the result of a more global visuospatial deficit. However, previous studies suggest an important influence of two elements on performance in visuospatial STM tasks, the mode of presentation (i.e. simultaneous and sequential), and the visuospatial arrangement (structured vs. unstructured). With regards to a recent proposal, the aim of this study was to examine the development of the two modes of presentation and the visuospatial arrangement of visuospatial information in STM in a hundred typically developing participants aged from 4 years old to adults. Moreover, we also examined how the model explains the pattern of visuospatial STM deficit in two neurodevelopmental syndromes with different profiles in terms of STM abilities, namely Williams syndrome and Down syndrome. We found distinct performance for sequential and simultaneous presentation only from 11 years old with better performance in simultaneous than in sequential presentation mode and a sensitivity to visuospatial arrangement that increases with age. Both syndromes presented deficits at different levels, people with Williams syndrome for visuospatial arrangement and with Down syndrome for simultaneous visuospatial information in STM. The results demonstrate the importance to consider the influence of preexisting visuospatial knowledge on STM abilities. A two processing route model of STM is an interesting framework to interpret the different results.

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Introduction

While the development of verbal short-term memory (STM) has attracted broad interest, the development of visuospatial STM has been much less investigated. In the last 20 years, significant efforts have been made to specify the nature of the processing involved in visuospatial STM. The continuity model (Cornoldi & Vecchi, 2003) proposed two continua, one vertical continuum, where WM tasks can be more or less passive or active (passive condition corresponding to the simple recall of information without active processing and manipulation) and one horizontal considering three components, a verbal component, a pure visual component processing visual information (i.e. colors, shapes) and a spatial component. This model considers that the spatial STM component may be further subdivided into two independent memory subsystems, depending on the mode of presentation of the stimulus: (1) the simultaneous component which is involved in the retention of simultaneously presented spatial information (i.e. remembering the position of dots presented all at once in a matrix), and (2) the sequential processing component which would be recruited when remembering the positions of elements presented sequentially (i.e. remembering the position of dots presented one by one in a matrix) (see for example Cornoldi & Vecchi, 2003; Mammarella, Pazzaglia, & Cornoldi, 2008).

Several studies provide evidence for different behavioral and developmental signatures for the simultaneous and the sequential components of spatial STM. Indeed, several data, mainly from patients, demonstrated that the mode of presentation could have a differential impact on performance in spatial STM tasks and that both, simultaneous and sequential components, could be considered as independent. Recently, Mammarella, Lucangeli, and Cornoldi (2010) found that children with nonverbal learning disabilities (with specific impaired visuospatial skills such as psychomotor, visuo-constructive skills and mathematics) presented specific difficulties in temporarily holding and manipulating spatial information in

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memory compared to typically developing children but only when spatial information was presented sequentially, and not when it was presented simultaneously. In typically developing children, Pickering, Gathercole, Hall, and Lloyd (2001) demonstrated greater development, in terms of performance, of STM abilities in simultaneous than in sequential STM task between 5 and 10 years of age (see also Logie & Pearson, 1997). In addition, recent neuropsychological studies have shown a dissociation between sequential and simultaneous processing in Down syndrome (Carretti & Lanfranchi, 2010) as well as in neglect patients (Wansard et al., 2015) showing a double dissociation. Thus, the mode of presentation seems to have a differential impact on performance and needs to be considered in cognitive models of visuospatial STM.

In addition to the mode of presentation, the arrangement of visuospatial information was found to have a significant impact on performance. In both typically developing children and in healthy adults, larger visuospatial STM spans were reported when the positions of the stimuli formed a regular, structured configuration, (i.e. a configuration respecting the gestalt principles of symmetry, repetition and continuation) than when the individual elements did not form any pattern (Imbo, Szmalec, & Vandierendonck, 2009; Kemps, 2001). Moreover, in a sequential visuospatial STM task, adolescents were found to take more benefit from structured arrangements than younger children, suggesting that perceiving a regularity in the stimuli arrangement supports the development of visuospatial STM performance (Imbo et al., 2009). This arrangement effect could reflect the positive influence of long-term memory knowledge on the temporary retention of visuospatial information, with the long-term visuospatial representation acting in support of the maintenance in STM. Moreover, structured arrangements could also provide better conditions for deploying memory strategies such as chunking or grouping of visuospatial information (i.e., the first three and the last at the top right corner).

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Interestingly, the arrangement effect was found to interact with the mode of presentation. In adults, Pieroni, Rossi-Arnaud, and Baddeley (2011) showed better performance when processing simultaneous than sequential visuospatial information only when the arrangement was structured (following a regular pattern respecting the Gestalt principles of symmetry or continuity). Likewise, in children, recent data suggest that the two presentation modes lead to a differential sensitivity to the arrangement of visuospatial information. Indeed, Attout, Noël and Rousselle (2018) found a more important arrangement effect with the simultaneous presentation mode than with the sequential presentation.

These consistent interactions between arrangement and presentation mode led Attout et al. (2018) to propose an updated model for visuospatial STM accounting for both the influence of visuospatial arrangement and the interaction with the mode of presentation (see Figure 1). This alternative model also postulates the existence of two processing routes for visuospatial information in STM. However, as a crucial difference to previous models, this model considers that the two presentation modes do not involve distinct memory subsystems but different access paths to a common configural representation in STM. The simultaneous-sequential dissociation may result from the way extrinsic visuospatial information is presented and then processed to be stored in STM. For both presentation modes, access to a configural representation in STM is facilitated when visuospatial information is embedded in a regular arrangement. This arrangement effect may result from the influence of long-term visuospatial knowledge/strategies on the temporary retention of a configural representation of visuospatial information (e.g. this arrangement is like the letter “U”, or like a fork). However, visuospatial information presented simultaneously could provide a more direct access to a configural, global representation in STM. By contrast, visuospatial information presented sequentially requires intermediate processing coding; first, for the local individual position in space and second, for the order of each position in the sequence. As a result, the arrangement effect is

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assumed to be smaller with the sequential mode of presentation. This may be because the global visuospatial representation in STM, which can benefit from the arrangement, is accessed indirectly after a series of processing coding for local position and the order of each element in the sequence.

<INSERT FIGURE 1 HERE>

The present study addressed two issues pertaining to the typical and atypical development of the two processing route model. To date, there is no data describing how these two processing routes for visuospatial information in STM develop in typical children. Moreover, until now, no model has included the arrangement effect, showing however a specific impact on performance in a visuospatial STM task. Therefore, in a first experiment, we examined the development of mode of presentation and arrangement effects in visuospatial STM in four groups of typically developing children aged from 4 to 15 years-olds and one group of adults (Experiment 1). This cross-sectional design will provide an in-depth characterization of the development of visuospatial STM taking into account the influence of the presentation mode and of the visuospatial arrangement. In particular, the developmental trajectory of the arrangement effect will be examined as a function of the modes of presentation to determine when children start being sensitive to visuospatial arrangement in each mode. Based on the model, an advantage of simultaneous over sequential presentation would indicate that the sequential mode requires additional resources for serial order processing (Attout, Ordonez Magro, Szmalec, & Majerus, 2019; McCormack, Brown, Vousden, & Henson, 2000). Furthermore, the arrangement effect is expected to increase with age due to children's growing ability to use some strategies such as chunking (some object locations within a room-sized environment are chunked together) as well as their increasing

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long-term visuospatial knowledge (knowledge of different letters, shapes and objects).

Finally, considering that the simultaneous presentation mode would provide more direct access to configural information, the arrangement effect is expected to be larger with this mode of presentation than with the sequential one.

In a second experiment, we examined how this model could be useful to characterize deficits in visuospatial STM in atypical development. Previous neuropsychological data shed light on specific visuospatial STM profiles by considering the arrangement of stimuli and/or the presentation mode. For example, a recent study from Attout et al. (2018) showed that participants with 22q11.2 deletion syndrome, who exhibit visuospatial processing deficits, took less advantage of the visuospatial arrangement to strengthen the trace in STM in comparison to children matched on chronological age, whatever the presentation mode. This lower influence of visuospatial arrangement could be attributed to their visuospatial impairment, resulting in limited pre-existing visuospatial knowledge, difficulties in processing online visuospatial information or to improper strategy use. The present study attempted to provide additional comparative data to complete this picture. Two other genetic syndromes presenting a STM deficit, namely, Williams and Down syndromes, will be contrasted to examine whether this model could help understand the nature of their difficulties in visuospatial STM. These two syndromes have been studied since they were found to be associated with different limitations of visuospatial skills and visuospatial STM deficits (Jarrold, Baddeley, & Hewes, 1999; Wang & Bellugi, 1994).

Experiment 1

Experiment 1 aimed to examine typical development of the visuospatial STM from childhood to adulthood by considering the influence of mode of presentation and arrangement effects on performance as a function of age.

