Spatial Exploration and Navigation in Down Syndrome and Williams Syndrome.

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Funding: This research was supported by ESRC Grant RES-062-33-0005 and Agence Nationale de la Recherche (ANR) Grant ANR-09-FRBR-035.

Declarations of interest: none

Author statement: Emily Farran: Conceptualization; Data curation; Formal analysis; Funding acquisition; Methodology; Supervision; Visualization; Writing - original draft.
Mark Blades: Conceptualization, Funding acquisition, Methodology, Writing - review & editing;
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Yannick Courbois: Conceptualization, Funding acquisition, Methodology, Writing - original draft

Acknowledgements: Thank you to the children who took part in our research, and for the parents and teachers who facilitated their participation. Thank you to Hayley White, Louise Ewing and Melanie Facon for help with data collection, and to Katie Gilligan-Lee, John Willoughby, Amelia Bennett, Mafalda Batista Da Costa, Iliana Misheva and Aan Ameen for help with data coding, and to Boris Gambet for programming the virtual environments.

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Abstract

We know little about the ability to explore and navigate large-scale space for people with intellectual disabilities (ID). In this cross-syndrome study, individuals with Down syndrome (DS), individuals with Williams syndrome (WS) and typically developing children (TD; aged 5 to 11 years) explored virtual environments with the goal of learning where everything was within the environment (Experiment 1) or to find six stars (Experiment 2).

There was little difference between the WS and DS groups when the goal was simply to learn about the environment with no specific destination to be reached (Experiment 1); both groups performed at a level akin to a subset of TD children of a similar level of non-verbal ability. The difference became evident when the goal of the task was to locate targets in the environment (Experiment 2). The DS group showed the weakest performance, performing at or below the level of a subset of TD children at a similar level of non-verbal ability, whilst the WS group performed at the level of the TD subset group. The DS, WS and TD group also demonstrated different patterns of exploration behavior. Exploration behaviour in DS was weak and did not improve across trials. In WS, exploration behavior changed across trials but was atypical (the number of revisits increased with repeated trials). Moreover, transdiagnostic individual difference analysis (Latent Profile Analysis) revealed five profiles of exploration and navigation variables, none of which were uniquely specific to DS or to WS. Only the most extreme profile of very poor navigators was specific to participants with DS and WS. Interestingly, all other profiles contained at least one individual with DS and at least one individual with WS. This highlights the

importance of investigating heterogeneity in the performance of individuals with intellectual disability and the usefulness of a data-driven transdiagnostic approach to identifying behavioral profiles.

Keywords: Navigation, Exploration, Spatial Cognition, Williams syndrome, Down syndrome, Intellectual Disability

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1. Introduction

Spatial navigation describes the skills required to know where you are in an environment and the ability to find places and to learn and retrace routes. These are essential skills for effective functioning in everyday life. Navigation can take place in spaces of different scales (Banta Lavenex & Lavenex, 2021), vista space and environmental space. *Vista space* can be viewed from a single location and the location of a target can be determined by a simple visual search (a room or a town square are vista space). In contrast, *environmental space* cannot be experienced from a single location, but requires travelling through interconnected spaces (a building, a neighborhood). Navigating environmental space can be described as a decision-making process, involving high-level cognitive systems and is typically referred to as *wayfinding* (Montello & Sas, 2006). Wayfinding can be separated into several components. In their taxonomy of human wayfinding tasks, Wiener et al. (2009) make the distinction between *aided* (using some form of external representations such as a map or signage) and *unaided* wayfinding. They also make the distinction between *undirected* and *directed* wayfinding. Undirected wayfinding refers to navigation with no spatial goal, for example, exploring an unfamiliar environment with the goal

to simply learn about its structure. Directed wayfinding refers to navigation behavior with a spatial goal such as reaching a landmark or destination.

Navigation impairments are common in people with Down syndrome (DS) and people with Williams syndrome (WS). DS is caused by partial or complete trisomy of chromosome 21 and is the most common genetic form of Intellectual Disability (ID) (Roubertoux & Kerdelhué, 2006). Experiments conducted in *vista space* demonstrated that individuals with DS could use egocentric coding to locate targets (they could encode a target location in relation to their body), but exhibited limited allocentric coding (encoding the relationships between multiple locations). Indeed, they could use basic allocentric coding for low resolution spatial tasks (remembering the location of a target among four locations) but not for high resolution spatial tasks (remembering the location of a target among 12 locations; see Bostelmann et al., 2018). The results are consistent with neuroanatomical evidence showing that there are abnormalities in hippocampal circuits in DS (Banta Lavenex & Lavenex, 2021; Bostelmann et al., 2018).

WS is a rare genetic syndrome that results from a deletion of approximately 28 contiguous genes due to a hemizygous microdeletion of 1.6Mb on chromosome 7q11.23 (Tassabehji, 2003). Banta Lavenex and Lavenex (2021) provided convincing evidence showing that individuals with WS could use egocentric coding to locate targets in vista space, but showed

marked deficits in all allocentric tasks (they were not able to encode the target locations in relation to other objects in the vista environment) (see also Julian et al., 2019). These behavioral findings are consistent with neuroimaging data showing impaired processing in the dorsal visual stream and in the hippocampus in WS (Banta Lavenex & Lavenex, 2021).

Investigation of environmental space in WS and DS has relied mostly on directed wayfinding tasks and has used virtual environments (VE) (but see Farran et al., 2010), i.e., virtual towns or mazes which feature local and distant landmarks. Directed wayfinding tasks are complex, thus implementing high level processes such as Executive Functions (Purser et al., 2012), which are also known to be impaired in WS (Menghini et al., 2010; Miezah et al., 2020) and DS (Tungate & Conners, 2021). Participants are asked to learn routes and to retrace them (Broadbent et al., 2015; Davis et al., 2014; N′Kaoua et al., 2019; Purser et al., 2015). In some studies, participants were also asked to find the shortest route between two places in order to assess configural knowledge (Courbois et al., 2013; Farran et al., 2015; Himmelberger et al., 2020). Results from these experiments showed that most participants with DS were able to learn routes, but they frequently made more errors than TD children of the same mental age (TD MA). Very few of them were able to gain configural knowledge in order to find a novel path to a known destination, with some using a strategy in which they combined known routes to reach the destination (Courbois et al., 2013; Farran et al., 2015; Himmelberger et al., 2020). Participants with WS

had better performance than participants with DS on route learning tasks (Farran et al., 2015; Purser et al., 2015). Farran et al. (2015) demonstrated that slightly more (35%) of their WS group succeeded in finding a shortcut between two known places (a measure of configural knowledge) relative to participants with DS (10%). Thus, despite their deficits in allocentric coding in vista space, individuals with WS display stronger *directed wayfinding* strategies than individuals with DS.

In most of the VE tasks mentioned above, participants were required to learn fixed routes and were not allowed to freely explore the environment (Courbois et al., 2013; Davis et al., 2014; Farran et al., 2012; Farran et al., 2015; Himmelberger et al., 2020; N′Kaoua et al., 2019; Purser et al., 2015). Yet, developmental research has shown that exploration involving self-locomotion through the environment, and children's opportunities to explore their surroundings, facilitate the development of spatial orientation and navigation skills (Campos et al., 2000; Oudgenoeg-Paz et al., 2015; Oudgenoeg-Paz & Mulder, 2021; Pullano & Foti, 2022; van den Brink & Janzen, 2013). Moreover, childhood exploration in large-scale environments may explain individual differences in adult wayfinding strategies. Indeed, self-report studies have shown that reduced exploration opportunities in childhood were associated with the use of a route strategy (using landmarks as references to learn a fixed route) over an orientation strategy in adults (using cardinal direction or geocentric cues) (Schug et al., 2022; Vieites et

al., 2020). Furthermore, there is evidence that exploration behavior in adults is related to navigation success. That is, Gagnon et al. (2018) and Munion et al. (2019) report that a higher rate of diffusion through an environment (the rate at which participants visit unique locations within the environment), pausing less and revisiting previously visited locations less were related to the ability to locate targets in large scale environments. Conversely, a “cautious” exploration behavior with a high number of pauses and a high number of revisits was associated with lower navigation performance (Munion et al., 2019).

In a recent developmental study, Farran et al. (2022) investigated exploration behavior in children aged 5 to 11 years using a desktop VE (the environment was an open space bounded by four walls, including 10 building blocks and 40 landmarks). The first experiment, Experiment 1, was designed to assess exploration behavior in an undirected wayfinding task. Participants explored the VE with the goal of learning where everything was within it (wayfinding without a specific destination). Experiment 2 assessed exploration behavior in a directed wayfinding task. Participants explored a VE with the goal of locating six stars in two experimental conditions; trials were carried out in a standard condition (unaided wayfinding) and an overhead map condition in which participants could view their location in real time on a map (aided wayfinding).

In the undirected wayfinding task, exploration of the environment was related to chronological age, and demonstrated an increasingly active strategy with age. That is, older children explored a wider area of the VE, created longer path lengths and paused less than younger children. This pattern of results suggested that the optimal solution for freely exploring an unfamiliar environment was to cover as wide an area as possible, and to take minimal time to pause. In the directed wayfinding task, navigation success (time per star collected) increased with age. It also increased with repeated trials and was stronger in the overhead map condition. Fewer pauses and visiting more areas of the environment were associated with navigation success for both conditions. Stronger target order consistency across trials (consistency in the order of star collection over consecutive trials, an indicator of route learning) was also related to navigation success. Furthermore, exploration behavior in Experiment 1 did not predict navigation success in Experiment 2, suggesting that undirected and directed wayfinding elicited different exploration strategies. Finally, individual difference analysis (Latent Profile Analysis) of the standard condition revealed three profiles. These reflected cautious explorers who were poor at navigating (low navigation success, low number of visited areas, high number of pauses), active and efficient explorers who were good at navigating (low number of pauses, high number of visited areas, high navigation success) and active and less efficient explorers who were average at navigating.

Taken together, the results from typically developing (TD) children suggest that with increasing age, children move from cautious and less efficient exploration to active exploration of the environment in which they make few pauses and cover more of the VE. Consistent with studies on adults (Gagnon et al., 2018; Munion et al., 2019), an active strategy during goal-directed exploration is associated with navigation success.

