WAYFINDING AND NAVIGATION: STRENGTHS AND WEAKNESSES IN ATYPICAL AND CLINICAL POPULATIONS

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WAYFINDING AND NAVIGATION: STRENGTHS AND WEAKNESSES IN ATYPICAL AND CLINICAL POPULATIONS

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EDITORIAL
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Editorial: Wayfinding and Navigation: Strengths and Weaknesses in Atypical and Clinical Populations

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Keywords: navigation, spatial ability, spatial memory, training, atypical development, Aging, impairments, blind

Editorial on the Research Topic

Wayfinding and Navigation: Strengths and Weaknesses in Atypical and Clinical Populations

Navigation is an essential activity of everyday life, related to both work, and leisure. For some populations with certain neurocognitive issues (e.g., those with injuries, genetic syndromes, or other clinical conditions) or characteristics (such as blindness or healthy old age), navigation is fundamental to their autonomy and access to the community. It is a complex activity that entails several stages, from planning a route to reaching a destination (Wiener et al., 2009). The encoding of environmental information in forming a mental representation or cognitive map (Tolman, 1948) and the retrieval and use of that information (Wolbers and Hegarty, 2010) rely on numerous cognitive functions—such as perception, memory, imagination, language, and decision-making—along with social and emotional processes (Dalton et al., 2019).

Our spatial memory of an environment is based on two fundamental frames of reference (Burgess, 2006). One is egocentric and involves mentally arranging the positions of objects in relation to ourselves (subject-to-object). The other is allocentric and establishes relations between objects to determine their respective locations (object-to-object). Navigation is recognized as a large-scale ability supported by small-scale spatial abilities, including the ability to mentally rotate objects or adopt different imaginary views (perspective taking), and processing skills such as visuospatial working memory (VSWM) (Hegarty et al., 2006). Motor abilities are also involved in environment learning (e.g., Voyer and Jansen, 2017). External means (such as navigation aids) can also improve our navigation efficiency. This is a core issue, for instance, in studies on the blind (e.g., Gallay et al., 2013). Brain structures provide the basis for our environment representations and there is neuropsychological evidence indicating that representations with allocentric properties are developed and stored in the medial temporal lobe, parahippocampal gyrus, and hippocampus. The posterior parietal lobe is involved in representations with egocentric properties, and the retrosplenial cortex, in switching between egocentric and allocentric properties of representation. Other brain structures play a part in wayfinding, for instance, the prefrontal cortex supports navigation planning activities (e.g., Lithfous and Després, 2013). The brain regions and networks involved in navigation mechanisms are often examined by considering individuals with brain damage or particular characteristics (e.g., hippocampal volume is smaller in Down syndrome than in matched typically-developing individuals).

With a collection of 15 studies, this special issue advances our knowledge of some navigation and related aspects. Several atypical development (AD) populations are examined in this issue, including: children and adults with William syndrome (WS), who are known to have stronger verbal than spatial abilities (Foti et al.); also comparing them with those with attention deficit hyperactivity disorder [ADHD]; Farran et al.); individuals with Down syndrome

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(DS; Meneghetti et al.; Himmelberger et al.), known to have stronger spatial than verbal abilities; individuals with autism spectrum disorders (ASD; Cardillo et al.) whose profile in the spatial domain varies. Some contributions examined adults with a cognitive disability (with heterogeneous pathogenesis; Delgrange et al.) who can have difficulty navigating due to their impaired intellectual functioning; and Korsakoff patients, whose memory disorders also affect their recall of spatial information (Janzen et al.).

Other papers examine healthy older adults (Muffato and De Beni) or those with impairments. Elderly people with vascular cognitive impairment (VCI), who have specific egocentric representation difficulties due to their parietal deficit, are compared with cases of early-stage Alzheimer's disease, whose temporal deficit causes specific allocentric representation difficulties (Lowry et al.). These studies mainly examined the allocentric versus egocentric properties of environmental representation using virtual environments (Lowry et al.; Farran et al.; Janzen et al.; Himmelberger et al.), videos of real environments (Muffato and De Beni), or real path learning (Meneghetti et al.). Some studies used spatial tasks to assess navigation-related aspects, such as perspective taking (Cardillo et al.), route planning (Bocchi et al.), and the peripersonal space (Foti et al.), or conducted semi-structured interviews on how respondents solved everyday navigation issues (Delgrange et al.). Two papers focus on individuals with brain lesions: one profiles the navigation-related difficulties of two right-brain-damaged patients using several small- to large-scale tasks (Bocchi et al.); the other concerns an imagery-based rehabilitation program for a patient with right temporal lobe damage suffering from topographical disorientation (Boccia et al.). Two other papers examine blind people and the use of haptic aids, one during navigation (Bharadwaj et al.) and the other for exploring a map before navigating in a real-world setting (Giudice et al.). Finally, two papers assess the malleability of healthy individuals' navigation skills: one involves specific training based on exploration, moving from small areas to larger and more complex environments (McLaren-Gradinaru et al.); the other specific training on the use of egocentric and allocentric navigation strategies (van der Kuil et al.). These findings offer insight into potential rehabilitation programs for individuals with navigation difficulties.

Although different aspects of navigation are examined, each of these studies uses different methods and considers specific populations. The results of these studies indicate that different populations (AD, older adults, Korsakoff patients) can mentally represent spaces and environments with egocentric properties (or sketched; Farran et al.; Meneghetti et al.; Janzen et al.; Muffato and De Beni; Cardillo et al.). Allocentric properties pose more of a challenge, with evidence that allocentric representation is no more impaired in AD than in VCI (Lowry et al.), and can be facilitated by a structured environment (Himmelberger et al.). Mental representations of spaces and environments seem to be supported by general cognitive functioning (as seen in older adults), visuospatial abilities such as mental rotation, VSWM, and the self-reported pleasure people experience when exploring an environment (Cardillo et al.; Foti et al.; Muffato and De Beni; Meneghetti et al.). Motor abilities seem to support spatial performance too, as seen in ASD (Cardillo et al.), but this relation was not found in individuals with WS or ADHD (Farran et al.). In semi-structured interviews, disabled people reported getting lost more frequently in complex environments, and having to ask others for help (Delgrange et al.).

Studies that have examined visual impairment with blind participants have shown the benefits of innovative haptic sources, such as using vibratory signals via a hip-worn belt to navigate, especially in typically noisy everyday environments (Bharadwaj et al.), or presenting vibro-audio maps before navigating (Giudice et al.). A case study contribution explores route planning difficulties in a patient with an occipitoparietal lesion, but not in a patient with a temporoparietal lesion (Bocchi et al.), shedding light on the neurocognitive mechanisms involved in navigation and related aspects of wayfinding. Finally, results obtained with training reveal that navigation skills are malleable, enabling more efficient strategies to be learned such as changing from an egocentric to an allocentric approach (McLaren-Gradinaru et al.; van der Kuil et al.), and enabling even those with topographical disorientation to navigate successfully (Boccia et al.).

To conclude, this special issue expands our understanding of navigation abilities in populations with different characteristics, and how they can be improved by appropriate intervention. Overall, it offers insights that will prompt us to continue to investigate navigation abilities, taking up the challenges faced by different populations and enabling us to create living environments that are more inclusive and accessible.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct and intellectual contribution to the work, and approved it for publication.

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ORIGINAL RESEARCH
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Cross-Domain Associations Between Motor Ability, Independent Exploration, and Large-Scale Spatial Navigation; Attention Deficit Hyperactivity Disorder, Williams Syndrome, and Typical Development

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In typical infants, the achievement of independent locomotion has a positive impact on the development of both small-scale and large-scale spatial cognition. Here we investigated whether this association between the motor and spatial domain: (1) persists into childhood and (2) is detrimental to the development of spatial cognition in individuals with motor deficits, namely, individuals with attention deficit hyperactivity disorder (ADHD) and individuals with Williams syndrome (WS). Despite evidence of a co-occurring motor impairment in many individuals with ADHD, little is known about the developmental consequences of this impairment. Individuals with WS demonstrate impaired motor and spatial competence, yet the relationship between these two impairments is unknown. Typically developing (TD) children (N = 71), individuals with ADHD (N = 51), and individuals with WS (N = 20) completed a battery of motor tasks, a measure of independent exploration, and a virtual reality spatial navigation task. Retrospective motor milestone data were collected for the ADHD and WS groups. Results demonstrated a relationship between fine motor ability and spatial navigation in the TD group, which could reflect the developmental impact of the ability to manually manipulate objects, on spatial knowledge. In contrast, no relationships between the motor and spatial domains were observed for the ADHD or WS groups. Indeed, while there was evidence of motor impairment in both groups, only the WS group demonstrated an impairment in large-scale spatial navigation. The motor-spatial relationship in the TD, but not the ADHD and WS groups, suggests that aspects of spatial cognition can develop via a developmental pathway which bypasses input from the motor domain.

Keywords: attention deficit hyperactivity disorder, Williams syndrome, motor development, spatial cognition, navigation

INTRODUCTION

The motor system is central to almost everything that we do. We use motor skills to interact socially, to produce language, in handwriting and in activities of daily living (e.g., eating, brushing hair). Motor activity is integral to early development; as motor ability develops, infants become more able to explore their environment and the objects within it. Here, we discuss the development of motor skills and their relationships to spatial cognition. For example, Clearfield (2004) demonstrated that infants who had been crawling or walking for more than 6 weeks were better able to use a landmark to find a goal location in a large octagonal space than infants with less crawling or walking experience. Furthermore, performance on the A-not-B task (Piaget, 1952), which has a large spatial component (alongside factors such as cognitive flexibility and object concept), has been linked to locomotor experience (Bertenthal et al., 1994). In this task, infants observe a toy being repeatedly hidden in one of two locations (A) and successfully find the toy. However, when they then observe the toy being hidden in location B, they perseveratively search in location A for the toy. This spatial error ceases to be made once infants have had sufficient crawling experience; for example, at 7.5 months, the length of time that an infant has been crawling or using a baby walker predicts their ability to solve this task (Bai and Bertenthal, 1992). This is thought to relate to the transition from body-centered spatial coding, to the ability to track landmarks and objects independent of the infant's own (changing) location once crawling has begun.

The relationship between motor ability and spatial cognition is not just limited to gross motor abilities. Soska et al. (2010), for example, demonstrate an association between visual-manual exploration skills and 3D object perception. The authors found that, in 4.5- to 7.5-month-old infants, the motor skills that are required to change the viewpoint of an object (rotating, fingering, and transferring objects between hands while simultaneously looking at them) were predictive of their ability to determine the spatial properties of 3D objects accurately when viewed from a single viewpoint. This demonstrates that the development of visual-manual skills facilitates the generation of knowledge surrounding object properties.

Beyond infancy, little is known about the relationship between motor skills, motor experience, and spatial knowledge. Longitudinal evidence has demonstrated that age of walking, as well as exploration through locomotion at 20 months, are both related to performance on the Block Design task of the Wechsler scales (Wechsler, 1999), a measure of spatial cognition, at 32 months (Oudgenoeg-Paz et al., 2015). Mental rotation performance has also been associated with motor competence in 5- to 6-year-olds (Jansen and Heil, 2010). It has also been reported that motor proficiency in childhood is related to extent of physical activity in adolescence (Barnett et al., 2009), and that the development of the strategies required for successful navigation of space is related to cumulative experience of physical exploration of the environment (Cornell et al., 2001). Thus, it seems likely that there is a developmental association between motor abilities and spatial ability beyond that observed in infancy.

One avenue for further investigating the relationship between motor function and spatial cognition is to explore the impact that impaired motor abilities have on spatial cognition. Evidence to-date is sparse, but has demonstrated that adolescents with physical disability show impaired spatial knowledge of their environment (Wiedenbauer and Jansen-Osmann, 2006) and that the extent of this spatial deficit is predicted by their mobility in infancy (concurrent motor ability was not measured) (Stanton et al., 2002). Furthermore, physical activity in children with developmental coordination disorder (DCD) is related to the extent of their motor impairment (Rivilis et al., 2011). Finally, Belmonti et al. (2015) report impaired spatial memory on a table-top task and a large-scale navigation task in children with motor deficits as a result of cerebral palsy. In summary, it is likely that children with motor impairments show delayed exploration of space in infancy, are less physically active, and do not explore their environment as actively as those without motor impairment, and that this has negative consequences for the development of spatial knowledge, in particular, large-scale spatial navigation ability.

Individuals with attention deficit hyperactivity disorder (ADHD), the most prevalent neurodevelopmental disorder during childhood (occurrence: 3–6%) (Polanczyk et al., 2007), present with primary characteristics of hyperactivity, impulsivity, and inattention (American Psychiatric Association [APA], 2013). ADHD is more prevalent in males than females, with a prevalence ratio estimated as between 1:3 and 1:16 females: males (Novik et al., 2006). In addition, a co-occurring motor impairment is evident in children with ADHD, with ∼50% meeting criteria for DCD (Brossard-Racine et al., 2011; Goulardins et al., 2013). There is mixed evidence that motor impairments in ADHD are related to severity of ADHD symptomatology (Kroes et al., 2002; Kopp et al., 2009; Farran et al., submitted). Despite this, it has been shown that the presence of motor deficits in ADHD contributes to poor psychosocial outcome in adults (Rasmussen and Gillberg, 2000). We do not know, however, whether motor deficits are associated with spatial cognition in this group.

Given the association in the typical population between motor competence and spatial cognition in infancy, here we investigate whether this association is evident in TD children into childhood. The studies with infants predominantly investigated large-scale spatial knowledge; this is one reason why we have chosen a large-scale spatial navigation task as the spatial measure for this study. Because little is known about the relationship between motor ability and large-scale spatial knowledge in the typical population beyond infancy, this will add to the body of knowledge surrounding typical development. We will also determine whether the same association between motor ability and spatial ability leads to impaired spatial cognition in those children with ADHD who present with a motor impairment. Therefore, we will explore the developmental relationship between early motor milestones and current motor abilities in children with ADHD, on spatial navigation. To-date spatial navigation has not been investigated in ADHD, and given that poor motor ability in individuals with physical disability is a limiting factor to the development of large-scale spatial knowledge (Stanton et al., 2002), this is our second reason for choosing a spatial navigation task as our measure of spatial ability.

In addition to a comparison of performance in our ADHD sample to that of typically developing (TD) children, we will also compare their performance to the performance of individuals with Williams syndrome (WS). Comparison between ADHD and TD children will determine whether the patterns of performance in the ADHD group are indicative of typical or atypical performance. By using cross-syndrome comparison with WS, we will also be able to differentiate between patterns of performance that are syndrome-specific to ADHD vs. a universal consequence of the presence of a motor deficit. WS is a rare genetic disorder, with an occurrence of 1 in 7,500 to 1 in 20,000, which occurs equally in males and females (Morris and Mervis, 1999; Strømme et al., 2002). Individuals with WS have mild to moderate learning difficulties and an IQ of ∼60 (see Farran and Karmiloff-Smith, 2012). Crucially, we chose WS as our comparison group because it shares deficits with ADHD in attention and motor skill. That is, with reference to attention, Rhodes et al. (2011) report that all of their 19 participants with WS met the criteria for ADHD on the Conners' Parent Rating Scale (CPRS; Conners et al., 1998). There is also consistent evidence for impaired motor ability in WS. This has been demonstrated with respect to: delayed motor milestones (Carrasco et al., 2005); impairments on standardized motor tasks (Tsai et al., 2008; Atkinson, 2017; Wuang and Tsai, 2017); atypical reaching movements, walking and stair decent (Elliott et al., 2006; Hocking et al., 2010; Cowie et al., 2012). Furthermore, impaired spatial cognition is a hallmark deficit of WS (Farran and Formby, 2012). With reference to large-scale spatial knowledge, impairments are consistently demonstrated in WS (e.g., Farran et al., 2010, 2015; Purser et al., 2015), but the contribution of motor impairments to this deficit is currently unknown. Using WS, we will determine whether different motor deficits (ADHD vs. WS) lead to different patterns of navigation ability and whether specific motor deficits (e.g., fine vs. gross motor) are more detrimental to navigation than others.

In the real world, it is difficult to dissociate the motor and non-motor demands of navigation; concurrent demands of locomotion (e.g., proprioceptive, vestibular demands) cause slow/disrupted movement and disturb effective navigation, making it difficult to uniquely measure spatial knowledge. Here, we will use desktop virtual reality; this neutralizes the inputs from the gross motor system, allowing a purer measure of the spatial aspects of navigation performance, while also maintaining ecological validity. Evidence has also shown that performance in virtual and real-world navigation tasks tap into the same cognitive mechanisms and that learning in a virtual environment (VE) transfers to the real world (Richardson et al., 1999; Coutrot et al., 2019).

