OXFORD
UNIVERSITY PRESS

Archives
of
CLINICAL
NEUROPSYCHOLOGY

How Flexible is the Use of Egocentric Versus Allocentric Frame of Reference in the Williams Syndrome Population?

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Editorial Decision 8 September 2017; Accepted 13 September 2017

Abstract

Objective: This study examined the spontaneous use of allocentric and egocentric frames of reference and their flexible use as a function of instructions.
Method: The computerized spatial reference task created by Heiz and Barisnikov (2015) was used. Participants had to choose a frame of reference according to three types of instructions: spontaneous, allocentric and egocentric. The performances of 16 Williams Syndrome participants between 10 and 41 years were compared to those of two control groups (chronological age and non-verbal intellectual ability).
Results: The majority of Williams Syndrome participants did not show a preference for a particular frame of reference. When explicitly inviting participants to use an allocentric frame of reference, all three groups showed an increased use of the allocentric frame of reference. At the same time, an important heterogeneity of type of frame of reference used by Williams Syndrome participants was observed.
Conclusion: Results demonstrate that despite difficulties in the spontaneous use of allocentric and egocentric frames of reference, some Williams Syndrome participants show flexibility in the use of an allocentric frame of reference when an explicit instruction is provided.

Keywords: Williams Syndrome; Frames of reference; Allocentric; Egocentric; Flexibility

Introduction

Williams Syndrome (WS) is a neurodevelopmental disorder caused by a microdeletion on chromosome 7q11.23 (Mervis, 2003). General intellectual functioning ranges from mild to moderate intellectual disability (IQ < 70) (Mervis & John, 2010). Cognitive functioning is marked by an important dissociation between relatively well-developed language abilities (Bellugi, Wang, & Jernigan, 1994; Paterson, Girelli, Butterworth, & Karmiloff-Smith, 2006) and severe deficits in visuo-spatial abilities (e.g., Bellugi, Lichtenberger, Jones, Lai, & St George, 2000; Bertrand, Mervis, & Isenberg, 1997; Mervis & John, 2010; Mervis, 2003), except for visuo-perceptual aspects such as face and object recognition (e.g., Bellugi et al., 2000; Bellugi, Sabo, & Vaid, 1988; Bertrand et al., 1997). Deficits in visuo-spatial abilities are considered to have a negative affect on academic achievement (Mervis & John, 2010). Difficulties in the acquisition of written language (Howlin, Elison, Udwin, & Stinton, 2010; Mervis, 2009; Volterra, Capirci, & Cristina, 2001), geometry and numerical processing (i.e., numerical abilities; Paterson et al., 2006) have been linked to visuo-spatial deficits observed in WS individuals. Furthermore, difficulties in activities of everyday-living, including practical and occupational skills, are also systematically reported from WS' families and care givers (Fisher, Lense, & Dykens, 2016; Mervis & Klein-Tasman, 2000). Compared to some specific genetic etiologies with intellectual disabilities, such as Prader-Willi and Down Syndrome, individuals with WS have been reported to be less likely to engage in any visuo-spatial leisure activities, such as puzzles, drawing, mazes tasks (Rosner, Hodapp, Fidler, Sagun, & Dykens, 2004).

doi:10.1093/arclin/acx097

Several studies using the developmental Visual Motor Integration test (VMI, Beery & Beery, 2004) have highlighted visuo-constructive deficits in the WS population when copying geometric figures (Bertrand et al., 1997; Dykens, Rosner, & Ly, 2001). These results have been confirmed by studies using block design tasks and block reconstruction procedures (e.g., Bertrand et al., 1997; Farran, Jarrold, & Gathercole, 2001; Semel & Rosner, 2003). Some studies have also documented deficits in navigation abilities, such as way-finding, learning a route (Farran, Blades, Boucher, & Tranter, 2010), or following a road according to a map (Semel & Rosner, 2003). In addition, difficulties in understanding projective terms (spatial prepositions) used to encode spatial relationships between elements or objects have been reported (e.g., Bellugi et al., 2000; Hayward & Tarr, 1995). As a result, WS individuals have difficulties processing spatial representations expressed through language, and more specifically through spatial vocabulary (e.g., behind, in front of, etc.). Similarly, individuals with WS have been shown to face important difficulties in the above-mentioned tasks requiring the encoding of spatial relationships between several objects and showed difficulties using an external object as a point of reference (Farran & Jarrold, 2003; Nardini, Atkinson, Braddick, & Burgess, 2008). The presence of visuo-spatial deficits in individuals with intellectual disabilities of various origins has been extensively reported (Simon, 2007; Vicario, Yates, & Nicholls, 2013), but little is known about the way frames of reference (FOR) are represented and used. This is an important shortcoming in literature because accurate FOR processing is a necessary component of many visuo-spatial tasks and learning abilities (Heiz & Barisnikov, 2015; Kim & Cameron, 2016; Zhou, McBride-Chang, & Wong, 2014).

