



PAPER

Egocentric and allocentric navigation strategies in Williams syndrome and typical development

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Abstract

Recent findings suggest that difficulties on small-scale visuospatial tasks documented in Williams syndrome (WS) also extend to large-scale space. In particular, individuals with WS often present with difficulties in allocentric spatial coding (encoding relationships between items within an environment or array). This study examined the effect of atypical spatial processing in WS on large-scale navigational strategies, using a novel 3D virtual environment. During navigation of recently learnt large-scale space, typically developing (TD) children predominantly rely on the use of a sequential egocentric strategy (recalling the sequence of left–right body turns throughout a route), but become more able to use an allocentric strategy between 5 and 10 years of age. The navigation strategies spontaneously employed by TD children between 5 and 10 years of age and individuals with WS were analysed. The ability to use an allocentric strategy on trials where spatial relational knowledge was required to find the shortest route was also examined. Results showed that, unlike TD children, during spontaneous navigation the WS group did not predominantly employ a sequential egocentric strategy. Instead, individuals with WS followed the path until the correct environmental landmarks were found, suggesting the use of a time-consuming and inefficient view-matching strategy for wayfinding. Individuals with WS also presented with deficits in allocentric spatial coding, demonstrated by difficulties in determining short-cuts when required and difficulties developing a mental representation of the environment layout. This was found even following extensive experience in an environment, suggesting that – unlike in typical development – experience cannot contribute to the development of spatial relational processing in WS. This atypical presentation of both egocentric and allocentric spatial encoding is discussed in relation to specific difficulties on small-scale spatial tasks and known atypical cortical development in WS.

Research highlights

- Large-scale navigation strategies in Williams syndrome were examined using a virtual environment.
- Where TD children rely on an egocentric strategy to spontaneously navigate, individuals with WS use an atypical view-matching strategy.
- Individuals with WS were found to have difficulties in both egocentric and allocentric spatial processing.
- WS difficulties are in line with known abnormalities in cortical areas for spatial coding.

Introduction

The development of large-scale spatial abilities is imperative to learning how to successfully navigate through

novel and diverse environments and essential in the pursuit of independent living. Use of an egocentric spatial frame of reference, involving the encoding of environmental locations in relation to the self, allows an individual to accurately retrace a route from one location to another. However, being able to use an allocentric spatial reference frame, that is, encoding the spatial relationships between landmarks (O'Keefe & Nadel, 1978), facilitates more complex navigation, such as the ability to make short-cuts or to understand how to relocate oneself when starting from a novel location in a familiar environment.

In typical development, both children and adults predominantly rely on the use of a sequential egocentric strategy during wayfinding, by recalling the temporal order of body turns at specific environmental locations

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(Bullens, Iglói, Berthoz, Postma & Rondi-Reig, 2010; Iglói, Zaoui, Berthoz & Rondi-Reig, 2009). However, using a virtual navigation task, Bullens and colleagues found that the ability to successfully employ an allocentric strategy to navigate develops progressively between 5 and 10 years of age, but is systematically utilized only when the task demands a more complex understanding of the spatial relationships in the environment. This is in line with research that suggests a developmental change between 6 and 8 years of age from a reliance on viewpoint-dependent spatial processing, or the use of stored views of spatial locations in relation to the self, to more flexible viewpoint-independent processing, or environment-centred spatial representations that allow accurate recall irrespective of the viewer's movements (Nardini, Thomas, Knowland, Braddick & Atkinson, 2009).

Williams syndrome (WS) is a rare genetic disorder resulting from a hemizygotic deletion of more than 25 genes on the long arm of chromosome 7 (Osborne, 2012). WS can be characterized by a distinctly uneven cognitive profile, with poor visuospatial processing relative to verbal abilities (Bellugi, Lichtenberger, Jones, Lai & St George, 2000; Jarrold, Baddeley & Hewes, 1998). Deficits on small-scale visuospatial tasks in WS have been well documented (e.g. Bellugi, Wang & Jernigan, 1994; Farran & Jarrold, 2003; Hoffman, Landau & Pagani, 2003). Individuals with WS have also been reported to exhibit specific difficulties on tasks requiring the encoding of spatial relationships between landmarks in a small-scale array (Bernardino, Mouga, Castelo-Branco & van Asselen, 2013; Nardini, Atkinson, Braddick & Burgess, 2008). In addition, difficulties on tasks requiring imagined rotations of the self and objects have been identified (Farran, Jarrold & Gathercole, 2001; Stinton, Farran & Courbois, 2008), indicative of particular difficulties in this disorder with encoding spatial locations of objects in relation to the self and other objects.

Imagined rotations of the self, in particular, require the ability to update egocentric spatial locations within an allocentric frame of reference, and this ability is thought to be supported by hippocampal and medial temporal lobe structures (Burgess, 2008; Lambrey, Doeller, Berthoz & Burgess, 2012; Vann, Aggleton & Maguire, 2009). Known structural and functional abnormalities of the hippocampus in WS (Meyer-Lindenberg, Mervis, Sarpal, Koch, Steele, Kohn, Marengo, Morris, Das, Kippenhan, Mattay, Weinberger & Berman, 2005) are therefore likely to be associated with difficulties observed in WS on tasks that require this kind of imagined rotation. In addition, given the known deficits in dorsal stream functioning in WS (Atkinson, Braddick, Anker, Curran, Andrew,

Wattam-Bell & Braddick, 2003; Atkinson, King, Braddick, Nokes, Anker & Braddick, 1997), difficulties in egocentric spatial coding supported by the dorsal stream in typical adults (Milner & Goodale, 1995) may also contribute to such deficits on imagined rotation tasks in these individuals.

In typical adults, increased activity in the hippocampal region is associated both with allocentric spatial coding (understanding the relationships between objects in an array) and with large-scale navigation (Burgess, Jeffery & O'Keefe, 1999; Burgess, Maguire & O'Keefe, 2002). Individuals with WS are, therefore, also likely to present with difficulties on such large-scale tasks. Moreover, in typical adults, the ability to imagine the self rotating predicts performance on navigation tasks that require the individual to constantly update self-to-object and object-object locations when moving through an environment, namely the ability to utilize allocentric spatial coding (Kozhevnikov, Motes, Rasch & Blajenkova, 2006). Thus, difficulties on small-scale tasks in WS suggest that large-scale spatial tasks, particularly those requiring allocentric encoding, are likely to be problematic for this group.

On large-scale spatial tasks, individuals with WS can successfully learn to navigate and accurately retrace their route both in real-world (Farran, Blades, Boucher, Tranter, Jarrold, Stinton & Courbois, 2010) and virtual environments (Farran, Courbois, Van Herwegen & Blades, 2012). However, in line with small-scale relational coding difficulties, Farran and colleagues found that in a real-world task, individuals with WS showed deficits on tasks requiring an understanding of spatial relationships in the environment. That is, when asked to point to the location of unseen landmarks from positions along a route, individuals with WS displayed a high number of errors compared to TD children, consistent with the predicted difficulties in allocentric encoding in WS.