Methods

Participants

Ninety-nine participants were assessed in the present experiment. As displayed in the Table 1, they were split into four different age groups including nineteen 4- to 5-year-old children, twenty-one 6- to 7-year-old children, nineteen 8- to 10-year-old children, twenty-one 11- to 15-year-old adolescents, and a group of 19 young adults between 20 and 29 years old. Children were recruited through local middle-class schools and by word of mouth while adults were university students recruited on a voluntary basis. A background questionnaire was used to identify and exclude children with a history of neurological disorders, neurodevelopmental delays, sensory impairments, or learning deficits. The experiment was conducted in accordance with the Declaration of Helsinki and the regional ethical committee for biomedical research approved the experimental protocol (Record number: B403201111579). For minors, all parents gave written consent before the cognitive tests were administered.

<INSERT TABLE 1 HERE>

Material

Visuospatial STM was assessed with two kinds of paper-pencil tasks inspired by Imbo et al. (2009): the simultaneous visuospatial STM task in which the elements were presented all at once, and the sequential visuospatial STM task in which the to-be-remembered positions were presented sequentially one at a time. In the simultaneous STM task, participants were presented with a matrix of two to ten black dots for 5 seconds. In the sequential STM task, participants were presented with a blank matrix and were instructed to remember the positions of a series of cells touched one by one (one per second), by the examiner. In both tasks,

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participants were instructed to recall the positions by placing black tokens in the right location on a blank matrix. Within each STM task, two visuospatial arrangements were contrasted: for half of the trials, the layout of the to-be-remembered position presented no regular pattern (called *unstructured* arrangement) while for the other half, the position of the presented elements formed a regular pattern respecting the Gestalt principles of symmetry or continuity (called *structured* arrangement; based on Imbo et al., 2009). In both STM tasks, trials were of growing complexity as the size of the matrix and the number of positions to be remembered increased during the task. The lower level of difficulty corresponded to a 2×2 matrix with two locations to be recalled and the higher level included a 4×5 matrix with 10 positions to be remembered. Participants had to succeed in two trials of the same difficulty to access to a higher level (larger matrix and/or larger number of locations). The task ended when the participant failed at two out of the three trials for a given difficulty level. The practice items corresponded to the first level, a 2×2 matrix with two locations to be recalled. No time restrictions were imposed. Each correct matrix was credited with one point.

Experimental procedure

Participants were tested individually at school or at home in a quiet room. Testing was completed in two sessions and the order of the tasks was counterbalanced for the presentation mode between the two sessions but also with regard to the arrangement within a session.

Statistical procedure

Considering the recent criticisms that have been raised relating to the use of frequentist statistical methods when making statistical inferences (see e.g. Dienes, 2011; Morey & Rouder, 2011; Wagenmakers, 2007), a Bayesian approach was used in combination with the more traditional frequentist approach.

All analyses report Bayes factors (BF), which can be considered as a relative measure of statistical evidence (Morey, Romeijn, & Rouder, 2016). The strength of evidence was

interpreted by following the guidelines proposed by Jeffreys (1961) : A BF of 1 provides no evidence, $1 < \text{BF} < 3$ provides anecdotal evidence, $3 < \text{BF} < 10$ provides moderate evidence, $10 < \text{BF} < 30$ provides strong evidence, $30 < \text{BF} < 100$ provides very strong evidence and $100 < \text{BF}$ provides extreme/decisive evidence for the presence of a given effect, including the null effect. When reporting BFs, BF10 indicates the evidence for H1 relative to H0 and BF01 indicates the reverse. Usually, authors also provide the $\text{BF}_{\text{inclusion}}$ value, which reflects the likelihood of all the models including a given effect (*with* model) as compared to any other model not including the effect (*without* model). Given that we used mainly repeated measures models, we reported the $\text{BF}_{\text{inclusion}}$ based on matched models, which gives more sensitive results and represents the sum of $P(M|\text{data})$ of all *with* models, divided by the sum of $P(M|\text{data})$ of all *without* models (as suggested by S. Mathôt in JASP). As such, a positive value would then indicate a higher likelihood of including the effect in the best fitting models. Bayesian analyses were conducted with version 0.9.0.1 of the JASP software package, using default settings for the Cauchy prior distribution (JASP Team, 2017).

We first conducted a Bayesian mixed repeated-measures analysis of variance (ANOVA) on the raw scores 2 (Presentation: sequential vs. simultaneous) x 2 (Arrangement: unstructured vs. structured) x 5 (Age groups). After that we examined the arrangement effect more specifically by subtracting the structured score from the unstructured score for each kind of presentation mode. Finally, we have also conducted an analysis on proportionalised scores of the arrangement effect (by dividing the arrangement effect by the total of structured and unstructured score) to obtain a clearer picture of the effects throughout development by taking into account the expected change in the span size with age.

Results

It is important to note that the visual inspection of the graphic and QQ plot of standardized residuals was good, all points being on the normal probability plot. A 2 (Presentation: sequential vs. simultaneous) x 2 (Arrangement: unstructured vs. structured) x 5

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(Age groups) Bayesian mixed repeated-measures analysis of variance (ANOVA) was computed. The results showed that, after comparison to the null model containing only the participant factor as a nuisance variable, the model that received the strongest evidence was the model containing the three main effects of Age, Presentation and Arrangement and the three double interactions, Presentation * Arrangement, Arrangement * Age and Presentation * Age ($BF_{10} = 7.84E+101$). This best fitting model was 7.13 times more likely than the same model without Presentation * Age ($BF_{10} = 1.10E+101$), showing strong evidence in favor of the model including the three main effects and the three double interactions.

Table 2 reports specific effect values (Inclusion Bayes Factor based on matched models), which reflect the likelihood of all the models including a given effect as compared to any other model not including the effect. Post-hoc analyses for main effects showed for the main effect of Presentation, that the simultaneous condition led to better performance than the sequential presentation mode (BF_{10} , $U=1452$); that the effect of Arrangement reflected an advantage for structured compared to unstructured visuospatial information (BF_{10} , $U=3.24E+48$) (see Figure 2); and that the main effect of Age group indicated improved performance with age ($BF_{10}=19.71$). For the interaction effects, Bayesian paired t-tests were conducted. First, at a developmental level, the interaction between Presentation and Age indicated that the advantage of the simultaneous over the sequential condition appears with age, the difference between both presentation modes becoming significant from the age of 11 years onwards (4-5 y.o., $BF_{10}=0.24$ and $BF_{01}=4.20$; 6-7 y.o., $BF_{10}=0.27$ and $BF_{01}=3.78$; 8-10 y.o., $BF_{10}=1.09$ and $BF_{01}=0.92$; 11-15 y.o., $BF_{10}=974.73$; Adults, $BF_{10}=3.50$). The interaction between Arrangement and Age indicated that the arrangement effect becomes larger with age. There is a significant difference between unstructured and structured arrangement at each age (4-5 y.o., $BF_{10}=69.53$; 6-7 y.o., $BF_{10}=221793.16$; 8-10 y.o., $BF_{10}=385399.96$; 11-15 y.o., $BF_{10}=2.90E+8$; Adults, $BF_{10}=2.72E+9$) (see Figure 2). Finally, with regard to the interaction

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between Presentation and Arrangement, Bayesian paired t-tests showed that both presentation modes led to similar performance when the visuospatial information was unstructured ($BF_{10}=0.13$ and $BF_{01}=7.62$) but there was a significant advantage for the simultaneous presentation mode when the visuospatial information was structured ($BF_{10}=10.5846.48$) (see Figure 3).

<INSERT TABLE 2 HERE>

<INSERT FIGURE 2 HERE>

<INSERT FIGURE 3 HERE>

Additionally, in order to better understand the development of the arrangement effect with regard to the presentation effect, we introduced the score of the arrangement effect for each kind of presentation in a 2 (Presentation) x 5 (Age groups) Bayesian mixed repeated-measures ANOVA (see Figure 4). The results showed that, after comparison to the null model containing only the participant factor as a nuisance variable, the model that received the strongest evidence was the model containing the two main effects of Age and Presentation and the interaction ($BF_{10}=1.53E+10$). This best fitting model was 8.5 times more likely than the same model without Presentation * Age ($BF_{10}=1.80E+9$), showing strong evidence in favor of the model including the two main effects and the interaction. The inclusion Bayes Factors based on matched models were in favor of moderate to strong evidence of the three effects (Presentation effect: $BF_{inclusion}=3401.13$; Age effect: $BF_{inclusion}=6.37E+5$; interaction effect: $BF_{inclusion}=8.47$). As already suggested by the previous ANOVA, post-hoc analyses for both main effects showed an increasing arrangement effect with age on the one hand and that the simultaneous condition led to a stronger arrangement effect (greater difference between

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structured and unstructured items) than the sequential presentation mode on the other hand. Interestingly, these analyses pointed to a crucial age where a switch operated in the arrangement effect by showing strong evidence in favor of a difference between the younger groups (4-5 y.o. and 6-7 y.o. groups) and the older groups (all BF_{10} , $U > 175$) while the two younger groups performed at a similar level (BF_{10} , $U = 0.67$) as did the two older groups (BF_{10} , $U < 0.38$). For the interaction effect between Age group and Presentation, Bayesian paired t-tests indicated that the advantage of the arrangement effect for the simultaneous over the sequential condition appears with age, the difference of the arrangement effect scores between both presentation modes becoming significant from the age of 8 years until the adult age where the arrangement effect scores return to similar in both presentation modes (4-5 y.o., $BF_{10}=0.49$ and $BF_{01}=2.05$; 6-7 y.o., $BF_{10}=1.52$ and $BF_{01}=0.66$; 8-10 y.o., $BF_{10}=460.22$; 11-15 y.o., $BF_{10}=30.64$; Adults, $BF_{10}=0.24$ $BF_{01}=4.19$).