More specifically, the ability to choose a suitable point of reference is important for the definition of spatial relations between objects and oneself in order to represent the environment. The majority of behavioral and neurocognitive studies indicate that egocentric and allocentric FOR are mainly used to determine the location of an object (see Galati et al., 2000; Howard & Templeton, 1966; Levinson, 1996; Lourenco & Frick, 2013; Newcombe, Huttenlocher, Drummey, & Wiley, 1998). The egocentric FOR concerns the location of an object in comparison to one’s own bodily position, for example “the apple is in front of me” (Galati et al., 2000). Developmental studies show that the egocentric FOR used by typically developing (TD) children, develops early (Taylor & Tversky, 1996) and is used automatically and naturally (Shelton & Mcnamara, 1997). The allocentric FOR uses the location of an external object in relation to its different constituent parts and to other objects in the environment (Klatzky, 1998), for example “the apple is on the table”. In contrast to the egocentric FOR, the allocentric FOR develops later in childhood (Huttenlocher, Newcombe, & Sandberg, 1994; Nardini, Burgess, Breckenridge, & Atkinson, 2006; Piaget & Inhelder, 1947; Rochat, 1995).

In their recent study, Heiz and Barisnikov (2015) assessed developmental trends in the use of egocentric and allocentric FOR. They used an adapted spatial reference task “Where is the donut?” (Taylor & Rapp, 2004) to assess children’s spontaneous use of egocentric and allocentric FOR, as well as their capacity to flexibly switch from one type of FOR to another. In the spontaneous condition, most of the children aged 7 and above used an allocentric FOR. In the allocentric condition, children aged 6 or less gave significantly more allocentric responses. In the egocentric condition, all participants gave more egocentric responses, independently of their age group. This study showed that simple instructions enable children to use allocentric and egocentric FORs earlier, more flexibly and more effectively than in spontaneous task conditions. Taylor and Rapp (2004) assessed adults with their spatial reference task “Where is the Donut?” and reported that adults with a typical development preferentially use an allocentric FOR if no specific instructions have been given and can flexibly use an egocentric FOR if they are instructed to. However, in contrast to literature on TD populations, we currently have very little knowledge about the way children and adults with WS code their spatial environment and which point of reference they preferentially use to define object locations.

Very few studies have assessed the ability to use egocentric and allocentric FOR in WS individuals despite the large number of studies on visuo-spatial skills, such as drawing (Bellugi et al., 1988; Bertrand et al., 1997; Dykens et al., 2001) and block design (Bihrle, Bellugi, Delis, & Marks, 1989; Farran et al., 2001), which more or less strongly rely on the use of an accurate FOR.

Nardini and colleagues (2008) used a spatial search task to determine whether spatial deficits in WS individuals (aged 5–42 years old) are restricted to specific FOR. These authors observed important difficulties especially in situations involving allocentric FOR. These results contrast those from a study by Bernardino, Mouga, Castelo-Branco and van Asselen (2013) who used three spatial judgment tasks that showed difficulties in situations involving egocentric or allocentric FOR. Similar results were observed by Broadbent, Farran and Tolmie (2014) using a virtual cross-maze task to assess navigational strategies employed by individuals with WS (mean age 21 years old). During spontaneous navigation, in contrast to TD controls, WS participants did not predominantly employ a sequential egocentric strategy (recalling the sequence of left-right body turns throughout the task) and exhibited deficits in allocentric spatial coding. However, the task used in this study required spatial memory and planning abilities that are weak in individuals with WS (Mandolesi et al., 2009). Discrepancies between different studies that have explored the use of different FOR in WS individuals could be due to methodological differences. Experimental designs vary in factors known to influence the choice of a particular FOR to perform a task (Coie, Costanzo & Farnill, 1973, cited by Verjat, 1994), such as the number of objects, the nature of spatial dimensions (e.g., above/below; left/right), and the response modalities (e.g., oral or manual). Furthermore, the majority of these tasks required complex abilities (e.g., spatial memory, mental imagery), known to be deficient in the WS population, which could have influenced these results. It is important to note that in contrast to findings from developmental literature, only a few studies have specifically and directly assessed the use of different FOR in the WS population. Therefore, it remains unclear whether individuals with WS are able to use egocentric and allocentric FOR and which of these FOR is used preferentially. Finally, it remains unknown how flexible the use of these FOR in individuals with WS is and whether it can be adapted to task requirements and instructions.

In the present study, we used the spatial reference task developed by Heiz and Barisnikov (2015) with a simple 2-object scenario allowing us to study the use of FOR in three conditions (spontaneous, allocentric, and egocentric) without a memory and verbal response demand. The first aim of the present study was to assess the spontaneous use of an egocentric or allocentric FOR in the WS population. The second aim was to examine the flexibility of the use of these FOR in the presence of verbal cues, favouring one of the two types of FOR. With the spatial reference task (Heiz & Barisnikov, 2015), we specifically explored the spontaneous and cued use of different FOR in children and adults with WS and two control groups of TD children and adults.