Using a radial-arm maze, Mandolesi, Addona, Foti, Menghini, Petrosini and Vicari (2009) examined the mnemonic components related to large-scale visuospatial difficulties in WS. The results showed that individuals with WS were impaired in acquiring procedural competencies and spatial working memory. This is in line with other findings implicating the role of a deficit in dorsal stream functioning on poor spatial working memory in WS (O'Hearn, Courtney, Street & Landau, 2009). Mandolesi and colleagues found that perseverative errors in exploration were also evident in some participants with WS, a further reflection of spatial memory and planning difficulties in this group. Others have also identified similar perseverative errors in WS during navigation (Farran *et al.*, 2012). Such difficulties in spatial working memory and use of inefficient exploration strategies in WS may

underlie some of the difficulties demonstrated on tasks requiring the recall of landmark locations (e.g. Farran *et al.*, 2010). Importantly, Mandolesi and colleagues found that some individuals with WS demonstrated an understanding of the spatial representation of the test layout when asked to draw the environment; the only indication in the literature of possible spatial relational knowledge in WS. In another real-world study, however, search strategies in WS were found to be disorganized and ineffective compared with TD children, and individuals with WS failed to develop an understanding of the environmental layout (Foti, Petrosini, Cutuli, Menghini, Chiarotti, Vicari & Mandolesi, 2011).

Our understanding of whether individuals with WS are able to develop spatial relational knowledge and utilize an allocentric spatial frame of reference in large-scale space, therefore, remains ambiguous. Although individuals with WS have demonstrated successful route-learning abilities, to date no study has specifically examined the navigational strategies employed in this group, and in particular the use of egocentric and allocentric spatial frames of reference. The aim of the present study was to examine navigational strategies in WS in a large-scale virtual environment (VE), compared to TD children between 5 and 10 years of age. Using a virtual cross-maze design, spontaneously employed navigational strategies were examined across groups, together with the ability to make use of allocentric spatial coding when required.

In typical adults and children, extended experience within an environment leads to enhanced understanding of the spatial relationships between landmarks (Anooshian & Young, 1981; Golledge & Spector, 1978; Siegel & White, 1975). Reduced opportunities for independent navigation and active exploration in real-world environments may inhibit individuals with physical disabilities from acquiring allocentric knowledge (Foreman, 2007; Foreman, Stanton, Wilson & Duffy, 2003). Findings have suggested that this may also be the case for individuals with learning difficulties (Farran *et al.*, 2010; Mengue-Topio, Courbois, Farran & Sockeel, 2010). Given the time restraints and physical demands involved in navigating a route multiple times, real-world tasks in WS may be limited by the scope of experience that each participant can gain from an environment. This suggests that difficulties in spatial relational knowledge identified in WS on previous real-world tasks (e.g. Farran *et al.*, 2010) may be a consequence of this lack of environmental experience, and that further experience in these environments may have led to the development of more complex spatial representations.

Given the potential of exploring a VE multiple times within a relatively short time period and without the

physical demands of real-world environments, the use of VEs to examine large-scale spatial difficulties may be a useful resource in WS. Furthermore, both in typical development and individuals with learning difficulties, learning in a VE has been shown to transfer successfully to real-world comparisons (Bailey & Witmer, 1994; Farrell, Arnold, Pettifer, Adams, Graham & MacManamon, 2003; Foreman *et al.*, 2003; Rose, Attree, Brooks, Parslow, Penn & Ambihapahan, 2000; Wilson, Foreman & Tlauka, 1996). As such, VEs can be taken as suitable equivalents to real-world tasks for the present study, and provide a unique opportunity to study spatial abilities in WS following extended experience in an environment.

Allocentric spatial strategies are thought to be supported in part by the hippocampal region (Iaria, Chen, Guariglia, Ptito & Petrides, 2007; King, Burgess, Hartley, Vargha-Khadem & O'Keefe, 2002; McNamara & Shelton, 2003; O'Keefe & Nadel, 1978), particularly the right hippocampus (Iglói, Doeller, Berthoz, Rondi-Reig & Burgess, 2010). Given the known hippocampal impairments in WS (Meyer-Lindenberg *et al.*, 2005), difficulties in the use of an allocentric strategy in WS were anticipated in the current study. Egocentric spatial representations, in contrast, are related to left hippocampal (Iglói *et al.*, 2010) and dorsal stream activation (Milner & Goodale, 1995), a finding supported by neuroimaging during the use of egocentric navigation strategies (Committeri, Galati, Paradis, Pizzamiglio, Berthoz & LeBihan, 2004; Galati, Lobel, Vallar, Berthoz, Pizzamiglio & Le Bihan, 2000; Neggers, Van der Lubbe, Ramsey & Postma, 2006). Deficits in dorsal stream functioning have been identified in individuals with WS (e.g. Atkinson *et al.*, 2003; Atkinson *et al.*, 1997; Nakamura, Kaneoke, Watanabe & Kakigi, 2002). As such, despite previous findings of successful performance on route learning tasks in WS (e.g. Farran *et al.*, 2010), it was hypothesized that, where TD children may predominantly employ a sequential egocentric strategy, individuals with WS may demonstrate alternative navigation strategies on such tasks, suggestive of an atypical pattern of coding spatial frames of reference in large-scale environments.

Methods

Participants

Sixty-eight typically developing (TD) children were recruited from three primary schools, and separated into four age groups; 5-, 6-, 8-, and 10-year-olds. Twenty-one individuals with Williams syndrome (WS) were recruited via the Williams Syndrome Foundation, UK. All WS

participants had received a positive diagnosis of WS, based on a 'fluorescence in situ hybridization' (FISH) test for deleted Elastin gene on the long arm of chromosome 7, as well as phenotypic diagnosis from a clinician.

All TD participants were tested in a quiet room within their schools, whilst WS participants were tested either at their home or in a testing room at the university. Ten participants from the TD group ($N = 7$, 5-year-olds; $N = 2$, 6-year-olds; $N = 1$, 10-year-old) and four from the WS group did not manage to complete the task successfully due to lack of motivation. Data for these participants were excluded from subsequent analyses. Six more TD 5-year-olds were therefore recruited to maintain comparable group sizes, all of whom completed the tasks successfully. Therefore, data were analysed from 64 TD children (5-year-olds: $N = 16$ [$M = 5.62$ years, $SD = .36$]; 6-year-olds: $N = 15$ [$M = 6.74$ years, $SD = .30$]; 8-year-olds: $N = 17$ [$M = 8.31$ years, $SD = .35$]; and 10-year-olds: $N = 16$ [$M = 10.08$ years, $SD = .33$]), and 17 participants with WS ($M = 21.85$ years, $SD = 8.49$). Verbal and non-verbal abilities were assessed using the British Picture Vocabulary Scale-III (BPVS-III; Dunn, Dunn, Styles & Sewell, 2009) and the Ravens Coloured Progressive Matrices (RCPM; Raven, Raven & Court, 2003), respectively.