Given the everyday importance of being able to navigate, and the evidence above that exploration is strongly associated with navigation success, it is important to study how individuals with neurodevelopmental conditions explore. In the current experiments, we used the same methodology as Farran et al. (2022). We studied exploration with no spatial goal (undirected wayfinding, Experiment 1) and exploration with the spatial goal of locating targets in the environment (directed wayfinding, Experiment 2). The latter included two conditions: a standard condition and an overheard map condition (unaided *versus* aided wayfinding). As in Farran et al. (2022), the exploration variables were the number of areas of the VE that were visited, the length of the path traveled, the number of revisits and the number of pauses. In Experiment 2, we also computed target order consistency (an indicator of route learning) and the time per star collected (an indicator of navigation success).

Based on the evidence above, we predicted:

1. With reference to undirected wayfinding (Experiment 1), we could not make any specific predictions since free exploration with no spatial goal has never been studied in WS or DS. For TD children aged 5 to 11 years, an efficient strategy is exemplified by exploring a wide area and making fewer pauses (Farran et al., 2022). Based on this, we anticipate that free exploration in DS and in WS will be at the same level as a subgroup of TD children of a similar non-verbal mental age. The use of the undirected wayfinding task (Experiment 1), in addition to the directed wayfinding task (Experiment 2) enabled us to determine the generalisability of any difficulties observed. Differences between the WS and DS group will determine whether the known relative wayfinding difficulties in DS are specific to goal-directed tasks or more general, and thus also extend to free exploration. The latter would be exemplified by the DS group showing less efficient exploration compared to the WS group.

2. With reference to directed wayfinding (Experiment 2), based on previous studies of directed wayfinding in route learning and configural knowledge tasks (Farran et al., 2015; Purser et al., 2015), we hypothesized that individuals with DS would have a

lower navigation success score than individuals with WS and a subgroup of TD children of a similar non-verbal mental age.

3. Given the known deficits in navigation in WS and DS discussed above, coupled with the links between navigation efficiency and exploration behavior that have been documented in both adult and developmental research (e.g., Gagnon et al., 2018; Oudgenoeg-Paz et al., 2015; Farran et al., 2022), we predicted that exploration behaviour would be related to navigation success in the WS and DS groups, and that exploration behaviour in WS and DS would reflect a cautious, less efficient strategy (fewer areas visited, shorter path lengths, and more pauses) as observed in young children. We also expected exploration behavior to be less efficient in the DS group than in the WS group.

4. Previous research on directed wayfinding has shown that both individuals with WS or DS could learn new routes in virtual environments (see Farran et al., 2015), we therefore predicted that navigation success would improve across trials in the two neurodevelopmental condition groups, to a similar extent as a subgroup of TD children of a similar level of non-verbal ability. Moreover, we expected exploration behaviour during the first

trial to be related to navigation success in subsequent trials for all groups.

5. Comparison between the standard and overhead map conditions will determine whether an external allocentric representation of the environment facilitates navigation in WS and in DS (aided wayfinding). There is very little research on map reading in these two neurodevelopmental conditions. Toffalini et al. (2018) found that individuals with DS did not benefit from seeing a sketch map before exploring the environment. Meneghetti et al. (2017) found that they benefited when the map was associated with a spatial description, but to a lesser degree than TD children. Farran et al. (2010) found limited evidence of map use in individuals with WS. Taken together, we anticipate that the DS and WS groups will show limited benefit of the overhead map, compared to the TD subgroup.

6. Finally, Latent Profile Analysis was employed to determine homogenous profiles of performance. The distribution of the individuals with DS and WS across different profiles was used to determine whether there were syndrome-specific profiles of behaviour or not. Based on prior research which demonstrated individual differences in navigation within these groups (Farran

et al., 2015), we predicted that profiles would not be syndrome-specific.

2. Experiment 1

In Experiment 1, participants had three minutes to freely explore a virtual environment.

2.1 Method

No part of the study procedures or analysis plans was preregistered prior to the research being conducted. We report how we determined our sample size, all data exclusions, all inclusion/exclusion criteria, whether inclusion/exclusion criteria were established prior to data analysis, all manipulations, and all measures in the study. Due to the rarity of our genetic syndrome groups, we report sensitivity power analyses (see Lakens, 2022). Data inclusion and exclusion criteria were established prior to data analysis and are reported.

2.1.1 Participants

There were three participant groups: 91 TD individuals (46 male, 55 female), 33 individuals with DS (17 male, 18 female), and 24 individuals with WS (7 male, 17 female). Ethical approval for Experiments 1 and 2 was obtained from the University ethics committee. All individuals with DS and individuals with WS had received both phenotypic and genetic diagnosis by their clinician. Participants were recruited via local schools and parent-support groups. Written consent was provided by parents/caregivers and

verbal consent was given by participants. Raven's Coloured Progressive Matrices (RCPM; Raven et al., 1998), a measure of non-verbal ability, and the British Picture Vocabulary Scale III (BPVS; Dunn et al., 2009), a measure of receptive vocabulary, were administered to participants. Legal copyright restrictions prevent public archiving of RCPM and BPVS assessments. These can be obtained from the copyright holders in the cited references. One factor ANOVAs with Group as the between participant factor were carried out on Chronological Age, RCPM raw score, BPVS raw scores and a measure of task-related keyboard and eye coordination, i.e. the time taken to complete the final familiarisation trial ('computer control'). The analyses demonstrated an effect of Chronological Age (F(2, 145)=114.39, p<.001, η_p^2=.61: Tukey comparisons demonstrated that the TD group were younger than the two atypical groups (p<.001 for both), with no difference between the DS and WS groups (p=.07). There was a group difference for RCPM (F(2, 143)=89.52, p<.001, η_p^2=.56); the TD group had higher RCPM scores than the DS and WS groups (p<.001 for both), and the WS group had higher RCPM scores than the DS group (p=.009). There was also a group difference for BPVS scores, F(2. 143)=54.83, p<.001, on account of low BPVS scores in the DS group compared to the WS group and the TD group (p<.001 for both), but similar scores for the WS and TD groups (p=.91). There was also an effect of computer control, F (2, 145)=16.44, p<.001, η_p^2=.19 due to better computer control in the TD group than the DS and WS (p<.05 for both).

There was no difference in computer control between the DS and WS group (p=.268). Participant information (mean, standard deviation and range) is given in Table 1.

Table 1 about here

2.1.2 Design and Procedure (as stated in Farran et al., 2022)

The virtual environment (VE) was created using Virtools 5.0 (Dassault Systems) and was presented on a 17-inch laptop[1]. Participants moved around the VE using the keyboard arrow keys (moving forward [up arrow key], rotating right [right arrow key], rotating left [left arrow key]; horizontal plane).

Familiarization trial: Participants were presented with a short empty corridor VE in which two changes in direction along right-angle turns were required to reach a target (a yellow star). The experimenter demonstrated how to move around the corridor VE using the keyboard controls. Once participants understood, it was their turn. Participants were told to navigate to a star and walk into it, which marked the end of the trial. Participants completed one familiarisation trial, or more if required, to ensure that they could move confidently and accurately in the VE. A second familiarisation trial was required by 82% (27 out of 33) DS participants, 50% (12 out of 24) WS participants and 66% (60 out of 91) TD participants. Each participant's

time taken to complete their final familiarisation trial was used as a measure of computer control.

Experimental trial: The experimental VE was constructed in a 300x300 virtual unit area bounded by four walls. The trial began at the ‘home’ location, which was the lower left corner of the VE and was coloured dark grey. Within the walls there were 10 building blocks (two 75x65units, two 60x45 units, and one each of 60x30 units, 75x75 units, 90x30 units, 30x75 units, 45x45 units, 45x60 units). A building block consisted of blocks of tall linked buildings (Figure 1). A total of 40 proximal landmarks were included; four proximal landmarks surrounded each building block (one on each wall), two on corners and two that were along a wall, but not at a corner. A distant landmark appeared along three of the four perimeter walls in a triangular formation to aid orientation within the VE. Participants were told that they had three minutes to learn as much as they could about where everything was in the VE. A blue bar on a white background was shown at the top of the screen that increased in length as the duration of the trial reduced.

Participant location was recorded as X and Y coordinates in whole units (the VE extended 300 x 300 units) every 200ms. These were used to derive four variables, based on variables used in previous research (Gagnon et al., 2018; Munion et al., 2019). First, the number of areas of the VE that were visited out of a maximum of nine. The nine areas were determined by

dividing the VE into a 3 by 3 grid of 100 unit x 100 unit square areas. Second, the length of the path traveled by the participant in virtual units. Third, the number of times that a participant's path intersected with itself or the participant immediately retraced part of their path (i.e., they made a 180 degree turn and retraced their steps). These two kinds of behaviors were summed together and labeled as revisits. Fourth, the number of times that the participant paused during their 3-minute exploration of the VE. A pause was categorized as having no change in X and Y coordinates for 2 seconds or more (note, that we also measured the absolute length of pauses. Length of pauses was consistent across age, and thus our variable of number of pauses also reflects dwell time in the VE).

Figure 1 about here

2.2 Results

2.2.1 Analysis plan

Anonymised data and analysis syntax for this study can be found at: https://osf.io/fcsug/ . The data were largely not normally distributed for the TD group (Kolmogorov-Smirnov, $p<.05$ for all), and were normally distributed for the DS and WS groups for two of the four variables only, path length and number of pauses ($p>.05$). For the number of areas visited, 66% of the TD children, 58% of the WS participants and 42% of DS participants visited eight or nine areas, with the distribution skewed

towards a high number of areas visited (nine being the maximum). For the number of revisits, for all three groups the data was positively skewed, with a long tail for higher numbers of revisits. Based on this, for correlational analyses, Pearson correlations and Spearman correlations are used for normal and non-normal data respectively. Because ANOVA is robust to violations of assumptions of normality (Blanca Mena et al., 2017), parametric ANOVA analyses were used. Correlations were carried out between chronological age and RCPM and each exploration variable, for each group, to investigate developmental changes in exploration approaches. Sensitivity power analyses demonstrated that the correlation analyses were powered to detect a medium to large effect size, with power at 80% and a critical alpha of 0.05 (TD: r = 0.21; DS: r=.34; WS: r = .40). ANOVA for each exploration variable was carried out to compare exploration performance of the WS and DS groups to typical development. It was important that the three groups were comparable for these analyses, and thus subsamples of the full groups were derived. We chose RCPM as our measure, i.e., a measure from the same domain as the experimental tasks. Due to a large number of low scores in the DS group, all genetic syndrome participants with an RCPM ≤10 were excluded from group comparison analyses (N=10 DS excluded; N=2 WS excluded). The TD subgroup was derived by including the N=20 TD participants with the lowest RCPM score. Sensitivity power analysis demonstrated that the ANOVAs were powered to detect a large effect size (f=.40), with power at

80% and a critical alpha of 0.05. The groups did not differ statistically by RCPM score (p=.063, although note that the score of the DS subgroup was marginally lower than that of the TD subgroup. WS vs TD: p=.794; DS vs TD: p=.061; WS vs DS: p=.208) or computer control (p=.136); descriptive statistics can be seen in Table 1.