The ability to navigate develops through three stages. First, an individual recognizes landmarks within an environment (landmark knowledge). This is followed by knowledge of the relationship between landmarks and turns of a specific route (route knowledge). Finally, configural information of the spatial relationship between landmarks and places within the environment is encoded (configural knowledge or a cognitive map) (Siegel and White, 1975). Note, however, that while these three stages are distinct, it is now considered that they do not necessarily follow a sequential pattern of emergence (see Montello, 1998). Individuals with WS are able to gain both landmark knowledge and route knowledge, but rarely encode a cognitive map of an environment (Farran et al., 2015). This limits their ability to deviate from a fixed learnt route, and thus has an impact on their ability to make short cuts or to reorient when lost.

An associated consequence of less sophisticated navigation skills is a strong reliance on landmarks for effective wayfinding. This is true of individuals with WS, but also young TD children, and thus appears to be a characteristic of immature navigation abilities (Farran et al., 2012, 2016; Lingwood et al., 2015; Purser et al., 2015). Landmarks are objects in the environment that are salient, either perceptually or on account of contextual information (Caduff and Timpf, 2008), and are an important aspect of spatial cognition. For example, in the classic reorientation task, 2-year-olds use landmark information to develop a geometric understanding of a rectangular environment (Learmonth et al., 2001), and we have already discussed the use of landmarks to crawl to a (hidden) target location in infants (Clearfield, 2004).

The ability to select useful landmarks is advantageous during spatial navigation. Landmarks at junctions are more useful than landmarks that are not near a decision point (Farran et al., 2012). Furthermore, proximal landmarks are more useful for developing route knowledge while distant landmarks are more useful for encoding configural information of the environment (Purser et al., 2015). TD children aged from 6 years, and individuals with WS, show stronger recall of landmarks at junctions than landmarks on path segments (Farran et al., 2012). This suggests that both TD children and individuals with WS recognize the usefulness of landmarks at decision points, and support the evidence for a reliance on landmarks for effective route learning. Here we will measure performance on the first two stages of navigation, landmark knowledge and route knowledge. Participants will be asked to learn a fixed route through a novel VE. We will measure the number of errors made while learning the route. Given the importance of landmarks to spatial cognition, and to determine whether participants rely on landmarks to navigate, we will also measure recall of landmarks along the learnt route. These will be divided into landmarks that featured at junctions and landmarks that did not feature at junctions, as an index of the ability to determine landmark usefulness. Alongside navigation performance, we will measure motor skills using a standardized battery of motor ability. In addition, for the atypical groups, we will also obtain parent reports of motor milestone achievement. Given the relationship between environmental factors, such as independent exploration, with motor ability and large-scale spatial knowledge, respectively, we will also measure this environmental factor in our groups.

This is the first study to determine whether the known association between motor and large-scale spatial ability in infancy (see Newcombe, 2019 for a review) extends to childhood and to atypical groups. If motor competence is related to spatial ability, we predict an association between motor ability and spatial ability across all participant groups. Furthermore, we predict that those individuals with a motor deficit (the WS group and a large number of the ADHD group) will show impaired spatial navigation abilities. Of significance, this study will broaden our understanding of the crucial processes that underlay the development of large-scale spatial navigation, with downstream implications for interventions designed to improve navigation performance.

MATERIALS AND METHODS

Participants

Fifty-one children with ADHD (regardless of ADHD subtype) aged 8–15 years were recruited into the study via parent support groups and social media. Three children with ADHD were excluded due to having a co-occurring diagnosis of a neurological condition (partial fetal alcohol syndrome, Tourette's syndrome, or microcephaly), all of which are associated with problems with movement which could have affected the results. A further two children with ADHD were excluded due to being on medication at the time of testing, which could have positively impacted their motor performance (Kaiser et al., 2015), while three further children with ADHD were excluded because they fell at or below the fifth percentile on our two IQ measures [British Picture Vocabulary Scale III (BPVS), Dunn et al., 2009; Matrices subtest of the British Ability Scales III (BAS), Elliot and Smith, 2011]. One child with ADHD had a co-occurring diagnosis of DCD. This child was not excluded from the analyses. A further 11 children with ADHD with diagnoses of one or more co-occurring disorders were not excluded because ADHD was their primary diagnosis and including these individuals provided a realistic representation of the ADHD population. Furthermore, recent research suggests that ADHD might share common early developmental pathways with other disorders, including autism (see Johnson et al., 2015), and excluding participants with co-occurring disorders would ignore this convergence. These included sensory processing disorder ($N = 2$), pervasive developmental disorder ($N = 1$), dyslexia ($N = 5$), autism ($N = 3$), Asperger's ($N = 1$), oppositional defiance disorder ($N = 2$), social communication disorder ($N = 1$), and obsessive compulsive disorder ($N = 1$). The final sample consisted of 43 children with ADHD, all of whom had a formal diagnosis of ADHD from a clinician, were medication naïve for at least 24 h prior to testing, had an IQ within the normal range, and received an ADHD index score (Conners' Parent Rating Scale – Revised Long version; CPRS-R:L; Conners, 1997) which supported their diagnosis of ADHD ($\geq$60).

Twenty participants with WS aged 12–50 years participated in the study. This broad age range is not unusual for this kind of study; this is due to the practical nature of recruiting participants with such a rare disorder, but also because the areas of deficit measured in this study are likely to have plateaued by 12 years (e.g., Farran and Formby, 2012), and thus any within group differences can be accounted for by individual differences rather than developmental factors. All participants with WS had been diagnosed based on phenotypic and genetic information. Genetic diagnosis was based on a fluorescent *in situ* hybridization (FISH) test (see Lenhoff et al., 1997). WS participants were recruited from the records of the Williams Syndrome Foundation, United Kingdom. CPRS-R:L (Conners, 1997) data were also collected for this group to provide an index of whether they displayed ADHD characteristics. Six parents/carers did not complete the questionnaire (**Table 1**); of the remaining 14 participants, nine received a CPRS-R:L ADHD-index score within the clinical range ($\geq$60) for ADHD.

Seventy-two TD children aged 5–11 years participated in the study. The TD sample was recruited from primary schools in the United Kingdom. The age range of the TD children was chosen based on the predicted range of abilities of the neurodevelopmental disorder groups on the motor battery [Bruininks-Oseretsky Test of Motor Proficiency Second Edition Short Form (BOT2-SF)]. One TD child scored below the fifth percentile on the two IQ measures and was excluded from the group, leaving a final TD sample of 71 children. All participants had normal or corrected to normal vision. Participant information is given in **Table 1**.

Design and Procedure

Ethical approval was obtained from the UCL Institute of Education Research Ethics Committee (approval number: REC 766; study title: Motor development and navigation in Attention Deficit Hyperactivity Disorder). Following written informed parental consent, the participants were tested individually either at their school, in the research lab, or at the participant's home. The order of tests was randomized for each participant, and the entire session lasted between 1 h 15 min and 2 h. The battery of tasks included those listed below in addition to two other tasks reported elsewhere (Farran et al., submitted).

Background Tasks

All participants completed the Matrices subtest of the BAS (Elliot and Smith, 2011), and the BPVS (Dunn et al., 2009) as measures of IQ. Standard scores for these tests are presented in **Table 1**. Standard scores for the BPVS III have a mean of 100 and a standard deviation of 15, while standard scores for the BAS III have a mean of 50 and a standard deviation of 10. In addition, parents/carers of the atypical groups completed the Long Form of the CPRS-R:L in order to derive ADHD index scores. Scores on subscales that are one standard deviation above the mean of 50 (i.e., scores of 60 or above) are considered to be in the clinical range. The test–retest reliability for the ADHD index is 0.72.

Motor Task: Bruininks-Oseretsky Test of Motor Proficiency Second Edition Short Form (Bruininks and Bruininks, 2005)

The BOT2-SF is a measure motor competence for individuals from 4 to 21 years. Raw composite scores and standard scores for this test are presented in **Table 1**; standard scores have a mean of 50 and a standard deviation of 10. The composite score is the sum of performance on eight subtests (comprised from 14 items). The fine motor control subtests are: Fine Motor Precision, Fine Motor Integration, and Manual Dexterity. The gross motor control subtests are: Bilateral Coordination, Balance, Running

TABLE 1 | Participant details.

	TD (N = 71)		WS (N = 20)		ADHD (N = 43)	
	M (SD)	Range	M (SD)	Range	M (SD)	Range
Chronological age (years)	8.410 (1.748)	5.020–11.460	27.619 (8.817)	12.860–50.670	11.403 (1.892)	8.010–15.600
Gender (m/f)	38/33 (53% male)		7/13 (32% male)		35/8 (81% male)	
BPVS-III standard score	103.282 (12.726)	70–128	77.000 (10.079)	70–107	98.302 (11.911)	81–123
BAS-III T-score	49.648 (11.899)	21–79	20.200 (0.696)	20–23	45.067 (12.876)	20–74
BOT2-SF standard score	57.320 (7.487)	41–70	28.500 (4.407)	20–37	43.020 (8.251)	28–65
BOT2-SF raw score	68.450 (9.202)	44–82	43.600 (12.796)	16–69	65.530 (10.110)	38–80
CPRS-R:L ADHD index	NA	NA	67.929 (15.598) (N = 14)	47–89	77.814 (7.863)	61–90

Note: The range of BOT2-SF raw scores of the TD group broadly covers the range of BOT2-SF raw scores of the atypical groups. This is with the exception of one participant with WS who had a BOT2-SF raw score of 16. This person was excluded for developmental trajectory analyses.

Speed and Agility, Upper Limb Coordination, and Strength. In addition to a Fine Motor and a Gross Motor score, a combined Motor Composite score can also be derived. The Short Form range has good test–retest reliability (0.80–0.87) and interrater reliability (0.98).

Motor Milestones Questionnaire

A parental questionnaire (developed by Sumner et al., 2016, which was based on Brouwer et al., 2006) was used to investigate the extent to which the children with ADHD and individuals with WS reached motor milestones. Parents were asked to give the age (in months) that 12 significant milestones were reached, six of which have been standardized against World Health Organization (WHO) data (WHO Multicentre Growth Reference Study Group, 2006). The data from these six milestones only are reported here (for details of the results of the full questionnaire, see Farran et al., submitted).

Environmental Measure: Independent Exploration (Based on Shaw et al., 2015)

A short questionnaire was read aloud to the individual and on occasion their parent to obtain information of experience of exploration. Participants were asked questions regarding the extent to which they were allowed to explore environments with others or by themselves. For example, they were asked about how and with whom they got to and from school and if they independently went to local shops or the park. They were also asked about the frequency of these behaviors. A composite exploration score was determined based on the sum of five independent activities and the frequency at which these activities took place in a typical week (max total score: 31). A binomial score was also calculated from this which determined whether the participant was permitted to explore independently (composite score $\geq$ 1 = binomial score 1) or not (score = 0).

Large-Scale Spatial Navigation

Two VEs were created using Vizard[1] and presented on a 17-in laptop computer. The VEs displayed mazes with either six or eight junctions/decision points. Each junction led to two paths, one correct and one incorrect. Incorrect path choices ended in a cul-de-sac, which had the same appearance as a T-junction when viewed from the preceding junction. Mazes were lined with brick walls and landmarks (objects) were placed on both correct and incorrect sections of the route (**Figure 1**). Landmarks were selected from a range of categories (e.g., animals, tools, furniture) for their high verbal frequency (Morrison et al., 1997) and for being easy to recognize. Landmarks within the maze were equally distributed to the left and right of the path. At the end of the maze was a gray duck, which once approached, ended the game.

The six-junction route had been previously used by Farran et al. (2012). It had 16 unique landmarks. Eight of the landmarks were near to junctions ("junction landmarks"). Eight of the landmarks were not near to junctions ("path landmarks"). Across these junctions, there were two left, two right, and two straight-ahead choices that led to the next correct path segment. A map of the maze layout is shown in **Figure 1**. The eight-junction route was created for this study. It had 20 unique landmarks; 10-junction landmarks and 10-path landmarks. Across the junctions, there were three left, three right, and two straight-ahead choices that led to the next correct path segment.

Corridor Task

Preceding the experimental mazes, participants were given the opportunity to practice navigating along a simple corridor which did not include decision points or landmark objects, but included two turns. Participants were instructed on how to navigate the VEs by using the four arrow keys on the keyboard. They then watched the experimenter navigate the corridor, before navigating it themselves. This involved simply following the path, which included two right-angle turns; there were no decisions to be made. If participants had difficulty controlling their navigation, they were given another walk of the corridor. No participants required more than two walks of the corridor.

Route Learning Task (Six-Junction Route)

The experimenter showed the participant the correct route through a six-junction maze. The experimenter instructed the participant to "Pay close attention to the route and to the objects that appear in the 'maze game' because you will have to go exactly the same way through the maze after I have shown you." After the experimenter had demonstrated the correct route, the

[1] http://www.worldviz.com

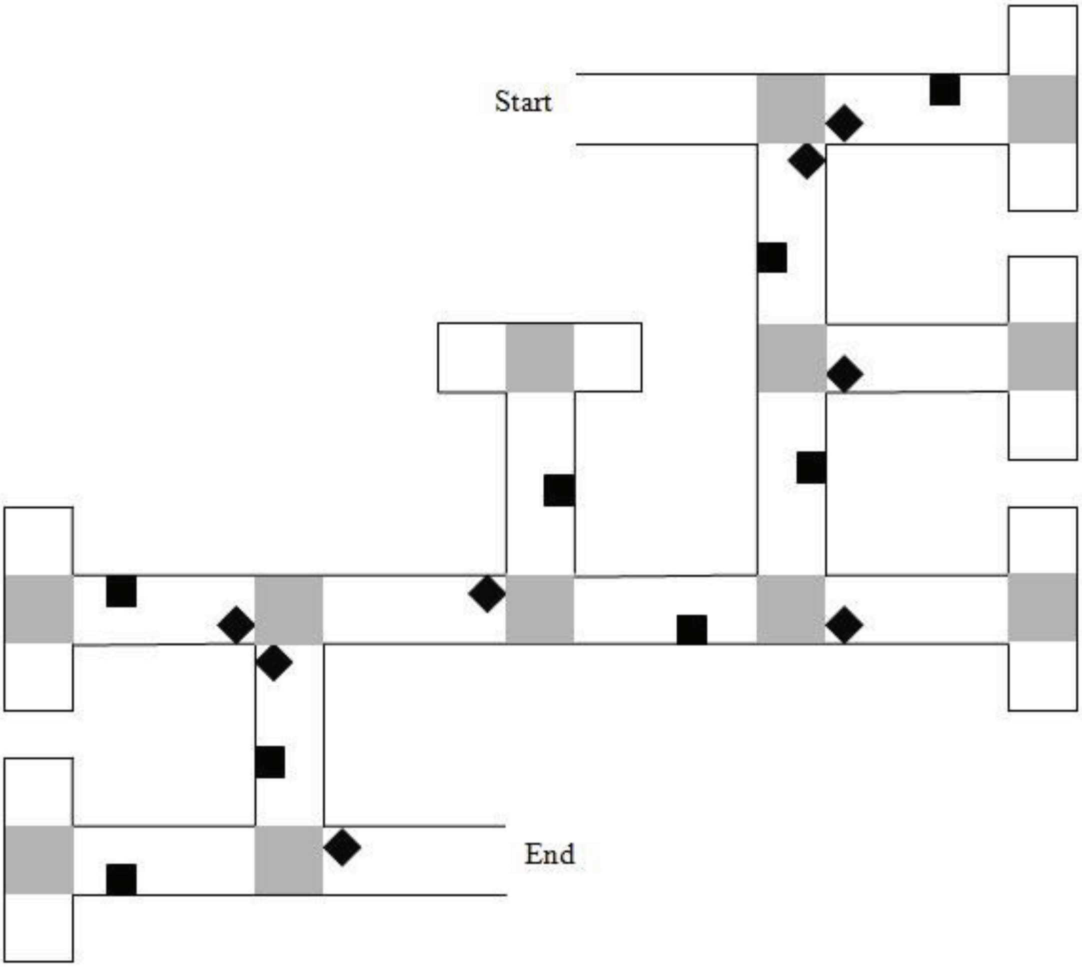

FIGURE 1 | Map of the six-turn maze layout. Gray squares represent "pebble" texture that was featured at junctions and at the end of cul-de-sacs. Black diamonds indicate junction landmarks. Black squares indicate path landmarks. Reprinted from Farran et al. (2012), with permission from Elsevier.

participants attempted to walk the correct route from start to finish using the arrow keys. If an incorrect path was selected, participants reached a cul-de-sac and were able to self-correct by turning around. Encouragement was given, but no help. If a participant turned toward the start of the maze, they were directed back to the junction where they made the error. Each trial terminated on reaching the gray duck and completing the route. Each walk through the maze from start to finish was labeled as a learning trial. Participants completed learning trials to a criterion of completing the maze from start to finish without error on two consecutive trials, or until they had completed 10 learning trials. The cumulative number of errors across learning trials was recorded. An error was defined as a deliberate incursion down an incorrect path; if the participant corrected his/her course before reaching half-way down an incorrect path section, no error was counted.