The same kind of analysis have also been conducted on the presentation effect to better appreciate the development of the presentation effect. We introduced the score of the arrangement effect for each kind of presentation in a 2 (Arrangement) x 5 (Age groups) Bayesian mixed repeated-measures ANOVA. The results showed that, after comparison to the null model containing only the participant factor as a nuisance variable, the model that received the strongest evidence was the model containing the two main effects of Age and Arrangement and the interaction ($BF_{10} = 1.67E+5$). The inclusion Bayes Factors based on matched models were in favor of moderate to strong evidence of the three effects (Arrangement effect: $BF_{inclusion}=4690.58$; Age effect: $BF_{inclusion}=4.29$; interaction effect: $BF_{inclusion}=9.94$). As already suggested by the previous ANOVA, post-hoc analyses for both main effects showed an increasing presentation effect with age on the one hand and again that the structured condition led to a stronger presentation effect (better performance for the simultaneous than for the sequential presentation mode) than the unstructured condition on

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the other hand. Interestingly, these analyses confirmed the crucial age of 11, where an important switch operated in the presentation effect by showing strong evidence in favor of a difference between the two younger groups and the 11-15 y.o. group (4-5 y.o., BF_{10} , $U=36.82$ and 6-7 y.o., BF_{10} , $U=71.96$) while the three younger groups performed at a similar level (BF_{10} , $U < 0.60$) as did the two older groups (BF_{10} , $U < 0.60$). The interaction effect between Age group and Arrangement reflected the same results than in the previous ANOVA on the arrangement effect.

<INSERT FIGURE 4 HERE>

Finally, in order to better understand the development of the arrangement and presentation effects by taking into account the expected change in the span size with age, we conducted the same 2 (Presentation) x 5 (Age groups) Bayesian mixed repeated-measures ANOVA with the proportionalised score of the arrangement effect. The results showed that, after comparison to the null model containing only the participant factor as a nuisance variable, the model that received the strongest evidence was the model containing the main effect of Presentation ($BF_{10} = 67.04$). This best fitting model was 5.5 times more likely than the same model with Age ($BF_{10} = 12.21$), showing strong evidence in favor of the model including only the main effect of presentation on the proportionalised scores. The inclusion Bayes Factors based on matched models were in favor of moderate to strong evidence of the main effect of Presentation but not Age or interaction effect (Presentation effect: $BF_{inclusion} = 48.90$; Age effect: $BF_{inclusion} = 0.18$; interaction effect: $BF_{inclusion} = 0.30$). This is in favor of a stronger effect of arrangement for simultaneous than sequential presentation whatever the age of participants for a same span size. The same analysis was conducted for the proportionalised score of the presentation effect with 2 (Arrangement) x 5 (Age groups)

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Bayesian mixed repeated-measures ANOVA. The results showed that, after comparison to the null model containing only the participant factor as a nuisance variable, the model that received the strongest evidence was the model containing the main effect of Arrangement ($BF_{10}=46.83$). The inclusion Bayes Factors based on matched models were in favor of moderate to strong evidence of the main effect of Arrangement but not Age or interaction effect (Arrangement effect: $BF_{inclusion}=47.81$; Age effect: $BF_{inclusion}=0.34$; interaction effect: $BF_{inclusion}=0.57$). This is in favor of a stronger effect of presentation for structured than for unstructured condition whatever the age of participants for a same span size.

Interim discussion

This first experiment was conducted in order to obtain a more precise idea of the development of the two processing pathways of visuospatial information in STM. Results showed evidence in favor of better performance for the simultaneous than for sequential presentation mode from the age of 11 years. Moreover, the arrangement effect was larger in older children and adolescents than in younger children. More specific analyses on the arrangement effect score pointed the age of 8 as a crucial time point for this effect, by showing a more significant gap between structured and unstructured visuospatial information to process from this age until the adult age and this for the simultaneous presentation mode but not for the sequential presentation mode. In adults, the arrangement effect score seems to be again in the same range for both presentation modes. It is important to note that when taking into account the span size of each participant, no longer difference between age groups appeared suggesting that this change was mainly quantitative but not necessarily qualitative. Moreover, no longer interaction was observed suggesting that this potential discontinuity for the arrangement effect in the simultaneous but not sequential presentation mode was largely led by the span size rather than by the age per se. In other words, our results demonstrated that for a same span size, the age was any longer an effect on the arrangement effect or the presentation effect.

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Moreover, in adults, presentation mode and arrangement effect were observed, even if the difference between both kinds of presentation mode decreased at this age.

Our results are in accordance with numerous previous studies as they show larger visuospatial spans when the stimuli are presented all at once (simultaneous presentation) than when they are presented sequentially and when the arrangement of the stimuli follows a regular, structured configuration than with random positions (Imbo et al., 2009; Kemps, 2001). However, our results are not completely in line with previous adults data (Pieroni et al., 2011) showing a larger arrangement effect for simultaneous than for the sequential presentation. Indeed, as in this previous study we demonstrated globally a larger arrangement effect for simultaneous than for the sequential presentation but not specifically for the adult group when we considered the raw data.

Our results reveal several key developmental findings. First, children as young as 4 years old are already sensitive to arrangement when holding visuospatial information in STM and this sensitivity increases with age (see also Imbo et al., 2009). Second, the presentation mode effect appears by the age of 11 years. This result is inconsistent with Carretti et al. (2015) who found an effect of presentation mode in children of 4- to 6- year-old. However, in their study, the task required participants to hold 2 to 8 positions with no stop rule whatever the age of the participant. Looking more carefully at their results, it appears that differences between both presentation modes were mainly observed for the higher span levels (from span 4) and this especially in the sequential condition (e.g. the proportion of recalled positions = 0.3), with no big difference under span 4, the span usually reached at this young age (5 years old children). Therefore, the difficulty level of the tasks could have obscured the true level of performance of young children, the amount of information to recall interfering with the possibility of properly recalling some of them. This methodological difference might have maximized the difference between the two presentation modes at a younger age or could

suggest that both presentation modes lead to similar performance in children when they are in their span range. Third, the age of 8 has been pointed to a crucial time point for the change in the processing of visuospatial information by showing a sharp increase in the individual arrangement effect for the simultaneous presentation mode. Other studies suggest that this age range is critical at the qualitative level for STM. In the verbal domain, this age range corresponds to a progressive intervention of more elaborate strategies such as the cumulative rehearsal strategy (Henry, Turner, Smith, & Leather, 2000; Poloczek, Henry, Messer, & Büttner, 2019). Moreover, numerous authors proposed that better STM performance for structured visuospatial information would be linked to semantic processes in long-term memory. Indirectly, the amount of information we can hold could be increased by enhanced chunking processes that facilitate the formation of gestalts in the pattern (e.g., Gobet & Simon, 1996; Mammarella, Giofrè, Caviola, Cornoldi, & Hamilton, 2014; Pieroni et al., 2011). Therefore, this requires not only some knowledge about visuospatial patterns, still developing until 8 years old for very basic perceptual skills (e.g. length, surface and orientation judgements) and until adolescence for the more elaborated skills (e.g. recognition of incomplete figures) (e.g. Schmetz, Rousselle, Ballaz, Detraux, & Barisnikov, 2018), but also sufficient free resources to be able to chunk and extract a more global pattern. This explanation in terms of better strategies is actually completely in line with the further results on the proportionalised effect, taking into account the span size of participants. We observed no age effect at this point, highlighting that the difference in the arrangement effect between the different age group was more linked to the span size than to the development per se and therefore probably more linked to some strategic and availability of cognitive resources aspects. Indeed, previous studies suggested that individual differences in effective strategy use was closely related to span performance (Dunlosky & Kane, 2007; Poloczek et al., 2019).

Additionally, a recent study found that the well-known visuospatial bootstrapping effect, corresponding to better performance in recalling verbal information when the participant has the opportunity to encode extra visuospatial information, appeared in 9 years old children and adults but not in 6 years old children, suggesting a difficulty in taking advantage of long-term memory to enhance STM performance (Darling, Goodall, Havelka, & Allen, 2014). Finally, in adults, the arrangement effect seems to be the same no matter if the visuospatial information is presented simultaneously or sequentially. These results are not completely in line with previous adults data (Pieroni et al., 2011) showing a larger arrangement effect for simultaneous than for the sequential presentation in this population. In this study we however observed identical size of arrangement effect for both presentation mode in adult, as if this significant advantage for the structured visuospatial information in the simultaneous presentation mode dropped at the adult age. In our study, unlike the study of Pieroni et al. (2011), position of the presented elements formed a regular pattern respecting the Gestalt principles of symmetry or continuity for the structured arrangement. Importantly, Pieroni et al. (2011) demonstrated that for the sequential presentation mode, the structured arrangement led to better recall only in the case of stimuli symmetrical along the vertical axis while for the simultaneous presentation mode, structured arrangement improves performance for all three types of symmetry tested (diagonal, vertical, horizontal). Our data did not allow us to go further in this analysis but maybe that the kind of symmetry used have influenced our result. Moreover, in this study, trials were of growing complexity as the size of the matrix and the number of positions to be remembered increased during the task. This increases have could interfered with the possibility to gain from the same strategies (use of long-term knowledge or chunking) in adults. This drop in arrangement effect for simultaneous presentation mode needs therefore to be addressed and confirmed in further studies.

