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Exploring spatial working memory performance in individuals with Williams syndrome: The effect of presentation format and configuration

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ABSTRACT

Williams syndrome (WS) is a neurodevelopmental disorder associated with an impaired capacity for visuospatial representation. Individuals with WS have a specific weakness in spatial processing, while visual components are relatively well preserved. This dissociation is apparent in working memory function too. The present study aimed to further investigate spatial working memory performance in individuals with WS, analyzing whether their impaired WM performance regards both simultaneous and sequential spatial formats, and whether presenting configurations differently might reduce their difficulties. These issues were examined by administering simultaneous and sequential spatial tasks, in which the information to be recalled was presented in random or arranged configurations.

Our results showed that individuals with WS performed less well than typically developing (TD) children in the spatial-simultaneous task, but not in the spatial-sequential one. The presence of a pattern enhanced the performance of both groups, but the difference between the two groups' performance in the spatial simultaneous task remained, albeit to a lesser degree.

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1. Introduction

Williams syndrome (WS) is a developmental disorder caused by a hemizygous deletion of approximately 26 genes on the long arm of chromosome 7 (7q11.23) (Peoples et al., 2000). The syndrome has an estimated prevalence in the range of 1 in 7500 to 1 in 20,000 population (Stromme, Bjornstad, & Ramstad, 2002). Most individuals with WS have intellectual disabilities, with an IQ in the mild to moderate range (Howlin, Davies, & Udwin, 1998; Udwin, Yule, & Martin, 1987). Their cognitive profile is characterized by strengths in some verbal abilities (Howlin et al., 1998; Udwin et al., 1987), contrasting with deficits in visuospatial cognition and visuospatial construction (Martens, Wilson, & Reutens, 2008). A more in-depth examination of the spatial skills of people with WS suggests that their visuospatial difficulties reflect deficiencies in the spatial functions supported by the parietal lobes (Atkinson et al., 2003; Hoffman, Landau, & Pagani,

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2003; Landau & Hoffman, 2007), while visual functions supported by the ventral visual stream seem to be relatively preserved (Landau, Hoffman, & Kurz, 2006; Musolino & Landau, 2012; Reiss, Hoffman, & Landau, 2005).

As concerns working memory (WM), previous studies have indicated that individuals with WS have a relatively well-preserved verbal WM and a relatively impaired visuospatial WM (e.g. Carney, Brown, & Henry, 2013; Klein & Mervis, 1999; Jarrold, Baddeley, & Hewes, 1999; Wang & Bellugi, 1994): the WM profile of individuals with WS is usually characterized by a domain-specific weaker performance in visuospatial tasks by comparison with typically developing (TD) children matched for mental age, whereas their performance in the verbal components is usually comparable with that of TD children (e.g. Costanzo et al., 2013). This was also confirmed in a recent study by Carney et al. (2013), who analyzed the developmental trajectories of individuals with WS and Down syndrome (DS) in verbal and visuospatial short-term memory by comparison with those of TD children. The intra- and inter-group comparisons showed the typical pattern of results when individuals with WS were examined (i.e. an impairment in the visuospatial component), but no significant developmental divergence in the verbal short-term memory domain between the group with WS and the TD children matched for mental age or chronological age. The WS group's verbal and visuospatial short-term memory skills developed, like their general cognitive development at similar rates to those of the TD group. These findings (Carney et al., 2013) thus seem to suggest that the weaker performance in the visuospatial domain usually seen in individuals with WS does not coincide with divergent trajectories of development.