Landmark Recall Task

After the participant had learnt the six-junction route to criteria, the landmark recall task commenced. The experimenter showed the participant the same maze but with all landmark objects shown as red balls (**Figure 2**). The experimenter navigated the route themselves and stopped at each junction to point out each red ball in the subsequent path section. Participants were asked what object the ball had been when they were walking around the maze. After an answer was given, the participants were then shown an image of the landmark object in its correct location, on another computer screen. This was conducted for all landmark objects that were visible from the correct path (eight landmarks on the correct path in addition to four landmarks that featured on incorrect path sections that could be viewed straight ahead before a correct turn to the left or right was executed). Landmark recall score was calculated as the number of correctly identified junction and path landmarks that featured at junctions (Max. = 6 for each landmark category, junction and path landmarks).

Naming Task

A naming task was administered after the landmark recall task, to ensure that the verbal labels used by the participants in the

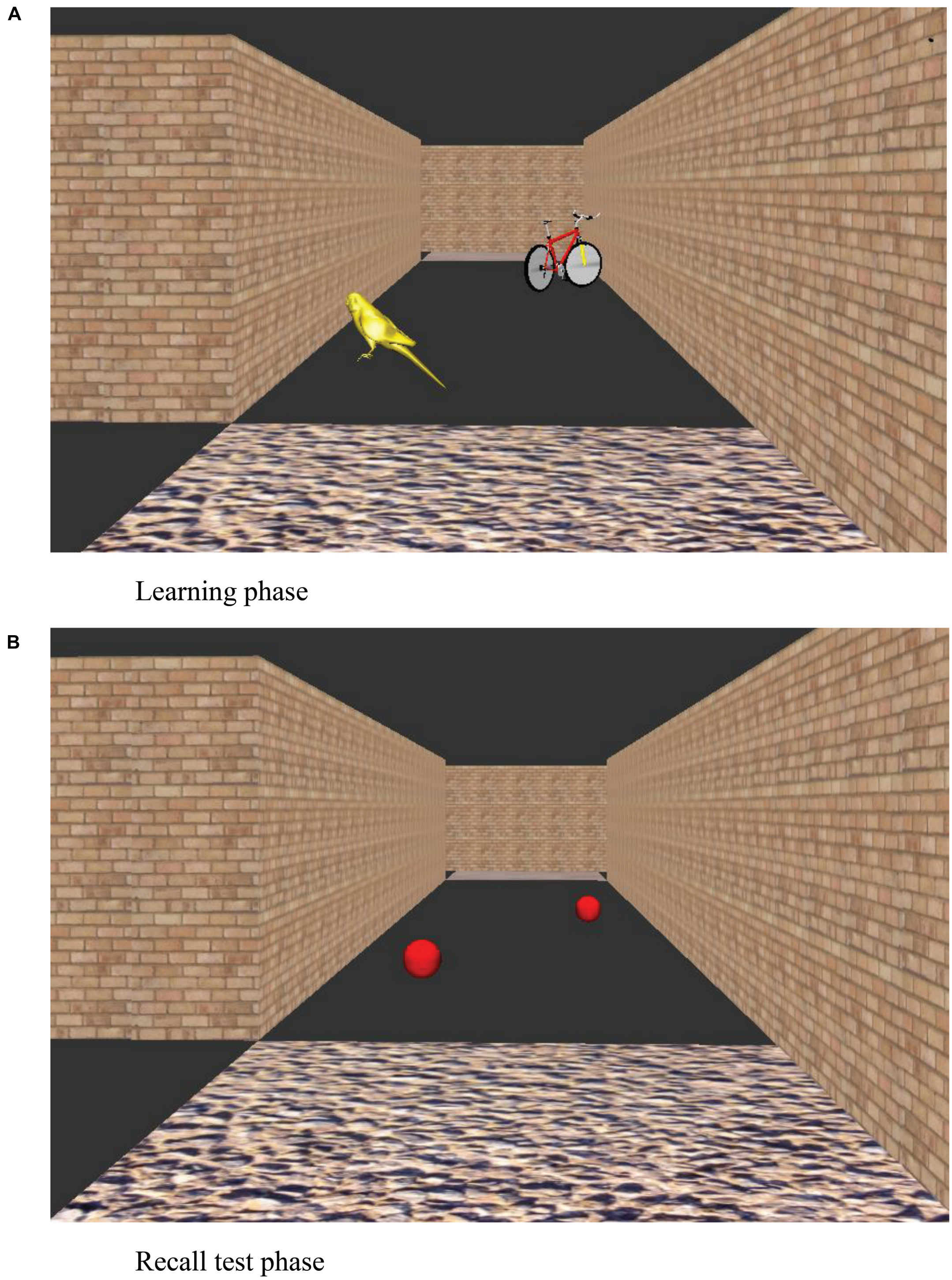

FIGURE 2 | View of a virtual environment during learning trials **(A)** and at the recall test phase **(B)**.

landmark recall task could be coded accurately (e.g., a participant might use the word "light" for "streetlamp"). Participants were shown images of each of the 16 landmarks in a pseudo-random order and were asked to name them. Participants received a score out of 16 on the naming task.

Route Learning Task (Eight-Junction Route)

Following the six-junction maze, in order to ensure a wide range of variability in route knowledge performance, participants were shown a longer eight-junction route in a different VE, and asked to walk the route themselves using the same procedure as used for the six-junction maze. For this route, participants simply completed three trials and the cumulative number of errors was recorded. No landmark recall task or naming task was completed. To determine overall maze error score, the cumulative number of errors across all learning trials in the six-junction maze and the three learning trials in the eight-junction maze was calculated for each participant.

RESULTS

Note that there were 11 children with ADHD who had co-occurring diagnoses that we had no reason to believe would impact the pattern of results. To exercise caution, all analyses listed below were run a second time, with these 11 children excluded. This only changed one result with respect to motor milestone data (noted in the manuscript). As such, results are presented with these children included in the analyses.

We are specifically interested in whether an impairment in motor competence has an impact on large-scale spatial knowledge. Given that large-scale spatial knowledge is associated with independent exploration (Cornell et al., 2001), we are also interested in whether poorer motor ability is associated with reduced independent exploration, and in turn, large-scale spatial knowledge. To this end, we have chosen two groups who are known to have motor impairments, ADHD and WS, the latter of which also presents with spatial deficits. The TD group was chosen to span the range of motor abilities of the ADHD and WS group in order that developmental trajectory analysis could be carried out on large-scale spatial performance, with motor ability as a measure of motor "mental age" (Thomas et al., 2009). Before detailing performance on the spatial navigation task, we first present the motor and independent exploration demographics of each group. Each participant completed the BOT2-SF and the independent exploration questionnaire, while the parents of the ADHD and WS samples also completed a motor milestone questionnaire.

Motor Performance: BOT2-SF

The BOT-SF is a standardized measure with standardized scores classified as falling within a number of zones. The TD sample fell within the "average" (N = 43), "above average" (N = 25), and "well above average" (N = 3) zones, indicative of no motor impairment. The ADHD group fell within the "well below average" (N = 2), "below average" (N = 18), "average" (N = 22), and "above average" (N = 1) zones; this indicates that 20 of the 43 participants with ADHD presented with a motor impairment ($\leq$16th percentile). The WS group fell within the "below average" (N = 7) and "well below average" (N = 13) zones and thus all presented with a motor impairment.

Motor Milestones

Motor milestone data were collected for ADHD and WS groups only. Data are presented for six motor milestones, and compared to percentiles based on WHO data (WHO Multicentre Growth Reference Study Group, 2006) in **Table 2**. Note that due missing data, the data presented are based on reduced Ns (Ns for each milestone for each group are given in **Table 2**).

Contrary to the findings from the BOT2-SF above, overall the ADHD group achieved motor milestones broadly within the typical range of achievement, with a slightly wider range of achievement than for the typical population. The WS group achieved all six motor milestones later than would be expected for a TD child (although note that the range of month of achievement for the WS group overlaps with the typical range). Motor milestone achievement was not related to concurrent motor ability (BOT2 Overall score) for either group [$p > 0.008$ for all (Bonferroni corrected alpha)]. Although note that when the 11 ADHD participants with comorbid conditions were excluded, age of independent walking correlated with both BOT2-SF fine motor score ($p = 0.007$) and the residuals (age partialled out) ($p = 0.002$) of this measure for this group.

Independent Exploration

An exploration score was not available for one TD participant (and thus N = 70 for the TD group). Descriptive statistics are presented in **Table 3**. Exploration score was related to age for the TD group [Spearman's rho (70) = 0.635, $p < 0.001$] and thus the TD group could be used as a method of standardization for the atypical groups. To do this, the TD group was split into three age groups [TD 5–6 years (N = 20), TD 7–8 years (N = 20), TD

TABLE 2 | Motor milestone month of achievement for the WS, ADHD-L, and ADHD-H groups compared to typical month of achievement.

	WHO Age in months at which milestone achieved		WS				ADHD			
	M (SD)	Range	*N*	*M*(SD)	P'tile	Range	*N*	*M*(SD)	P'tile	Range
Sit without support	6.0 (1.1)	3.8–9.2	9	12.222 (5.911)	>99th	3–24	30	6.100 (1.589)	50th	3–10
Crawl hands and knee	8.5 (1.7)	5.2–11.4	6	15.500 (5.612)	>99th	5–21	28	8.607 (2.254)	50th	3–13
Stand with assistance	7.6 (1.4)	4.8–11.4	7	16.286 (6.130)	>99th	10–24	31	9.000 (2.758)	90th	3–18
Stand without support	11.0 (1.9)	6.9–16.9	5	24.600 (9.370)	>99th	12–36	33	11.136 (2.356)	50th	7–19
Walk with assistance	9.2 (1.5)	6–13.7	6	21.167 (9.131)	>99th	12–36	33	11.288 (2.414)	90th	6–19
Walk without support	12.1 (1.8)	8.2–17.6	13	24.615 (8.921)	>99th	15–42	36	13.125 (2.831)	75th	9–24

WHO = World Health Organization. WHO Milestones data from the WHO Multicentre Growth Reference Study Group (2006). Note: Ns differ across cells due to missing data.

9–11 years ($N = 30$)] for comparison with the atypical groups. Note that the TD 9–11-year-old group also did not differ from the ADHD group for chronological age and thus represented an age-matched comparison group (ADHD: $p = 0.530$). The data for the TD 5–6-year-olds, the TD 7–8-year-olds, and the WS group were not normally distributed due to a large number of zero scores (Kolmogorov–Smirnoff test, $p < 0.05$ for all). As such, a Friedman ANOVA was carried out with group as a between participants factor. This demonstrated a main effect of group, $\chi^2(4) = 35.732$, $p < 0.001$. Mann–Whitney-U paired comparisons demonstrated that the WS group explored to a greater extent than the TD 5–6-year-olds ($p < 0.001$), but at a similar level to the 7–8-year-olds ($p = 0.284$) and 9–10-year-olds ($p = 0.270$). The ADHD group was exploring more than the TD 5–6-year-olds ($p < 0.001$) and the TD 7–8-year-olds ($p = 0.002$), but at the same level as the 9–11-year-olds ($p = 0.723$). Thus, the ADHD groups were exploring at the level appropriate for their chronological age, while the WS groups were exploring at the level of a 7–11-year-old child, even though they were adults.

The relationship between exploration score and BOT2-SF overall motor score was determined using Spearman correlations. Because age was related to exploration score for the TD and ADHD group ($p < 0.05$ for both), the residuals of exploration score (age partialled out) were also used for these two groups to determine the relationship after accounting for age-related variance. This demonstrated a relationship between exploration score and motor ability for the TD and ADHD groups only (TD: $p < 0.001$; ADHD: $p = 0.014$; WS: $p = 0.112$), which was accounted for by variance in age (TD: $p = 0.131$; ADHD: $p = 0.223$).

Due to the large number of zero scores, a binomial score was also calculated which determined whether the participant was permitted to explore independently or not. The percentage of participants who received a score of 1 (i.e., they were permitted to explore independently) is also shown in **Table 3**.

Spatial Navigation

Two primary dependent variables were derived from the navigation task, maze error score and landmark recall. Maze error score is a measure of an individual's ability to learn a route, i.e., route knowledge. Landmark recall score provides information about strategy use when learning the route, i.e., did participants use landmarks as an aid to learning the route, and were landmarks at junctions considered strategically more useful than landmarks on paths?

Spatial navigation was analyzed with respect to variation in motor competence across our participants using developmental trajectory analysis. Developmental trajectory analysis is used to ascertain whether the trajectory of performance across the range of mental ages (in this case motor mental age) of each group differs in: mean value; intercept; or slope (rate of development). To determine which measures of motor ability were most suitable as a measure of motor "mental age," correlational analyses were carried out for each group between the two spatial measures, maze errors and landmark recall (for all 12 landmarks) and five motor measures [BOT-2 gross motor score, BOT2 fine motor score, walking unsupported (atypical groups only), hands and knees crawling (atypical groups only), exploration score] (**Table 4**). On account of significant input from chronological age to BOT-2-SF gross and fine motor scores and exploration scores for the TD and ADHD groups ($p < 0.05$ for all), correlations were also included for the residuals of these three measures for these two groups (age partialled out). This constituted up to 16 correlations per atypical group and 12 correlations for the TD group, thus we used Bonferroni corrected critical alphas (atypical groups: $p \leq 0.003$; typical group: $p \leq 0.004$). Due to the very small sample size for crawling for the WS group ($N = 6$), these correlations would not be informative and so are not reported.

Associations Between Spatial Navigation and Motor Performance

None of the motor scores or exploration score correlated with *landmark recall* for any of the groups ($p > 0.003$ for all). Despite medium effect sizes for the BOT2-SF measures for the ADHD group (**Table 4**), there were no (Bonferroni corrected) significant correlations with *maze error* for the ADHD and WS groups ($p > 0.003$ for all). Maze error correlated with BOT2-SF fine motor scores for the TD group ($p \leq 0.004$; Gross motor score: $p = 0.005$). Correlations with the residuals demonstrated that any association between BOT2-SF gross motor score and maze error in the TD group was mediated by age, $r(71) = -0.179$, $p = 0.135$. This was not the case for BOT2-SF Fine motor score, $r(71) = -0.395$, $p = 0.001$.

Maze Error Score

As shown in **Table 4**, BOT2-SF fine motor ability demonstrated a small ($r = 0.10$) to medium ($r = 0.30$) effect size (Cohen, 1992) for all groups for maze error score, albeit only to (Bonferroni corrected) significance for the TD group. As such, BOT2-SF fine motor ability was deemed the best measure of "mental age" for developmental trajectory analysis (Thomas et al., 2009). Developmental trajectory analysis can be influenced by outliers, thus we used an exclusion criteria of maze error scores that were three standard deviations above the group mean. One participant in the ADHD-L group met this exclusion criteria only [this changed the correlation reported in **Table 4** to $r(42) = -0.164$].

TABLE 3 | Exploration scores for each participant group.

	TD 5–6 (*N* = 20)		TD 7–8 (*N* = 20)		TD 9–11 (*N* = 30)		WS (*N* = 20)		ADHD (*N* = 43)	
	Median	Range	Median	Range	Median	Range	Median	Range	Median	Range
Exploration score (Max:31)	0	0–9	3	0–21	6.5	0–21	3.5	0–22	7.0	0–23
% Permitted to explore	10.0%		57.1%		90.0%		80.0%		86.0%	

TABLE 4 | Bivariate correlations with maze error and landmark recall.

Group		BOT-2 raw motor score				Motor milestones		Exploration	
		Raw		Residuals (age partialled out)				Raw	Residuals (age partialled out)
		Gross	Fine	Gross	Fine	Crawling	Walking		
TD	Maze error	−0.332	−0.481*	−0.172	−0.395	NA	NA	−0.034(N = 70)	0.145(N = 70)
	Landmark recall	−0.061	−0.027	−0.096	−0.029			−0.095	−0.106
WS	Maze error	0.118	−0.163	NA	NA	NA	−0.203 (N = 13)	−0.157	NA
	Landmark recall	−0.063	0.399				−0.054 (N = 13)	0.184	
ADHD	Maze error	−0.308	−0.364	−0.186	−0.192	0.027	0.079	0.169	−0.359
	Landmark recall	0.123	0.151	0.075	0.097	−0.110	0.027	0.131	0.181

*$p \le 0.004$ (TD critical alpha); No correlations met the disorder group critical alphas of $p \le 0.003$. Note: Ns are reported where the data was not available for the full sample.

In order for the ranges of the covariates to be largely overlapping, one WS participant who only achieved a fine motor score of 1 was excluded [this changed the correlation reported in **Table 4** to $r(19) = -0.252$]. In order that any differences in intercepts were meaningful, BOT2-SF fine motor score was rescaled such that the intercept was at the lowest BOT2-SF fine motor score of the participants. This does not change the analysis, but enables meaningful interpretation of the intercept.