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Moreover, when we taking account the span size (proportionnalised analyses), this modulation of the arrangement effect by the presentation mode was still observed while the interaction with Age group disappeared, suggesting that this difference of arrangement in function of presentation mode during the development could be largely driven by the use of strategies rather than by the development of age per se.

Overall, our results indicated a growing sensitivity to visuospatial arrangement and a growing advantage for the simultaneous over the sequential presentation mode with the increase of the span size. These data add support to the recent model of visuospatial STM, which proposes two routes for the processing of simultaneous and sequential visuospatial information (Attout et al., 2018). Not only do these results demonstrate that the intervention of preexisting visuospatial knowledge facilitates the recall of information (arrangement effect) but they also suggest that children accumulate more and more visuospatial configural knowledge and probably develop strategies (such as chunking, grouping) to organize, and then support the maintenance of visuospatial information in STM. They also demonstrated that holding the spatial coordinates of a sequence of events is more difficult than when visuospatial information is accessible all-at-once probably because the sequential presentation mode requires supplementary steps to integrate a series of individual local positions into a global configural visuospatial representation in STM. By contrast, the simultaneous presentation mode became easier to process with age as this mode enables a more direct access to a global visuospatial representation in STM.

Experiment 2

A second issue is to determine whether the two processing routes model could be useful to characterize the deficits in visuospatial STM in atypical development and to understand the nature of the underlying difficulties. Here, we focused on two specific genetic

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syndromes, Williams syndrome (WS) and Down syndrome (DS) which have been found to be associated with different limitations of visuospatial skills and visuospatial STM deficits (Jarrold, Baddeley, & Hewes, 1999; Wang & Bellugi, 1994). People with WS, a rare genetic condition caused by the microdeletion of several genes on chromosome 7q11.23, were shown to experience difficulties in a wide range of visuospatial tasks such as the block design subtest of the Wechsler scales, puzzle, drawings, 3D-geometry or line orientation judgment, all tasks requiring good visuo-perceptive and constructive skills (Farran & Jarrold, 2003; Wang, Doherty, Rourke, & Bellugi, 1995). They were found to be particularly poor at processing global (relative to local) visuo-perceptual information (Bellugi, Wang, & Jernigan, 1994). In long- and short-term memory, a selective deficit in processing visuospatial information relative to verbal information has been observed (Carney et al., 2013; Jarrold, Baddeley, & Phillips, 2007). More specifically, for visuospatial STM, two studies showed that people with WS presented a specific deficit in a simultaneous condition but not in a sequential presentation condition (Carretti, Lanfranchi, De Mori, Mammarella, & Vianello, 2015; Lanfranchi, De Mori, Mammarella, Carretti, & Vianello, 2015), suggesting that they do not experience difficulties with encoding serial local positions but that they have difficulties in maintaining visuospatial information presented all at once in STM. They could take advantage of the structured presentation of visuospatial information but less than control participants matched on nonverbal mental age, a deficit which becomes apparent when memory load increases. Their reduced benefit from the visuospatial arrangement could possibly be due to limitations in preexisting visuospatial long-term knowledge or in the use of more effective strategies. Using a similar methodology, these results should be replicated in the present experiment. Accordingly, we expected: (1) a more prominent deficit in the simultaneous mode of presentation, which requires direct access to a configural representation, than for the sequential presentation, which allows the configural representation to be constructed one

position at-a-time, and (2) a lower or absent arrangement effect in people with Williams syndrome.

The second population of interest here was participants with Down syndrome, a genetic anomaly resulting from a complete or a partial third copy of chromosome 21. This very common genetic syndrome is mainly characterized by a verbal impairment, with visuospatial skills preserved relative to their mental age (Abbeduto et al., 2001; Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 2015). In the case of visuospatial skills, individuals with DS differ from people with WS as they mainly present difficulties with processing local information details, while processing global visuospatial information is better preserved. Several authors claimed that people with DS preferentially use global processing to comprehend visuospatial information (Grieco et al., 2015; Paterson, 2001). Their STM profile is characterized by better performance in visuospatial than in verbal STM relative to their mental age (Hulme & Mackenzie, 1992; Jarrold et al., 1999; Laws, 2002). Moreover, some dissociations were reported in visuospatial STM. For example, spatial memory (e.g. serial order reconstruction of two shapes or Corsi block test) seems to be preserved in individuals with DS while visual memory (e.g. colour memory task) is impaired relative to typical developing children matched on mental age (Laws, 2002; Vicari, Bellucci, & Carlesimo, 2006). However, most of these studies used the well-known Corsi block test where (patients need to reproduce a sequence of up to nine separated blocks touched by the experimenter) and thus, visuospatial STM was only tested in the sequential presentation mode. More recently, some studies examined their performance in spatial working memory tasks by distinguishing the presentation format (simultaneous and. sequential) relative to control participants matched on non-verbal IQ (Carretti & Lanfranchi, 2010; Carretti, Lanfranchi, & Mammarella, 2013; Lanfranchi, Carretti, Spanò, & Cornoldi, 2009). As in Williams syndrome, results showed that individuals with DS performed similarly to their control group when visuospatial information

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was presented sequentially but not simultaneously. Moreover, their deficit was found to be restricted to structured visuospatial arrangement in the simultaneous presentation mode.

To sum up, in spite of the fact that individuals with WS and DS are assumed to process visuo-perceptual information very differently (i.e. a global perceptual processing advantage in people with DS vs. a local perceptual processing advantage in people with WS), they seemed to present a very similar cognitive profile in visuospatial STM. Both groups of patients have been found to be significantly impaired when holding structured visuospatial information presented simultaneously in their STM and have difficulties with taking as much advantage of visuospatial structure in comparison to controls. In terms of the alternative two routes processing model, this pattern of performance may result from impaired access to a configural representation from visuospatial information presented simultaneously. The difference between those two genetic syndromes with regard to the visuospatial STM is that participants with WS seem to be also impaired when holding simultaneously presented unstructured arrangement while individuals with DS are not.

Here, we propose to examine the arrangement and presentation effects and their interactions as a way to explore the nature of the visuospatial STM deficit in those two genetic syndromes associated with divergent profiles of local-global perceptual dominance. Each group was matched with a control group of typically developing children on a non-verbal IQ measure. To acquire a more complete picture of the STM profile and replicate the dissociation between verbal and visuospatial STM in the participants, measures of verbal executive components of working memory and verbal STM were also administered to participants.

Methods

Participants. Twenty-one participants with WS aged between 5 and 52 years old (Mean = 21.95 years old, 10 females) participated in this study. They were recruited through

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the Williams Syndrome Foundation and the department of paediatric cardiology of the University Hospital. Diagnosis was confirmed with the fluorescent in situ hybridization test (FISH) or by the Multiplex Ligation-dependent Probe Amplification method (MLPA). Each participant with WS was paired to a typically developing (TD) child matched on the performance in a non-verbal IQ task, the Concept Identification subtest from a Weschsler test (± 2 gap points of the raw score). We choose this task to match our groups since it reflects reasoning abilities, without over-emphasis on the verbal or visuospatial domains. The TD_{WS} control group was composed of 21 typically developing children aged between 3 and 9 years old (Mean = 5.82 years old, 15 girls) with no history of neurological or psychiatric disorders, hearing or visual impairments (uncorrected), or neurodevelopmental disorders.

Twenty-two children and adults with DS aged between 7 and 29 years old (Mean = 10.53 years old, 15 females) participated in this study. They were recruited through associations and the department of pediatric cardiology of the Hospital Center. Each participant with DS was paired to a typically developing child matched on performance in the same non-verbal IQ measure, the concept identification from a Weschsler test (± 2 gap points of the raw score). The TD_{DS} control group was composed of 22 typically developing children aged between 3 and 8 years old (Mean = 4.11 years old, 11 girls) and none of them reported a history of neurological or psychiatric disorders, hearing or visual impairments (uncorrected), or neurodevelopmental disorders. We note that 8 typically developing children belonged to both the TD_{DS} and the TD_{WS} groups.

Materials

We assessed verbal and non verbal intellectual abilities from IQ scales as well as the three main components of working memory defined in Baddeley and Hitch's model (Baddeley, 1986; Baddeley & Hitch, 1974) : the visuospatial STM as a measure of interest and the central executive component and the verbal STM as complementary measures.