Another aspect to consider regarding the visuospatial component is that it is not a unitary system, and this might have an influence when individual differences are examined. For example, according to the Logie (1995) model, visuospatial WM consists of a visual store (known as the visual cache) and a rehearsal mechanism (known as the inner scribe). Consistent with this distinction, there is a large body of evidence to support a dissociation between visual and spatial memory, based on studies using the selective interference paradigm (Della Sala, Gray, Baddeley, Allamano, & Wilson, 1999; Klauer & Zhao, 2004; Quinn & McConnell, 1996), and on neuropsychological research (Carlesimo, Perri, Turriziani, Tomaiuolo, & Caltagirone, 2001; Farah, Hammond, Levine, & Calvanio, 1988; Luzzatti, Vecchi, Agazzi, Cesa-Bianchi, & Vergani, 1998), and developmental studies (Gathercole & Pickering, 2000; Hamilton, Coates, & Heffernan, 2003; Logie & Pearson, 1997; Pickering, Gathercole, & Peaker, 1998; Pickering, Gathercole, Hall, & Lloyd, 2001). More recently, Pazzaglia and Cornoldi (1999), and Mammarella, Pazzaglia, and Cornoldi (2008) likewise suggested to distinguish between three components: a visual component in charge of processing shapes and colors, and two kinds of spatial component, both involved in memorizing patterns of spatial locations, but presenting them in a different format and consequently using different spatial processes – simultaneous in one case, sequential in the other. Evidence collected with various groups of children support the distinction between visual and spatial-simultaneous processes (Mammarella, Cornoldi, & Donadello, 2003), and between spatial-simultaneous and spatial-sequential processes (Mammarella et al., 2006).

The literature on individuals with WS also points to possible differences in the pattern of performance depending on the visuospatial WM components considered, although the results are still not clear. For example, Vicari, Bellucci, and Carlesimo (2006) found more severe spatial than visual WM impairment in individuals with WS, while other studies found a significant impairment in both visual and spatial WM (e.g. Rhodes, Riby, Park, Fraser, & Campbell, 2010; Sampaio, Sousa, Fernandez, Henriques, Goncalves, 2008). More recently, Lanfranchi, De Mori, Mammarella, Carretti and Vianello (in press) examined how individuals with WS perform in spatial WM tasks in more depth, distinguishing between simultaneous and sequential presentation formats, as well as between passive and active tasks. Using passive tasks, they found that individuals with WS performed less well than TD children of the same mental age in a spatial-simultaneous WM task, but not in a spatial-sequential one; when active tasks were used, on the other hand, the WS individuals performed worse in both spatial-sequential and spatial-simultaneous tasks. The authors suggested that individuals with WS perform less well in passive spatial-simultaneous WM tasks due to an impaired processing of global aspects of visuospatial stimuli (Bihrle, Bellugi, Delis, & Marks, 1989; Hoffman et al., 2003), while their poor performance in active tasks, both spatial-sequential and spatial-simultaneous, would be a consequence of their difficulty in performing tasks that require both the storage and the manipulation of information, as seen in other etiologies of intellectual disability.

Similar findings had already been reported by Lanfranchi, Carretti, Spanò, and Cornoldi (2009), Carretti and Lanfranchi (2010), and Carretti, Lanfranchi and Mammarella (2013) in individuals with DS, who revealed a specific impairment in spatial-simultaneous tasks, but not in spatial-sequential tasks. Subsequent studies demonstrated, however, that this weakness in the spatial-simultaneous condition persisted even when spatial locations were grouped to form a pattern (Carretti et al., 2013), showing that individuals with DS are less able than TD children of the same mental age to take advantage of a configuration, thus suggesting a role for encoding strategies.

In the light of those findings, the aim of the present study was to compare the performance of individuals with WS in a spatial-sequential and spatial-simultaneous WM task to see whether the advantage of spatial-simultaneous over spatial-sequential presentation formats in WM tasks seen in the typically-developing population (Allen, Baddeley, & Hitch, 2006; Blalock & Clegg, 2010; Lecerf & de Ribaupierre, 2005), and in Down syndrome (Carretti & Lanfranchi, 2010; Carretti et al., 2013), exists in WS too. We also aimed to explore whether individuals with WS are capable of benefiting from configured material, in both spatial-sequential and spatial-simultaneous tasks. For this purpose, spatial-sequential and spatial simultaneous tasks were devised in two versions, i.e. in random configurations (in which filled cells were separated by spaces and did not give rise to any patterns), and in pattern configurations (in which the filled cells were grouped to form a visual pattern).

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