2.2.2 Development of exploration behaviours

Correlations between chronological age and RCPM and: path length; number of pauses; number of areas visited; number of revisits; computer (keyboard and eye coordination) (critical alpha: p≤.003) were carried out, per group. As shown in Table 2, for the WS group performance was not related to Chronological Age or non-verbal ability. For the DS group, all correlations were non-significant with the exception of a negative association between the number of revisits and Chronological Age, and between computer control and both path length and areas visited. For the TD group, path length increased, the number of pauses decreased and the number of areas visited increased with increasing Chronological Age, whilst path length and the number of areas visited increased with increasing non-verbal ability. There was also a negative association between computer control and path length.

Table 2 about here

2.2.3 Group comparisons of exploration behaviours

ANOVA for each VE exploration variable included Group as a between participant factor (3 levels; WS-subgroup, DS-subgroup, TD-subgroup). Groups behaved similarly for number of pauses ($F<1$); and number of areas visited ($F<1$) (Figure 2). There was a main effect of Group for path length ($F(2, 60)=3.18$, $p=.049$, $\eta_p^2=.10$) and number of revisits ($F(2, 60)=6.87$, $p=.002$, $\eta_p^2=.19$). Post-hoc Tukey paired comparisons revealed that the DS group had shorter path lengths (mean [SD]: 1111.76 [520.91]) than the TD group (mean [SD]: 1597.45 [860.27]) ($p=.041$); WS mean [SD]: 1277.45 [436.80]) all other comparisons, $p>.05$). For the number of revisits the TD group (mean [SD]: 5.75 [3.78]) made significantly more revisits than the DS (mean [SD]: 2.43 [1.86], $p=.002$) or WS group (mean [SD]: 3.18 [3.08], $p=.02$), whilst the DS and WS group did not differ, $p=.69$.

Figure 2 about here

2.3 Discussion

This experiment was designed to assess exploration behavior in an undirected wayfinding task (with no specific destination to reach). Exploration behavior of the TD group is discussed fully in Farran et al. (2022). In summary, for the TD group, exploration of the environment was related to chronological age, and demonstrated an increasingly active strategy with age. That is, older children explored a wider area of the VE, created longer path lengths and paused less than younger children. This strategy is beneficial for efficient free exploration.

In the WS and DS groups, most of the exploration variables were not related to chronological age (except for the number of revisits that decreased with age in the DS group). Whilst interpretation is limited by low power, this suggests that experience acquired with age does not improve exploration behaviors in these individuals.

Moreover, in contrast to typical development, their patterns of exploration differed. The number of areas visited and the number of pauses were similar to the TD comparison subgroup, whilst individuals with DS had shorter path lengths than their TD peers (although note that the DS subgroup were less well matched to the TD subgroup, and time per target comparison with the TD subgroup only just met the significance cut-off), and both the WS and DS subgroups made fewer revisits than the TD comparison subgroup. This demonstrates that there is little difference between the two neurodevelopmental condition groups when the goal is simply to learn about the environment with no specific destination to be reached (undirected wayfinding). The differences in their exploration behavior, relative to the TD children, might be the consequence of the absence of a tangible goal to achieve. People with intellectual disability are sometimes described as having limited motivation (Cuskelly & Gilmore, 2014) and a more goal-directed task may increase their exploration behaviors. The DS group also demonstrated an association between

computer control and both path length and the number of areas visited; whilst the groups did not differ on their level of computer control, it is possible that limitations in computer control further negatively impacted motivation in this group. Note that a correlation between computer control and path length does not necessarily indicate a limitation of computer control on the ability to travel within the time limit because the same correlation was observed for both the TD and the DS group, despite significantly different path lengths.

In Experiment 1, we did not measure learning and so do not know whether the patterns of exploration observed support spatial learning. Experiments 1 and 2 were designed to be complementary studies of free exploration / undirected wayfinding (Experiment 1) and exploration with a goal / directed wayfinding (Experiment 2). In Experiment 2, participants were required to collect six stars within a VE and bring them back home within three minutes. The number of stars collected were shown on a scoreboard on the side of the screen. As in Farran et al. (2022), there were two conditions: a standard condition (unaided goal-directed wayfinding) and an overhead map condition (aided goal-directed wayfinding). Learning was measured across five trials.

3. Experiment 2

3.1 Method

No part of the study procedures or analysis plans was preregistered prior to the research being conducted. We report how we determined our sample size, all data exclusions, all inclusion/exclusion criteria, whether inclusion/exclusion criteria were established prior to data analysis, all manipulations, and all measures in the study. Due to the rarity of our genetic syndrome groups, we report sensitivity power analyses (see Lakens, 2022). Data inclusion and exclusion criteria were established prior to data analysis and are reported.

3.1.1 Participants.

The same participants took part in Experiment 2 as in Experiment 1.

3.1.2 Design and Procedure (as stated in Farran et al., 2022)

Experiment 2 was conducted immediately after Experiment 1, in the same session (up to forty minutes, with breaks). Participants attempted to locate six targets (stars) within a VE with and without the presence of an overhead, 'satellite navigation'-style view map of the environment, with five trials per condition. Two VEs were created which were counterbalanced across overhead map and standard conditions. VE 1 and VE 2 contained ten buildings, three distant landmarks and 40 proximal landmarks within a 300x300 unit environment bounded by four walls (as in Experiment 1). Different landmarks were used in VE1 and VE2, and these differed from those used in Experiment 1[2].

There were six stars in each condition and at least one (and a maximum of two) appeared in each quadrant of the VE for VE 1 and VE 2 (Figure 1). Each star was placed next to a proximal landmark. To ensure that participants recognised that they were in different VEs between conditions the perimeter walls of the VEs were uniquely coloured; for VE 1 the walls were red, for VE 2 the walls were blue. Participants were told that they were on a treasure hunt to find six stars and bring them back to 'home' within three minutes (time remaining indicated by the blue bar at the top of the screen). When they collected a star it disappeared from within the VE and a congratulatory sound was played. The number of stars collected was shown in a scoreboard on the left side of the screen, the stars were shown as grey on the scoreboard when they were yet to be collected and yellow once collected (Figure 1). Equal numbers of participants were assigned VE 1 or VE 2 for the standard condition, with the remaining VE being used for the overhead map condition. The standard (without map; five trials, three minutes each) condition was always presented first to avoid carryover effects of strategies learnt using the presence of the map. For the overhead map condition (five trials, three minutes each), participants were told that this time there would be a map on the screen that might help them (see Figure 1). The map appeared at the bottom right and depicted: the outline of the buildings present in the environment (as white blocks); the participant's current location (as a blue dot which moved in real-time contingent with the participant's location); the 'home' location (a dark grey

square); the three distant landmarks for the VE (it did not display proximal landmarks); and the stars that had been collected (stars were not visible on the map prior to being collected). For both conditions participants started each trial at ‘home’ and then attempted to find each star before returning to ‘home’ within a three minutes time limit. The trial ended after three minutes regardless of this goal being achieved. Participants completed five trials per condition. Star locations were identical across the five trials of each condition.

Dependent variables for each trial were recorded with respect to spatial knowledge and to exploration patterns. Exploration variables were as in Experiment 1 (note, that we also measured the absolute length of pauses. Consistent with the variable ‘number of pauses’, ‘length of pauses’ also reduced with age, and thus our variable of number of pauses also reflects dwell time in the VE). The first spatial knowledge variable was the number of stars collected divided by the time that each participant spent in the VE, a ‘time per target’ variable. Time in the VE either timed out at 180 seconds (if the participant was still looking for stars and/or had not returned to the home location) or stopped at the point at which the participant had collected all 6 stars and returned to the home location. The second spatial knowledge variable captured participant’s route creation across the five trials. This was a measure of consistency in the order of star collection over consecutive trials, and thus a measure of the extent to which star collection

became systematic as the participant learnt their way around the environment. The following formula was used: Sum of differences x [(6-sequence length)/6]. The first part of the formula was calculated by representing each star as a number, and documenting the star order collection for each trial (e.g. Trial 1 star order collection: 2, 3, 5, 1, 4, 6; Trial 2 star order collection: 3, 5, 1, 6, 4, 2; Trial 3… , etc..), and subtracting these values between consecutive trials. If a star was not collected, the subtraction was replaced with a value of 6. The absolute values of these six differences were then summed such that a value close to zero represents a more consistent order (e.g. For the example trials above, the absolute subtracted values would be: 1, 2, 4, 5, 0, 4; and the sum would be: 16). The second part of the formula captures common sequences that are shifted in the overall order of star collection from one trial to the next. In the examples above, both Trials 1 and 2 share a sequence of '3, 5, 1', but the sequence falls in a shifted position for each trial. This common sequence is not captured by simple subtraction. Thus, for shared sequences of 3 or more stars (between two consecutive trials) we credited this by multiplying scores by the proportion of the sequence that was not shared across two consecutive trials, i.e. (6-sequence length)/6. For the example trials above, this would by 16 x (6-3)/6= 8. Thus, the lower the target consistency score, the higher the consistency in target order between trials.

3.3 Results

3.3.1 Analysis plan

Anonymised data and analysis syntax for this study can be found at: https://osf.io/fcsug/. Four children from the TD group, five individuals with DS and three individuals with WS could not be included because they did not complete all ten trials across the two navigation conditions. For two individuals with DS and one individual with WS, no stars were collected on one of the ten trials. For these 3 cells, to create a time per star datapoint, the time used was their time in the maze (180 seconds). This is conservative as it is the equivalent of crediting these individuals with having found 1 star. The associational analyses therefore included 87 children from the TD group, 21 individuals with WS and 28 individuals with DS, whilst group difference analyses included 20 children from the TD subgroup, 19 individuals with WS and 20 individuals with DS (subgroups had similar levels of RCPM score, $p=.081$ [WS vs TD: $p=.942$; DS vs TD: $p=.092$; WS vs DS: $p=.187$] and computer control, $p=.173$). Sensitivity power analyses demonstrated that the correlation analyses were powered to detect a medium to large effect size, with power at 80% and a critical alpha of 0.05 (TD: $r = 0.21$; DS: $r=.37$; WS: $r = .43$). ANOVAs were powered to detect a small to medium effect size ($f=.08$ to $f=0.20$).