IQ measures. Verbal intelligence was assessed using the Vocabulary and Similarity subtests while non-verbal intelligence was assessed using the Concept identification and the Block design from the Wechsler *Preschool and Primary Scale of Intelligence*-3rd edition (WIPPSI-III; Wechsler, 2004) or the Wechsler Intelligence Scale for Children-4th edition (WISC-IV; Wechsler, 2005), depending on the child's age.

Central executive component. The category-span task developed by Noël (2009) was used to examine the central executive component and to some extent on verbal STM skills. In this task, one-syllable words, related alternately to food or animals, were read at the rate of one word per second to the participants and they had to recall them category by category (starting with the food words and then the animal words). Sequences started with 2 words, reaching a maximum of 9 words. For this task, as well as for the other span tasks, participants had to succeed in two trials of the same difficulty level (or number of items) to access a higher level (span +1). The task ended when the participant failed at two (or more) out of the three trials for a given difficulty level. Each correct response was credited with one point. The dependent variable corresponded to the total of points credited. To help participants understanding the instructions, pictures of a forest and a plate with cutlery were presented to support the recall of the animal and food names, respectively.

Verbal STM. Phonological loop capacity was assessed in a forward letter span task. Participants were instructed to listen to a sequence of letters and to repeat them immediately after in the same order. Letters were read at the rate of one per second. No repetition was allowed. Sequences consisted of monosyllabic consonants with no repetition within any sequence. The first sequences included two letters and were then followed by sequences of increasing length (3 to 9 letters). For each sequence length, there was a maximum of three trials, out of which only the two best trials were scored. Participants who succeeded at repeating two sequences of n letters were given sequences of $n + 1$ letters at the next trial.

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The task was stopped when a participant failed at two out of the three trials for a given sequence length. Each correct response was credited with one point.

Visuospatial STM. The visuospatial STM was assessed using the same visuospatial STM tasks as in the first experiment.

Experimental procedure

Participants were tested individually at school or at home in a quiet room. Testing was completed in two sessions and tasks were proposed in a Latin square order. The experiment was conducted in accordance with the Declaration of Helsinki and the regional ethical committee for biomedical research approved the experimental protocol (Record number: B403201111579). All parents gave written consent before the cognitive tests were administered. Analyses compare each genetic syndrome group with the corresponding control group.

Results

Table 3 reports descriptive statistics for IQ measures, short-term and working memory tasks in participants with WS, DS and typically developing participants matched on nonverbal IQ.

<INSERT TABLE 3 HERE>

Williams syndrome analysis. A series of Bayesian independent t-tests showed strong evidence for the alternative hypothesis that the WS group was older than the TD_{WS} group ($BF_{10}=65929$). They also demonstrated moderate evidence in favor of similar performance in both groups on the vocabulary ($BF_{01}=5.02$), the similarities subtest ($BF_{01}=7.24$) and concept identification ($BF_{01}=4.17$) while no evidence against and anecdotal evidence in favor of better performance for the TD_{WS} group than in the WS group on the block design subtest was observed ($BF_{10}=1.63$; $BF_{01}=0.61$). For the two verbal memory measures, there was moderate

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evidence in favor of no difference between both groups (verbal STM: $BF_{01}=3.3$; Central executive: $BF_{01}=3.896$).

A 2 (Presentation mode: sequential vs. simultaneous) x 2 (Arrangement: unstructured vs. structured) x 2 (Group: WS vs. TD_{ws}) Bayesian repeated measures ANOVA was computed. The results showed that, after comparison to the null model, the model that received the strongest evidence was the model containing the two main effects of Arrangement and Group, and the interaction between Arrangement and Group ($BF_{10}=2.72E+9$) as compared to a model including only the two main effects, Arrangement and Group ($BF_{10} = 6.47E+8$) (see Figure 5).

<INSERT FIGURE 5 HERE>

We report in Table 4 specific effect values (Inclusion Bayes Factor based on matched models), which reflect the likelihood of all the models including a given effect (*with* model) as compared to any other model not including the effect (*without* model). Consistent with the Bayesian repeated measures ANOVA, the model providing the strongest evidence involved the arrangement effect and the arrangement x group interaction. It is important to note that no evidence in favor of a main effect of group was observed. Post-hoc analyses for the main effect of arrangement showed as expected better performance for the structured than for the unstructured arrangement (BF_{10} , $U=6.42E+8$). For the interaction effect between Group and Arrangement, Bayesian independent t-tests indicated evidence in favour of a difference between both groups in the structured conditions ($BF_{10}=4.18$) but not for unstructured stimuli ($BF_{10}=1.88$).

In order to deeper understand the differential arrangement effect, we conducted a 2 (Presentation) x 2 (Group) Bayesian mixed repeated-measures ANOVA with the score of the

arrangement effect (see Method of the first experiment). Results showed that, after comparison to the null model containing only the participant factor as a nuisance variable, the model that received the strongest evidence was the model containing the two main effects of Group and Presentation ($BF_{10}=8.22$). The inclusion Bayes Factors based on matched models were in favor of moderate evidence of the main effect of Presentation ($BF_{inclusion}=3.08$) but not for the main effect of Group ($BF_{inclusion}=2.68$) or the interaction effect ($BF_{inclusion}=0.44$). However, post-hoc analyses for both main effects showed an anecdotal evidence for a difference between the arrangement effects in function of both presentation modes (BF_{10} , $U=2.76$) but a moderate evidence in favour of a difference between both groups for the arrangement effect (BF_{10} , $U=6.56$).

Finally, we have also conducted an analysis on proportionalised scores of the arrangement effect to appreciate the differences between both groups in the arrangement effect by taking into account the span size of each individual (see Method section of the first experiment). We observed with a 2 (Presentation) x 2 (Group) Bayesian mixed repeated-measures ANOVA no evidence in favour of a model differing from the null model ($BF_{10}=1.37$) suggesting that when we take into account the span size, both groups did not differ from each other. The Bayes factors across matched models showed however a moderate evidence in favour of a main effect of Presentation ($BF_{inclusion}=3.08$) but not for Group or interaction effects (respectively, $BF_{inclusion}=2.68$ and 0.44).

<INSERT TABLE 4 HERE>

Down syndrome analysis. Bayesian independent t-tests were run to compare both groups of participants for descriptive measures (see Table 3). They showed strong evidence for a difference in chronological age between both groups ($BF_{10}=6.54E7$), the participants

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with DS being older than the control participants. They also demonstrated anecdotal to no evidence in favor of lower performance for the DS group relatively to the TD_{DS} on the vocabulary ($BF_{10}=2.55$), the similarities ($BF_{01}=1.56$), the block design ($BF_{01}=3.32$) and the concept identification subtest ($BF_{01}=3.30$). For the two verbal memory measures, there was strong to very strong evidence in favor of a difference between both groups (verbal STM: $BF_{10}=407809$; Central executive: $BF_{10}=28.85$), with lower performance for the participants with DS.

Performance in visuospatial STM tasks was analyzed using a 2 x 2 x 2 Bayesian repeated measures ANOVA with Presentation mode (sequential vs. simultaneous) and Arrangement of the patterns (unstructured vs. structured) as within-subject factors and Group (DS vs. TD_{DS}) as a between-subjects factor. After comparison to the null model, the results showed that the model receiving the strongest evidence was the model comprising all the main effects (Presentation, Arrangement and Group) and the interaction between Presentation and Group ($BF_{10}=11804.70$) as compared to a model including only the three main effects without the interaction between Presentation and Group ($BF_{10} = 1572.78$). As shown in Table 5, the $BF_{inclusion}$ based on matched models indicated a strong evidence in favor of the model including the Arrangement effect and the Presentation x Group interaction and a substantial evidence against the interaction between Group and Arrangement and the triple interaction. Overall, these results provide evidence that the data are better explained by a model containing the main effects and the presentation by group interaction while there was no evidence for an interaction between arrangement and group or a triple interaction in comparison to the null model, suggesting that both groups presented a similar arrangement effect. Post-hoc analyses for the main effect of arrangement showed as expected better performance for the structured than for the unstructured arrangement (BF_{10} , $U = 3.27E+8$). For the interaction effect between Group and Presentation, Bayesian independent t-tests indicated

no evidence in favour of a difference between both groups for the simultaneous ($BF_{10}=0.42$) or the sequential presentation mode ($BF_{10}=0.36$).

<INSERT TABLE 5 HERE>

To appreciate the interaction between Presentation and Group we compared the presentation effect (computed by subtracting simultaneous from sequential presentation) in both groups, as for the arrangement effect computed in previous analyses. We conducted a 2 (Arrangement) x 2 (Group) Bayesian mixed repeated-measures ANOVA with the score of the presentation effect. Results showed no evidence in favour of a model differing from the null model ($BF_{10}=1.52$), suggesting that there was no difference between both groups for the presentation effect. However, the interaction between Presentation and Group in the global ANOVA could be due to the fact that controls tended to present a positive presentation effect with better performance in simultaneous presentation than in sequential presentation (Mean difference [simultaneous-sequential] = +0.95) while DS group exhibited a negative presentation effect with lower performance in simultaneous presentation than in sequential presentation (Mean difference [simultaneous-sequential] = -2.86) (see Figure 6).