Initial ANOVA of group means revealed that maze error differed across groups, $F(2,129) = 17.288$, $p < 0.001$, $\eta_p^2 = 0.211$. Tukey paired comparison demonstrated that this was due to higher maze error score in the WS group relative to all other groups ($p < 0.001$ for both), with no differences across the remaining groups ($p > 0.05$). ANCOVA with BOT2 fine motor as a covariate demonstrated a significant impact of BOT2 fine motor score [$F(1,126) = 10.541$, $p = 0.001$, $\eta_p^2 = 0.079$]. There was also a significant group difference in the intercept of maze error scores [$F(2,126) = 4.304$, $p = 0.016$, $\eta_p^2 = 0.064$], such that at the lowest motor ability, the ADHD group had lower maze error scores than the TD and WS groups (ADHD and WS: $p = 0.036$; ADHD and TD: $p = 0.003$; TD and WS: $p = 0.281$). Note that this difference in intercept remained when BOT2-SF fine motor score was replaced by the residuals (age partialled out) of this variable ($p = 0.024$). The slope of the relationship between motor ability and maze error score did not differ across groups [$F(2,126) = 1.85166$, $p = 0.159$, $\eta_p^2 = 0.029$].

Naming Score

Participant's naming scores were sufficiently high that we could be confident that all participants were able to provide verbal labels for the landmarks [mean (SD) out of 16: TD: 15.521 (0.790); WS: 15.200 (1.001); ADHD: 15.628 (0.757)], thus enabling accurate scoring of landmark recall. Naming score was consistent across groups, $F(2,131) = 1.904$, $p = 0.153$, $\eta_p^2 = 0.028$. Where participants named the item inaccurately (e.g., "jelly" for "cake," or "bat" for "tennis racket"), we accepted this answer in the landmark recall task as accurate.

Landmark Recall

As observed in **Table 4**, effect sizes were often below the cut-off for a small effect, which indicates that motor ability was not related to landmark recall. As such, it was not possible to carry out developmental trajectory analysis. Landmark recall was also not related to chronological age ($p > 0.05$ for all groups). Consequently, landmark recall was analyzed using ANOVA with a between-participant factor of Group (TD, ADHD, WS) and a repeated measures factor of landmark type (junction landmarks, path landmarks). There was a main effect of group, $F(2,131) = 3.413$, $p = 0.036$, $\eta_p^2 = 0.050$, due to poorer landmark recall in the WS group, compared to the TD group ($p = 0.043$) only (all other p's > 0.05). The effect of landmark type enables us to draw conclusions about strategy use in each group. As shown in **Figure 3**, there was a main effect of landmark type, $F(1,131) = 39.300$, $p < 0.001$, $\eta_p^2 = 0.231$, due to weaker recall of path landmarks than junction landmarks. This effect did not interact with group, $F < 1$, which indicates consistent use of landmarks to learn the route across all groups. To further determine whether the use of a landmark strategy was associated with success at learning routes, we investigated the relationship between landmark recall score and maze error score for each group. This demonstrated that maze error score was not related to landmark recall score for any group: TD: $r(71) = -0.029$, $p = 0.808$; WS: $r(20) = -0.409$, $p = 0.073$; ADHD: $r(42) = -0.274$, $p = 0.075$.

DISCUSSION

In this study, we demonstrated that the relationship between motor competence and large-scale spatial cognition observed in infancy (Clearfield, 2004) is also observed in TD children aged 5–11 years. This contrasted to no relationship between motor competence and large-scale spatial cognition in children with ADHD or individuals with WS. Furthermore, while the WS group demonstrated impairments in both the motor and spatial domains, the ADHD group did not show any deficits in large-scale spatial cognition, despite evidence of impairment in the motor domain. We suggests that a motor impairment does not necessarily lead to a deficit in large-scale spatial cognition, and that spatial ability can develop independent of the motor domain.

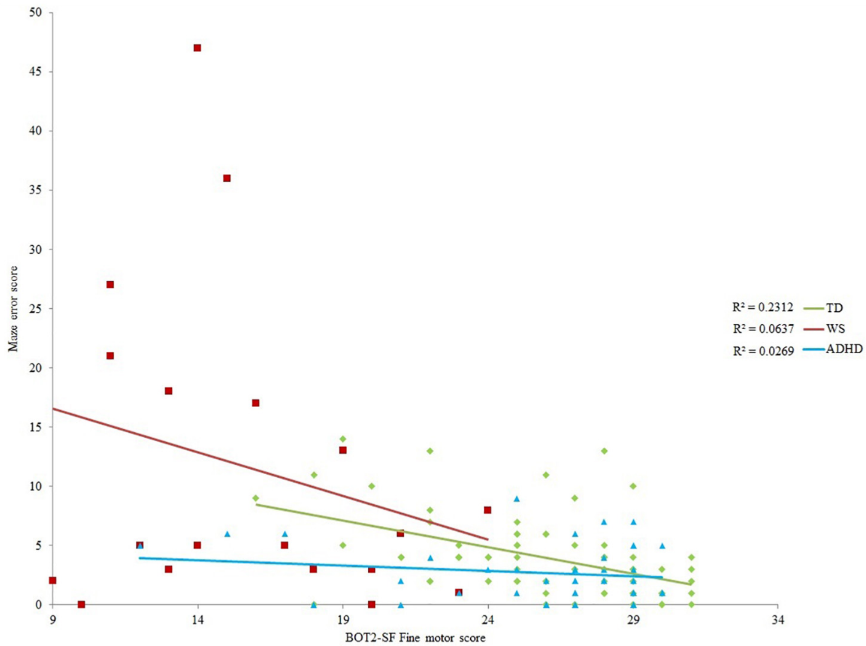

FIGURE 3 | The relationship between maze errors and motor ability (BOT2-SF fine motor), by group.

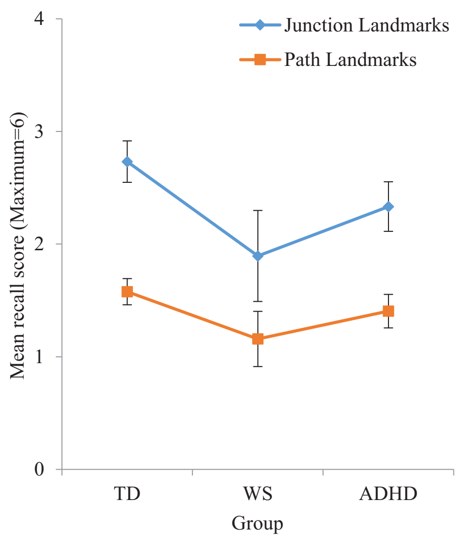

FIGURE 4 | Mean numbers of junction and path landmarks correctly recalled during test phase. Error bars represent standard error.

On account of the novelty of our dataset within the TD literature, we first discuss the novel findings with respect to the TD group only, before comparison across the TD, ADHD-L, ADHD-H, and WS groups.

Typical Development

The findings from the spatial navigation task replicate previous studies (e.g., Farran et al., 2012). That is, all TD children were reliant on landmarks to remember the route as evidenced by stronger memory for landmarks that featured at junctions (i.e., more useful landmarks) compared to landmarks that featured on path sections.

Having demonstrated successful spatial navigation in the TD children, we were interested in how motor ability related to this ability. We found that, at least for some aspects of motor ability, there is a relationship between performance in the motor and spatial domains. That is, for TD children aged 5–11 years, stronger fine motor ability is associated with fewer errors on a route learning task (23% of variance explained), even after controlling for variation in age. The relationship between gross motor ability and spatial ability, however, simply reflected age-related variation. Spatial ability in infancy has been assessed in relation to the development of both gross motor milestones (e.g., Clearfield, 2004) and fine motor skills (Soska et al., 2010), both reporting an impact of motor skill on spatial understanding. Similarly, motor ability is related to mental rotation ability in 5- to 6-year-olds (Jansen and Heil, 2010). Our findings support and extend the findings of Jansen and Heil (2010) by demonstrating that the relationship between motor ability and spatial cognition in infancy is evident across the primary school years with respect to large-scale spatial cognition.

The interpretation that has been put forward for the association between motor and spatial abilities in infancy relates predominantly to the development of self-movement either through crawling or walking; with the new ability to move comes the requirement for the infant to focus their attention on their spatial environment, which has a positive impact on spatial cognition (Clearfield, 2004). Soska et al. (2010) further our understanding of the importance of new attentional perspectives; they explain that fine motor skills such as transferring an object from hand to hand and rotating an object while looking at it, enables the infant to learn about objects from different viewpoints. This leads an infant to understand the three-dimensional nature of objects. The association observed in this study in relation to fine motor ability in childhood expands our understanding of this cross-domain relationship.

Oudgenoeg-Paz and Rivière (2014) situate the motor-spatial relationship within the theory of embodied cognition. They explain that sensory-motor interaction with the environment facilitates spatial development. Our finding of a relationship between fine motor ability and spatial ability supports an embodied cognition explanation. It is likely that the association observed in 5–11-year-olds is not a direct consequence of a step change in awareness of the spatial environment (as in infants), but represents the continuation of the relationship observed by Soska et al. (2010) in infancy. That is, it is a result of increased understanding of space via physical manipulation of objects and manipulation of the relationships between objects within the environment, which requires fine motor skills. We suggest that this likely benefits skills such as the ability to perform mental transformations and perspective taking, both of which are spatial skills that feed into navigation performance (Broadbent, 2015). In summary, this is the first study to demonstrate the importance of motor ability for large-scale spatial cognition, in the typical population, beyond infancy.

Children were asked about their independent exploration, such as whether they were allowed to walk home from school alone or to cross roads alone. While only 10% of TD 5–6-year-olds indicated that they had performed at least one of these independent acts in the preceding week, 60% of TD 7–8-year-olds reported independent exploration. This contrasts to 5–10% of children aged 7–8 years reported by Shaw et al. (2015) and hence questions the reliability of the self-reports of the children in the TD group (although note that the schools in this study were in inner London and so a local shop might be relatively close in comparison to other locations). Shaw et al. (2015) used parent report with 512 parents of the United Kingdom 7- to 15-year-olds. Despite this, we did see the anticipated relationship between increasing independent exploration and age, which supports the validity of the measure. Exploration score, did not, however, relate to motor ability, or to either of the spatial measures (maze error score or landmark recall score). This does not support the embodied cognition notion that motor action enables exploration, which in turn impacts cognition (Smith and Gasser, 2005). It also contrasts with Cornell et al. (2001) who demonstrated a relationship between exploration of the local environment and spatial navigation ability. In addition to the potential limitation in reliability mentioned above, it is possible that the exploration measure did not capture the kind of exploration that is employed by this age range. Independent exploration outside of the home is relatively limited for UK children due to cultural and safety reasons (Shaw et al., 2015). Perhaps a measure adapted from Oudgenoeg-Paz et al. (2015) in which exploration is measured in a safe environment would provide a more sensitive and reliable measure of this variable.

Neurodevelopmental Disordered Groups

The primary aim of the cross-syndrome comparison between individuals with ADHD and individuals with WS was to determine whether the presence of a motor deficit dictates that (large-scale) spatial cognition will also be impaired. This was based on the known relationship between the achievement of motor milestones and spatial abilities in the typical population, as well as findings that physical disability can negatively impact large-scale spatial knowledge (Stanton et al., 2002). The findings from the TD group in this study, discussed above, also demonstrate a relationship between motor ability and spatial cognition, in children aged 5–11 years.

If a motor deficit has a cascading downstream negative impact on spatial abilities, then the poorest spatial navigation performance should have been observed in those with a motor impairment (the WS group, and approximately half of the ADHD group, i.e., those ADHD participants on the left half of **Figure 4**). This prediction was not borne out. In fact, at the lowest level of motor ability (the intercept), the ADHD group had statistically *lower* maze errors than the TD group—although this finding must be interpreted with caution due to the low amount of variance in route learning errors explained by motor ability in this group. Motor ability explained 6% (WS) and 3% (ADHD) of variance in route learning errors. This demonstrates that motor competence is not a significant contributor to large-scale spatial ability for these groups. Furthermore, only the WS group demonstrated a deficit in spatial navigation. Spatial navigation in the ADHD group was on a par with than that of the TD group, which indicates that there is no large-scale spatial impairment in this group. This cross-syndrome difference between the WS and ADHD groups, coupled with the lack of significant association between motor and spatial competence across both of the disorder groups suggests that, in contrast to the spatial deficits observed in children with physical disability (Stanton et al., 2002), a motor impairment need not lead to an impairment in large-scale spatial cognition. This is, however, within the context of a small sample size for the WS group. Nevertheless, the effect sizes presented in **Table 4** do not suggest that any non-significance relates to lack of power in this sample.

The above finding has two possible interpretations. First, perhaps motor milestone achievement plays a larger role in the development of spatial cognition, than later motor competence. That is, if motor milestones are achieved late, then this could be critical for the development of early spatial ability, with cascading negative impact on the development of the spatial domain. Note that the parents/carers of the WS group report delayed motor milestone achievement of their children, but the parents/carers of the ADHD group report that their children achieve motor milestones at a broadly typical time, regardless of their concurrent motor ability. There was a hint that the age of walking onset in ADHD is related to concurrent motor competence, as this was a significant association when the children with comorbid diagnoses were excluded. It is possible that the motor difficulties experienced by some children with

ADHD stem from more subtle motor (or attention) deficits in infancy (see Farran et al., submitted for further discussion), which were not measured here. This requires further investigation using more sensitive measures, such as investigation of motor quality of walking in this group. Nonetheless, walking was not achieved substantially later than TD children in this group. This contrast between WS and ADHD groups with respect to motor milestones mirrors the pattern of impaired spatial ability in the WS group, but not the ADHD group. This is also consistent with delayed motor milestone achievement in children with physical disability, who also demonstrate impaired navigation performance (Stanton et al., 2002). Despite this, motor milestone achievement was not significantly related to spatial ability in either the ADHD or WS group. Of course, this could be due to a lack of power (there was missing data for this measure). Furthermore, although this kind of retrospective report has been shown to be as reliable as concurrent assessments (Langendonk et al., 2007), it is possible that the retrospective nature of this measure impacted reliability in our sample, particularly given that many of the WS group were adults. Further data are therefore required to support or refute this hypothesis.

The second possible interpretation is that, while a relationship is observed between motor ability and spatial ability in both infancy (Clearfield, 2004), and TD children (Jansen and Heil, 2010, this study), motor competence might not be a prerequisite for the development of large-scale spatial competence. That is, if the usual developmental pathway is limited, then over developmental time, it is possible that large-scale spatial skill development is redirected to alternative pathways, i.e., a pathway which is less reliant on input from the motor system and more reliant on other mechanism that are important to spatial navigation (spatial, memory, and executive function mechanisms). This has been observed for the language domain, where individuals with WS demonstrate language acquisition before the use of joint attention, an ability which was initially thought to be a prerequisite for the acquisition of language (Laing et al., 2002). If spatial ability can develop without input from the motor system, this suggests that the motor impairment and the spatial impairment observed in WS are unrelated, and also explains why the ADHD group demonstrates a large range of motor abilities, but typical large-scale spatial abilities. That is, poor motor competence in approximately half of the ADHD sample and all of the WS group was not a limiting factor to the development of spatial navigation abilities, and the disparity in spatial ability between these two groups was independent of their motor ability. To further support this hypothesis, it would be interesting to employ a wider battery of both small-scale and large-scale spatial tasks, and to investigate this relationship longitudinally, from infancy, in these groups.

This is the first investigation of large-scale route knowledge in individuals with ADHD. This group demonstrated typical route knowledge, i.e., the ability to learn a route from A to B. Of interest, both of the neurodevelopmental disorder groups employed the same, typical strategy to remember the route. That is, they used landmarks to determine which way to turn. However, despite the use of a typical strategy, the WS group recalled fewer landmarks overall and took longer (more errors and hence more trials) to learn the route than the other groups, and performed at the level below a typical 5–6-year-old. This is broadly consistent with previous research and reflects their hallmark deficit in spatial cognition (e.g., Farran et al., 2012; Purser et al., 2015).

While we used a relatively pure spatial navigation task by design, it is also entirely possible that children with ADHD might experience navigation difficulties on account of the attentional and sensory integration of additional demands that are present in real-world navigation (locomotion demands, proprioceptive and auditory information, a richer visual array). This is unlikely given that VEs have been shown to tap into the same cognitive mechanisms as real-world environments (Coutrot et al., 2019). Nonetheless, a deficit in real world, but not virtual navigation, in ADHD, would point toward difficulties in integrating information rather than a purely spatial deficit.

We included an environmental measure that might have impacted large-scale spatial knowledge in our groups, independent exploration. This did not demonstrate a relationship with large-scale navigation performance in either of the groups, perhaps due to the impact of non-motor variables related to dangers in the outside world which might have limited participants' opportunity to explore. As discussed earlier, a "safe" measure of exploration might have been more sensitive. Comparison across the groups showed only subtle, albeit significant, differences. The WS group showed an exploration at the level of 7–11-year-olds, despite being adults. It is likely that independent exploration is restricted in WS due to their low IQ and hypersociability, which make them particularly vulnerable (Farran and Karmiloff-Smith, 2012). The ADHD group explored at the level of 9–11-year-olds, and thus at a level commensurate with their chronological age.