The time per target variable, and many of the exploration variables were mainly not normally distributed (Kolmogorov-Smirnov, $p<.05$). For correlational analyses, Pearson correlations and Spearman correlations are used for normal and non-normal data respectively. Because ANOVA is

robust to violations of assumptions of normality, parametric analyses were applied throughout (Blanca Mena et al., 2017). Target order consistency scores data were broadly normal (Kolmogorov-Smirnov, $p>.05$ for the majority of variables); parametric analyses were applied.

Correlations were carried out, for each group, between chronological age, RCPM, BPVS, computer control and: path length; number of pauses; number of areas visited; number of revisits, target consistency and time per target (critical alpha: $p\leq.002$). All variables were created by summing the variables across the five trials of the standard condition to create a single variable for each measure. To investigate learning across the five trials, a series of ANOVAs were carried out with Trial number (or Trial pair, for the target consistency variable) and Condition (overhead map, standard) as within participant variables and Group (3 levels; WS-subgroup, DS-subgroup, TD-subgroup) as the between participant variable. Dependent variables were: time per target, consistency in target order collection, path length, number of pauses, number of areas visited and number of revisits.

Correlations between consistency and time per target were carried out to investigate the impact of route learning strategy on navigation success. We used partial correlations controlling for age and computer control for the TD group and partial correlations controlling for computer control only for the two neurodevelopmental condition groups, since these variables were not related with chronological age in these groups.

To determine whether performance on Trial 1, when the environment was unfamiliar, was associated with subsequent navigation success, correlations were carried out between the four exploration variables and time per target during the first trial and time per target (as a measure of navigation success) during the subsequent trials, for the standard condition only. The latter variable was a sum of time per target for the last four trials. Where data was not normally distributed, Spearman correlations were carried out. As above, we used partial correlations controlling for age and computer control for the TD group and partial correlations controlling for computer control only for the two neurodevelopmental condition groups. To correct for multiple comparisons, a critical alpha of $p \leq .01$ was used.

Finally, Latent Profile Analysis (LPA) was used to understand individual differences in the profiles of exploration and navigation variables in our developmental sample of TD children and to better understand whether the profiles of the DS and WS individuals represent typical or atypical approaches to exploration and navigation. LPA is a data-driven exploratory approach to understanding heterogeneity in a sample in which all participants are entered, agnostic to their group membership. LPA organises the sample into profile subgroups. Each profile represents a homogenous subset of the sample, who are characterised by the same pattern of responses on the chosen set of variables (Berlin et al., 2014). Here, we are using it to understand whether latent profiles of our set of

exploration and navigation variables are syndrome-specific, or group agnostic. All variables were created by summing the variables across the five trials to create a single variable for each measure.

3.3.1 Navigation efficiency: Target collection

ANOVA of time per target was carried out with Trial number (5 levels) and Condition (overhead map, standard) as within participant variables and Group (WS-subgroup, DS-subgroup, TD-subgroup) as between participant variables. There was a main effect of Group, $F(2, 56)=4.25$, $p=.02$, $\eta_p^2 =.13$. Tukey comparisons revealed this was due to higher time per target for the DS group than the WS ($p=.031$) and the TD group ($p=.049$). The WS group did not differ from the TD group, $p=.973$. There was a main effect of Trial, which is best reported as a linear contrast, $F(1, 56)=13.51$, $p<.001$, $\eta_p^2 = .19$ due to lower time per target with increasing trials. There was no main effect of Condition ($F<1$), and no significant interactions ($p>.05$ for all).

3.3.2 Navigation strategy: Consistency in order of target collection

ANOVA of consistency in target collection order was carried out with Trial pair (four levels: Trial 2 to 1; Trial 3 to 2; Trial 4 to 3; Trial 5 to 4) and Condition (2 levels) as within participant factors and Group (3 levels) as a between participant factor. This demonstrated a main effect of Group, $F (2, 56)=3.19$, $p=.049$, $\eta_p^2 =.10$. Tukey pairwise comparisons revealed that this

was due to marginally lower consistency in the DS group compared to the WS group (p=.067), and no differences between the DS and TD group (p=.105) or the WS and TD group (p=.970), and thus should not be considered further. There was also a main effect of Trial pair, which is best reported as a linear contrast, F(1, 56)=8.64, p=.01, η_p^2 =.13 (Figure 3). The main effect of Condition was not significant, F<1. The three-way interaction of Group by Condition by Trial pair was significant, F(6, 168)=2.65, p=.02, η_p^2 =.09. Further exploration revealed a Condition by Trial pair interaction for the WS group only (p=.02). This was due to the effect of Trial pair for the WS group being driven by the overhead map condition only (overhead map condition: p=.01; standard condition: p=.60). The main effect of Trial pair in the overhead map condition for the WS group was due to lower consistency scores for Trial 4 – Trial 3 compared to Trial 2 - Trial 1 pair only (p=.034), indicative of some increase in the consistency of the order of target collection as trials progressed. All remaining interactions were non-significant (p>.05 for all).

Correlations for the full samples, between consistency and time per target were computed in order to see whether the strategy of creating a route by collecting the targets in the same order contributed to navigation success. These correlations were significant in the three groups (partial correlation controlling for age and computer control in the TD group, r = .68; p<.001;

partial correlation controlling for computer control in the DS and WS group, respectively: r = .78; p<.001; r = .70; p<.001).

Figure 3 about here

3.3.3 Exploration patterns: number of pauses; number of areas visited; number of revisits

To determine developmental changes in exploration and navigation variables, correlations were carried out between cognitive ability, age and computer control, with exploration and navigation variables. As shown in Table 3, in the two neurodevelopmental condition groups, performance was not related to chronological age or cognitive ability, but there were associations between computer control and target consistency and time per target for the DS group, but not the WS group. For the TD group, the number of pauses and the time per target decreased with increasing chronological age, whilst target consistency increased. Similar to the DS group, computer control was related to time per target.

For each of four exploration variables, ANOVA was carried out with Condition (2 levels) and Trial (5 levels) as within participant factors, and Group (3 levels) as a between participant factor (Table 4). The four dependent variables were: number of areas visited, path length, the number of revisits and the number of pauses over 2 seconds.

Tables 3 and 4 about here

The main effect of Group was not significant for any of the four variables. There was a main effect of Trial for the number of areas visited and the number of pauses (Figure 3). For the number of areas visited, this is best described as a linear contrast, due to an increased number of areas visited with increased trials, $F(1, 56) = 8.32$, $p=.006$, $\eta_p^2=.13$. There was a significant interaction between Trial and Group for the number of pauses and thus the main effect of Trial for the number of pauses is best explained via this interaction. There was also a significant Trial by Group by Condition interaction for the number of revisits, and thus the above interactions for pauses and revisits are described in tandem. This revealed that the DS group did not show any change in the number of pauses or the number of revisits (consistently between conditions) across trials ($p>.05$ for both). The WS group demonstrated an effect of Trial for number of pauses and number of revisits, which were best explained as linear contrasts (pauses, $F(1, 18)=6.73$, $p=.02$, $\eta_p^2=.27$; revisits, $F(1, 18)=6.70$, $p=.02$, $\eta_p^2=.27$). There was an interaction between Condition and Trial for the WS group only for revisits ($F(4, 72)=5.71$, $p<.001$, $\eta_p^2=.24$). This demonstrated that whilst there was an effect of Trial for both Conditions, it was significantly linear in the overhead map condition only and plateaued at Trial 3 for the standard condition (standard (simple main effect): $F(4, 72)=4.16$, $p=.004$, $\eta_p^2=.19$

(Trial 3>1,2, p<.05 for both); overhead map (linear contrast): F(1, 18)=8.96, p=.008, η_p^2=.33). The TD group showed no effect of Trial for revisits (consistently between conditions), F<1, and a linear reduction in pauses across trials, F(1, 19)=36.78, p<.001, η_p^2=.66. There was an effect of Condition for the number of revisits due to more revisits in the overhead map condition than the standard condition. For the number of pauses, Condition interacted with Group. This was due to an effect of Condition (more pauses in the standard condition than the overhead map condition) in the TD group (p<.001), but not in the WS or DS groups (p>.05 for both).

3.3.4 Associations between exploration behaviour during the first trial and subsequent navigation success

Given the minimal additional impact of the overhead map condition over the standard condition, associations with navigation success are investigated for the standard condition only.

Table 5 about here

As shown in Table 5, in the TD group, low navigation success (high time per target) in the last four trials was associated with short path length, few areas visited, few revisits and high number of pauses during the first trial. In the WS group, it was associated with short path length and high number of pauses. No correlation was significant for the DS group. Interestingly, the mean time per target during the first trial was associated with the mean

time per target for subsequent trials for the TD and WS groups. This suggests that individual differences in navigation success were already apparent during the very first trial in these groups.

3.3.5 Latent Profile Analysis (LPA)

The four exploration variables, target consistency score and time per target variables were entered into the LPA, ignoring group membership. LPA was conducted using R version 4.0.3 with R studio version 1.3. Taking an exploratory approach, models with up to six latent classes were fit to the data (Table 6). The model with the optimal number of classes was determined using the following criteria: low Bayesian Information Criterion (BIC) and sample-adjusted BIC (SABIC), indicative of better model fit; significant Bootstrap Likelihood Ratio Test (BLRT); high entropy values (see Berlin et al., 2014).

Model fit: Observing BIC values in Table 6, the lowest BIC and SABIC values are for model 5, and these values increase from model 5 to model 6, suggesting that model 5 has the least unexplained variance. With reference to BLRT, a significant p value indicates a significant difference between k profiles and k-1 profiles. LPA solutions were significant for models 1 to 5 (p=.01), but non-significant for model 6, thus indicating that model 6 is not an improvement on model 5. Entropy varies from zero to one. The closer

entropy is to one, the fewer classification errors. Entropy for all models was above 0.9 for models 1, 2, 3 and 5, but showed a drop from model 5 to model 6. Consideration of the smallest class was also taken into account. The smallest class in the 3, 4 and 5 class model was the same size (N=7), thus although small it was deemed a reliable class (Lubke & Neale, 2006). Taken together, these findings are not supportive of a 6 class model, thus the 5 class model was accepted.