Finally, we have also conducted an analysis on proportionalised scores of the presentation effect to appreciate the differences between both groups in the presentation effect by taking into account the span size of each individual. We observed with a 2 (Arrangement) x 2 (Group) Bayesian mixed repeated-measures ANOVA that the model that received the strongest evidence was the model containing the main effect of Group ($BF_{10} = 5.80$; $BF_{Inclusion}=5.85$), illustrated by a larger difference between the two presentation modes (higher performance for the simultaneous condition) in the control group than in the DS group. This suggests that when we take into account the span size, both groups differ from each other.

<INSERT FIGURE 6 HERE>

Interim discussion

This second experiment aimed at using the two processing route model to characterize and understand the nature of the deficits in two genetic syndromes known to be associated with atypical development of visuospatial STM. People with Williams or Down syndrome showed two distinct profiles. First, as regards WS, we found strong evidence for a model including an effect of arrangement and an interaction between Group and Arrangement. Indeed, the WS group showed lower performance than controls but only for the structured arrangement as the controls benefitted more from the structured arrangement than the participants with WS. However, when we take into account the global span size of each participant, both groups presented similar performance and took advantage of the structured conditions in a similar way. By contrast, with respect to participants with DS, we found moderate evidence in favor of a model containing a presentation by group interaction, the control group performing better in the simultaneous than in the sequential presentation mode, while the reverse kind of profile was observed for participants with DS. Importantly, this was particularly true when we took into account the span size for each participant.

Our results with WS are consistent with the assumption that the visuospatial impairment in WS (Bellugi et al., 1994) could result in a lower sensitivity to the visuospatial arrangement in STM. This profile is similar to the one found with the same tasks in another genetic syndrome associated with visuospatial impairment namely, the 22q11.2 deletion syndrome (Attout et al., 2018). Indeed, participants with 22q11.2 deletion syndrome also took less advantage of the visuospatial arrangement to support their representation in STM for both simultaneous and sequential presentation modes in comparison to children matched on

chronological age. This pattern of performance could be attributed to their visuospatial impairment which results in limited pre-existing visuospatial knowledge, difficulties with processing online visuospatial information and/or improper strategy use. However, when we used a proportionalised arrangement effect on their global span size, we did not observe any difference between both groups. This suggests that the difficulties in taking advantage of the structured patterns in the simultaneous condition could reveal a mere delay, rather than an atypical development. This point leads to possible guidelines for the rehabilitation of the visuospatial STM in patients with WS. First, these results suggest that they are likely to benefit from specific training on visuospatial strategies by prompting to link some patterns to known forms or by focusing on the verbalization of forms. Second, a more specific training on the sequential-spatial component, a crucial component in different learning abilities and a strength of this population, could be prioritized, especially since this component can be successfully and specifically trained by a metacognitive approach for example (Caviola, Mammarella, Cornoldi, & Lucangeli, 2009; Cornoldi, Carretti, Drusi, & Tencati, 2015).

On the other hand, our results only partially reproduce results from two previous studies conducted in people with WS (Carretti et al., 2015; Lanfranchi et al., 2015) which also reported reduced benefit of the visuospatial arrangement for people with WS compared to the control group matched on nonverbal mental age. These studies reported a deficit in the simultaneous but not in the sequential presentation mode. However, as we already explained earlier, the method was not exactly the same and the WS populations studied in their studies was younger than us. Therefore, if the difficulties to take advantage of arrangement is associated to a development delay, as suggested by our additional analyses, the young age of WS participants could lead to worse performance in a visuospatial STM domain mastered earlier than the sequential processing of information (see results from the first experiment).

To sum up, our data suggest that people with WS have a reduced ability to take advantage of the arrangement of visuospatial information to support the short-term retention compared to control participants matched on non-verbal abilities. With regard to the two processing route model (Attout et al., 2018), this deficit may reflect lower positive influence of visuospatial long-term knowledge or strategies whose acquisition itself relies on efficient visuospatial abilities. This result is thus consistent with the usual visuospatial profile of WS (with weak performance in all tasks requiring perception of visuospatial relationship) and the weaker performance than typical children in the block design subtest in our sample (no evidence against and anecdotal evidence in favor of better performance for the TD_{WS} group than in the WS group).

With regard to the participants with DS, our results are partially in line with previous research as they show a similar level of performance in sequential STM task than typically developing children but some difficulties with holding visuospatial information presented simultaneously in the visuospatial STM (Carretti & Lanfranchi, 2010; Carretti et al., 2013; Lanfranchi et al., 2009; Laws, 2002; Vicari et al., 2006). Here, we observed an advantage for the simultaneous condition with regard to the sequential one for the control group but the reverse profile of performance in the DS group. Importantly, this was particularly true when we took into account the span size for each participant, suggesting an atypical development rather than a simple delay. With regard to the two processing route model, these results suggest that accessing a configural representation might be impaired when visuospatial information is presented all at once. One hypothesis is that their global perceptual processing dominance might not be enough to achieve the same level of precision in the access to a configural representation of the visual array in STM as their controls. It is also important to note that the encoding time of the simultaneous grids were constant whatever the level of difficulty while the encoding time for the sequential presentation increased with span level.

Therefore, a difference in processing speed could explain that the DS participant tend to be less efficient in simultaneous than in sequential presentation mode. However, in a previous study, they found the same profile of specific difficulties to process simultaneous information by controlling for speed of visuospatial processing (Vicari et al. 2006). Finally, another hypothesis could be that the simultaneous presentation mode might consume too many resources, with too many positions to process at the same time for these children, assumption in line with the idea that this specific presentation mode difficulty could be more significant at a same span size for both groups. As the presentation mode effect is small here, these hypotheses deserve further investigation. It maybe that a DS group with a higher mental age could demonstrate a more observable difference at the presentation mode level since, as observed in our first study in typical developing children, the difference between both presentation modes appeared only from 11 years old.

However, two studies also underlined that the deficit in simultaneous visuospatial STM task was mainly observed with structured arrangements (Carretti et al., 2010; 2013), a finding which was not observed in this study, as the interaction between arrangement and group was not retained in the best fitting model. In this respect, it should be noted that in Carretti et al. (2013), the interaction between presentation and arrangement was significant and they showed no effect of arrangement in the sequential visuospatial STM task, which clearly compromises the opportunity to observe any reduction of an arrangement effect. In addition, in both studies, the sizes of the matrices and the spans were smaller than in this present study and no stop criterion were used, making it difficult to compare the results directly.

General discussion

The present study addressed two main issues about the development of the two processing routes of visuospatial information in STM in typically developing children and in people with

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a neurodevelopmental syndrome. In a first experiment, we examined the development of the presentation mode and arrangement effects in visuospatial STM tasks in five groups, four of typically developing children from the age of 4 years and a group of adults. And in a second experiment, we examined how this model could be used to characterize deficits in visuospatial STM in atypical development. Results indicated that distinct performance in sequential and simultaneous modes of presentation appears from the age of 11 years with an advantage seen in the simultaneous presentation mode. This effect was also present in people with WS but not in participants with DS. The absence of a presentation mode effect before the age of 11 calls into question the opportunity to track this effect in populations with a lower IQ, as often done in the literature (see for example Carretti et al., 2010; 2013; 2015; Lanfranchi et al., 2015). However, In Experiment 2, the typically developing participants matched to participants with DS were aged from 3 to 9, which accounts for the fact that the presentation effect was quite flat and not significant. However, at a same span size level, both groups presented dissimilar presentation effect.

The two processing routes model predicted not only a differential development of simultaneous and sequential presentation modes, just like the Continuity model, but also a differential impact of arrangement effect for both kinds of visuospatial WM “components”. Our data also showed that children become more and more sensitive to visuospatial arrangement with age but with a more pronounced arrangement effect for the simultaneous mode of presentation. Moreover, this observation was mainly true from the age of 8, identified as a crucial time point for the change in the processing of visuospatial information by showing a sharp increase in the individual arrangement effect for the simultaneous presentation mode, this modification being more characterized by qualitative than quantitative changes. Indeed, our results suggest that at a same span size level, the age was any longer an effect on the arrangement effect or the presentation effect. These further analyses suggest that

this change was mainly driven by the use of strategies rather than by the development of age per se. It is also important to note that our results found a similar arrangement effect, even if this one was more pronounced for simultaneous presentation mode during childhood, in adults. Given that this interaction could be more related to the span size than to the age per se, one possible explanation could be the expertise of adults and this especially in sequential processing of information in STM. It is possible that with expertise, the additional step of order processing, as proposed in the alternative two processing routes model of visuospatial STM (Attout et al., 2018) and which could alter or slow down the access to the configural representation in children, become less consuming and more automatic in adults. In this line, a recent neuroimaging study found an age-related activity in the fronto-parietal network increasing in school age children (from 6 to 12 years) to process order information in verbal STM (by judging if two words were in a same order than in the memoranda list) while it was not the case to process item information in STM (by judging if a word was in the memoranda list or if it is a phonologically neighbor of the word) (Attout et al., 2019), suggesting that the order processing of information continues to develop with age.