In summary, we investigated spatial navigation in ADHD for the first time. This demonstrated a typical level and pattern of abilities in this group, which was not impacted by whether the individual displayed a motor impairment or not. Furthermore, cross-syndrome comparison between ADHD and WS demonstrated that a motor impairment in these groups is not associated with large-scale spatial navigation ability. Finally, although our data suggest that the timepoint of motor milestone achievement does not impact the development of large-scale spatial abilities, this conclusion is given with caution due to the large amount of missing motor milestone data in our sample. Indeed, our findings contrast with those of Stanton et al. (2002) who demonstrated a relationship between motor ability in infancy and large-scale spatial navigation in individuals with physical disability.

CONCLUSION

This is the first study to demonstrate that the relationship between motor ability and large-scale spatial cognition observed in typical infants (Clearfield, 2004) extends to TD children aged 5–11 years. This supports an embodied cognition view of development and suggests that this cross-domain relationship is present across the primary school years. In contrast, motor ability and large-scale spatial ability were not related in any of the neurodevelopmental disorder groups. This suggests that a motor impairment does not necessarily lead to a deficit in large-scale spatial cognition, i.e., spatial ability *can* develop via an alternative developmental pathway, with little or no input from the motor domain. With respect to each group, in the first study to measure large-scale spatial ability in ADHD, we demonstrated that despite a motor impairment, the children with ADHD and low motor ability displayed competent, age-appropriate, navigation abilities. Furthermore, we measured two of the most impaired domains in WS, motor ability and spatial ability, within the same study for the first time; our findings demonstrated that these two deficits are unrelated in this group. Knowledge that the developmental pathway for spatial cognition is atypical in WS has implications for how best to train navigation abilities to improve independence in this group.

DATA AVAILABILITY

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

Ethical approval was obtained from the UCL Institute of Education Research Ethics Committee (approval number: REC 766; study title: Motor development and navigation in Attention Deficit Hyperactivity Disorder). Following parental consent, the participants were tested individually either at their school, in the research lab, or at the participant's home.

AUTHOR CONTRIBUTIONS

EF conceived of the study, with input from AK-S and EH. AB, HD'S, and LM collected and coded the data. EF analyzed the data and wrote the manuscript with input from EH, AB, HD'S, and LM.

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frontiers in Human Neuroscience

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Topographical Disorientation: Clinical and Theoretical Significance of Long-Lasting Improvements Following Imagery-Based Training

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Neuropsychological studies on acquired topographical disorientation have provided useful insights into the contribution of different brain regions to human navigation. However, little is known about the possibility to restore navigational skills after brain damage. Here we describe the case of No Longer Lost (NLL), a 49-year-old man who complained of severe topographical disorientation following traumatic brain injury. Extensive neuropsychological evaluation at baseline revealed selective episodic memory deficits and topographical disorientation. NLL underwent 8-week imagery-based treatment (IBT) inspired by current cognitive models of human spatial navigation. After IBT, NLL improved topographical skills and episodic memory. From a clinical point of view, the present study describes a model-based intervention for topographical disorientation. From a theoretical point of view, it provides new insights into the cognitive models of human spatial navigation and straightforward evidence about common phylogenetic roots of brain mechanisms devoted to spatial navigation and memory.

Keywords: acquired topographical disorientation, brain damage, spatial navigation, topographical memory, episodic memory

BACKGROUND

To orient themselves within the environmental space – namely, the space beyond the sensory horizon (Wolbers and Wiener, 2014) – individuals have to process "online" information about their own position and facing direction (Sulpizio et al., 2017, 2018), as well as to recall previously acquired "offline" information about the environment (Wolbers and Hegarty, 2010). Physiological evidence suggests that "online" information about position and facing direction is coded by distinct neural populations, namely by hippocampal place cells and head-direction cells, respectively (O'Keefe and Dostrovsky, 1971; Taube, 1998; Ekstrom et al., 2003; Vass and Epstein, 2013; Sulpizio et al., 2014). "Offline" environmental knowledge may be represented as Landmark, Route, and Survey representation (Siegel and White, 1975). Landmark representation roughly corresponds to the figurative memory of environmental objects, through which individuals "beacon" toward salient landmarks. Route representation concerns the memory of paths connecting landmarks and is organized according to an egocentric frame of reference. Survey representation implies the encoding of directions and distances between landmarks regardless of the individual's position, resembling a map-like representation of the environment. Recent neuroimaging evidence supports this model (Boccia et al., 2016).

Following the BBB model (Byrne et al., 2007), neural populations within the posterior parietal lobe – namely, "egocentric parietal window" (p. 345) – maintain the head-centered egocentric map of the space and provide exclusive access into long-term navigational memory stored in the medial temporal lobe; in the medial temporal lobe, the parahippocampal gyrus develops allocentric (survey) representations and the hippocampus stores long-term spatial memories; finally, a transformation circuit in the retrosplenial cortex allows to transform the spatial representations stored in an allocentric format into an egocentric (route) format, and vice versa. Evidence in nonhuman primates (Kravitz et al., 2011) and humans (Boccia et al., 2014, 2017a), and neuropsychological studies (Aguirre and D'Esposito, 1999) support this model. Specifically, lesions of the posterior parietal cortex yield to egocentric disorientation, namely a deficit in representing the location of objects with respect to the self (Holmes and Horrax, 1919; Levine et al., 1985; Stark et al., 1996). Instead, lesions of the retrosplenial cortex lead to heading disorientation: patients lose the sense of direction and are unable to direct toward locations beyond the vista space (Takahashi et al., 1997), namely the space that can be explored at a glance (Montello, 1993; Wolbers and Wiener, 2014). Landmark recognition deficits are widely reported following lesions in the lingual gyrus: patients fail in recognizing and representing salient environmental stimuli, even in absence of perceptual deficits (Aguirre and D'Esposito, 1999). These neuropsychological findings provide important information about the unique and causal contribution of each specific node of the parieto-medial temporal lobe network of spatial navigation in humans. However, evidence for a redundant code within this network (Ekstrom et al., 2017; Boccia et al., 2019) suggests the possibility that, in presence of a lesion in a specific node, compensation mechanisms may allow the recovery of the impaired process.

Here we describe the case of NLL (acronym for No Longer Lost), a patient with acquired topographical disorientation, who underwent a novel imagery-based neuropsychological rehabilitation for spatial navigation, tailored on the patient's neuropsychological profile and the theoretical framework described above.

CASE PRESENTATION

NLL, a 49-year-old man working as a realtor, suffered from an extensive head trauma (including facial trauma) and coma, which lasted about 1 week, following a motor vehicle accident. NLL referred to the IRCCS Santa Lucia in Rome 4 years after his accident due to persistent topographical disorientation, even in highly familiar environments, and memory deficits. The CT performed immediately after the event, and the MRI performed 4 years later, revealed a lesion of the right temporal lobe extending also to subcortical areas (**Supplementary Figure 1**). No neuropsychological evaluation has been performed immediately after the accident or in the following 4 years.

NLL reported that topographical disorientation greatly impacted over his life and professional duties, and that soon after the event he realized he was unable to recognize landmarks and routes, and adopted several compensatory strategies (e.g., he wrote little numbers on the outside wall of the houses he had to visit with customers, he mentally counted the number of bus stops he had to pass to get to the correct place). Also, he reported to be unable to imagine landmarks and routes connecting them, in both familiar and novel environments. Furthermore, he reported episodic memory deficits: he was impaired in recalling events, including, for example, the customers he met or the houses he visited; he was unable to retrieve memories about previous travels reporting that "it is as if I've never been there . . . There is no trace about that in my memory." He also reported to be unable to talk about relevant daily events, for example with his father, when he called NLL in the evening, because he could not recall them.

The study was designed in accordance with the principles of the Declaration of Helsinki and was approved by the ethical committee of the IRCCS Fondazione Santa Lucia. Informed consent was obtained from the patient.

Neuropsychological Assessment at Baseline

General Neuropsychological Assessment

NLL performed well within the normal range in tests assessing attention, intelligence, executive functions, language, and working memory (**Table 1**). He was impaired in tests evaluating visuo-spatial learning and delayed recall (*Corsi Block Tapping Test*, CBT; Corsi, 1972; **Table 2**) and visual, spatial, and verbal memory in ecological contexts (RBMT-3; Wilson et al., 2008; **Table 3**).

Assessment of Navigational Abilities

Navigational abilities were tested using the *DiViNa Developmental Topographical Disorientation Battery* (DDTDB), which is extensively described in previous works (Bianchini et al., 2010, 2014; Palermo et al., 2014) and is based on theoretical models of human navigation (Siegel and White, 1975; Wang and Spelke, 2002). NLL's scores were compared with those of men in our database or previous published studies (**Table 2**), by using *t*-test modified procedure (Singlims.exe; Crawford and Garthwaite, 2002). Age and education were included as covariates (BTD_Cov.exe; Crawford et al., 2011) when necessary.

Walking Corsi Test (WalCT; Piccardi et al., 2008, 2013)

Both short-term and long-term memory in navigational vista space (i.e., learning and delayed recall) were assessed. NLL performed as well as controls in short-term memory and delayed recall. However, he was impaired in learning spatial positions (**Table 2**).

Cognitive Map Test (CMT; Iaria et al., 2007)

We assessed both the formation/learning (CMT-L) and the recall (CMT-R) of a cognitive map, asking NLL to travel in a virtual city, in which there were six landmarks (i.e., a cinema, a restaurant, a bar, a hotel, a pharmacy, and a flower shop) (for full description

TABLE 1 | General neuropsychological assessment.

Test	NLL's score (baseline)	ES[a]/PR[b]/SS[c]	Cut-off
Attention			
Trail making test			
A	55 s	2[a]	
B	90 s	4[a]	
B−A	35 s	4[a]	
Stroop			
Errors	0	4[a]	
Time	14	4[a]	
TEA			
Alertness – tonic	298	10[b]	
Phasic	237	38[b]	
Median	0.239	99[b]	
Go-NoGo median	543	42[b]	
False alarms	0	>46[b]	
Divided attention median	740	12[b]	
Omissions	**5**	**4**[b]	
False alarms	**2**		
Working memory median	684	28[b]	
Omissions	3	<16[b]	
False alarms	0	>76[b]	
PASAT			
Stimulus interval 3000			
Correct answers	46		44.2
Errors	7		
Omissions	7		
Percentage of errors	23		
Stimulus interval 2600			
Correct answers	47		38.69
Errors	6		
Omissions	7		
Percentage of errors	22		
Stimulus interval 2200			
Correct answers	47		34.77
Errors	3		
Omissions	10		
Percentage of errors	22		
Stimulus interval 1800			
Correct answers	37		28.07
Errors	0		
Omissions	23		
Percentage of errors	38		
Intelligence			
Raven progressive matrices	36	4[a]	
Executive functions			
Wisconsin card sorting test			
Errors (%)	15	61[b]	
Perseverative errors (%)	8	63[b]	
Non-perseverative errors (%)	7	58[b]	
Categories completed	6	>16[b]	

(Continued)

TABLE 1 | Continued

Test	NLL's score (baseline)	ES[a]/PR[b]/SS[c]	Cut-off
Number of attempts on the first category	12	>16[b]	
Inability to maintain the set	0	>16[b]	
Learning to learn	+	>16[b]	
Tower of London			
Correct responses (total)	7	118[c]	
Moves (total)	8	120[c]	
Planning time (total)	88	114[c]	
Execution time (total)	175	106[c]	
Total time	263	100[c]	
Time-limit breaks	0	104[c]	
Rule breaks	0	108[c]	
Language			
Fluency			
Phonemic	38	4[a]	
Semantic	61	4[a]	
Alternate	46	4[a]	
Shifting	0.93	4[a]	
Memory			
Digit span (forward)	7	4[a]	
Rey auditory verbal learning test			
Immediate recall	44	3[a]	
Delayed recall	8	2[a]	
Recognition	12		
Rey–Osterrieth figure			
Copy	36	4[a]	
Immediate recall	20	3[a]	
Delayed recall	21	3[a]	
Oblivion	−1	4[a]	
SMIRNI			
Words	24	10–25[b]	
Buildings	25	25–50[b]	
Faces	25	25–50[b]	
Space and object perception			
VOSP			
Screening	20		15
Incomplete letters	20		17
Silhouettes	26		16
Objects decision	19		15
Progressive silhouettes	**13**		**14**
Dot counting	10		8
Position discrimination	20		8
Number of location	10		7
Cube analysis	10		6

Tests are standardized for use with Italian-speaking individuals. Pathological scores are marked in bold. [a]Equivalent scores adjusted for sex, age, and years of education. [b]Percentile rank. Performances below the 5th percentile and equivalent score equal to zero should be considered pathological. [c]Standard scores; performances below 84 should be considered pathological.

of the task and virtual environment, see Iaria et al., 2007). NLL performed worse than controls on CMT-L, but comparably to controls on CMT-R (**Table 2**).

Route Strategy and Landmark Recognition

Experimenter shows a path in real environmental space, asking the participant to pay attention to all environmental objects. Immediately after, the participant is blindfolded and brought back to the starting point; then she/he is asked to discriminate among distractors (N = 8) the specific landmarks (N = 8) encountered along the path (maximum score = 16). Finally, the participant has to reproduce the path shown by the examiner (for similar procedure, see Conson et al., 2018). The score is calculated by summing the segments correctly retraced (maximum score = 7). NLL correctly performed landmark recognition (**Table 2**) and retraced the route without errors (**Table 2**), even though he claimed he had no clear navigational goal and was unable to imagine the successive steps of the path.

Throwback to the Starting Point

Participant is asked to go back to the starting point of a route previously shown by the examiner. The score corresponds to the number of segments correctly performed (maximum score = 6). NLL's performance was errorless.

Map-Following Task

The participant is provided with a map of a real environmental navigational space on which a starting point and a goal are depicted, and asked to draw the shorter path to reach the goal. Then the participant is brought to the starting point and asked to use the map to reach the goal, following the path he/she drew. The score corresponds to the number of segments correctly performed following the path participant drew (maximum score = 4). NLL hesitated at crossroads, but his performance (3/4 correct segments) did not differ from that of controls (**Table 2**), and he correctly recognized his errors and difficulties.

Mental Imagery Skills

NLL complained of deficits in imaging landmarks and routes; thus, we tested his mental imagery skills by using the *Complete Visual Mental Imagery Battery* (CVMIB) (Palermo et al., 2016), which allows to assess systematically the ability to generate, maintain, inspect, and transform visual mental images (for a complete description of tests and normative data, see Palermo et al., 2016). For each subtest (Buildings, Objects, Color 1, Color 2, and Color 3, Inspection of Objects and Letters, Folding, and the Mental Rotation), the sum of the correct answers was computed (**Table 4**). NLL performed worse than controls on Object and Color 2 subtests (**Table 4**); he performed within the normal range in the remaining subtests (**Table 4**). Also, Road Map Test (*RMT*; Money et al., 1965) and *3D Mental Rotation Task* (*MRT*; Thurstone, 1937) were performed comparably to controls.

Intervention Hypothesis

The neuropsychological assessment revealed a specific deficit in the formation of the cognitive map. Cognitive maps are pivotal for spatial orientation, since they allow individuals to reach any destination within the environmental space. Individuals unable to form a cognitive map get lost more frequently than individuals who are able to form and use cognitive maps of the environment (Iaria and Barton, 2010). Based on NLL's complaints, as well as on evidence on the pivotal role of Mental Imagery in environmental navigation, we submitted him to an imagery-based treatment (IBT), tailored on NLL's difficulties and aimed at improving and restoring navigational skills, with possible generalization to the episodic memory domain. The IBT was inspired by the imagery-based intervention proposed by Kaschel et al. (2002) for memory rehabilitation. Evidence from current cognitive models of spatial navigation and psychophysiological findings were integrated in the IBT (Siegel and White, 1975; Wang and Spelke, 2002; Wolbers and Wiener, 2014). Specifically, IBT followed the developmental hierarchical model proposed by Siegel and White (1975), neurophysiological evidence about head-direction and place coding (O'Keefe and Dostrovsky, 1971; Taube, 1998) and evidence about the impact of the spatial scale (Wolbers and Wiener, 2014). Thus, we developed training activities allowing to progressively move from the figurative memory of landmark representation in the vista space, to the egocentric route-based and allocentric survey-based representation of the environmental space, retracing ontogenetic acquisition stages proposed by Siegel and White (1975). IBT is extensively described in the **Supplementary Material**. In brief, in a first phase NLL was motivated to the imagery training and acquired the ability to rapidly generate mental images. Then, he underwent a second phase during which he was asked to generate and retrieve navigational mental images of landmarks, routes, and environmental map-like representations (i.e., survey representations).