Table 6 about here

Table 7 details the means and standard deviations for the six variables for each of the 5 profiles. Profile 1 included a small number of participants with extreme scores on all variables with the exception of pauses. Participants had very high time per target (indicative of very poor navigation), very high target consistency scores (indicative of very low consistency across trials), average number of pauses, very short path lengths, visits to very few areas and very few revisits. Profile 2 represented average performance across all six variables. Profile 3 consisted of cautious participants indicated by a high number of pauses. This was coupled with particularly short path lengths, visiting average to low numbers of areas and low numbers of revisits. This profile also showed low navigation success (average to high time per target). Profile 4 consisted of active explorers indicated by infrequent pausing. They showed average path lengths, average numbers of areas

visited and average numbers of revisits. This strategy was coupled with the strongest navigation across the 5 profiles (lowest time per target) and the strongest target consistency (lowest target consistency scores). Profile 5 consisted of above average to strong navigation (time per target) and average target consistency scores, and a relatively active strategy indicated by average to low numbers of pauses. This was coupled with long path lengths, high numbers of areas visited and high numbers of revisits. In summary navigation was most successful for profile 4, followed by profile 5, with average navigation success in profile 2. Navigation was poor in profile 3 and extremely poor in profile 1. The standardised means for each measure are plotted per profile in Figure 4. Figure 5 shows the extent to which the profiles reflected diagnostic group, TD developmental level and gender.

Table 7, Figure 4 and Figure 5 about here

4. Discussion

In Experiment 2, participants were required to collect six stars and bring them back to the home location within three minutes. Wayfinding was analyzed through the time per target variable (hereafter referred to as ‘navigation success’) and the measure of consistency in target collection order (capturing route creation across the five trials). Exploration behavior was analyzed with four variables: path length; number of pauses; number of

areas visited; number of revisits. We were also interested in the association between the exploration variables during the first trials and navigation success during the subsequent trials.

4.1 Exploration and wayfinding in typical development

Exploration behavior of the TD group is discussed fully in Farran et al. (2022).

In summary, wayfinding efficiency increased with age and the strategy of creating a route by collecting the targets in the same order contributed significantly to navigation success in the TD children. Partial correlations controlling for CA and computer control revealed that TD participants who took less time to collect the targets during the first trial also took less time to collect the targets in the following trials. This suggests that individual differences in navigation efficiency were already present during the first exploration. Moreover, participants who explored the VE using an active strategy in Trial 1 (with longer path lengths, more visited areas, and less pauses) took less time to collect the targets in subsequent trials. This result could be seen in the light of adult research showing that higher rate of diffusion through an environment and pausing less were related to the ability to locate targets in large scale environments (Gagnon et al., 2018; Munion et al., 2019). The overhead map improved wayfinding efficiency of TD children in Farran et al. (2022), but had little impact on the TD comparison subgroup. Indeed, the map reduced the number of pauses

without increasing wayfinding efficiency. This is probably due to the young age of this TD subgroup.

4.2 Exploration and wayfinding in Down syndrome

Consistent with other studies using VE, the participants with Down syndrome had lower navigation performance than the WS participants (Farran et al., 2015; Purser et al., 2015). They took more time to collect the targets than the WS group and the TD group (although note that the DS group were less well matched to the TD group, and time per target comparison with the TD group only just met the significance cut-off). However, as with the TD children and WS group, the strategy of creating a route by collecting the targets in the same order contributed significantly to their navigation success. Consistent with other studies using VE, all three groups (including participants with DS) improved their performance across trials (Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015; Himmelberger et al., 2020; Purser et al., 2015). Finally, the overhead map condition did not improve their navigation success or modify their exploration pattern. This suggested that the map did not improve their configural knowledge of the spatial display, as in Toffalini and al. (2018).

Wayfinding efficiency and exploration behavior were related to computer control in the DS group more than the two other groups. Computer control was negatively related to path length, number of areas visited (Experiment

1), time per target and target order consistency (Experiment 2). Thus, difficulties or slowness in hand-eye coordination may have impaired spatial exploration and navigation in the DS group. Note, however, that the TD group also showed associations between computer control and path length (Experiment 1) and time per target (Experiment 2) which suggests that associations with computer control are not unique to DS. Purser and al. (2015) used an equivalent measure of keyboard control in a route learning task with a virtual maze. In their study, the correlation between computer control and number of errors in route learning was low and non-significant. The link between computer control and spatial navigation seems to depend on the DV (i.e. errors rates in Purser et al. *versus* path length in the current study) and/or on the task parameters (route learning in a path-maze *versus* unconstrained exploration in a large-scaled virtual environment). The open spaces and unconstrained nature of the current study might be more impacted by variation across participants in their ability to control their movement through the environment, whereas previous route-learning studies used a relatively constrained set of paths, which could be argued to add some environmental constraint on movement. This suggestion requires further research to support, however, due to the different DVs used across studies.

Exploration behaviour in the DS group was akin to the TD comparison subgroup and WS comparison subgroup. However, contrary to the WS group and the TD group, number of pauses and number of revisits did not

vary across trials, suggesting that the DS participants did not improve their exploration behavior with repeated trials. Furthermore, unlike the TD children, for the DS group, the relationships between exploration variables during the first trial and navigation success in the following trials were weak and non-significant (partial correlations controlling for computer control). This suggested that their exploration behaviour during the first trial did not enable them to form a spatial representation of the environment that could help them to locate the targets more rapidly.

4.3 Exploration and wayfinding in Williams syndrome

Navigation efficiency was higher in the WS group than in the DS group. Navigation success improved across trials and, as for the other groups, the strategy of creating a route by collecting the targets in the same order contributed significantly to navigation success. The overhead map did not improve navigation success but seemed to improve the consistency of target order collection across trials. This result could mean that participants with WS were able to translate the direction that they were travelling on the map, to their egocentric view and direction of travel during navigation in order to locate next targets. This contrasts to Farran et al. (2010), who found limited evidence of map use in individuals with WS and thus further research is needed to investigate this inconsistency between studies.

Computer control was not related to wayfinding efficiency and exploration behavior in the WS group (and effect sizes were generally small). In the

standard condition, exploration behavior during the first trial was related to navigation success in the following trials. As in typical development, participants with WS who walked a longer path length and paused less during the first trial took less time to collect the targets thereafter. Moreover, exploration behavior in the WS groups changed across trials, with a reduction in the number of pauses and an increase in the number of revisits (although note that the increase in revisits across trials was linear in the overhead map condition, but plateaued at Trial 3 in the standard condition). The reduction in the number of pauses was also observed in the TD comparison subgroup. This reduction is likely due to their progressive development of a spatial representation of the environment with repeated trials. As their representation of the environment developed, participants had less need to stop to get their bearings or to encode new spatial information (such as a landmark).

The increase in revisits across trials was unique to the WS group. In an experiment where participants were required to search for targets in a garden with rewarded location (vista space), Foti et al. (2011) found that participants with WS revisited more previously inspected locations than TD MA children. According to Foti et al. (2011) this behavior could be due to a visuo-spatial memory deficit or to impaired Executive Functions in WS individuals. However, in the current study, these deficits are unlikely to explain the increase in revisits across trials. It is possible that this increase reflects the use of an increasingly more active strategy for exploring the

environment, with repeated trials (an explanation which is also consistent with the reduction in the number of pauses across trials). Another possibility is that the increase in revisits is an artifact of a deficit in allocentric coding in WS; a lack of allocentric knowledge of the environment and thus limited understanding of the configuration of the environment would force an individual to rely on developing a fixed route (i.e. route knowledge, via egocentric coding). This could entail developing a route which involves revisits to the same place, or simply revisiting to seek an egocentric viewpoint that they recognise, for reorientation purposes. Note that the increase in revisits plateaued earlier for the standard condition than the overhead map condition, where arguably the map provided some allocentric information. This fits with the effect of target consistency in WS, which was observed for the overhead map condition only; a continued increase in revisits could reflect the progressive development of a consistent route (albeit not an efficient route).

4.4 Individual differences

Important individual differences have been reported from previous studies on navigation in neurodevelopmental conditions (Courbois et al., 2013; Farran et al., 2015). Indeed, in our study, participants with DS and participants with WS were distributed across five profiles. Profile 1, including 7 participants with very low navigation performance and low

exploration behavior (high time per target, low target consistency, short paths, visiting few areas and few revisits), mainly constituted participants with DS (and one participant with WS). These participants exhibited qualitatively different behaviour relative to participants in other profiles; they were probably disoriented in the VE and/or could have a low level of motivation to complete the task. Most of the participants with DS were distributed across profile 2 and profile 3. Profile 3 is qualitatively different from all other profiles because it represents the profile with the highest number of pauses despite average to low time per target. This suggests that this profile consisted of “cautious” participants (high numbers of pauses) with low exploration activity (short path lengths, few areas visited, low numbers of revisits) and low to average navigation efficiency (low to average navigation success, low target order consistency). Ten participants out of 28 with DS (35.7%) and 3 out of 21 participants with WS (14.28%) constituted this profile. This profile also included the youngest TD children (mean age: 7 years and 11 months) and was associated with females (14 females out of 16 TD children). Participants in profile 2 were average across the six variables. This profile included eight participants with DS (28.57%), 8 participants with WS (38.09%), and TD children who were slightly older than the profile 2 children (8 years, 4 months) and who were mainly females (11 females out of 12 TD children). Finally, 3 participants with DS (14.28%) and 9 participants with WS (42.85%) were distributed across the two most successful profiles (profile 4 and profile 5), which also included the older TD

children and were associated with males. These participants were active explorers (low numbers of pauses), but participants in profile 5 walked longer distances and made more revisits than participants in profile 4. Whilst we cannot rule out that these two profiles differ by degree (see Bauer, 2022), it could be argued that longer path lengths and higher revisits (profile 5) reflect predominant use of a route knowledge strategy (reliance on egocentric coding), compared to more reliance on configural knowledge (allocentric coding) in profile 4 (see Farran et al., 2022, for further discussion of these two 'active' strategies).

None of the five profiles were syndrome specific. Only the most extreme profile of very poor navigators was specific to DS and WS participants. Interestingly, all other profiles contained at least one individual with DS and at least one individual with WS. However, a larger proportion of participants with DS were found in the poor navigation profiles (profiles 1 and 3) while the participants with WS were more likely to be found in the successful navigation profiles (profiles 4 and 5). This is consistent with the Farran et al.'s (2015) study showing that only 10% of DS participants were able to find a shortcut in a VE, compared to 35 % of WS participants.

Wide individual differences are not uncommon in groups with genetic syndromes (Karmiloff-Smith et al., 2016). Whilst this can be observed in group level data by looking at data visually (e.g., the spread of data in violin plots or the size of error bars), to better understand the data it is important to investigate this heterogeneity. A data-driven transdiagnostic approach

can be used to go beyond simply observing individual differences. It is extremely useful for identifying whether the heterogeneity reflects a broader degree of difference, or whether there are qualitatively distinct behavioural profiles, and furthermore whether distinct profiles map onto group labels or not (Astle et al., 2022; Farran & Scerif, 2022).