Virtual environment (VE)

The interactive VE cross-maze was developed using the Vizard Development Edition software (www.worldviz.com), and presented on a 17" laptop screen. Adapted from a five-arm maze used in other studies (e.g. Bullens *et al.*, 2010; Iglói *et al.*, 2009), the VE cross-maze task presented participants with a more simple environmental layout within which individuals were able to use either a 'sequential egocentric' or 'allocentric' spatial strategy to navigate, or a combination of the two (a 'mixed' strategy). For a bird's-eye schematic image of the cross-maze layout, see Figure 1.

The VE consisted of four paths (A, D, G, and J) extending from a central square. The central square was made up of eight paths, made distinct by a turn or a junction (B, C, E, F, H, I, K and L). The alleys of the central square and the extending pathways all had red brick walls and therefore appeared identical from any starting position and all paths between decision points were the same length. Surrounding the environment were six distal landmarks, consisting of three different landmarks appearing twice each (two trees, two city landscapes and two red towers). Each distal landmark appeared twice around the environment so that participants would have to encode landmarks in terms of their relationship to each other and the paths, and not use

them as directional cues. Participants navigated through the environment using the keyboard arrow-keys.

Design and procedure

Learning phase

Participants were asked to navigate from the starting position at the end of path A to find a 'hidden exit' at the end of path G. Participants were unable to see the end of any other pathway from any starting position within the environment. During the learning phase, participants were first shown the optimal route to the hidden exit by following a grass path. Once the target location was found, a 'Yippee!' sound was played and the program window closed. The participant was then returned to the starting position in path 'A', without a grass path, for the first learning trial. Here, participants were required to walk the route from memory. During the learning trials (maximum 10 trials) participants had to navigate down the correct path to reach the hidden target without error on two trials to move on to the testing phase. Invisible walls were positioned one-quarter of the way down incorrect paths to guide learning. Participants were therefore able to look down the incorrect turns but not travel down them further than this point. If participants incorrectly navigated to the point of hitting an invisible wall, this was counted as an error.

Testing spontaneous navigation strategies

Following successful learning, participants were told that the invisible walls had now been removed and so they could travel down any path they wanted. Participants were instructed that for each test trial, they must try to reach the hidden exit in path G by the shortest route possible. Participants then completed 12 test trials.

To intermittently test the strategy that the participant was using, the 12 test trials were interspersed with four 'strategy test' trials (trials 3, 7, 9, and 12); where instead of the normal starting point in path A, the participant was unsuspectingly placed at the end of path J as a starting location. From both starting positions (J and A), the views contained similar distant landmarks, including a tree, cityscape and red tower. Therefore, participants who spontaneously relied on the use of an egocentric strategy may not have encoded the spatial layout, even if they could identify similar landmarks, and would have followed the same sequence of body turns as usual (Figure 1b). Participants spontaneously using an allocentric strategy, however, would have identified the different spatial relationships and used an allocentric strategy to

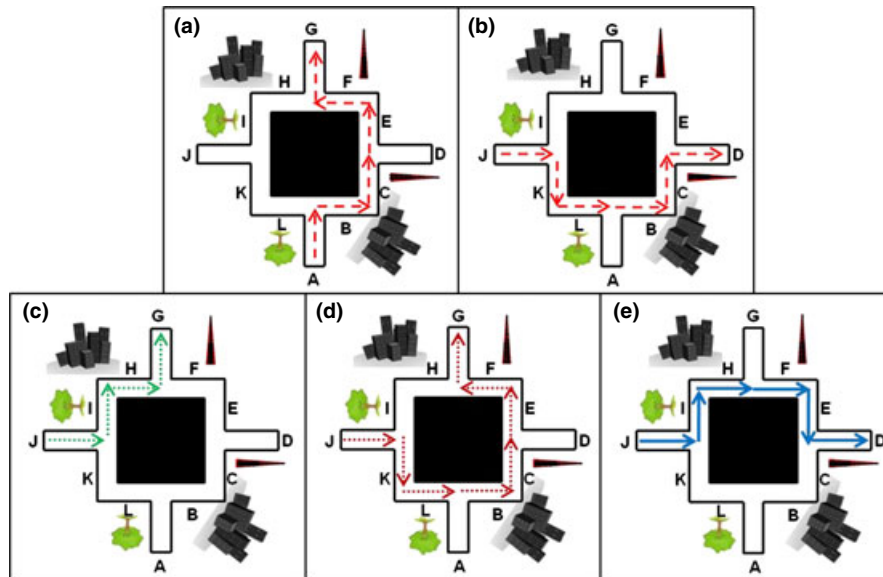


Figure 1 Schematic layout of cross-maze with routes taken by participants using different strategies: (a) route taken during learning and eight normal test trials, (b) route taken using a Sequential Egocentric strategy, (c) Allocentric, (d) Mixed, (e) Mirrored Egocentric.

navigate to the correct exit (Figure 1c). On the four spontaneous strategy tests, the ends of both path G and path D revealed the reward sound as positive feedback for the use of either of these spontaneous strategies. Therefore, participants who used an egocentric strategy would not be alerted to the change in starting position, and they would continue to use their spontaneous strategy on subsequent test trials.

Enforced allocentric strategy trials

To examine whether participants were able to navigate using an allocentric strategy when required, they were asked to navigate in the same cross-maze to a hidden exit from different starting points. Participants were first told that the hidden exit had moved to a new place. They were then placed in path J and asked to follow the grass path to the hidden exit now in path A. Participants then completed the learning trials without the grass until they had successfully navigated to the new hidden exit on two trials, i.e. they had reached the learning criterion. Following learning, participants were informed that they would now have to find this hidden exit from different starting positions and were encouraged to take note of the environmental landmarks to remind them of the location of the hidden exit. Participants completed six test trials from different starting positions (three from path G and three from path D, presented in a random order). The reward sound was played only at the end of path A, following the use of an allocentric strategy.

Layout knowledge test

Following the allocentric strategy test, to further examine allocentric understanding of the environmental layout, participants were asked to choose the correct layout of the environment from a set of six map options (Figure 2).