Materials and Methods

Participants

Sixteen participants with WS (seven male) aged between 10 and 41 years (mean age = 21.6, *SD* = 9.5, range: 10.1–41.8 years) took part in this study. All participants with WS had been diagnosed through genetic analysis (fluorescence in situ hybridization—FISH test). They were recruited via the *Association Suisse du Syndrome de Williams-Beuren*, Switzerland, and the *Association France Rhône-Alpes*, France.

The first control group TD_CA was comprised of 16 typically developing individuals, of which seven were male (mean chronological age = 22.5, *SD* = 9.6, range: 10.2–41.2 years; mean RCPM score = 29.94 ± 3.79) matched with the WS participants by chronological age (CA) ($t_{(30)} = 0.06$, $p = .995$). The second control group TD_MA was comprised of 16 TD children, of which six were male (mean chronological age = 6.6, *SD* = 2.3, range: 4.3–10.1 years) matched with the WS participants for non-verbal intellectual ability, as measured by the Raven Colored Progressive Matrices (RCPM; Raven, Raven, Court, & Raven, 2003). All control children had a percentile score over the 50th percentile. The TD_MA (mean RCPM score = 17.63 ± 7.89) and the WS groups (mean RCPM score = 16.88 ± 8.37) did not differ on raw scores on the RCPM ($t_{(30)} = -0.334$, $p = .741$) but as expected, there was a significant difference for chronological age ($t_{(30)} = 6.36$, $p < .001$).

All TD children were recruited from regular primary schools in the city of Geneva, Switzerland. All adult participants were recruited from the social circles of researchers and through advertisements. Finally, all participants were native French speakers.

The ethics committee of the Department of Psychology at the University of Geneva and the Cantonal Authorities for Primary Education of Geneva approved the study. Informed consent was obtained from the parents of child participants and from the children themselves. Direct written informed consent was obtained from the adult participants.

Materials

The material consisted of a computerized spatial reference task (Heiz & Barisnikov, 2015). It included 108 trials, each one consisting of a stimulus-picture and four response-pictures appearing simultaneously.

The stimulus-picture illustrated two objects: a reference object and a “to-be-located object” whose location had to be determined. The reference object, with intrinsic sides, was one of eight everyday objects: a chair, a bicycle, a frying pan, a pitcher, a violin, a toy, a bottle, and a hairbrush. This object was presented in three different possible orientations: left, right, or back (The front position was not used, because when the object of the stimulus-picture is oriented in the front position, egocentric and the allocentric responses are the same.) (see Fig. 1 for an example).

The located object, without intrinsic sides, was a soccer ball; this object looks the same from different representations (2D or 3D) and could be placed in one of two locations, on the left or on the right side of the reference object. This resulted in a total of six different configurations (3 orientations × 2 locations), with each configuration being tested six times.

Fig. 1. Example of the three different orientations of the reference object (left, right, and back) (Heiz & Barisnikov, 2015).

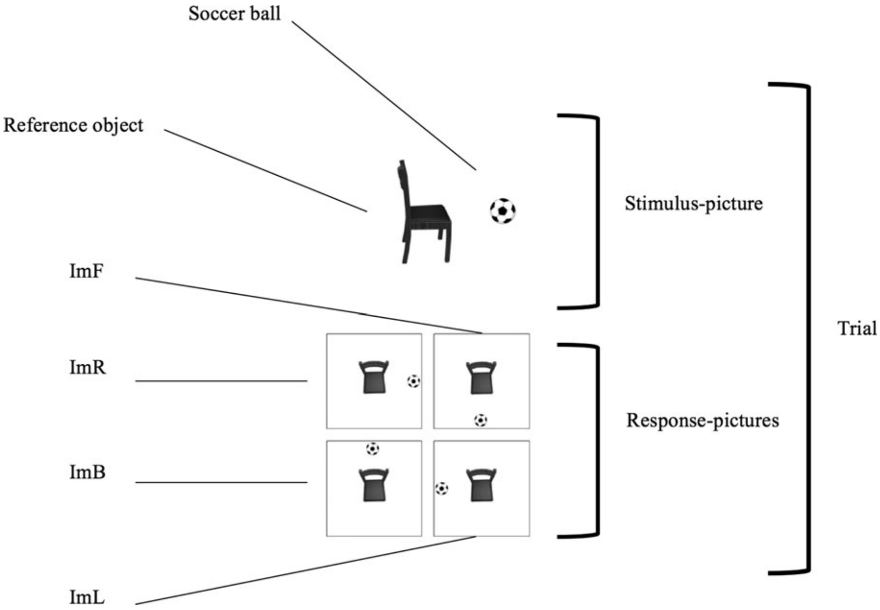

Fig. 2. Example of a trial: in this example, ImR is the egocentric response-picture and the ImF is the allocentric response-picture. ImB and ImL are incorrect responses. All elements were displayed at the same time (Heiz & Barisnikov, 2015).

In the response-pictures, the reference object was always presented in the same orientation and seen from above. The soccer ball could be placed in four different locations, either on the left (ImL), right (ImR), front (ImF), or back (ImB) of the object. A large fixation cross separated each trial. Fig. 2 shows an example of a trial.