Transdiagnostic approaches are also a first step to better understand the mechanisms underlying this variability; currently these are mostly unknown. Interindividual variability in neurodevelopmental conditions affects numerous domains such as intelligence, language, memory, and social cognition. It is likely that genetic, epigenetic, early neural development and environmental factors play a role in how the phenotype expresses itself in each DS or WS individual (Karmiloff-Smith et al., 2016; Kozel et al., 2022; Thomas et al., 2020). Moreover, having a neurodevelopmental condition changes the environment in which children develop, in terms of parental expectations and their interactions with their child (Cebula et al., 2010; Hodapp, 1997). With respect to spatial navigation, there are also large individual differences in the general population (Hegarty et al., 2022; Wolbers & Hegarty, 2010). It is likely that factors underlying these differences are also involved in DS and WS spatial navigation. Beyond spatial abilities, anxiety and navigation specific anxiety have a negative impact on navigation in neurotypical adults (Lawton, 1994; Wolbers & Hegarty, 2010). Based on parental reports, Farran et al. (2022)

did not find that navigation anxiety predicted navigation competence in participants with WS and participants with DS, but they noted important individual differences in anxiety and in navigation competence in both groups. However, in their study, navigation competence was indirectly assessed with a questionnaire comprising 15 questions. Further research should study the relationship between navigation anxiety and navigation performance using spatial exploration tasks, as in Gagnon et al.'s (2018) experiment with neurotypical adults. Greater childhood independence also explains individual differences in children's navigational skills (Newcombe et al., 2022) and, later, in adult wayfinding strategy (Vieites et al., 2020). Perceived social vulnerability leads many parents of children with neurodevelopmental conditions to restrict their child's navigation experience (Farran et al., 2022), which would in turn impact the wayfinding performance and the level of navigation anxiety of their children. This is supported by evidence that parents' concerns over their children's independent wayfinding predict the children's wayfinding competence and confidence in DS (Yang et al., 2018).

4.5 Limitations

This study is not without limitations. First, gender was not a planned comparison in this study and could not be analysed here because it would have unduly limited the group sizes in the two neurodevelopmental condition groups. However, we note that, in the TD group, the distribution of males and females across the latent profiles varied significantly between

profiles 2 and 3 (more females) and profiles 4 and 5 (more males). This suggests that gender differences should be investigated in future research on exploration strategies. Second, whilst the majority of this paper concerns associational analyses and used all participants, group comparisons are also of interest and were best conducted using matched groups. To create comparison subgroups, the participants with the lowest RCPM scores in the DS and WS groups were excluded. This risks these groups not being representative samples; this could inflate our understanding of the level of ability of the DS and WS groups. Furthermore, matching across three groups is difficult; whilst the DS-subgroup was well matched to the WS-subgroup, they had marginally lower RCPM scores than the TD-subgroup. This is not problematic for interpretation of the majority of comparisons, as there were few group effects. However, there were two significant comparisons between the DS-subgroup and TD-subgroup groups that are discussed with caution. Third, we did not measure heading direction. This could have differentiated pauses with and without a rotational element. Rotational movement while pausing could be indicative of efficient planning behaviour. Fourth, our correlational analyses were only powered to detect medium to large effect sizes (see Farran & Scerif for discussion of power in studies with genetic syndromes) and thus for these analyses, whilst statistical significance is reported, observation of effect size could be argued to be more useful than observation of p-values. Fifth, we did not measure participant's experience with navigational computer games. As

such, we cannot determine whether any effects were influenced by experience with navigation games. However, we have no reason to think that one neurodevelopmental condition group would have more experience in computer games than the other. Future research should include a measure of navigational computer games. Finally, consistency in the order of target collection was measured solely using the sequence in which the stars were collected. Future investigation of consistency over consecutive trials could use methods which also take into account the spatial and temporal information of each route. For example, methods that have been used with eye track data could be adapted (for example, the ScanMatch method in Anderson et al.., 2015).

5. Conclusion

The aim of this research was to study unconstrained exploration behavior in people with WS and people with DS. We used the same methodology as Farran et al. (2022) in order to study free exploration (undirected wayfinding) of a large-scale environment (Experiment 1) and exploration with the goal of locating targets in the environment (directed wayfinding, Experiment 2). Our results support previous findings on wayfinding in people with WS and people with DS. Exploration and wayfinding variables were not related to chronological age in either group, suggesting that experience acquired with age does not improve exploration behaviours in individuals with these neurodevelopmental conditions. Moreover, in line with the results of other comparative studies, navigation efficiency was

higher in the WS group than in the DS group (Farran et al., 2015; Purser et al., 2015).

There was little difference between the WS and the DS participants in the undirected wayfinding task (Experiment 1). The difference became evident when the goal of the task was to locate targets in the environment. In accordance with our expectation, individuals with WS and individuals with DS did not have the same exploration pattern, even though the strategy of creating a route by collecting the targets in the same order contributed significantly to navigation success in both groups. Exploration behaviour in DS seemed disorganized and did not improve across trials. Unlike TD children and WS participants, the relationships between exploration variables during the first trial and navigation success in the following trials were weak and non-significant (partial correlations controlling for computer control). In the WS group,
participants who explored the VE using an active strategy in Trial 1 of Experiment 2 (with longer path lengths and less pauses) took less time to collect the targets in subsequent trials. However, the WS pattern of exploration was atypical. The number of revisits increased with repeated trials for this group. This might be explained by their reliance on egocentric coding, and limitations brought about by known deficits in visuo-spatial memory and executive functions. Further research is needed to clarify this issue.

The weak exploration behaviour in DS is difficult to interpret. From a spatial information processing standpoint, they should perform better than participants with WS since they can rely on two systems to encode spatial information: egocentric and low resolution allocentric (Bostelmann et al., 2018). Nevertheless, wayfinding is a complex behaviour involving high-level cognitive functions, such as decision making, planning, flexibility, in addition to spatial information processing (Patai & Spiers, 2021). Moreover, these cognitive functions can be altered by acquired spatial anxiety (Gagnon et al., 2018). Therefore, a number of factors could explain the weak exploration behaviour in DS; this is consistent with evidence that limitations in long-term memory (Purser et al., 2014) and executive function (Farran et al., 2015) negatively impact wayfinding in this group. Further research is needed to clarify this issue.

Finally, there were wide individual differences within the two neurodevelopmental condition groups. WS and DS participants were distributed among the five profiles, with a larger proportion of DS in the poor navigation profiles and a larger proportion of WS in the successful navigation profile. Moreover, in both groups individual differences in navigation success were already apparent during the very first trial. These differences highlight the importance of investigating heterogeneity in the performance of individuals with intellectual disability and the usefulness of a data-driven transdiagnostic approach to identifying behavioural profiles (Astle et al., 2022; Farran & Scerif, 2022).

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Table 1. Descriptive statistics for the cognitive test battery. BPVS = British Picture Vocabulary Scale; RCPM = Raven's Coloured Progressive Matrices.

	Mean	SD	Range
Age (years;months)			
TD (N=91)	9;03	1;08	5;08 – 11;11
DS (N=33)	19;03	5;05	11;08 – 32;10
WS (N=24)	21;11	8;05	11;02 - 46;09
TD subgroup (N=20)	7;10	1;06	6;00 – 11;07
DS subgroup (N=21)	21;05	5;06	11;08 – 32;10
WS subgroup (N=22)	22;00	8;09	11;02 – 46;09
BPVS (raw)			

TD (N=91)	119.54	22.12	29-156
DS (N=31)*	64.10	36.52	5-145
WS (N=24)	122.00	26.19	54-168
TD subgroup (N=20)	102.50	19.27	55-134
DS subgroup (N=21)	71.23	36.40	14-145
WS subgroup (N=22)	123.00	20.72	76-147
RCPM (raw)			
TD (N=91)	28.54	5.37	10-36
DS (N=31)*	13.97	5.97	4-29
WS (N=24)	18.54	6.21	7-31
TD subgroup (N=20)	20.50	4.25	10-26
DS subgroup (N=21)	17.05	4.48	11 - 29
WS subgroup (N=22)	19.55	5.44	11 - 31

Computer			
TD (N=91)	20.84	9.30	8.32-50.41
DS (N=33)	40.34	31.73	12.48- 139.41
WS (N=24)	33.00	14.68	13.26-65.94
TD subgroup (N=20)	25.10	9.88	11.52-50.41
DS subgroup (N=21)	37.56	29.81	12.48- 123.50
WS subgroup (N=22)	32.68	13.54	13.66-65.94

Table 2: Experiment 1. Correlations between VE exploration variables and developmental variance. CA=Chronological Age; RCPM=Raven Coloured Progressive Matrices; Computer = familiarisation trial (keyboard and eye coordination).

Group	Development variable	VE exploration variables			
		Path length	Pauses	Areas visited	Revisits
DS	CA	r=-.30	r=-.48	≠ r=-.02	≠ **r=-.55**
	RCPM (N=31)	r=.06	r=-.02	≠ r=.22	≠ r=-.35
	BPVS (N=31)	r=.11	r=.26	≠ r=.04	≠ r=.02
	Computer	≠ **r=-.54**	≠ r=-.42	≠ **r=-.58**	≠ r=-.22
WS	CA	r=-.31	r=.18	≠ r=-.19	≠ r=-.06
	RCPM	r=.26	r=-.06	≠ r=.31	≠ r=.08

	BPVS	r=.19	r=-.38	+r=.02	+r=-.10
	Computer	+r=-.05	+r=.21	+r=.22	+r=-.04
TD	CA	+**r=.43**	+**r=-.40**	+**r=.34**	+r=.25
	RCPM	+**r=.32**	+r=-.18	+**r=.39**	+r=.07
	BPVS	+**r=-.34**	+r=.27	+r=-.29	+r=-.26
	Computer	+**r=.34**	+r=.27	+r=-.29	+r=-.26

+Spearman correlations. Significant correlations in bold (p<.003).

Table 3. Experiment 2 - standard condition: Correlations between VE exploration and navigation variables and developmental variables. CA=Chronological Age; RCPM=Raven Coloured Progressive Matrices. Computer = familiarisation trial (keyboard and eye coordination).