Results

Verbal and non-verbal abilities

To examine the difference across groups on BPVS and RCPM scores, one-way analyses of variance (ANOVA) were conducted for each, with group (five levels; 5y, 6y, 8y, 10y, and WS) as a between-subjects factor (see Table 1). This demonstrated an uneven cognitive profile in WS, characteristic of the disorder (Bellugi *et al.*, 2000; Jarrold *et al.*, 1998), with non-verbal abilities at a level no different from TD 5-year-olds, and relatively higher verbal abilities, significantly greater than TD 5- and 6-year-olds, at the level of TD 8- and 10-year-olds.

Learning trials

Following the original learning trial with the grass path, the number of trials (including two correct criterion trials) taken to successfully reach criterion (required to advance to the testing phase) was examined in each group.

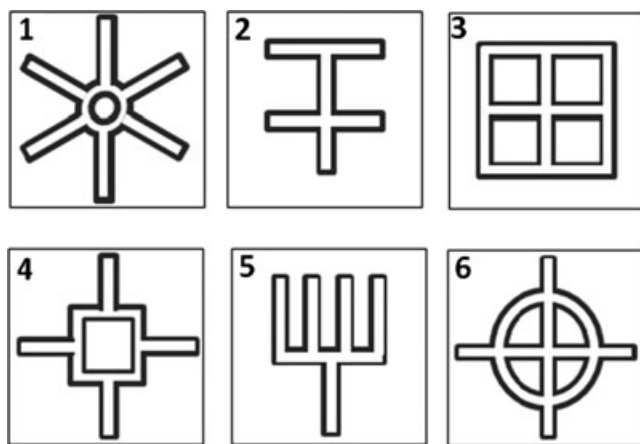


Figure 2 Layout knowledge test: six environment layout choices.

Results of a one-way ANOVA showed a significant effect of age on mean number of trials taken to learn the route to the hidden target, $F(4, 80) = 4.589, p = .002$. Tukey-corrected post-hoc tests showed that TD 10-year-olds ($M = 2.75, SD = .68$) required significantly fewer trials to learn the route than the 5-year-old ($M = 4.38, SD = 1.71, p = .033$), 6-year-old ($M = 4.67, SD = 1.88, p = .009$) and WS ($M = 4.76, SD = 2.08, p = .003$) groups, but not 8-year-olds ($M = 3.76, SD = .97$). No significant difference in number of trials was found across any other groups ($p > .05$, for all).

Performance on test trials

To examine whether performance on the eight trials in the test phase – not including the four spontaneous strategy trials – was successful for each participant, the percentage of trials without error (direct route taken to the exit, without traversing more than half-way down incorrect paths) was calculated for each group.

Results of a one-way ANOVA showed steady performance on the eight test phase trials following learning in each group, with no significant difference in percentage of correct test trials found across groups [5 years, $M = 77.34\%$ ($SD = 20.01\%$); 6 years, $M = 71.67\%$ ($SD = 26.50\%$); 8 years, $M = 71.32\%$ ($SD = 21.99\%$); 10 years, $M = 80.47\%$ ($SD = 21.39\%$); WS, $M = 72.22$ (19.91%), $F(4, 81) = .562, p = .691$.

Spontaneous strategy trials

Strategy types

The spontaneous strategies used by participants in each of the four strategy trials were grouped into four different categories. As in previous findings using VEs (Bullens *et al.*, 2010; Iglói *et al.*, 2009), three of the strategies observed were (a) ‘Sequential Egocentric’ (Figure 1b), where participants repeated an identical sequence of body turns in both the normal and strategy tests; (b) ‘Allocentric’ (Figure 1c), where participants used environmental cues and an understanding of the environment layout to reach the correct hidden exit by the shortest route; and (c) ‘Mixed’ (Figure 1d), where participants demonstrated a change from a sequential egocentric strategy at the start of the trial to the use of environmental cues within one trial. Some participants in the current study also demonstrated the use of a fourth strategy that we have labelled (d) ‘Mirrored Egocentric’ (Figure 1e), where during the strategy tests participants traversed along a route towards the hidden target that was the mirror image of the sequential egocentric route. This strategy potentially demonstrated an understanding of the symmetrical layout of the environment and thus an alternative route to the hidden exit, but without consideration of the positioning of landmarks. Participants who did not reach the hidden exit at the end of path D or G using any of these strategies, or who got lost

Table 1 Mean (*SD*) participant scores on BPVS-III and RCPM

	Group					ANOVA		
	5 years ($N = 16$)	6 years ($N = 15$)	8 years ($N = 17$)	10 years ($N = 16$)	WS ($N = 17$)	F (df)	p	Post-hoc ^a
BPVS-III ^b	78.94 (13.49)	91.33 (14.25)	112.35 (15.78)	130.81 (15.18)	123.65 (22.38)	27.546 (4, 80)	< .001	5 = 6 < 8, 10 and WS 8 < 10
RCPM ^c	19.38 (3.85)	24.33 (4.91)	28.00 (5.02)	30.38 (3.12)	16.76 (3.29)	31.825 (4, 79)	< .001	WS = 8 and 10 5 < 6, 8, and 10 6 < 10 WS < 6, 8 and 10

^aTukey-corrected post-hoc tests, ‘=’ refers to no significant difference at .05 level, and ‘<’ denotes $p < .01$. ^bBPVS-III: British Picture Vocabulary Scale-III raw scores. ^cRCPM: Ravens Coloured Progressive Matrices (RCPM) raw scores.

during the spontaneous tests, were allocated the category (e) 'Non-specific' for those trials. For mean percentage of each type of strategy used across the four spontaneous strategy trials see Figure 3a.

Data were analysed to examine the differences between groups on the percentage of each strategy type used on spontaneous trials. As data did not meet normality assumptions (Kolmogorov-Smirnov, $p < .05$) for any groups, Kruskal-Wallis tests were conducted, with post-hoc Mann-Whitney tests. Results showed a significant difference across groups on percentage of egocentric strategy use, $\chi^2(4) = 13.946$, $p = .007$, due to the WS group using an egocentric strategy significantly less often than all other groups (5-year-olds, $p = .006$; 6-year-olds, $p < .001$; 8-year-olds, $p = .048$; and 10-year-olds, $p = .026$). A significant difference across groups on percentage of mixed strategy use was also found, $\chi^2(4) = 13.292$, $p = .010$, with individuals with WS using a mixed strategy significantly more often than all other groups (5-year-olds, $p = .025$; 6-year-olds, $p = .005$; 8-year-olds, $p = .006$ and 10-year-olds, $p = .004$). Results also showed a significant difference across groups on percentage of mirrored strategy use, $\chi^2(4) = 14.634$, $p = .006$. Post-hoc tests showed that 10-year-olds used a mirrored strategy significantly more often than 5-year-olds ($p = .037$), 6-year-olds ($p = .010$), and WS ($p = .006$), and

8-year-olds used a mirrored strategy significantly more often than 6-year-olds ($p = .048$) and WS ($p = .036$). No significant group differences were found on percentage of allocentric or non-specific strategies used ($p > .05$ for both).