Procedure

The task was presented on a Windows touch screen computer (19-in. color monitor) running Eprime2.0® Software. Participants were informed that they would be presented with the stimulus-picture, two objects (the reference object and the to-be-located soccer ball), and four response-pictures. They were then instructed to select the correct response-picture as fast as possible according to the instructions, by touching the selected response-picture on the screen.

The task was presented three times to each participant in three different conditions: spontaneous, egocentric, or allocentric. Each task began with a training phase consisting of five trials intended to verify that the task instructions were understood.

All participants began with the spontaneous condition, then, half continued with the allocentric condition followed by the egocentric condition. The order of the condition was reversed for the other half of the participants. When starting the task, participants were randomly allocated to one of the two conditions. There was a pause of at least 20 min between the different conditions in order to avoid the effects of fatigue. The instructions given for the three conditions were the following:

- Spontaneous condition: "Select among the four small pictures the one that matches the big one, the one that shows the soccer ball in the right place". Participants' responses were classified depending on the type of picture they selected: allocentric, egocentric or incorrect for the other two picture types.
- Egocentric condition: "Select among the four small pictures the one that matches the big one, the one that shows the soccer ball in the right place in relation to yourself". Response scoring was the same as for the spontaneous condition.
- Allocentric condition: "Select among the four small pictures the one that matches the big one, the one that shows the soccer ball in the right place in relation to the object". Response scoring was the same as for the previous two conditions.

All the instructions were given in French.

Statistical analyses

Firstly, a Chi-Square goodness-of-fit test ($p < .005$) was performed on individual responses to classify each participant according to their type of response in each of the three experimental conditions. Participants were classified as having an (1) *egocentric response* (highest egocentric responses); (2) *allocentric response* (highest allocentric responses); or (3) *incorrect response* (highest incorrect responses). These classifications were used to perform a contingency table analysis (Groups × FOR) within each experimental condition.

Subsequently, the percentages of allocentric, egocentric and incorrect (not egocentric and not allocentric) responses were calculated for each instruction and each participant.

We aimed to determine the extent to which participants were able to change their use of FOR as a function of the task condition. A 3 (group) × 2 (condition: spontaneous vs. allocentric or spontaneous vs. egocentric) mixed ANOVAs were conducted on the percentages of allocentric responses in the spontaneous and allocentric conditions, and on the percentages of egocentric responses in the spontaneous and egocentric conditions, respectively. Additional post hoc analyses were performed in order to determine the source of any significant effects, with a Bonferroni adjustment applied to p values where necessary.

Finally, in order to examine the possible heterogeneity of results within the WS population, a cluster analysis was run. Firstly, the analysis was run on the basis of individual response patterns on the percentage of allocentric responses given by participants in the spontaneous and allocentric instructions condition. Secondly, it was run on the percentage of egocentric responses given by participants in the spontaneous and egocentric instructions condition.

All analyses were run on SPSS.

Results

Group Comparisons

Spontaneous instructions. In the spontaneous instructions condition, a contingency table analysis (table 5 × 3) showed that most WS individuals (9/16 participants) gave an incorrect response (see Table 1), five WS individuals preferentially used the *allocentric FOR* and only two WS individuals preferentially used the *egocentric FOR*. For the TD_CA group, the contingency table showed that all participants preferentially used the *allocentric FOR* (16/16). For the TD_MA group, participants either preferentially used the *allocentric FOR* (7/16) or gave incorrect response (6/16), while only three TD_MA individuals preferentially used the *egocentric FOR*.

Table 1. Number of subjects in each group who preferentially used the allocentric vs. egocentric vs. no preference FOR in the spontaneous instructions condition

Group	FOR		
	Allocentric	Egocentric	Incorrect
WS	5	2	9
TD_CA	16	0	0
TD_MA	7	3	6
Total	28	5	15

A χ^2 test of independence showed a significant association between the two variables “group” and “type of response” ($\chi^2_{(4)}$ = 18.56, p = .001). Cramer’s V was 0.44, showing that nearly 19% of variation in one variable could be explained by variation in the other variable, arguing for a link between the variables “group” and “type of response”.

Allocentric instructions. In the allocentric instructions condition, a contingency table analysis (table 5 × 3) showed that the majority of participants preferentially used the *allocentric FOR*, in all groups (see Table 2).

A χ^2 test of independence revealed a significant association between the two variables “group” and “type of response” ($\chi^2_{(4)}$ = 6.99, p = .041). Cramer’s V was 0.27; showing that nearly 7% of variation in one variable could be explained by variation in the other variable.