Post-treatment Neuropsychological Assessment

After 8 weeks of treatment, the tests in which NLL performed worse than controls were repeated. For all navigational, spatial, and memory tests, alternate versions were used. The only test for which alternate versions were not available is the CVMIB. However, no feedback is provided to participants during the execution of the test and possible test/re-test effects are unlikely. Effectiveness, namely the measure reflecting the potential improvement achievable during rehabilitation, was calculated for each test, as it follows:

$$\text{Effectiveness} = \frac{\text{Post treament score} - \text{Baseline score}}{\text{Maximum score} - \text{Baseline score}} * 100$$

Learning of spatial positions within reaching and navigational vista spaces significantly improved, with an effectiveness of 89.79 and 82.97% on the CBT and the WalCT, respectively; after IBT performances on both tests were comparable with those of the control group (**Table 2**). Also, after IBT NLL did not differ from the control group on an alternate version of the CMT (**Table 2**). His performance on an

TABLE 2 | Performances on DDTDB.

	Baseline		Post-treatment		Follow-up	
	NLL's score	**Crawford analysis**	**NLL's score**	**Crawford analysis**	**NLL's score**	**Crawford analysis**
Corsi Block Tapping Test (CBT)						
Span	5[a]	$t = 0.047$, $p = 0.481$*				
Learning	46[a]	$t = -3.187$, $p = 0.001$*	134[a]	$t = 1.065$, $p = 0.146$*	127[a]	$t = 0.727$, $p = 0.235$*
Delayed recall	3[a]	$t = -1.681$, $p = 0.050$*	6[a]	$t = -0.280$, $p = 0.390$*	8[a]	$t = 0.654$, $p = 0.258$*
Walking Corsi test (WalCT)						
Span	5[a]	$t = 0.000$, $p = 0.500$*				
Learning	50[a]	$t = -3.995$, $p < 0.001$*	128[a]	$t = 0.582$, $p = 0.282$*	101[a]	$t = -1.002$, $p = 0.161$*
Delayed recall	8[a]	$t = 0.412$, $p = 0.341$*	6[a]	$t = -1.650$, $p = 0.053$*	8[a]	$t = 0.412$, $p = 0.341$*
Cognitive map test (CMT)						
Learning (CMT-L)	1500s[b]	$z = 2.686$, $p = 0.047$#	720s[k]	$z = -0.806$, $p = 0.461$#	1380s[b]	$z = 2.188$, $p = 0.091$#
Recall (CMT-R)	17.44s[c]	$z = -1.706$, $p = 0.139$#	38.72s[l]	$z = -1.340$, $p = 0.436$#	19.27s[c]	$z = -1.563$, $p = 0.169$#
Road map test	28[d]	$t = -0.345$, $p = 0.372$*				
3D mental rotation						
Accuracy	38[e]	$z = 1.046$, $p = 0.251$#				
Response time	6955.33ms[f]	$z = 1.641$, $p = 0.148$#				
Navigational tasks in real environment						
Landmark recognition	15[g]	$z = 0.202$, $p = 0.461$#				
Route strategy	7[h]					
Throwback	6/6[i]					
Map-following task	3/4[j]	$z = -0.827$, $p = 0.342$#	4/4[j]		4/4[j]	

*For each task and assessment (baseline, post-treatment, and follow-up) NLL's raw score is reported, along with the result of the Crawford analysis [*Crawford and Garthwaite (2002); #Crawford et al. (2011)]. [a]Normative data from Piccardi et al. (2013). [b]Normative data from 29 males (M = 542.18, SD = 275.91). [c]Normative data from 29 males (M = 20.76s, SD = 8.49). [d]Normative data from six males (M = 29.00, SD = 2.68). [e]Normative data from 29 males (M = 34.74, SD = 7.03). [f]Normative data from 29 males (M = 8935.90s, SD = 355.26). [g]Normative data from 13 males (M = 12.77, SD = 2.62). [h]Normative data from 13 males (M = 6.08, SD = 0.95). [i]Normative data from five males (M = 5.80, SD = 0.45). [j]Normative data from 13 males (M = 3.77, SD = 0.60). [k]Normative data from six males (M = 580s, SD = 256.43). [l]Normative data from six males (M = 21.19s, SD = 8.66).*

alternate version of the map-following task was errorless (four out of four segments correctly executed), with an effectiveness of 100%. Improvement was stable 8 months later (**Table 2**).

Interestingly, after IBT NLL performed within the normal range on ecological memory tests (RBMT-3) in which he initially performed below the cut-offs (effectiveness on General Memory Index = 32.81%), suggesting that improvement on navigational skills also generalized to episodic memory (**Table 3**).

Immediately after the IBT, NLL also performed at ceiling on the Object Generation and Color subtests of the CVMIB, with an effectiveness of 100% in both subtests.

TABLE 3 | Rivermead Behavioral Memory Test-3.

Subtest	NLL's score (baseline)	NLL's score (post-treatment)
Names	6	7
Belongings	6	8
Appointments	4	4
Picture recognition	14	14
Story recall – immediate	**5.5**	9
Story recall – delayed	**4.5**	8.5
Face recognition – delayed	14	10
Route recall – immediate	13	13
Route recall – delayed	**8**	13
Messages – immediate	6	6
Messages – delayed	6	6
Orientation and date	14	13
Novel task – immediate	**18**	48
Novel task – delayed	**6**	17
GMI	83	104

Pathological scores are marked in bold. GMI, general memory index.

DISCUSSION

When NLL came to our observation he was seriously worried about his topographical disorientation and memory impairment. At baseline, he showed deficits in topographical and visuo-spatial learning and an inability to form a cognitive map of environmental space. We also found a specific deficit in generating mental images of objects and in maintaining them. Performances on landmark recognition, route learning, and map-following tasks were well within the normal range, even if they were performed with evident efforts. Neuropsychological evaluation highlighted a deficit in learning and retrieving structured verbal material (i.e., stories), despite an intact ability to learn and recall unstructured verbal material, (i.e., word lists). IBT yielded a significant improvement in all the areas

TABLE 4 | Complete visual mental imagery battery at baseline.

Process	Subtest	Maximum score	Control (Mean and SD)[a]		NLL's score (baseline)	Crawford analyses (*t* and *p*)	
Generation	Buildings	20	19.28	1.04	18	−1.209	0.119
	Objects	20	19.60	0.78	18	−2.016	0.027
Maintenance	Color 1	10	9.57	0.57	10	0.741	0.233
	Color 2	10	9.82	0.39	9	−2.066	0.024
	Color 3	10	9.57	0.74	9	−0.757	0.228
Inspection	Objects	20	18.67	1.18	18	−0.558	0.291
	Letters	20	19.50	1.10	20	0.447	0.329
Transformation	Folding	20	16.75	2.93	12	−1.593	0.062
	Mental rotation	20	16.71	2.66	14	−1.001	0.163

[a]*Normative data are derived from Palermo et al. (2016).*

in which NLL was impaired. Indeed, after 8 weeks of IBT, NLL's performances did not differ from controls' ones in visuo-spatial and topographical learning, suggesting that IBT fostered memory for positions within both reaching and vista space; he also performed without differences from controls on CMT, suggesting that NLL recovered the ability to form a cognitive map of the environmental space. Accordingly, after IBT he ameliorated his ability to use a map for navigating the environmental space, performing at ceiling on the map-following task. Interestingly, also episodic memory improved over the original performance: after IBT NLL's performances on learning and recalling a story fell well within the normal range. These results deserve great attention in the light of possible clinical applications to topographical disorientation and episodic memory deficit as well as for their theoretical implications.

Despite the single case methodology, present findings provide a unique Contribution To The Field of neuropsychological rehabilitation of topographical disorientation. To the best of our knowledge, there is no evidence about effective neuropsychological rehabilitation of topographical disorientation. The only papers describing the rehabilitation of this disorder, indeed, focused on the acquisition of compensatory strategies (Incoccia et al., 2009; Bouwmeester et al., 2015; Svoboda et al., 2018); in some cases (Svoboda et al., 2018), the intervention was just aimed to allow patients to autonomously navigate in very familiar environments; in others, no generalization occurred for untreated materials (Davis and Coltheart, 1999). However, topographical disorientation may have a great impact on individual functioning. As it happened for NLL, people with topographical disorientation experience great difficulties which may prevent full recovery of autonomies. Before IBT, NLL used compensatory non-spatial strategies to cope with his daily life activities and professional duties, which likely resulted in non-pathological performances on ecological assessment of navigational skills (i.e., performance on landmark recognition, route learning, and map-following tasks). However, the compensation did not fully prevent NLL from getting lost and experiencing daily life difficulties. Indeed, he failed in processing and acquiring proper spatial information, such as positions, as also demonstrated by pathological scores on WalCT. After IBT, NLL was able to learn and recall spatial positions within the environmental, vista, and reaching spaces, suggesting that the neuropsychological rehabilitation protocol we developed was able to restore the spatial mechanisms disrupted by traumatic brain injury.

Spatial information is processed by a redundant code in the brain and the interaction between different areas is crucial for successfully navigating within the environmental space (Ekstrom et al., 2017). IBT likely taps on the wide network of areas involved in generating mental images of environmental space, including the hippocampus, the retrosplenial cortex and the parahippocampal place area (Boccia et al., 2015, 2017b). Considering that spatial navigation and mental imagery of familiar places arise from the interaction between these brain areas (Boccia et al., 2016, 2017b, 2019) it is possible that IBT, fostering the interaction between these regions, allowed to restore mechanisms of environmental navigation on account of the redundant code within this network (Boccia et al., 2019). This interpretation is consistent with lesion location and extension of NLL, which mainly involved the right temporal lobe, sparing other nodes of the parieto-temporo medial network of spatial navigation (Boccia et al., 2017a).

Besides the severe topographical disorientation, NLL was also affected by a severe episodic memory deficit. Interestingly, the effect of IBT generalized to episodic memory. Previous evidence supports the use of imagery-based trainings for neuropsychological rehabilitation of memory (Piras et al., 2011). Most randomized control trials (RCTs) used visual imagery and visualization of stories (Chiaravalloti et al., 2005) of relevant everyday materials (Kaschel et al., 2002). Here we found that imagery-based training of spatial abilities may improve performance on the recall of a story. Also, NLL reported that recall of relevant everyday events was significantly improved after IBT. He said "… now I'm able to tell my father what happened during the day when he calls me in the evening." This result ties well with the recent hypothesis that mechanisms of planning and memory have their phylogenetic roots within mechanisms of spatial navigation in the physical world (Buzsaki and Moser, 2013). According to this view episodic memory has evolved from mechanisms of environmental navigation, and shares the same neuronal algorithms used to navigate within the real environment. In this vein, the improvement we found for episodic memory may be due to the restoring of imbalanced spatial "root" mechanisms. Also, this result is consistent with the idea that "spatial navigation serves as a model system to identify key coding principles governing cognitive spaces" (Bellmund et al., 2018, p. 362).

An alternative explanation for NLL's disabilities is related to his visual imagery deficits: a lesion in the right temporal region may cause the slight deficit we observed at baseline in generating and maintaining mental images; accordingly, NLL complained of a deficit in imaging landmarks and routes. This deficit might have caused a mixed spatial and episodic memory deficit. In this light, the recovery of spatial and episodic memory deficits may be mediated by the recovery of mental imagery skills we detected at the post-treatment evaluation. However, it has to be noted that we observed only a slight imagery deficit at baseline, in contrast to NLL's dramatic disability in navigating within highly familiar and novel environments and in remembering his own life events.

Even if compelling, present results deserve caution, since the effects we detected are based on the observation of a single case. Thus, further systematic investigations of imagery-based rehabilitation protocols for spatial navigation and memory (especially by means of RCTs) are needed to draw definite conclusions.

DATA AVAILABILITY

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Local ethical committee of the IRCCS Fondazione Santa Lucia in Rome. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

All the authors significantly contributed to the work, read, and approved the final version of the manuscript. AD and MPC performed the neuropsychological assessment. MB, SD, AD, and CG conceived the IBT. SD and AB conducted the IBT. MB and AB ran the statistical analyses and wrote the first draft of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2019.00322/full#supplementary-material

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Comparing Tactile to Auditory Guidance for Blind Individuals

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The ability to travel independently is crucial to an individual's quality of life but compromised by visual impairment. Several navigational aids have been developed for blind people to address this limitation. These devices typically employ auditory instructions to guide users to desired waypoints. Unfortunately, auditory instructions may interfere with users' awareness of environmental sounds that signal dangers or provide cues for spatial orientation. Accordingly, there is a need to explore the use of non-auditory modalities to convey information for safe and independent travel. Here, we explored the efficacy of a tactile navigational aid that provides turn signals *via* vibrations on a hip-worn belt. We compared the performance of 12 blind participants as they navigated a series of paths under the direction of the tactile belt or conventional auditory turn commands; furthermore, we assessed the effect of repeated testing, both in the presence and absence of simulated street sounds. A computer-controlled system triggered each turn command, measured participants' time-to-path-completion, and detected major navigational errors. When participants navigated in a silent environment, they performed somewhat worse with the tactile belt than the auditory device, taking longer to complete each trial and committing more errors. When participants navigated in the presence of simulated street noises, the difference in completion time between auditory and tactile navigation diminished. These results suggest that tactile navigation holds promise as an effective method in everyday environments characterized by ambient noise such as street sounds.

Keywords: visual impairment, waypoint, navigation, haptic, vibration, blindness, spatial orientation and wayfinding

INTRODUCTION

In order to navigate safely and efficiently, blind individuals attend to nonvisual environmental stimuli, such as street sounds, while making use of mobility aids. Common mobility aids include the white cane and guide dog, which provide information about the user's immediate surroundings, and Global Positioning System (GPS) devices, which provide location and heading information. Despite the usefulness of these aids, much research is needed to develop more effective navigational devices for blind individuals (Loomis et al., 1994; Giudice and Legge, 2008).

Assistive navigational devices for blind people typically employ auditory instructions to guide users (Loomis et al., 2005; Gaunet, 2006). Despite their obvious usefulness, these instructions may prevent users from perceiving simultaneous environmental sounds that signal dangers or provide cues for spatial orientation—for instance, sounds made by passing cars, by nearby pedestrians, or by other sources. The presence of two simultaneous sources of auditory information places demands on attentional processing and raises the possibility of physical acoustic interference. Imagine navigating without vision in the midst of a busy, unfamiliar environment. A "turn left" auditory GPS command might interfere with the sound of a car driving by, with potentially lethal consequences. Alternatively, a car may be honking nearby, such that you're unable to decipher the auditory commands emanating from your GPS device. When two or more sound sources are simultaneously active, distinguishing them is difficult, as their acoustic waveforms sum into a composite waveform prior to entering the ear (Bregman, 1990; Darwin, 1997).

In light of these considerations, the most suitable navigational aid in acoustically rich environments may be one based upon the sense of touch. Conveying navigational commands *via* touch would decouple the two sources of information—navigational commands and environmental sounds—preventing both physical and attentional interference between them. The present study was designed to test this proposition and to investigate blind individuals' ability to process simple navigational instructions *via* the skin.

The somatosensory system would seem to offer a reliable communication channel for navigational purposes. The skin has a large available surface area; the point-to-point mapping from the skin to the somatosensory homunculus naturally conveys spatial information (Nakamura et al., 1998); and vibrations applied in sequence to adjacent skin locations can be accurately interpreted as directional information (Raj et al., 1998; Chiasson et al., 2002; Cholewiak et al., 2004; Van Erp et al., 2005; Jones and Ray, 2008; Barber et al., 2015).

In light of these promising characteristics, tactile-directed navigation has been a focus of research and development for many years (Bach-y-Rita, 1967; Ertan et al., 1998; Tsukada and Yasumura, 2004; Van Erp et al., 2005; Johnson and Higgins, 2006; Gustafson-Pearce et al., 2007; Pielot and Boll, 2010; Flores et al., 2015; Jimenez and Jimenez, 2017). Ertan et al. (1998) successfully guided sighted participants through indoor test paths by passing directional commands to a wearable vest containing an array of vibrotactile actuators. Similarly, Tsukada and Yasumura (2004) developed the "ActiveBelt" to guide participants to waypoint destinations, with tactile commands based on GPS. In order to assess the efficacy of tactile displays, Van Erp et al. (2005) and Pielot and Boll (2010) compared participants' navigational performance with a tactile display to that with a visual display. Both studies showed promising results for the potential use of tactile displays as hands-free guidance systems. In a study, with standing, stationary participants, Gustafson-Pearce et al. (2007) demonstrated that visually impaired and sighted participants could accurately follow tactile turn commands delivered through a vest; they further showed that, in the presence of simulated street sounds, participants made fewer errors in response to tactile than to auditory commands.