Data were also analysed to examine whether one strategy was used significantly more often than any other within each group separately. Results of Friedman's ANOVAs (five levels; egocentric, allocentric, mixed, mirrored, and non-specific) showed a significant effect of strategy type used on spontaneous trials in all groups: 5-year-olds, $\chi^2(4) = 22.932$, $p < .001$; 6-year-olds, $\chi^2(4) = 29.434$, $p < .001$; 8-year-olds, $\chi^2(4) = 10.076$, $p = .039$; 10-year-olds, $\chi^2(4) = 15.300$, $p = .004$; and WS, $\chi^2(4) = 21.543$, $p < .001$. Post-hoc Wilcoxon Signed Ranks tests showed that this was due to a significantly greater percentage of egocentric compared to allocentric and non-specific strategies in all TD groups; 5 years ($p = .002$ and $p = .014$); 6 years ($p = .002$ and $p = .003$); 8 years ($p = .015$ and $p = .022$); and 10 years ($p = .023$ and $p = .003$). A significantly greater percentage of egocentric than mixed was found in 6- ($p = .007$) and 8-year-olds ($p = .043$), and a significantly greater percentage of egocentric than mirrored for 5- and 6-year-olds ($p = .001$ for both). Only the 6-year-olds showed significantly less use of a mirrored strategy than allocentric ($p = .034$) or mixed ($p = .024$). In contrast, the WS group used a mixed strategy significantly more often than egocentric ($p = .030$), allocentric ($p = .041$) and mirrored ($p = .001$) strategies, and a mirrored strategy significantly less often than allocentric ($p = .015$) and non-specific ($p = .006$).

To examine the consistency of performance in each group across the four strategy trials, the percentage of participants who used the same strategy on at least three out of the four trials was calculated. The majority of participants in each of the TD groups were consistent in their use of strategy type (5-year-olds = 93.3%, 6-year-olds = 66.7%, 8-year-olds = 64.7%, and 10-year-olds = 75.0%). However, consistent strategy use across the four trials was not observed in participants with WS, with the majority of participants in this group (70.6%) using different strategies across trials.

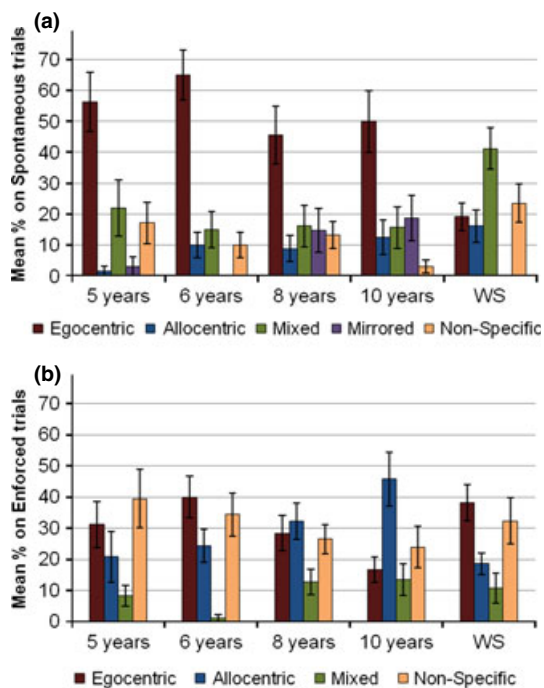


Figure 3 Percentage of participants in each group using each strategy during (a) the four spontaneous strategy trials, (b) the six enforced allocentric trials.

Allocentric score for spontaneous trials

An allocentric score from the four spontaneous trials was calculated for each participant. For each of the four trials, zero points were awarded for the use of a sequential egocentric strategy, or non-specific strategy, and two points were given for each allocentric strategy used. Given that the use of a mixed strategy may have demonstrated a change from an egocentric to an allocentric strategy within one trial, one point was

awarded for each mixed strategy used. One point was also allocated for each mirrored egocentric trial as this may have resulted from an understanding of the shape of the environment, a component of allocentric knowledge. Participants could therefore receive a maximum score of eight allocentric points across the four spontaneous trials. Total allocentric score was then converted to a percentage for analysis, for comparison to the corresponding score for enforced allocentric trials. For mean percentage allocentric score for each group, see Table 2.

Data were normally distributed for 8- and 10-year-olds and WS groups only (Kolmogorov-Smirnov, $p > .05$). However, given that normality was present for more than half of the groups, and ANOVA can be robust to violations of normality assumptions, parametric tests are reported. Non-parametric equivalents of the following analyses were also conducted, with comparable results. The result of a one-way ANOVA showed a significant difference across groups on percentage allocentric score on spontaneous trials, $F(4, 80) = 3.413$, $p = .013$. Tukey-corrected post-hoc tests showed that participants with WS had a significantly higher allocentric score than TD 5- and 6-year-olds ($p = .027$ and $p = .032$, respectively). No other significant differences were found between groups ($p > .05$).

Enforced allocentric trials

Strategy types

The strategy used on each of the six enforced allocentric trials was identified for each participant. Strategies were identified in the same way as for the spontaneous trials and consisted of sequential egocentric, allocentric, mixed, and non-specific strategies. For mean percentage of each strategy type used across the six enforced allocentric strategy trials see Figure 3b.

As for spontaneous trials, data were analysed to examine the differences between groups on the percentage of each strategy type used during enforced trials. As data violated normality assumptions for all groups (Kolmogorov-Smirnov, $p < .05$), Kruskal-Wallis tests were conducted, with post-hoc Mann-Whitney tests. Results showed a significant difference across groups on

percentage of egocentric strategies used, $\chi^2(4) = 9.528$, $p = .049$, with 6-year-olds and WS groups using an egocentric strategy significantly more than 10-year-olds ($p = .006$ and $p = .009$, respectively). Results also showed a significant difference across groups on percentage allocentric strategy used, $\chi^2(4) = 10.052$, $p = .040$, with 5-year-olds and WS groups using an allocentric strategy significantly less than 10-year-olds ($p = .013$ and $p = .014$, respectively).