A mixed ANOVA for the variables “group” and “type of instruction” (spontaneous vs. allocentric) on the percentage of allocentric responses revealed a significant group effect ($F_{(2,\ 45)}$ = 26.114, MSE = 383.817, p < .001, η^2 = .537) but no effect of the type of instruction ($F_{(1,\ 45)}$ = 0.705, MSE = 767.634, p = .406, η^2 = .015). However, a significant interaction was observed ($F_{(2,\ 45)}$ = 4.714, p = .014, η^2 = .173). Tukey Post-Hoc tests confirmed a difference between the WS group and the TD_CA group. Furthermore, they showed no significant difference between the WS and TD_MA groups. A significant interaction was also observed ($F_{(2,\ 45)}$ = 18.853, p < .001, η^2 = .456). The “type of instruction” had an effect only on the TD_MA group. In this group, Bonferroni-adjusted pairwise post-hoc comparisons revealed significantly better performances ($t_{(15)}$ = −3.091, p = .007) following the allocentric instructions compared to the spontaneous instructions (see Fig. 3).

Egocentric instructions. In the egocentric instructions condition, a contingency table analysis (table 5 × 3) showed that many individuals with WS (7/16) gave incorrect responses, five preferentially used the *egocentric* FOR and four preferentially used the *allocentric* FOR (see Table 3). The two control groups preferentially used the *egocentric* FOR (≥11/16).

A χ^2 test of independence revealed a significant association between the variables “group” and “type of response” ($\chi^2_{(4)}$ = 14.52, p = .006). Cramer’s V was 0.39, with nearly 13% of variation of a variable explained by variation in the other variable.

A mixed ANOVA for the variables “group” and “type of instruction” (spontaneous vs. egocentric) on the percentage of egocentric response revealed a main effect of the instruction’s condition, ($F_{(1,\ 45)}$ = 73.752, MSE = 809.105, p < .001, η^2 = .621).

Table 2. Number of subjects in each group who preferentially used the allocentric vs. egocentric vs. no preference FOR in the allocentric instructions condition

Group	FOR		
	Allocentric	Egocentric	Incorrect
WS	8	3	5
TD_CA	13	3	0
TD_MA	11	1	4
Total	32	7	9

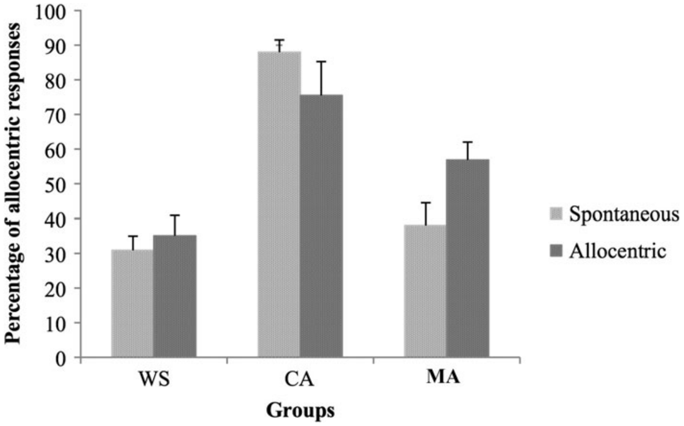

Fig. 3. Percentages and standard errors of allocentric responses in the spontaneous and allocentric instructions conditions for each group.

Table 3. Number of subjects in each group who preferentially used the egocentric vs. allocentric vs. no preference FOR in the egocentric instructions condition

Group	FOR		
	Egocentric	Allocentric	Incorrect
WS	5	4	7
TD_CA	13	3	0
TD_MA	11	4	1
Total	29	11	8

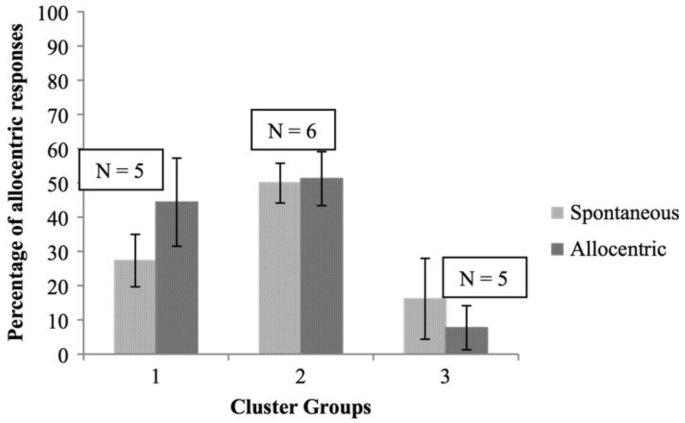

Fig. 4. Percentages of allocentric responses in the spontaneous and allocentric instructions condition for each cluster group.

In general, all participants gave more egocentric responses in the egocentric instructions condition ($M = 60.96\%$, $SD = 17.59$) as compared to the spontaneous instructions condition ($M = 24.43\%$, $SD = 10.82$) (see Fig. 4).

A significant interaction was also observed ($F_{(2,\ 45)} = 18.853$, $p < .001$, $\eta^2 = .456$). Bonferroni-adjusted pairwise post-hoc comparisons revealed an advantage for the egocentric versus spontaneous condition in the TD_CA group ($t_{(15)} = -8.529$, $p < .001$) and TD_MA ($t_{(15)} = -3.447$, $p = .004$), with a particularly large advantage for the TD_CA group (see Fig. 4). A trend towards a significant difference was found for the WS group ($t_{(15)} = -1.909$, $p = .076$).