Most recently, Flores et al. (2015) and Jimenez and Jimenez (2017) compared navigation performance with a tactile display to that with an auditory device. In a study with blind participants, Flores et al. (2015) used an automated participant localization system to precisely transmit directional commands to both navigational devices. They found that navigation was slower but more accurate in the tactile condition. Jimenez and Jimenez (2017) transmitted either auditory or vibrotactile navigational commands to blindfolded sighted participants. They found that navigation was slower and more error-prone in the tactile condition. While both these studies found that navigation was slower when directed *via* tactile displays, neither study assessed improvement in performance with repeated testing or how performance was affected by conditions of high sensory load. We wondered whether tactile navigation, as it is less familiar to users, might be slower initially but become more efficient with practice.

Here, we extend this body of research. We compared the performance of blind participants as they navigated under the direction of auditory commands or tactile belt commands. Additionally, we assessed the effect of repeated testing in a controlled environment, in the presence or absence of simulated street sounds. For the reasons outlined above, we predicted that: (1) in the absence of street sounds and with sufficient practice, tactile navigation would be at least as effective as auditory navigation; and (2) in the presence of street sounds, tactile navigation would be superior to auditory navigation. The study's results, while intriguing, corresponded only partially to these predictions.

MATERIALS AND METHODS

Participants

We conducted experiments with 14 blind adults (11 men and three women, ranging in age from 21 to 60 years; mean, 39.9 years). Exclusion criteria ensured that blind participants did not have impairments known to affect tactile sensation, that blindness was of peripheral origin, that the participants' degree of vision did not exceed residual light perception (ability to perceive light but not form), and that no participant had diabetes, hearing problems, balance difficulties, tremor, epilepsy, multiple sclerosis, stroke, neurological disorders, learning disabilities, dyslexia, attention deficit disorders or cognitive impairments. All participants gave signed consent (consent form read aloud by an investigator) and received monetary compensation for their participation. All procedures were approved by the McMaster University Research Ethics Board.

The participants had no more than residual light perception, but their visual histories were quite varied. At one extreme were participants born with normal vision who then progressed through a stage of low vision (defined here as the ability to perceive both light and form) to reach residual light perception (perception of light but not form). At the other extreme were participants born with residual light perception

TABLE 1 | Participant characteristics.

	Participant Characteristics			
	Congenitally blind	Early blind	Late blind	Total
No vision	0	2	2	4
Residual light perception	3	0	5	8
			Total	12

or less. Defining childhood as the period between birth and 12 years of age, we classified four participants as congenitally blind (residual light perception or less at birth), two as early-blind (normal or low vision at birth declining to residual light perception or less by the end of childhood), and eight as late blind (normal or low vision throughout childhood, declining to residual light perception or less in adulthood). Nine participants had residual light perception at the time of testing and five had no light perception. Two participants were excluded from the analysis due to incomplete data as they were unable to complete the full experiment. Characteristics of the 12 included participants are summarized in **Table 1**.

Equipment

The experiment was conducted in a 16.5 ft. wide by 51 ft. long room in the Psychology Building on the campus of McMaster University. Sturdy foam mats (2 ft × 2 ft) were arranged throughout the room to delineate walkways. Four distinct walking paths were defined and equated for difficulty, each containing 10 (90°) turns with similar lengths (one path was 127 ft and the rest were 128 ft; **Figure 1A**). The equivalent difficulty of these paths was confirmed by the similar times required by participants to complete them (one-way ANOVA on completion time: $F = 0.249$, $p = 0.862$).

Following the consent procedures, the tactile belt was fitted to the participant. The belt attached to the torso with the help of Velcro straps. It contained ten vibratory coin motors, each of diameter 10 mm, arranged in pairs at regular intervals (**Figure 1B**). The belt had elastic segments that allowed it to fit each participant comfortably and rigid segments that allowed for proper anchoring of the circuitry and wiring; due to the belt's elasticity, the spacing between the motors scaled with the participant's waist size. The coin motors, similar to those in smartphones, vibrated by spinning an imbalanced mass at a high speed. The peak-to-peak displacement of the vibration produced by the motors was 1.5 mm, at a frequency of 55 Hz. The direction of the sweeping vibration was controlled by LabVIEW 2014 on a Windows PC and communicated to the belt using a Bluetooth 4.0. Once a control was initiated, motors centered on the participant's midline immediately began vibrating to cue participants for an upcoming turn. The motors subsequently vibrated consecutively at adjacent positions (0.3 s per location, 0 s ISI) in order to create a directional signal. Depending on the direction fed by the Bluetooth control, the vibration traveled from either midline to left or midline to right (**Figure 1B**). At the beginning of every tactile trial, the vibration traveled around the participant's torso twice to prompt the participant to begin the task.

Attached to the same belt were two small Bluetooth audio speakers. The smaller, circular speaker (diameter 8 cm × thickness 3.5 cm) was attached to the front of the belt on the participant's midline. Similar to the tactile belt, this speaker output navigational—"turn left" or "turn right"—female speech commands corresponding to the Bluetooth controls sent from the same LabVIEW program. To enable direct comparison between the command types, the duration of the auditory commands was also 1 s, and the "turn" portion of the auditory commands was analogous to the midline tactile vibration of the belt. Additionally, this speaker output a "please start" command at the beginning of every auditory trial to prompt the participant to begin.

The second, rectangular, speaker (14.5 × 7 × 2 cm) was attached to the back of the belt, behind the participant. Its function was to output background street noise, which was broadcast from an Android-device *via* Bluetooth. The street noise was played from this speaker mounted on the back of the belt, rather than a fixed speaker in the room because the use of a fixed speaker would provide spatial cues that could artificially facilitate the participant's learning of the path. The streets sound recording played on continuous run during a trial and was not synchronized in any way to the location of the participant or to the turn commands emitted from the front speaker. The full sound recording was of approximately 14.5 min duration; the recording was stopped at the end of each trial and restarted, where it left off, at the beginning of the next trial. The recording consisted of various sound events (e.g., conversation, car horn, a truck backing up, dog barking, car skidding). The average sound event duration was 7.3 s (SD 5.3 s). Events could overlap in time (e.g., the sounds of children playing might overlap with the sound of bicycle bells or adults speaking). During the full 14.5 min recording, 50 silent intervals were interspersed between sound intervals. The mean silent interval was 7.9 s (SD 4.4 s).

Sound pressure level (dB SPL) was measured with an i436 omnidirectional professional microphone (MicW) using SoundMeter X, v 10.1 running on an iPhone 6 s. With the microphone placed approximately 80 cm above the speaker to simulate the distance from a participant's belt to ears, the overall max level in repeated measurements averaged 76.1 dB for the street sounds, 77.5 dB for the turn left command, and 71.8 dB for the turn right command.

The participant localization system consisted of a grid of 6 laser beams distributed throughout the room (**Figure 1C**). The laser beams (5 mW, 650 nm, 20 mA red laser diodes) traveled parallel to the floor at a height of approximately 1 meter and passed through the participant's walking path at a distance of 3 feet prior to an edge of a mat connected to an intersection. Sensors (CDS Cell 690 nm 0.17 ∼2 kOhm @ 21 lux) detected the moment each beam was broken by the participant and relayed the change in voltage to the LabVIEW program *via* an NI USB-6008 I/O board. Upon receipt of the voltage signal, LabVIEW issued the Bluetooth signals to either the front speaker or belt, as appropriate to the experimental condition (system

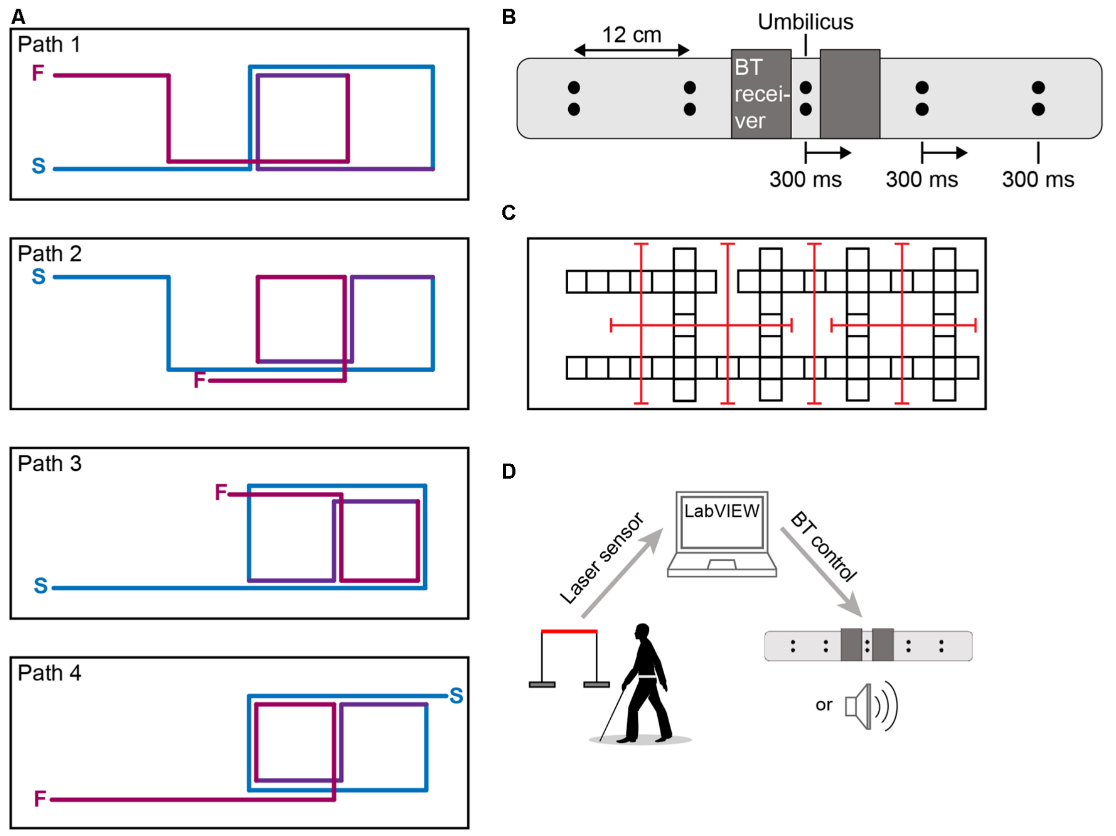

FIGURE 1 | Apparatus. **(A)** Four walking paths equated for difficulty (10 turns, ~127 ft in length; S = path start; F = path finish). Lines slightly offset and colored for clarity only. **(B)** Vibrotactile navigational belt schematic. Each dot represents a coin motor. Vibrotactile stimulation began below the umbilicus and moved towards the left or the right (a rightward movement is shown for illustration). **(C)** Room layout. Each square represents a 2 ft. × 2 ft. mat. Red lines show the participant localization laser transmitter-receiver grid. **(D)** Systems overview. Image of person walking with cane is modified from ID 45773820 © Anastasia Popova | Dreamstime.com.

overview; **Figure 1D**). Due to hardware latencies, the time delays between beam break and auditory command onset, and beam break and tactile vibration command onset, were 298 ± 5 ms and 153 ± 34 ms, respectively.

The LabVIEW program recorded the times taken to successfully complete each path and the errors made during waypoint navigation (i.e., any instances during which the participant missed a turn, turned in the wrong direction, or walked off the course in any way). In the event of an error, the participant was told to stop walking and was returned to the start of the path to begin again. Additionally, the navigational behavior of participants was recorded by two cameras connected to a separate Windows 8 based PC.

Navigational Testing

The testing consisted of four conditions in a 2 [navigational command type: tactile (T) vs. auditory (A)] by 2 [ambient noise: quiet (Q) vs. background street sounds (S)] repeated-measures experimental design (**Table 2**). This design allowed us to assess the efficacy of navigation with tactile commands compared to conventional auditory commands, under conditions of either quiet or background street sounds. Participants were given a 20-min practice phase prior to the commencement of testing.

TABLE 2 | Testing conditions.

	Tactile belt (T)	Auditory device (A)
Quiet (Q)	TQ, 5 Trials: Time and error	AQ, 5 Trials: Time and error
Sound (S)	TS, 5 Trials: Time and error, comprehension	AS, 5 Trials: Time and error, comprehension

During practice, they had the opportunity to become familiar with the tactile belt and auditory device and the foam mats that made up the paths and to gain a basic understanding of the navigational task. They did so by navigating through a practice path that was significantly different and shorter than the test paths. During the practice and all testing sessions, investigators stood silently at strategic locations within the room, in order to intervene if necessary (i.e., to tell participants to stop walking if they were off-path and in danger of colliding with a wall or other object).

For all participants, Quiet testing with both navigational command types preceded Sound testing. This was done for the safety of the participants, such that the participants faced less difficult tasks initially and would not be distracted from hearing the voices of the investigators, should it be necessary for safety reasons for the investigators to intervene. Half of the participants

TABLE 3 | Order of testing by participant.

Participant	Order of testing (Condition-Path) by participant			
	Testing order			
	1	2	3	4
1	AQ-1	TQ-2	AS-3	TS-4
2	TQ-1	AQ-2	TS-3	AS-4
3	TQ-2	AQ-1	TS-3	AS-4
4	AQ-2	TQ-1	AS-3	TS-4
5	TQ-1	AQ-2	TS-4	AS-3
6	AQ-1	TQ-2	AS-4	TS-3
7	TQ-2	AQ-1	TS-4	AS-3
8	AQ-2	TQ-1	AS-4	TS-3
9	TQ-3	AQ-4	TS-1	AS-2
10	AQ-3	TQ-4	AS-1	TS-2
11	TQ-3	AQ-4	TS-2	AS-1
12	AQ-3	TQ-4	AS-2	TS-1

completed conditions in the order A-Q, T-Q, A-S, T-S; the other half completed conditions in the order T-Q, A-Q, T-S, A-S (**Table 3**). Participants were required to successfully complete five trials in each condition before proceeding to the next condition. Each condition used a different path from among four designed paths. The paths corresponding to the four conditions were counterbalanced across participants so that all paths were equally used for each condition. Participants were required to take a minimum 2-min rest after each trial within a condition, and a 5-min rest period between conditions.

Participants were instructed to walk at their normal, comfortable speed and to use their white cane as they typically would while navigating a path. They were instructed to walk straight along a path until receiving a turn command, and to make 90° left or right turns upon receipt of the corresponding left or right command. Participants were told to try to avoid errors (i.e., stepping off the path or making a wrong turn). Except for the mode of command (vibrations delivered by the belt or auditory instructions issued *via* a belt-attached portable speaker), the protocol was identical in the tactile and auditory command conditions.

At the end of each trial in a Sound condition, a co-investigator questioned the participant as to what street sounds (s)he heard within the timeframe of the last trial. All recall questionnaires were identical, asking if the participant heard a specific event. Participants were required to choose from "yes," "no," or "unsure" for every question. On any particular trial, sounds may have included dogs barking, car horns, large truck back-up beeps, ambulance/police sirens, bike bells, and/or people talking.

At the end of the experiment, the participant was asked to respond to a series of follow-up questions regarding their experience with the tactile system in comparison to the auditory commands, in order to provide ideas for the future development of the device.

Navigational Data Collection and Analysis

During navigation, the time at which the participant broke each laser beam was automatically recorded by the computer program. Major navigational errors were defined as failing to respond to navigational instructions or making wrong turns. These errors were picked up by the beam/sensor system and automatically recorded by the computer. Minor navigational errors were defined as stepping off the path such that more than half a foot was off the path. These errors were recorded by two co-investigators who were situated in the corners of the room and observed the participant visually. The number of minor errors recorded by the two co-investigators was averaged if the tally differed among the co-investigators. If the difference was large, the video recording of the corresponding trial was viewed to determine the correct number of minor errors. The path completion time was recorded by the computer as the time elapsed between the first and final beam breaks.

We performed repeated-measures ANOVAs with type III sum of squares and two-tailed t-tests using SPSS Statistics version 25 (IBM) for Windows with an alpha level of 0.05, in order to assess the effects on the dependent measures of command type (i.e., Tactile vs. Auditory), ambient noise (Quiet vs. Sound), and repeated testing (i.e., trial number). For the purpose of the three-way repeated-measures ANOVA on minor errors, if a particular trial number was terminated due to a major error and consequently re-run one or more times, we averaged the number of minor errors from the runs.

RESULTS

We assessed the ability of blind participants to navigate paths using either tactile or auditory commands and in the absence or presence of background street sounds. We measured navigational performance as the time taken to complete each trial and the number of errors committed.

Participants Committed More Major but Not Minor Errors When Using the Tactile Belt

All 12 participants completed the five trials per condition successfully within the allotted experimental time. However, the navigation task was somewhat challenging, as indicated by the observation that every participant had at least one major error (i.e., missed turns or wrong turns); consequently, every participant was required to repeat a trial at least once. The mean number ($\pm$ SE) of major errors produced by the participants across all conditions was 4.0 ± 0.8. At the extremes, P5 and P11 committed only one error each, whereas P3 and P6 committed eight errors each. For minor errors, the mean was 29.8 ± 4.1, with the extremes being P7 with three errors and P12 with 51 errors.