To examine whether one strategy was used significantly more often than any other within each group separately, Friedman's ANOVAs (four levels: egocentric, allocentric, mixed, or non-specific) were conducted. Results showed a significant difference between strategy type used during enforced trials in 6-year-olds, $\chi^2(3) = 19.872$, $p < .001$, and WS, $\chi^2(3) = 13.800$, $p = .003$ groups, and a trend in 5-year-olds, $\chi^2(3) = 7.626$, $p = .054$. Post-hoc Wilcoxon Signed Ranks tests showed that for 6-year-olds, an egocentric strategy was used significantly more often than a mixed strategy ($p = .001$). Allocentric and non-specific strategies were also used more often than a mixed strategy in this group ($p = .007$ and $p = .003$, respectively). Similarly, 5-year-olds used an egocentric strategy significantly more often than a mixed strategy ($p = .016$). Individuals with WS, however, demonstrated the use of an egocentric strategy significantly more often than mixed ($p = .006$) and allocentric ($p = .003$) strategies, both of which would have resulted in successful navigation to the hidden exit. The predominant use of egocentric searching during the enforced trials in WS was, therefore, indicative of an ineffective navigation strategy in this group.

Allocentric score for enforced allocentric trials

An allocentric score from the six enforced trials was calculated for each participant using the same criteria as for spontaneous trials. Participants could therefore receive a maximum score of 12 allocentric points across the six trials. As before, allocentric scores were then converted to percentages for analysis. For mean percentage allocentric score for each group, see Table 2.

Data were normally distributed for all groups, except TD 5-year-olds. However, given that normality was

Table 2 Mean (SD) percentage allocentric score for spontaneous and enforced trials for each group

	Group				
	5 years ($N = 16$)	6 years ($N = 15$)	8 years ($N = 17$)	10 years ($N = 16$)	WS ($N = 17$)
Spontaneous strategy trials	14.06 (20.35)	14.17 (18.22)	24.26 (20.48)	29.69 (28.46)	36.76 (18.47)
Enforced allocentric trials	25.00 (32.06)	23.33 (19.47)	38.73 (22.81)	52.60 (31.58)	24.51 (11.96)

present for all but one of the groups in the sample, parametric tests are reported. Non-parametric equivalents of the following analyses were also conducted, with comparable results. Results of a one-way ANOVA showed a significant difference across groups on percentage allocentric score on enforced trials, $F(4, 80) = 4.251, p = .004$. Tukey-corrected post-hoc tests showed that TD 10-year-olds had significantly higher allocentric scores than TD 5-year-olds ($p = .019$), TD 6-year-olds ($p = .012$) and the WS group ($p = .014$).

Differences in allocentric score on spontaneous trials compared to enforced allocentric trials were examined. A two-way mixed ANOVA was performed on percentage allocentric score, with a between-participants factor of group (five levels: 5, 6, 8, 10 and WS) and within-participants factor of trial type (2 levels: spontaneous or enforced). Results showed a significant main effect of group, $F(5) = 4.278, p = .004$, and a significant main effect of trial type, $F(1) = 7.709, p = .007$. A significant interaction was also identified, $F(4) = 3.306, p = .015$. Results of post-hoc paired-samples t -tests for each group showed a significant increase in allocentric score from spontaneous to enforced trials for TD 8-year-olds, $t(16) = -2.397, p = .029$, and TD 10-year-olds, $t(15) = -2.400, p = .030$. In contrast, a significant decrease in allocentric score was demonstrated in participants with WS, $t(16) = 2.235, p = .040$. No significant difference was found for 5- or 6-year-olds.

To determine whether there were any effects of gender, spontaneous and enforced allocentric scores for males and females were also compared. Results found no significant differences between males and females on spontaneous or enforced allocentric scores in any group ($p > .05$ for all).

Layout knowledge test

The correct layout from a choice of six was chosen by 31.3% of participants in the 5-year-old group, 33.3% of 6-year-olds, 52.9% of 8-year-olds, 62.5% of 10-year-olds, and 35.3% of participants with WS. Even the oldest TD children did not, therefore, perform at an exceptionally high level on this task. However, this is in line with what could be expected based on the percentage of older TD participants who were able to successfully employ either an allocentric or mixed navigation strategy on enforced trials (8-year-olds = 45.1%, and 10-year-olds = 59.4%).

Performance in each group was compared to the level that would be expected due to chance (16.67%). Results of chi-squared tests showed no significant difference from chance among 5-year-olds, 6-year-olds, or participants with WS, ($p > .05$ for all). In contrast, the correct layout was chosen significantly more often than chance in 8-year-olds $\chi^2(1) = 5.106, p = .024$, and 10-year-olds,

$\chi^2(1) = 7.535, p = .006$. Figure 4 displays the proportion of each maze layout option, including the correct choice (layout 4) chosen by participants in each group.

Independent samples t -tests were used to examine whether children in the oldest two TD groups who chose the correct layout had higher allocentric scores than those who were incorrect. The results found that TD 10-year-olds who chose the correct layout had significantly higher percentage enforced allocentric scores (correct: $M = 66.67 [SD = 30.17]$, incorrect: $M = 29.17 [SD = 17.28]$), than those who were incorrect, $t(13.99) = 3.160, p = .007$. No significant differences were found for spontaneous allocentric scores, or for either allocentric score in TD 8-year-olds ($p > .05$ for all).

Relationships between allocentric score, age and cognitive abilities

Given the wide age range in the WS group, it may have been that older participants with WS were more likely to use an allocentric strategy than younger participants with WS, due to greater life experience in navigating, and thus increased opportunities to develop a more efficient navigation strategy. Therefore, the relationships between age and allocentric scores were calculated for each group separately.

Results showed that, although age was significantly correlated with both spontaneous and enforced allocentric score in the TD group when collapsed across age groups (spontaneous, $r(64) = .288, p = .021$; enforced, $r(64) = .390, p = .001$), no significant relationships were found for any TD group separately ($p > .05$ for all), nor between age and allocentric scores in the WS group ($p > .05$ for both).

The relationship between performance on cognitive measures and ability to employ an allocentric strategy during navigation was also examined. Correlations were calculated for each group separately between BPVS-III and RCPM raw scores and both spontaneous and enforced allocentric scores. Results showed that for 6-year-olds, percentage enforced allocentric score was significantly positively related to BPVS-III raw score, $r(15) = .685, p = .005$. Conversely, for 10-year-olds, the percentage enforced allocentric score was significantly positively related to RCPM raw score, $r(16) = .514, p = .042$. No other significant correlations were found in 5-year-olds, 8-year-olds and WS groups ($p > .05$ for all).