Cluster Analyses

Allocentric instructions. A hierarchical cluster analysis using Ward's method produced three clusters corresponding to three patterns of responses, as determined by the percentage of allocentric responses given by the participants in the spontaneous and allocentric instructions conditions ($F_{(2,\ 13)} = 5.513$, MSE = 88.48, $p = .018$, $\eta^2 = .459$) (see Fig. 5). The first cluster, with five participants, was characterized by a minimum of 40% of allocentric responses. The second cluster, with six participants, was characterized by a marginally significant increase of allocentric responses in the allocentric instructions condition ($F_{(1,\ 5)} = 6.181$, MSE = 83.411, $p = .055$, $\eta^2 = .553$). The third cluster, with five participants, was characterized by a low percentage of allocentric responses in the spontaneous and allocentric instructions condition.

Egocentric instructions. A second hierarchical cluster analysis using Ward's method was run on the percentage of egocentric responses given by participants in the spontaneous and egocentric instructions conditions. The analysis was significant and yielded three clusters ($F_{(2,\ 13)} = 12.957$, MSE = 175.047, $p = .001$, $\eta^2 = .666$) (see Fig. 6). The first cluster, with three participants, was characterized by a high increase of egocentric responses in the egocentric instructions condition. The second cluster included only one participant and was characterized by a high percentage of egocentric responses in the spontaneous and egocentric instructions conditions. The third cluster, with 12 participants, was characterized by a low percentage of egocentric responses in spontaneous and egocentric instructions conditions.

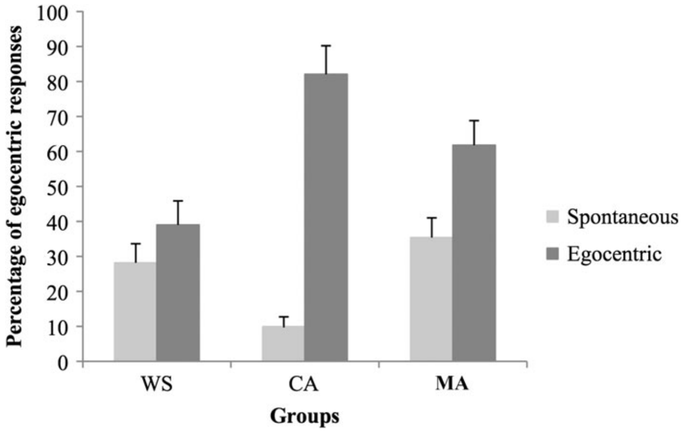

Fig. 5. Percentages and standard errors for egocentric responses in the spontaneous and egocentric instructions conditions.

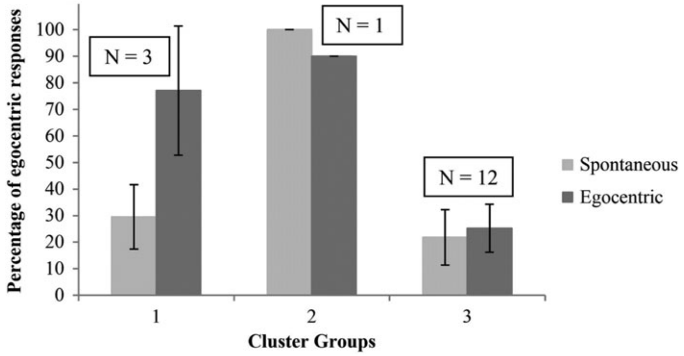

Fig. 6. Percentages of egocentric responses in the spontaneous and egocentric instructions conditions for each cluster group.

Discussion

The present study aimed to assess the use of egocentric and allocentric FOR by means of a spatial reference task, in either spontaneous conditions or in conditions where participants were explicitly instructed to use a specific type of FOR. The results of the WS participants were compared to those of two TD control groups matched individually on chronological age (CA) and on non-verbal abilities (MA).

Results revealed that the majority of the WS participants (9/16) did not show a preference towards a particular FOR and gave a majority of incorrect responses, while five WS individuals spontaneously used an “allocentric” FOR and two used an “egocentric” FOR. In contrast, the majority of participants in both control groups spontaneously used an allocentic FOR. Secondly, results showed that specific instructions helped participants in all three groups to make a more appropriate and consistent choice of FOR. Allocentric instructions seemed to help some WS participants to make use of an allocentric FOR. At the same time, an important variability in performances was observed in all three conditions in the WS group.

In the spontaneous instructions condition, the majority of WS participants gave incorrect responses showing difficulties using an egocentric or allocentric FOR. Our results are in line with Bernardino and colleagues’ (2013) results reporting that individuals with WS have deficits using both FOR. Similarly, Broadbent and colleagues (2014) observed difficulties in WS individuals using both FOR during spontaneous conditions of a virtual maze task.