Also, participants with WS showed a deficit in taking advantage of the arrangement of the visuospatial information. This underlines the importance of considering both parameters, the presentation mode and the arrangement when assessing visuospatial STM abilities and not only the presentation mode as in the Continuity model. Moreover, the model proposed, by integrating numerous previous data (Imbo et al., 2009; Kemps, 2001; Pieroni et al., 2011), that the arrangement effect could be directly dependent of the kind of presentation mode, the simultaneous or the sequential presentation. Indeed, a crucial difference to previous models is that this model considers that the two presentation modes do not involve distinct components but different access paths to a common configural representation in STM, the visuospatial STM where the access could be facilitated when visuospatial information is embedded in a

regular arrangement. As predicted by the two processing routes model, our results showed that extracting a global visuospatial configuration to increase STM performance was facilitated in the simultaneous condition and only from 8 years old for the raw scores. This could explain why previous data failed to show a specific deficit in one component for children with a global memory deficit (without a massive spatial skills deficit) such as, in mathematical learning disabilities children, where authors observed a global deficit to process visuospatial information whatever the presentation mode or the arrangement of the information (Mammarella, Caviola, Giofrè, & Szűcs, 2018).

Furthermore, it also suggests that the possibility to enhance the performance in visuospatial STM by using some strategies and/or long-term visuospatial knowledge seems compromised or need to be considered cautiously in a series of neurodevelopmental disorders presenting a visuospatial deficit such as Williams or 22 q11.2 deletion syndromes. However, the suggestion of a simple delay to process structured information in visuospatial STM for the WS seems really encouraging at the rehabilitation level.

Therefore, the two processing routes model for the maintenance of simultaneous and sequential visuospatial information in STM highlights interesting perspectives that help characterize both typical and atypical development. These two routes seem to follow independent developmental trajectories with better performance for the simultaneous than for sequential presentation mode from the age of 11 years. In addition, these results support the idea of an important influence of visuospatial ability and long-term visuospatial knowledge which differs depending on the presentation mode. While the continuity model did not embed this interaction between presentation mode and arrangement effect, the alternative two processing routes model of visuospatial STM proposed by Attout et al. (2018) thus bring new insights into the nature of the deficit in visuospatial STM in a series of neurodevelopmental disorders.

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Disclosure statement

The manuscript represents original material that has not been published previously and that is not being considered for publication elsewhere. All authors have substantially contributed to this work and have approved the final version of the manuscript. The manuscript agrees with APA ethical standards. There is no conflict of interest in connection with this work.

References

- Abbeduto, L., Pavetto, M., Kesin, E., Weissman, M., Karadottir, S., O'Brien, A., & Cawthon, S. (2001). The linguistic and cognitive profile of Down syndrome: Evidence from a comparison with fragile X syndrome. *Down Syndrome Research and Practice*, 7(1), 9-15. doi:10.3104/reports.109
- Attout, L., Noël, M.-P., & Rousselle, L. (2018). The effect of visual arrangement on visuospatial short-term memory: Insights from children with 22q11.2 deletion syndrome. *Cognitive Neuropsychology*, 1-9. doi: 10.1080/02643294.2018.1461616
- Attout, L., Ordonez Magro, L., Szmalec, A., & Majerus, S. (2019). The developmental neural substrates of item and serial order components of verbal working memory. *Human Brain Mapping*, 40, 1541-1553. doi: 10.1002/hbm.24466
- Baddeley, A. D. (1986). *Working memory*. Oxford, England: Oxford University Press.
- Baddeley, A. D., & Hitch, G. J. (1974). Working memory. In G. H. Bower (Ed.), *The psychology of learning and motivation* (pp. 47-90). San Diego, CA: Academic Press.
- Bellugi, U., Wang, P. P., & Jernigan, T. L. (1994). Williams syndrome: An unusual neuropsychological profile. In B. Broman & J. Grafman (Eds.), *Atypical cognitive deficits in developmental disorders: Implications for brain function* (Vol. 23, pp. 23-56). Hillsdale, NJ: Lawrence Erlbaum Associates.
- Carney, D. P., Henry, L. A., Messer, D. J., Danielsson, H., Brown, J. H., & Rönnerberg, J. (2013). Using developmental trajectories to examine verbal and visuospatial short-term memory development in children and adolescents with Williams and Down syndromes. *Research in Developmental Disabilities*, 34(10), 3421-3432. doi: 10.1016/j.ridd.2013.07.012

- Carretti, B., & Lanfranchi, S. (2010). The effect of configuration on VSWM performance of Down syndrome individuals. *Journal of Intellectual Disability Research*, 54(12), 1058-1066. doi: 10.1111/j.1365-2788.2010.01334.x
- Carretti, B., Lanfranchi, S., De Mori, L., Mammarella, I. C., & Vianello, R. (2015). Exploring spatial working memory performance in individuals with Williams syndrome: The effect of presentation format and configuration. *Research in Developmental Disabilities*, 37, 37-44. doi: 10.1016/j.ridd.2014.10.031
- Carretti, B., Lanfranchi, S., & Mammarella, I. C. (2013). Spatial-simultaneous and spatial-sequential working memory in individuals with Down syndrome: the effect of configuration. *Research in Developmental Disabilities*, 34(1), 669-675. doi: 10.1016/j.ridd.2012.09.011
- Caviola, S., Mammarella, I. C., Cornoldi, C., & Lucangeli, D. (2009). A metacognitive visuospatial working memory training for children. *International Electronic Journal of Elementary Education*, 2(1), 122–136.
- Cornoldi, C., & Vecchi, T. (2003). *Visuo-spatial working memory and individual differences*. Hove, UK: Psychology Press.
- Cornoldi, C., Carretti, B., Drusi, S., & Tencati, C. (2015). Improving problem solving in primary school students: The effect of a training programme focusing on metacognition and working memory. *British Journal of Educational Psychology*, 85(3), 424–439. <https://doi.org/10.1111/bjep.12083>
- Darling, S., Parker, M. J., Goodall, K. E., Havelka, J., & Allen, R. J. (2014). Visuospatial bootstrapping: Implicit binding of verbal working memory to visuospatial representations in children and adults. *Journal of Experimental Child Psychology*, 119, 112–119. <https://doi.org/10.1016/j.jecp.2013.10.004>

DEVELOPMENT OF VISUOSPATIAL STM

- Dienes, Z. (2011). Bayesian versus orthodox statistics: Which side are you on? *Perspectives on Psychological Science*, 6(3), 274-290. doi: 10.1177/1745691611406920
- Farran, E. K., & Jarrold, C. (2003). Visuospatial cognition in Williams syndrome: reviewing and accounting for the strengths and weaknesses in performance. *Developmental neuropsychology*, 23(1-2), 173-200. doi: 10.1080/87565641.2003.9651891
- Gobet, F., & Simon, H. A. (1996). Templates in chess memory: A mechanism for recalling several boards. *Cognitive Psychology*, 31(1), 1-40.
<https://doi.org/10.1006/cogp.1996.0011>
- Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). *Down syndrome: Cognitive and behavioral functioning across the lifespan*. Paper presented at the American Journal of Medical Genetics Part C: Seminars in Medical Genetics.
- Henry, L. A., Turner, J. E., Smith, P. T., & Leather, C. (2000). Modality effects and the development of the word length effect in children. *Memory*, 8(1), 1-17.
<https://doi.org/10.1080/096582100387678>
- Hulme, C., & Mackenzie, S. (1992). Working memory and severe learning disabilities: Hove, UK: Lawrence Erlbaum.
- Imbo, I., Szmalec, A., & Vandierendonck, A. (2009). The role of structure in age-related increases in visuo-spatial working memory span. *Psychologica Belgica*, 49, 275-291. doi: 10.5334/pb-49-4-275
- Jarrold, C., Baddeley, A., & Hewes, A. (1999). Genetically dissociated components of working memory: Evidence from Downs and Williams syndrome. *Neuropsychologia*, 37(6), 637-651. doi: 10.1016/S0028-3932(98)00128-6
- Jarrold, C., Baddeley, A. D., & Phillips, C. (2007). Long-term memory for verbal and visual information in Down syndrome and Williams syndrome: performance on the Doors

- and People test. *Cortex*, 43(2), 233-247. doi: 10.1016/S0010-9452(08)70478-710.1016/S0010-9452(08)70478-7
- Jeffreys, H. (1961). *Theory of probability* (3 ed.). Oxford: Oxford University Press, Clarendon Press.
- Kemps, E. (2001). Complexity effects in visuo-spatial working memory: Implications for the role of long-term memory. *Memory*, 9(1), 13-27. doi: 10.1080/09658210042000012
- Lanfranchi, S., Carretti, B., Spanò, G., & Cornoldi, C. (2009). A specific deficit in visuospatial simultaneous working memory in Down syndrome. *Journal of Intellectual Disability Research*, 53(5), 474-483. doi: 10.1111/j.1365-2788.2009.01165.x
- Lanfranchi, S., De Mori, L., Mammarella, I. C., Carretti, B., & Vianello, R. (2015). Spatial-sequential and spatial-simultaneous working memory in individuals with Williams Syndrome. *American journal on intellectual and developmental disabilities*, 120(3), 193-202. doi: 10.1352/1944-7558-120.3.193
- Laws, G. (2002). Working memory in children and adolescents with Down syndrome: evidence from a colour memory experiment. *Journal of Child Psychology and Psychiatry*, 43(3), 353-364. doi: 10.1111/1469-7610.00026
- Logie, R. H., & Pearson, D. G. (1997). The inner eye and the inner scribe of visuo-spatial working memory: Evidence from developmental fractionation. *European Journal of Cognitive Psychology*, 9(3), 241-257. doi: 10.1080/713752559
- Mammarella, I. C., Caviola, S., Giofrè, D., & Szűcs, D. (2018). The underlying structure of visuospatial working memory in children with mathematical learning disability. *British Journal of Developmental Psychology*, 36(2), 220–235.
<https://doi.org/10.1111/bjdp.12202>
- Mammarella, I. C., Giofrè, D., Caviola, S., Cornoldi, C., & Hamilton, C. (2014). Visuospatial

working memory in children with autism: The effect of a semantic global organization.