Discussion

The aim of this study was to examine navigation strategies employed in a large-scale virtual environment

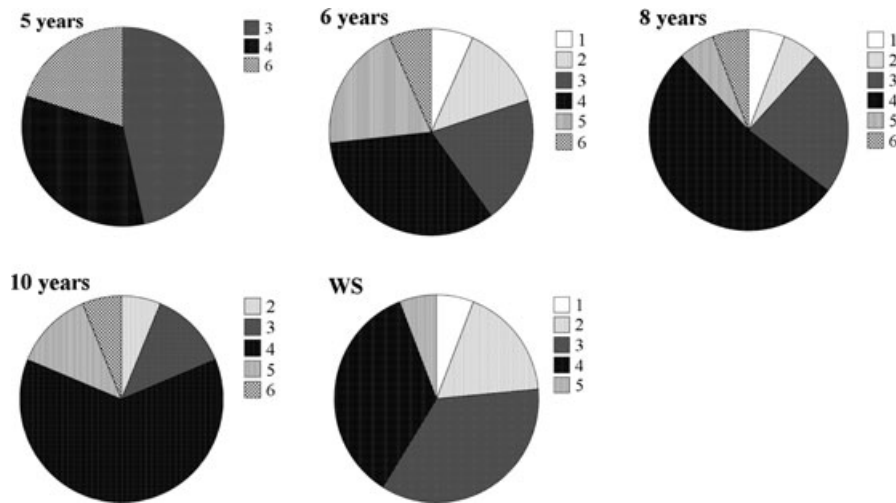


Figure 4 Proportions of each maze layout option chosen by participants in each group.

by typically developing children aged 5–10 years and individuals with Williams syndrome. The study examined the strategies that were employed spontaneously during navigation and also the ability to use an allocentric strategy on trials that required an understanding of interrelationships between features within the environment.

The types of strategies used to navigate through the cross-maze were in line with those identified in previous studies of similar design (Bullens *et al.*, 2010; Iglói *et al.*, 2009). In particular, typically developing children between 5 and 10 years predominantly relied on the use of a sequential egocentric strategy – retracing the same sequence of body turns – to navigate through a route. The results also identified some spontaneous use of allocentric strategies, particularly in the older TD children, and an increase with age in the spontaneous use of a mirrored strategy, indicative of an increasing sophistication in the use of different spatial frames of reference with development. This, in turn, suggests that the use of a mirrored strategy was a reflection of partial allocentric knowledge, rather than a backwards retracing of the route from the end to the start. Backwards retracing would have reflected a misunderstanding of the task instructions and thus would be more likely seen in the youngest TD group.

These findings also reflect performance seen in typical adults (Iglói *et al.*, 2009), suggesting that from at least 5 years of age a predominant reliance on the use of an egocentric strategy does not go through a significant developmental change. The results did, however, highlight a developmental increase in the ability to employ an allocentric spatial strategy when necessary, suggesting an age-related increase in successful and

efficient navigation. During enforced allocentric trials where participants were required to encode the spatial relationships within the environment, independent of previously experienced viewpoints, only 8- and 10-year-old TD children showed a reliable use of this spatial ability. Conversely, Bullens *et al.* (2010) showed above chance performance on allocentric spatial tasks in TD children as young as 5 and 6 years. Although the current cross-maze was designed as a less complex environment than that used in previous studies, including fewer decision points, our results may be a reflection of more stringent scoring criteria, with trials counted as incorrect when a participant made incorrect initial turns or returned later to a correct path following indirect exploration. This is reflected in the high number of incorrect ('non-specific') trials performed across groups. However, the more stringent scoring criteria used in the current study are likely to more accurately bring to light the ability of each participant to use an allocentric strategy from the starting point of a trial, without error; thus, producing a clearer summary of performance.

The findings from the layout choice test also show that only the oldest TD participants were able to accurately select the correct layout more often than chance, indicative of having developed an allocentric spatial representation of the environment. Indeed, this was reflected in higher enforced allocentric scores in TD 10-year-olds who correctly chose the layout compared to those who made an incorrect selection. The second highest choice across all groups was the alternative square layout with four paths, suggesting that participants in each group had developed some understanding of the basic shape of the environment. However, given that this was the most commonly selected layout by

individuals in the youngest TD and WS groups, the conceptualization of traversing and making turns around a square-shaped route may have been the most prominent and important feature for these participants, rather than the correct global spatial representation that would have taken into account all aspects of the environmental layout.

In addition to replicating previous findings in TD children, these results provide insight into the nature of large-scale spatial navigation in WS. Profound impairments on visuospatial tasks relative to verbal abilities are reported consistently in WS (e.g. Bellugi *et al.*, 2000), including difficulties with large-scale spatial tasks and wayfinding (e.g. Smith, Gilchrist, Hood, Tassabehji & Karmiloff-Smith, 2009).

Our results demonstrate that individuals with WS were able to successfully learn a route through the environment after a similar number of trials as 5–8-year-old TD children. This is in line with previous route-learning studies, in which individuals with WS were able to learn a route sequence and recall this successfully after a few attempts, albeit at a slower rate than TD children (e.g. Farran *et al.*, 2010; Farran *et al.*, 2012). Therefore, although route-learning may be impaired relative to chronological age in WS, the performance of individuals with this disorder may be in line with TD children of similar non-verbal ability.

In contrast to route-learning ability in WS, trials examining the type of spontaneous strategies used during wayfinding highlighted an atypical pattern of performance in this group. Unlike TD children, for whom a sequential egocentric strategy was predominant, individuals with WS instead primarily demonstrated the use of a mixed strategy. On such trials, participants began by using a body-based sequential egocentric strategy to navigate, but then switched to using environmental landmarks as a guide to the correct path. Use of this strategy could therefore be a reflection of a change from an egocentric to an allocentric strategy within one trial, following the late realization of a change in starting position. Thus, participants were rewarded for partial use of an allocentric strategy when this occurred. This therefore elucidates the high allocentric score on spontaneous trials in the WS group relative to TD groups. Two possible explanations could be put forward for this finding in WS. First, it may have been that participants with WS were more able than TD children to spontaneously switch to using an allocentric strategy to find the correct path, or second, individuals with WS would start by relying on the use of a sequential egocentric strategy, but then following a difficulty in determining subsequent turns, would switch to using a view-matching strategy, relying on distant landmarks to guide navigation. If the

first turn made by participants in the WS group was due to random choice, there would likely be an equal number of 'mixed' and 'allocentric' strategies reported during spontaneous strategy trials. However, the higher proportion of 'mixed' strategies in this group is indicative of the ability to remember only the initial egocentric turn. It should be noted, however, that the use of a 'mixed' strategy was not used consistently across the four strategy trials in this group. Therefore, although this may have been the predominant strategy used in the WS group, inconsistency by participants with WS on this task suggests a more unsystematic and extemporaneous approach to navigation.

Poor performance in WS on 'enforced allocentric' trials that required the use of an allocentric navigation strategy for success also suggest that the use of a mixed strategy on spontaneous tasks was unlikely to be a reflection of allocentric spatial knowledge in this group. Indeed, participants with WS showed a significant decrease in allocentric score from spontaneous to enforced trials. This indicates that although landmarks are significantly utilized in guiding the retracing of a route in WS (likely through the use of a view-matching strategy), developing a representation of the interrelationships between landmarks and the location of the exit following movement of the self may be particularly problematic.