In the allocentric instructions condition, some WS participants improved with the use of an allocentric FOR. These results contrast those of previous studies showing persistent difficulties using an allocentric FOR in conditions favouring this FOR. Either because an egocentric FOR was not available (Nardini et al., 2008) or because the task could not be performed with a body-centered FOR (Bernardino et al., 2013; Farran & Jarrold, 2005). In the experimental design of the present study, contrary to previous studies, scenes depicting both FOR were available when performing the task with explicit allocentric instructions. Furthermore, the positive affect of the use of specific instructions to favor the use of allocentric FOR indicates the existence of some degree of flexibility of FOR use in some WS participants, similarly to the TD control group (MA).

This latter group improved performances compared to those in the spontaneous instructions condition where they preferentially used egocentric FOR or gave incorrect responses. These results suggest that children demonstrate flexibility in the use of allocentric FOR at a much younger age than evidenced in the spontaneous instructions condition. This is also in accordance with a developmental study (Heiz & Barisnikov, 2015) showing a significant improvement in performances in an allocentric instructions conditions in TD children (aged 4, 5, and 6) relative to those in spontaneous instructions conditions.

It is to be noted that no significant differences in performances were found between our WS participants and the TD_MA control group, which could be seen as a developmental delay in the WS group. However, such a delay could not be established for all WS subgroups showing a distinct pattern of performances, which is discussed below.

Additionally, the TD_CA group used more allocentric FOR than the WS group did. The results of the TD_CA group, comprised of a majority of adults (M = 22;5 years old), are in accordance with several studies reporting that adults preferentially use an allocentric FOR, also known as an intrinsic FOR (Carlson-Radvansky & Jiang, 1998; Taylor & Rapp, 2004). However, they showed the ability to easily switch to the use of an egocentric FOR depending on the spatial situation (Taylor & Rapp, 2004). It has been demonstrated and suggested that adults can alter between the 2 FOR and invest more cognitive effort to process non-preferred frames of reference (Carlson, West, Taylor, & Herndon, 2002; Taylor & Rapp, 2004). Nevertheless, similarly to previous literature, our results show that the TD_CA group can flexibly shift between an egocentric and an allocentric FOR depending on the instructions provided.

Some of the WS participants were also able to switch between the use of one or another FOR according to instructions. In contrast to literature reporting persistent visuo-spatial deficits in the WS population, our results show that when precise and specific instructions are provided (verbal cues), these individuals can flexibly use both types of FOR. These encouraging results could open the perspective for an intervention strategy benefiting from verbal cues to guide and support the use of specific FOR, namely the allocentric and/or egocentric FOR.

Similarly to the TD_MA control group, the allocentric instructions helped some WS participants to improve their performances. However, an important heterogeneity in the WS participants' performances was observed. These results indicate the need for more individual and qualitative analyses of the affect of specific instructions on WS individual performances. Thus, three subgroups with distinct performance patterns were observed (see Fig. 5): the first subgroup (5/16) showed a similar performance pattern to the TD_MA controls; the second subgroup (six WS) outperformed the first subgroup in the spontaneous condition. Unlike these two WS subgroups, the third subgroup (five WS) showed particular difficulties in using an allocentric FOR in both conditions. These difficulties could be interpreted in light of Farran and colleagues' (2001; 2003) findings suggesting that WS individuals have a major deficit in understanding categorical spatial locations (e.g., above/below) and in coordinating spatial relations relative to different reference points or landmarks. Similarly, Nardini and colleagues (2008) suggested that the highly impaired use of an allocentric FOR ("array-move" condition) in WS individuals could be partly related to impairment in the use of visual landmarks to code locations. However, considering the small number of WS individuals in each subgroup, the existence of distinct patterns of performances needs to be confirmed with a larger number of participants. At the same time, our results show that the use of a cluster analysis can be very informative about the nature of visuo-spatial processing styles in WS individuals. In contrast, WS average group performances proved to be less pertinent for understanding the way WS individual's process spatial information.

In the *egocentric instructions condition*, WS participants, similarly to the control groups, gave more egocentric responses compared to the spontaneous instructions condition. The few studies that proposed an "imposed" egocentric condition showed contrasting results. Nardini and colleagues (2008) reported that WS individuals were able to use body-FOR in both conditions. However, Bernardino and colleagues (2013) reported that difficulties in the spontaneous use of an egocentric FOR remained constant in the "imposed" egocentric condition. Broadbent and colleagues (2014) reported difficulties in the WS group when using an egocentric FOR in the spontaneous instructions condition; however, they did not propose an "imposed" egocentric condition. Discrepancies between these studies could be due to methodological differences. Experimental designs varied with respect to the number of objects used, the type of material and its presentation mode (computerized 2D or 3D model, virtual maze), and the response modalities (oral or manual). Furthermore, the majority of these tasks required complex abilities (e.g., spatial memory, mental imagery), known to be deficient in the WS population, which could have influenced these results.