Group	Developmen t variable	VE exploration variables	Navigation

		Path length	Pauses	Areas visited	Revisits	Target consistency	Time per target
DS	CA	+-.12	-.23	-.03	+-.19	.18	+-.23
	RCPM	+.32	-.24	.33	+-.19	-.19	+-.41
	BPVS	+.42	-.04	.50	+.25	-.34	+-.39
	Computer	+-.51	+-.04	+-.58	+-.34	**+.63***	**+.60***
WS	CA	+-.24	+.42	+-.19	+-.02	+.27	.57
	RCPM	.36	-.21	+.22	.32	-.41	+.35
	BPVS	-.13	.04	+.33	-.23	-.23	+.02
	Computer	+-.14	+.19	+.14	+-.11	+.34	+.13
TD	CA	+.08	**+-.52***	+.24	+.04	**+-.53***	**+-.55***

	RCPM	[+].20	[+]-.29	[+].16	[+].02	[+]-.20	[+]-.28
	BPVS	[+].11	[+]-.29	[+].10	[+]-.09	[+]-.26	[+]-.27
	Computer	[+]-.21	[+].32	[+]-.19	[+]-.04	[+].17	**[+].36***

[+] *Spearman correlations. Significant correlations in bold. p<.002*

Table 4: ANOVA results for each of the four exploration variables.

	Main effects			Interactions			
	Group	Condition	Trial	Condition x Group	Trial x Group	Condition x Trial	Condition x Trial x Group
d.f.	2, 56	1, 56	4, 224	1, 56	4, 224	4, 224	8, 224
DV							
areas visited	F=2.94, p=.06, η_p^2=.10	F=13.19, p=.01 , η_p^2=.05	F<1	F=1.13, p=.34 , η_p^2=.04	F=1.88, p=.12, η_p^2=.03	F<1	F<1
path length	F=2.56, p=.09, η_p^2=.08	F=1.34, p=.26, η_p^2=.02	F<1	F<1	F<1	F<1	F<1
revisits	**F=5.80, p=.003, η_p^2=.17**	F=1.09, p=.36 , η_p^2=.02	F=1.50, p=.23, η_p^2=.05	F=2.07, p=.04, η_p^2=.07	F=2.48, p=.05, η_p^2=.08	F=4.01, p=.003, η_p^2=.10	
pauses	F=1.62, p=.21, η_p^2=.06	F=2.47, p=.12 , η_p^2=.04	F=4.60, p<.001, η_p^2=.15	F=4.92, p=.01, η_p^2=.15	F=2.26, p=.02, η_p^2=.08	F<1	F<1

Table 5: Standard condition, correlation between exploration behaviour during the first trial and navigation success in subsequent trials for the three groups (partial correlation controlling for age and computer in the TD group, computer in the WS group and computer in the DS group).

	Time per target T1	Path length T1	Pauses T1	Areas T1	Revisits T1
Mean time per target TD	**+.41**	**+-.53**	**+.59**	**+-42**	**+-.35**
Mean time per target DS	+.38	+-.18	+.18	+-20	+-.08
Mean time per target WS	**+.57**	**-.68**	**+.60**	+-22	-.50

+Spearman correlations. Significant correlations in bold. p<.01

Table 6: Model fit statistics for latent profile analysis models

Model	BIC	SABIC	Entropy	BLRT p value	N assigned to each Profile (P)
1-profile	8141.59	8103.63	1	NA	P1=136
2-profile	7839.57	7779.47	0.93	0.01	P1=48, P2=88
3-profile	7661.59	7579.34	0.95	0.01	P1=7, P1=84, P3=45
4-profile	7630.69	7526.29	0.88	0.01	P1=7, P2=45, P3=42, P4=42
5-profile	7590.84	7464.30	0.91	0.01	P1=7, P2=28, P3=29, P4=26, P5=46
6- profile	7621.94	7473.26	0.84	0.92	P1=7, P2=22, P3=26, P4=29, P5=28, P6=24

Table 7: Means and standard deviation of each variable per profile

	Profile 1 **N=7**	**Profile 2** **N=28**	**Profile 3** **N=29**	**Profile 4** **N=26**	**Profile 5** **N=46**
Measures			Mean (SD)		
Time per target	86.89 (21.41)	34.24 (5.23)	41.47 (7.88)	20.69(7.03)	27.17 (4.37)
Target consistency	103.00(17.24)	56.08(14,68)	70.30(19.33)	31.13(11.54)	43.71(14.11)
Pauses	73.57(29.74)	66.93(14.89)	110.97(17.04)	13.15(8.99)	20.85(13.15)
Path length	4518.63 (970.88)	8526.16 (1005.15)	6566.74 (726.36)	8552.00 (1227.56)	11312.38 (971.05)
Areas	30.57(3.74)	42.50(2.03)	39.07(2.70)	42.19(2.45)	43.69(1.53)
Revisits	11.00(4.16)	20.68(7.74)	16.41(7.70)	20.31(8.41)	36.43(11.43)

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Figure 1: Plan view of maze layouts for Experiments 1 and 2, with example images depicting local landmarks, star (use in Experiment 2 only), building block, distant landmarks.

Figure 2: Experiment 1 effect of Group plotted for exploration variables (number of pauses; number of areas visited; path length; number of revisits)

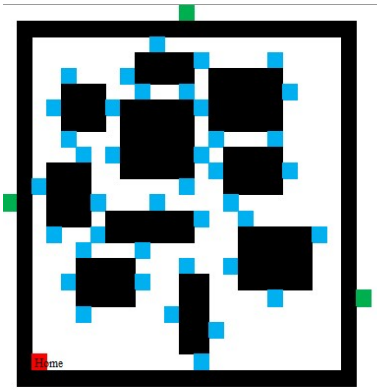
Figure 3: Experiment 2 effect of Trial plotted by Group for: time per target; target order consistency score and exploration variables (number of pauses; number of areas visited; path length; number of revisits).

Figure 4. Latent Profile plot of Z scores (means; standard error) for each measure.

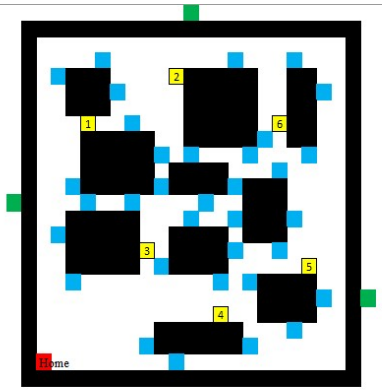
Figure 5: Number of participants as a function of diagnostic groups, TD

developmental age or genders a function of profile

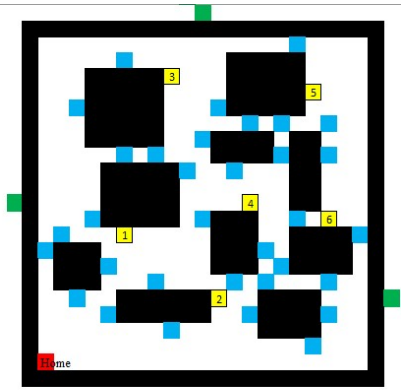
Figure 1: Plan view of maze layouts for Experiments 1 and 2, with example images depicting local landmarks, star (use in Experiment 2 only), building block, distant landmarks.



Experiment 1

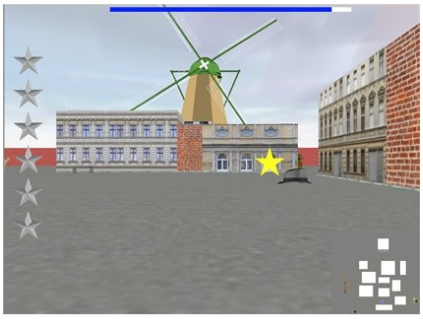


Experiment 2: VE 1



Experiment 2: VE 2

Experiment 1: Example local landmark and building block



Experiment 2 (VE1, Overhead map condition): Example local landmark adjacent to star 4, building blocks, distant landmark in view

Figure 2: Experiment 1 effect of Group plotted for exploration variables (number of pauses; number of areas visited; path length; number of revisits)

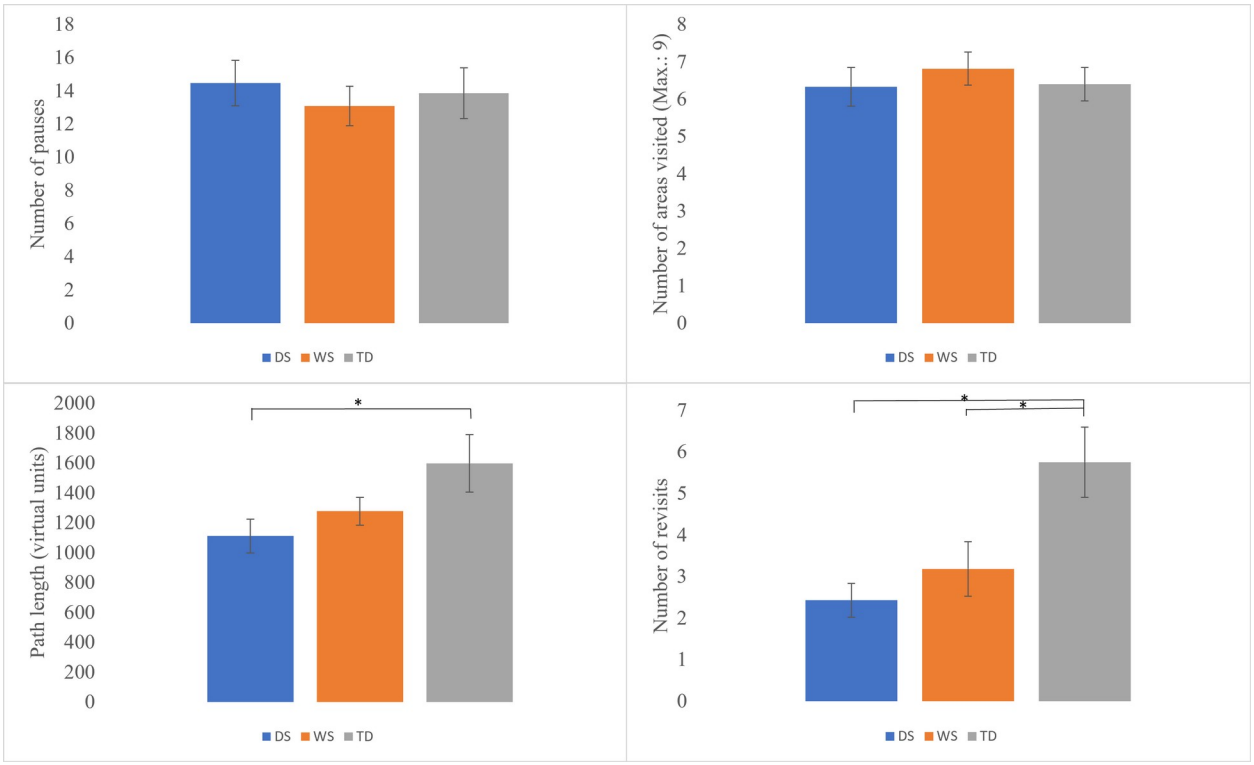


Figure 3: Experiment 2 effect of Trial plotted by Group for: time per target; target order consistency score and exploration variables (number of pauses; number of areas visited; path length; number of revisits).