This use of a mixed strategy and possible switch to a reliance on view-matching was less evident, however, on enforced trials. This may have been due to difficulties in switching from an egocentric to a view-matching strategy when beginning from multiple starting places on these trials. Alternatively, the second route may not have been consolidated as well as the first, and therefore have been under competition from the previously learnt route. However, this is unlikely as participants in all groups successfully learnt the second route before continuing onto the enforced allocentric trials. While it was not an aim of the current study, it would be interesting to determine the extent to which individuals with WS perform successfully during an enforced egocentric task. The use of a task where external landmarks are removed may be a useful tool for examining this further.

Allocentric coding difficulties in WS are in line with previous results from both small (e.g. Bernardino *et al.*, 2013; Nardini *et al.*, 2008) and large-scale (Farran *et al.*, 2010) tasks, and findings of atypical search strategies and difficulties developing a cognitive spatial map in real-world tasks (Foti *et al.*, 2011). On a small-scale task requiring an understanding of the intrinsic spatial relationships between landmarks within an array, Nardini *et al.* (2008) showed that even adults with WS had difficulties in applying this strategy, despite it being seen

to develop in TD children as young as 5 years of age. Similarly, impairments in the use of egocentric and allocentric frames of reference in WS were identified using 3D spatial judgment tasks (Bernardino *et al.*, 2013).

Given the multiple opportunities to navigate the VE in this study, allocentric coding difficulties in WS are unlikely to be due to reduced navigation experience, as seen in typical development (e.g. Anooshian & Young, 1981; Siegel & White, 1975). As such, it is unlikely that poor performance on large-scale tasks requiring relational knowledge in previous studies (Farran *et al.*, 2010) would have been alleviated with continued environmental experience.

Findings from spontaneous trials in particular suggest a greater unprompted reliance on landmarks in WS than in TD children, who showed a predominant use of a strategy that did not take into account the presence of landmarks. This reliance on view-matching during wayfinding in WS may be related to underlying difficulties in spatial working memory, previously noted in WS (Mandolesi *et al.*, 2009; Vicari, Bellucci & Carlesimo, 2003, 2006). The use of an egocentric strategy relies on the ability to continuously update the location of the self in reference to the starting position both temporally and spatially, and is hence supported by spatial working memory. Research examining the use of landmarks during navigation in WS, alongside performance on perceptual view-matching tasks, may therefore further explicate the spontaneous strategies relied on in WS. Poor performance on perceptual-matching tasks would demonstrate difficulties in recalling and visually matching patterns, and would thus rule out view-matching as a possible wayfinding strategy; however, findings suggest that perceptual matching may be relatively unimpaired in WS (Hoffman *et al.*, 2003). Indeed, Vicari *et al.* (2006) found that performance in WS was in line with mental-age matched controls on a perceptual object matching task, but not on a perceptual spatial orientation task, suggesting that different perceptual abilities may be differentially affected in WS, particularly when a spatial element is included in a task.

Results from spontaneous and enforced trials in this study imply that processing of both allocentric and egocentric spatial codes may be atypical in WS. Even though route-learning ability was in line with TD children of similar non-verbal ability, the differential use of strategies during navigation compared to the same group of TD children suggests that this is not simply a reflection of developmental delay in WS, but of a differential developmental trajectory. It is likely, therefore, that individuals with WS use alternative

compensatory strategies to navigate, often resulting in inefficient search techniques.

Difficulties in the use of egocentric navigation are in line with a particular vulnerability of the parietal portion of the dorsal stream known to be associated with WS (e.g. Atkinson & Braddick, 2011; Atkinson *et al.*, 1997; Meyer-Lindenberg, Kohn, Mervis, Kippenhan, Olsen, Morris & Berman, 2004). In typical development, the dorsal visual stream is specialized for the guidance of action (Milner & Goodale, 1995), and the atypical development of the dorsal stream in WS is thought to be an enduring feature across development, persisting into adulthood (Atkinson, Braddick, Rose, Searcy, Wattam-Bell & Bellugi, 2006). Activation of the left hippocampus also plays a role in the use of a sequential egocentric representation during navigation (Iglói *et al.*, 2010). Vulnerability of the hippocampus in WS (Meyer-Lindenberg *et al.*, 2005) is therefore likely to contribute to impairments in episodic memory, namely, difficulties in retracing the complete sequence of turns through an environment identified in this group.

Turning to allocentric coding and the associated role of the hippocampus (e.g. Iaria *et al.*, 2007; Iglói *et al.*, 2010; King *et al.*, 2002), impairments in global spatial representations are also in line with known atypical function and metabolism of this region in WS (Meyer-Lindenberg *et al.*, 2005). This said, the ability to update the location of the self following movement within an allocentric frame of reference is thought to rely on successful translation between egocentric and allocentric spatial codes supported by the parietal and retrosplenial regions (including the hippocampus), respectively (Burgess, 2008; Lambrey *et al.*, 2012; Vann *et al.*, 2009). Therefore, attributing the difficulties in egocentric and allocentric spatial coding in WS to deficits in individual brain regions may be somewhat simplistic, and a more detailed understanding of the relationships between behaviour and atypical functioning in these brain areas in WS is required. Further studies involving functional brain imaging during large-scale navigation tasks in WS may help to elucidate the specific cortical mechanisms associated with the performance and types of navigation strategies employed in this group.

Difficulties in the coding of both egocentric and allocentric spatial reference frames indicate that it may not only be the more complex spatial tasks that pose a challenge for individuals with WS. It is unclear, however, whether the use of view-matching and reliance on landmarks during navigation in WS is the result of a spontaneous atypical strategy, or whether this has been taught as a compensatory strategy to some individuals. Further examination of this and the development of

navigation abilities with age and experience in WS may provide insight into possible interventions to enhance the use of more efficient navigation strategies. This said, no relationship was found between age or cognitive ability and allocentric score in WS in this study, and so it may be unlikely that experience and age are influencing factors in increased navigational ability in this group. However, further research into the reliance on landmarks in WS compared to TD children during route learning may provide insight into whether this is an important strategy for individuals with WS.

In summary, during spontaneous navigation through a newly learnt VE, TD children between 5 and 10 years of age predominantly relied on the use of a sequential egocentric strategy. Individuals with WS, however, predominantly relied on an atypical navigation strategy, which likely involved an early switch from the use of an egocentric strategy to a reliance on landmarks as indicators to the correct pathway. On trials that required an understanding of the spatial relationships between landmarks in the environment, however, only TD children aged 8–10 years demonstrated the ability to use an allocentric strategy and develop a global mental representation of the environmental layout. These results are indicative of deficits both in allocentric and egocentric spatial coding in WS, even following extensive experience in an environment, resulting in the use of more time-consuming and less efficient strategies for wayfinding in large-scale environments.

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