Nevertheless, the analyses of the performances of our WS individuals revealed three subgroups with distinct patterns of performances. While the improvement in the use of an egocentric FOR in the first subgroup (three participants) was comparable to those of the TD_MA control group, the second subgroup (one participant) showed better performances in both conditions (egocentric and spontaneous), comparable to the TD_CA controls. In contrast, the third subgroup was comprised of a majority of participants (12/16) with difficulties using their own body as a FOR. Those who failed to use the egocentric FOR might have a poor representation of their body schema. A progressive shift from a body-centered to an object-centered FOR seems to take place between the ages of 5 and 7 (Heiz & Barisnikov, 2015; Nardini et al., 2006) or between the ages of 7 and

10 (Bullens, Iglói, Berthoz, Postma, & Rondi-Reig, 2010; Piaget & Inhelder, 1947). Little is known about body-based representations in the WS population. The few studies that assessed these abilities reported poor body schema knowledge in WS children and adults. Dykens and colleagues (2001) reported that WS participants drew poor human figures with a smaller number of portrayed body parts in comparison to the TD_MA control group. A low recognition of and naming of body parts and difficulties in identifying the right and the left parts of own or other bodies has also been reported (Heiz, Cengic, Fernandes, & Barisnikov, 2015). According to a recent study (Saj, Heiz, & Barisnikov, 2017), these body representation difficulties in WS participants may be explained by a significant left deviation of their bodily sagittal plan, which allows an individual to determine a spatial location (e.g., on my left/right). The lack of a well-established representation of left and right directions might, according to Bernardino and colleagues (2013), introduce additional difficulties for WS participants. Broadbent and colleagues (2014, 2015) also showed that unlike TD children, for whom a sequential egocentric strategy is predominant, individuals with WS begin a spatial navigation task by using a body-based strategy (going in the correct direction), but then rapidly switch to an environmental view-matching strategy (non-specific). Although our participants were able to distinguish if an object was located on the left or on the right (two opposite sides) of their body at the beginning of the task, they exhibited difficulties with this distinction throughout the rest of the task. Similarly, our participants were able to distinguish if an object was on one side or the other of their body, on the left or on the right, during the training phase, but exhibited general difficulties with this distinction throughout the rest of the task. Therefore, the lack of a fully developed body representation in the WS population could also lead to difficulties in maintaining a stable representation of spatial location (with spatial terms) and to coordinate different spatial information (between body and objects).

Nevertheless, our results are the first to show that a two-object experimental design with simple instructions helps to improve flexibility in the choice of a specific FOR. These findings are in contrast to literature reporting persistent visuo-spatial deficit in WS individuals. Consequently, our results could contribute significantly to intervention strategies for visuo-spatial processing difficulties in WS individuals. Precise and specific instructions could help individuals with WS to use a viewpoint according to the situation and to subsequently improve their ability to process spatial information. Genetic syndromes marked by similar visuo-spatial deficits (e.g., Turner syndrome, 22q11deletion syndrome) and learning difficulties (Simon, 2007; Vicario et al., 2013) may also benefit from this kind of cueing strategy.

However, these important results do have some limitations. It is to be noted that the comparison between groups showed no significant differences in performances between some of our WS participants and the TD_MA control group, which could be seen as a developmental delay. Although the RCPM is a reliable measure in obtaining a non-verbal functioning score in WS individuals and can be used to match WS individuals to other participant groups (Van Herwegen, Farran, & Annaz, 2011), there is an important difference in chronological age between these groups that limits the interpretation of the present results. Consequently, a distinct pattern of performances exhibited by different WS subgroups could not be compared to those of the TD_MA controls.

Although a qualitative analysis of WS performances highlights the existence of different patterns of performances, these results need to be confirmed with a larger number of WS participants. The reason for this being that the statistical power of the different subgroups was too weak to perform further analyses of different patterns of performances observed. Although no affect of age on WS performances was observed, a larger number of WS individuals grouped by age could also provide more accurate information on the developmental trajectory of the use of specific FOR. This could also allow more precise comparisons between children and adult groups when examining the influence of experience (e.g., work place, everyday life activities) on spatial abilities, for example.

In conclusion, results demonstrate that despite difficulties in the spontaneous use of allocentric and egocentric FOR, some WS participants show flexibility in the use of allocentric FOR when an explicit instruction is provided. In contrast, they exhibit particular difficulties in the use of an egocentric FOR, which may be the result of a poor representation of their body schema. This could be the primary cause of their deficit in representing spatial locations (e.g., left/right) and establishing spatial FOR, which are both necessary for spatial task solving (e.g., navigation; drawing). Clinical assessments should consider the existence of contrasted patterns of performances among WS participants, which could also lead to the development of adapted intervention strategies.

Funding

This work was supported by the Swiss National Science Foundation (grant no 10019_166047/1).

Conflict of Interest

None declared.

Acknowledgments

We would like to thank all WS participants and their families. We would also like to thank Ana Da Costa Fernandes and Lejla Cengic our master students and the interns of the Child Clinical Neuropsychology Unit (Department of Psychology, University of Geneva, Switzerland) for their assistance in the data collection.

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