Research in Developmental Disabilities, 35(6), 1349–1356.

<https://doi.org/10.1016/j.ridd.2014.03.030>

Mammarella, I. C., Lucangeli, D., & Cornoldi, C. (2010). Spatial working memory and arithmetic deficits in children with nonverbal learning difficulties. *Journal of Learning Disabilities*, 43(5), 455-468. doi: 10.1177/0022219409355482

Mammarella, I. C., Pazzaglia, F., & Cornoldi, C. (2008). Evidence for different components in children's visuospatial working memory. *British Journal of Developmental Psychology*, 26(3), 337-355. doi: 10.1348/026151007X236061

McCormack, T., Brown, G. D. A., Vousden, J. I., & Henson, R. N. A. (2000). Children's serial recall errors: Implications for theories of short-term memory development. *Journal of Experimental Child Psychology*, 76(3), 222-252. doi: 10.1006/jecp.1999.2550

Morey, R. D., Romeijn, J.-W., & Rouder, J. N. (2016). The philosophy of Bayes factors and the quantification of statistical evidence. *Journal of Mathematical Psychology*, 72, 6-18. doi: 10.1016/j.jmp.2015.11.001

Morey, R. D., & Rouder, J. N. (2011). Bayes factor approaches for testing interval null hypotheses. *Psychological methods*, 16(4), 406-419. doi: 10.1037/a0024377

Noël, M. P. (2009). Counting on working memory when learning to count and to add: a preschool study. *Developmental Psychology*, 45(6), 1630-1643. doi: 10.1037/a0016224

Paterson, S. (2001). Language and number in Down syndrome: The complex developmental trajectory from infancy to adulthood. *Down Syndrome Research and Practice*, 7(2), 79-86. doi:10.3104/reports.117

- Pickering, S. J., Gathercole, S. E., Hall, M., & Lloyd, S. A. (2001). Development of memory for pattern and path: Further evidence for the fractionation of visuo-spatial memory. *The Quarterly Journal of Experimental Psychology Section A*, 54(2), 397-420. doi: 10.1080/713755973
- Pieroni, L., Rossi-Arnaud, C., & Baddeley, A. (2011). What can symmetry tell us about working memory. In A. Vandierendonck & A. Szmalec (Eds.), *Spatial working memory* (pp. 145-158): Psychology Press, Hove, UK.
- Poloczek, S., Henry, L. A., Messer, D. J., & Büttner, G. (2019). Do children use different forms of verbal rehearsal in serial picture recall tasks? A multi-method study. *Memory*, 27(6), 758–771. doi: 10.1080/09658211.2018.1563615
- Schmetz, E., Rousselle, L., Ballaz, C., Detraux, J. J., & Barisnikov, K. (2018). The BEVPS: A new test battery to assess visual perceptual and spatial processing abilities in 5–14 year-old children. *Applied Neuropsychology: Child*, 7(4), 317–333. <https://doi.org/10.1080/21622965.2017.1336710>
- Vicari, S., Bellucci, S., & Carlesimo, G. A. (2006). Evidence from two genetic syndromes for the independence of spatial and visual working memory. *Developmental Medicine and Child Neurology*, 48(2), 126-131. doi: 10.1017/S0012162206000272
- Wagenmakers, E.-J. (2007). A practical solution to the pervasive problems of p values. *Psychonomic bulletin & review*, 14(5), 779-804. doi: 10.3758/bf03194105
- Wang, P. P., & Bellugi, U. (1994). Evidence from two genetic syndromes for a dissociation between verbal and visual-spatial short-term memory. *Journal of Clinical and Experimental Neuropsychology*, 16(2), 317-322. doi: 10.1080/01688639408402641
- Wang, P. P., Doherty, S., Rourke, S. B., & Bellugi, U. (1995). Unique profile of visuo-perceptual skills in a genetic syndrome. *Brain and Cognition*, 29(1), 54-65. doi: 10.1006/brcg.1995.1267

DEVELOPMENT OF VISUOSPATIAL STM

Wansard, M., Bartolomeo, P., Bastin, C., Segovia, F., Gillet, S., Duret, C., & Meulemans, T.

(2015). Support for distinct subcomponents of spatial working memory: A double dissociation between spatial–simultaneous and spatial–sequential performance in unilateral neglect. *Cognitive Neuropsychology*, 32(1), 14-28. doi: 10.1080/02643294.2014.995075

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Table 1. Characteristics of the groups from typically developing children

Groups	4-5 y.o.	6-7 y.o.	8-10 y.o.	11-15 y.o.	Adults
N	19 (12 F)	21 (15 F)	19 (12 F)	21 (12 F)	19 (11 F)
Mean Age in months (SD)	61 (7)	82 (8)	114 (11)	157 (16)	293 (30)
Range of Age	48-70	73-95	96-128	132-189	246-348

Note. y.o.= years old; N= number of participants; F= females

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Table 2. Bayes factors for all effects of the model across matched models.

Effects	Bayes factors
Presentation	876.30
Arrangement	3.15E+58
Age	2.96E+32
Presentation * Arrangement	154.57
Presentation * Age	8.72
Arrangement * Age	2.24E+9
Presentation * Arrangement * Age	0.65

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Table 3. Descriptive and visuospatial STM measures for all groups.

	WS group Mean (SD)	TD_{WS} group Mean (SD)	DS group Mean (SD)	TD_{DS} group Mean (SD)
Age (months)	263.38 (136.20)	78.57 (27.49)	204.1 (77.26)	59.5 (17.93)
IQ				
Vocabulary	21.52 (5.26)	20.14 (7.64)	10.09 (6.26)	14.05 (4.83)
Similarities	17.24 (5.58)	14.43 (6.55)	8.57 (7.70)	11.77 (7.50)
Block design	20.24 (6.43)	23.71 (7.23)	19.09 (5.51)	19.41 (5.89)
Concept identification	12.90 (4.01)	12.48 (3.71)	9.32 (4.89)	9.64 (4.76)
Verbal STM	5.10 (1.73)	5.10 (1.22)	2.59 (0.73)	4.82 (1.33)
Central Executive	4.48 (1.91)	4.62 (1.83)	1.50 (1.57)	3.36 (1.94)
Visuospatial STM				
Simultaneous				
Unstructured	7.95 (3.35)	9.76 (3.39)	6.55 (3.51)	7.32 (3.21)
Structured	9.62 (4.95)	13.24 (5.42)	7.73 (4.85)	9.41 (4.16)
Sequential				
Unstructured	8.14 (3.26)	10 (4.17)	7.77 (2.18)	6.82 (3.94)
Structured	9.24 (4.46)	12.19 (4.79)	9.36 (3.34)	8.96 (4.62)

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Table 4. Bayes factors for all effects of the model across matched models.

Effects	Bayes factors
Presentation	0.21
Arrangement	4.16E+8
Group	1.37
Presentation * Arrangement	0.72
Presentation * Group	0.26
Arrangement * Group	4.22
Presentation * Arrangement * Group	0.44

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Table 5. Bayes factors for all effects of the model across matched models.

Effects	Bayes factors
Presentation	0.39
Arrangement	13212.68
Group	0.40
Presentation * Arrangement	0.22
Presentation * Group	7.85
Arrangement * Group	0.36
Presentation * Arrangement * Group	0.28

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Figure 1. Alternative two processing routes model of visuospatial STM.

Figure 2. Interactions between (a) Age and Presentation mode and (b) Age and Arrangement.

Means and standard errors are displayed.

Figure 3. Interaction between Presentation mode and Arrangement. Means and standard errors are displayed.

Figure 4. Interaction between Age and Presentation mode for the Arrangement effect. Means and standard errors are displayed.

Figure 5. Illustration of the interaction between Arrangement and Group for WS and TD_{WS} groups in visuospatial STM tasks.

Figure 6. Illustration of the interaction between Presentation and Group for DS and TD_{DS} groups in visuospatial STM tasks.