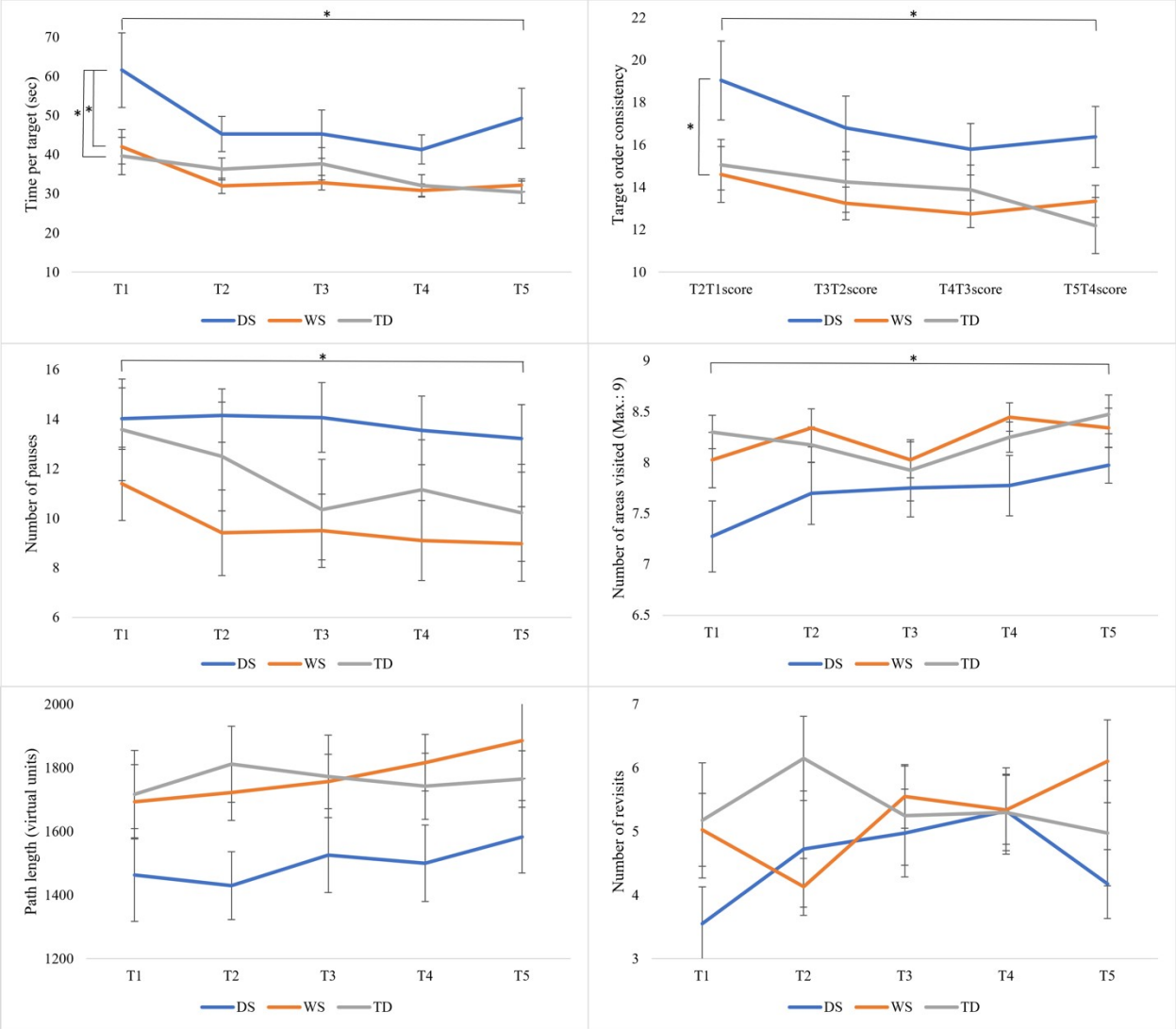


Figure 4. Latent Profile plot of Z scores (means; standard error) for each measure.

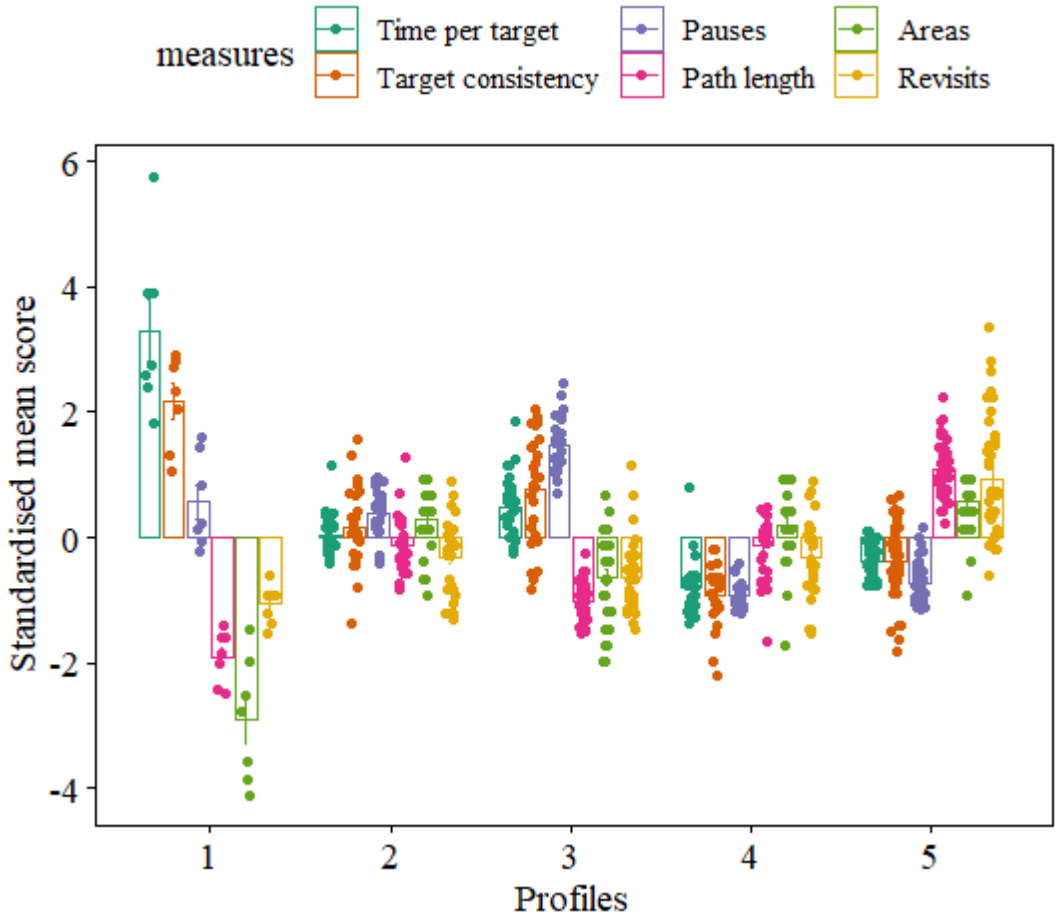
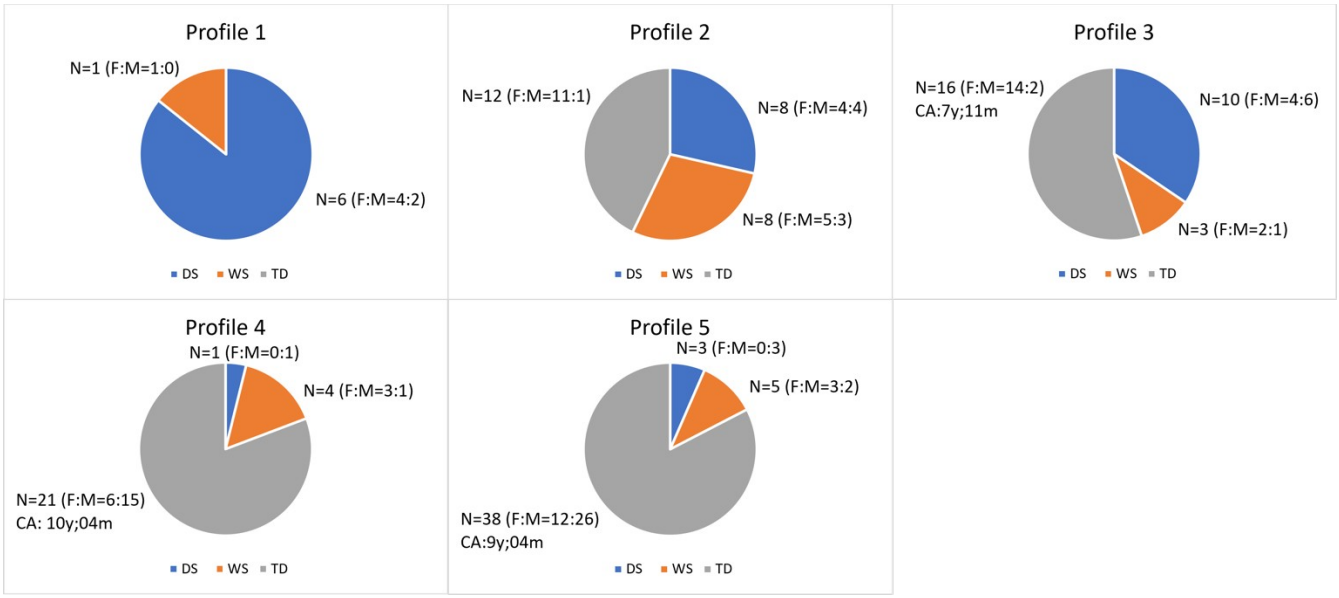


Figure 5: Number of participants as a function of diagnostic groups, TD developmental age or genders a function of profile



[1] The maze code can be found at https://osf.io/fcsug/ but the program can only be opened (and run) with the Virtools software. However, we have since built a replica of the standard condition using Unity that can be found at https://osf.io/fcsug/

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