The average number of major and minor errors committed per participant in each testing condition is shown in **Table 4**. Participants committed more major errors under tactile than auditory commands, in both the Quiet and Sound conditions. Additionally, participants tended to commit more major errors, within each command type condition, with the addition of background street sound. Nevertheless, a two-way (command type $\times$ ambient noise) repeated-measures ANOVA on committed major errors indicated a significant effect of command type ($F = 5.046$, $p = 0.046$) only, with no significant

TABLE 4 | Major and minor errors.

Major and minor errors (Mean ± SE) by condition averaged across participants			
Ambient noise	**Command type**	**Major errors**	**Minor errors**
Quiet (Q)	Tactile (T)	1.25 ± 0.46	7.88 ± 1.54
	Auditory (A)	0.33 ± 0.22	8.46 ± 1.49
Sound (S)	Tactile (T)	1.58 ± 0.43	6.96 ± 1.05
	Auditory (A)	0.83 ± 0.37	6.50 ± 1.15

effect of ambient noise ($F = 1.244$, $p = 0.288$). We did not analyze the effect of trial number on major errors, as the number of major errors committed was too small to support such an analysis.

In contrast to the result with major errors, the number of minor errors did not differ significantly between the two command types. A 2 × 2 × 5 (command type × ambient noise × trial) three-way repeated-measure ANOVA on minor errors indicated no effect of command type ($F = 0.203$, $p = 0.660$) or trial ($F = 0.224$, $p = 0.924$) and no significant two-way interactions. Participants committed significantly fewer minor errors in the street sound condition ($F = 6.943$, $p = 0.022$), perhaps reflecting an effect of practice, as the Sound condition occurred in the second half of the experiment.

In summary, participants committed fewer major but not minor errors when using auditory navigational commands, and performance did not significantly worsen with the addition of background street sounds.

Recall Performance Was Equivalent for the Two Command Types

We next compared the ability of participants to recall events from the background street sounds (Sound conditions) while navigating with either device. Most participants performed well on the recall questionnaire signifying that they were actively attending to the background street noise. A two-way (command type × trial) repeated-measures ANOVA verified that there was no significant effect of either command type or trial ($F = 0.441$, $p = 0.520$; $F = 0.224$, $p = 0.923$) on the number of correct responses. These results indicate that participant recall was equivalent across navigational devices, signifying that participants were equally and actively attending to the background street noise while navigating with both devices. Additionally, participants' ability to recall events from their immediate environment did not change with practice or increasing number of trials.

Improvement With Practice Was Statistically Similar in Auditory and Tactile Navigation

The time taken by the participants to complete each of the five navigational trials in the four conditions is shown in **Figure 2**. Completion times consistently diminished as a function of trial number, indicating that participants improved with practice in every condition.

For each participant and each condition, we determined the best-fit line relating completion time to trial number by linear regression; the slopes of these best-fit lines indicate the improvement in completion time with practice (**Figure 3**). We investigated whether the slopes differed across conditions. A two-way (command type × ambient noise) repeated-measures ANOVA on slope revealed a significant effect of ambient noise ($F = 23.886$, $p < 0.001$) with no significant effect of command type ($F = 0.008$, $p = 0.931$) and no significant ambient noise × command type interaction ($F = 0.615$, $p = 0.450$). The slopes in the Quiet conditions were steeper than in the Sound conditions, indicating greater improvement with repeated testing in the Quiet conditions. This difference in the rate of improvement may have occurred because the Quiet conditions came first in the experiment. The non-significant effect of command type indicates a similar rate of navigational improvement with the two devices.

In the Absence of Ambient Noise, Auditory Navigation Was Faster Than Tactile Navigation

The participants' mean completion times for each trial of each condition are shown in **Figure 4A**. A 2 × 2 × 5 (command type × ambient noise × trial) three-way repeated-measure ANOVA on the completion times revealed a highly significant effect of trial ($F = 28.639$, $p < 0.001$), a significant effect of command type ($F = 6.678$, $p = 0.025$), and a significant effect of ambient noise ($F = 5.066$, $p = 0.046$). The ANOVA further revealed a significant command type × ambient noise interaction ($F = 7.004$, $p = 0.023$).

Figure 4A suggests that the significant interaction was due to a larger difference in completion times between auditory and tactile command types in Quiet than in Sound. To investigate this, we conducted two separate *post hoc* two-way (command type × trial) repeated-measures ANOVAs, one for Quiet and one for Sound. These ANOVAs revealed a significant effect of command type in Quiet ($F = 7.676$, $p = 0.018$) but not in Sound ($F = 4.028$, $p = 0.070$). As expected, the effect of trial was highly significant in both cases ($F = 22.413$, $p < 0.001$ and $F = 8.480$, $p < 0.001$, respectively).

Collectively, these analyses indicate that participants navigated more slowly when using the tactile belt than the auditory device, particularly in the Quiet conditions.

Auditory Navigation Was More Compromised by the Introduction of Background Street Noise

Figure 4A indicates that participants' performance was disrupted (i.e., completion time jumped upward) with the introduction of ambient noise (see dotted line connecting trials 5 and 6). Interestingly, the data suggest that ambient noise adversely affected navigation with the auditory device more than it did navigation with the tactile device. Two *post hoc* pairwise comparisons confirmed this impression. The increase in completion time from Auditory trial 5 ($M = 46.68$, $SD = 11.52$) to Auditory trial 6 ($M = 53.96$, $SD = 11.88$) was highly significant ($t_{(11)} = 8.82$, $p < 0.001$), whereas the increase in completion time from Tactile trial 5 ($M = 52.21$, $SD = 12.03$) to Tactile

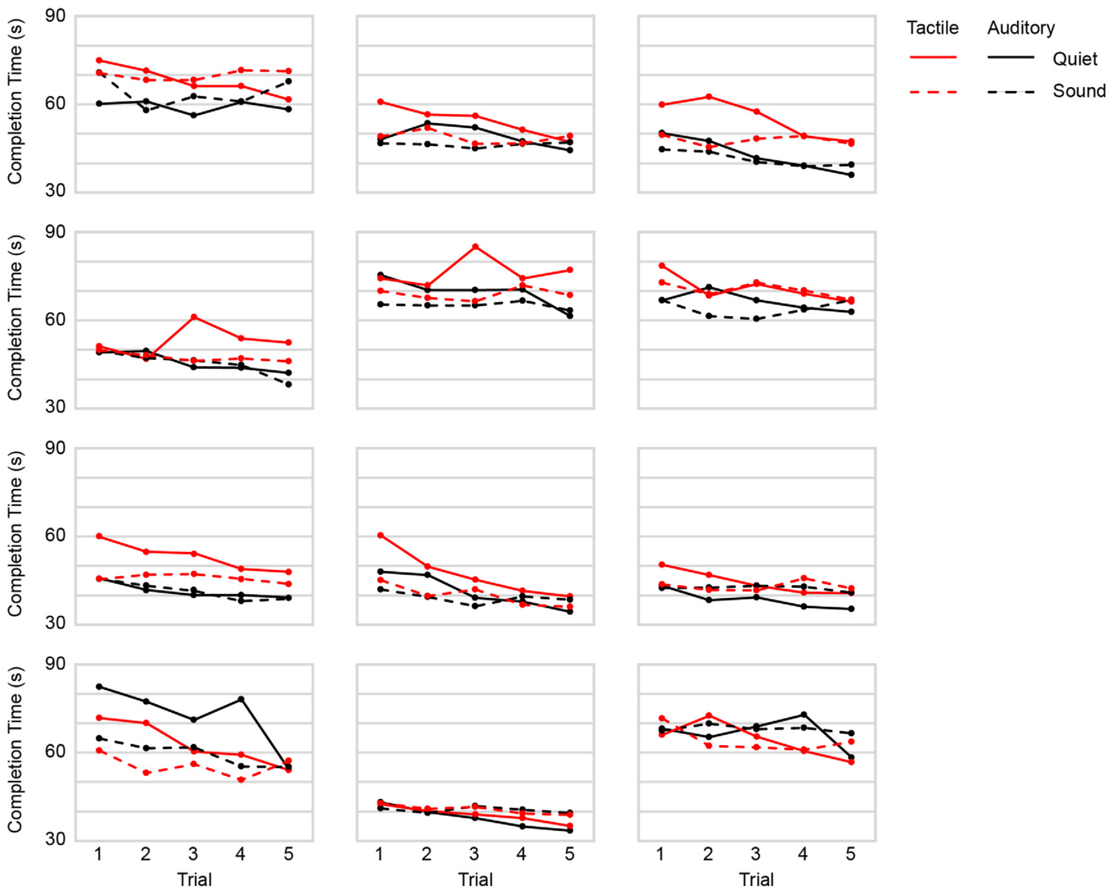

FIGURE 2 | Completion times as a function of trial by condition. Panels: individual plots of all participants (n = 12). Red: tactile navigational commands. Black: auditory navigational commands. Solid: quiet. Dashed: street sound.

trial 6 (M = 55.98, SD = 12.22) was only marginally significant ($t_{(11)} = 2.18$, $p = 0.052$).

Additionally, **Figure 4A** suggests that completion times under Auditory commands—but not Tactile commands—remained compromised after the introduction of ambient noise, even with repeated testing over five trials. This was confirmed with two *post hoc* pairwise comparisons. The increase in completion times from Auditory trial 5 (M = 46.68, SD = 11.52) to Auditory trial 10 (M = 50.20, SD = 12.80) was significant ($t_{(11)} = 3.30$, $p = 0.007$), whereas the difference in completion time from tactile trial 5 (M = 52.21, SD = 12.03) to tactile trial 10 (M = 52.59, SD = 12.41) was not significant ($t_{(11)} = 0.25$, $p = 0.808$).

To further investigate these trends, we replotted the data from **Figure 4A** after dividing each participant's completion times by the completion time on the first trial of the corresponding command type (**Figure 4B**). The normalized completion times for the two navigational devices appear to follow very similar courses as participants improved with practice in the Quiet conditions, but the completion times appear to worsen more for the auditory device in the Sound conditions. This was confirmed using a 2 × 2 × 5 (command type × ambient noise × trial) three-way repeated-measures ANOVA on the normalized completion times. The ANOVA revealed a significant command type × ambient noise interaction ($F = 9.978$, $p = 0.009$) and a significant effect of trial ($F = 32.300$, $p < 0.001$) but no significant effect of command type ($F = 2.210$, $p = 0.165$) or ambient noise ($F = 4.491$, $p = 0.058$). To further investigate the command type × ambient noise interaction, we conducted two *post hoc* two-way (command type × trial) repeated-measures ANOVAs, one for Quiet and the other for Sound. In keeping with the visual impression provided by **Figure 4B**, these ANOVAs revealed no significant effect of command type in Quiet ($F = 0.085$, $p = 0.776$) but a significant effect of command type in Sound ($F = 5.815$, $p = 0.035$).

These results indicate that background street noise compromised participants' ability to navigate with auditory commands more than it compromised their ability to navigate with tactile commands.

Straightaway and Turn Speeds Under Auditory Navigation Were More Compromised by Background Street Noise

We next sought to determine how participants' navigational behavior changed to account for the changes in completion

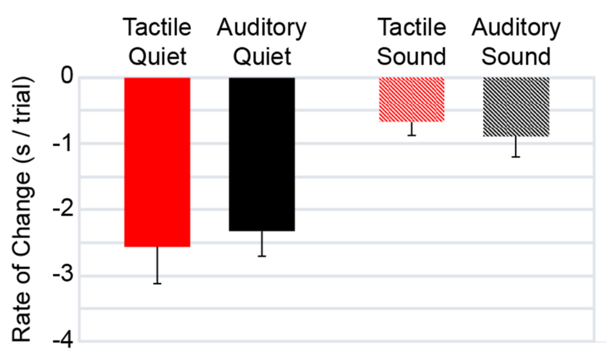

FIGURE 3 | Rate of change in completion time by condition. Bars: mean regression slopes derived from completion times as a function of trial (Note: negative slopes indicate improvement—i.e., reduction—in completion time). Red: tactile navigational commands. Black: auditory navigational commands. Solid: quiet. Hatched: street sound. Errors bars: 1 SE.

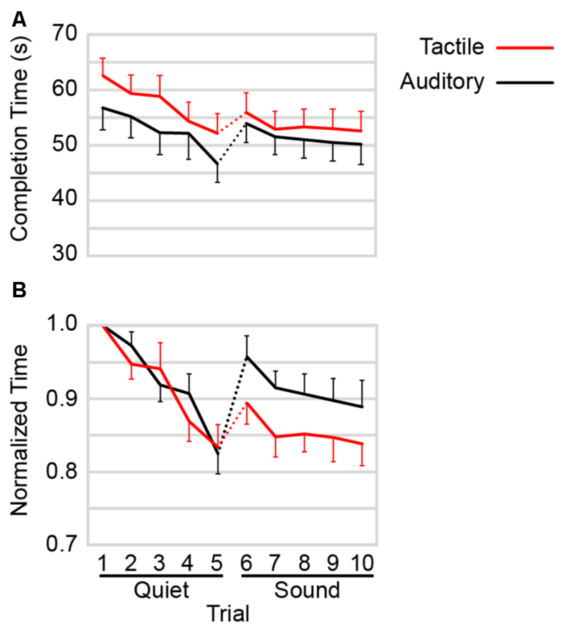

FIGURE 4 | (A) Mean completion time as a function of trial by command type. Data from **Figure 2** were replotted by averaging across all participants ($n = 12$) for each condition. **(B)** Mean of completion times normalized against trial 1. For each participant, completion time on each trial was divided by the completion time from trial 1 of the corresponding command type. Red: tactile navigational commands. Black: auditory navigational commands. Dotted lines: background street sounds introduced into the experiment. For visual clarity, error bars show +1 SE and −1 SE, respectively for the highest and lowest points at each comparison distance.

times from Quiet to Sound observed in **Figure 4**. To this end, we focused on how participants adjusted their walking speed on the straightaway and turn components of the paths (**Figure 5**). Not surprisingly, the data indicate that participants tended to walk more rapidly on the straight portions of the path than they did when turning. Separate 2 × 2 × 5 (walking direction × ambient noise × trial) repeated-measures ANOVAs on the auditory and tactile navigation speeds revealed a highly significant main effect of walking direction (i.e., straight vs. turn) in both cases (auditory navigation: $F = 33.794$, $p < 0.001$; tactile navigation: $F = 28.698$, $p < 0.001$).

Both turning and straightaway speeds under auditory commands diminished by a larger magnitude—relative to tactile—with the introduction of background street noise (**Figure 5**). With the introduction of background street noise, turning speeds under auditory commands decreased (trial 5 to trial 6) from 2.52 ± 0.61 to 2.17 ± 0.41 ft/s, whereas turning speeds under tactile commands decreased from 2.13 ± 0.45 to 1.98 ± 0.36 ft/s (**Figure 5A**). Similarly, straightaway speeds under auditory commands decreased from 2.94 ± 0.71 to 2.40 ± 0.60 ft/s, whereas straightaway speeds under tactile commands decreased from 2.60 ± 0.71 to 2.27 ± 0.63 ft/s (**Figure 5B**). Thus, when participants were being guided by auditory commands, their walking slowed more noticeably with the introduction of background street noise.

These trends were confirmed by two 2 × 2 × 5 (command type × ambient noise × trial) three-way ANOVAs on turning and straightaway speeds. For turning speeds, the ANOVA revealed significant effects of command type ($F = 16.276$, $p = 0.002$), ambient noise ($F = 7.107$, $p = 0.022$), and trial ($F = 22.248$, $p < 0.001$), with a marginally significant command type × ambient noise interaction ($F = 4.637$, $p = 0.054$). Similarly, for straightaway speeds, the ANOVA revealed significant effects of command type ($F = 14.143$, $p = 0.003$) and trial ($F = 22.105$, $p < 0.001$), with a significant command type × ambient noise interaction ($F = 5.757$, $p = 0.035$); the effect of ambient noise on straightaway speeds was not significant ($F = 0.008$, $p = 0.930$).

To further investigate these trends, we replotted the data from **Figures 5A,B** after dividing each participant's speeds by the speed on the first trial of the corresponding command type (**Figures 5B,C**). The normalized speeds for auditory and tactile navigation followed similar courses as participants improved with practice in the Quiet, but the speeds appeared to worsen more for the auditory device in Sound. Confirming this visual impression, a 2 × 2 × 5 repeated-measures ANOVA on the normalized turn speeds revealed a significant command type × ambient noise interaction ($F = 5.084$, $p = 0.046$). A 2 × 2 × 5 repeated-measures ANOVA on the normalized straightaway speeds revealed a non-significant interaction trend ($F = 3.885$, $p = 0.072$).

Collectively, these results suggest that ambient noise caused more interference with auditory than tactile navigation.

The Participant Report Supported the Potential Usefulness of the Tactile Device

Results from the end-of-experiment questionnaire are displayed in **Table 5**. Participants responded strongly disagree, disagree, neutral, agree, or strongly agree to the following statements: (1) Overall, I think the belt would be helpful for navigation; (2) The signals given by the belt were clear and easy to feel; (3) I would find it easy to integrate the belt into my usual

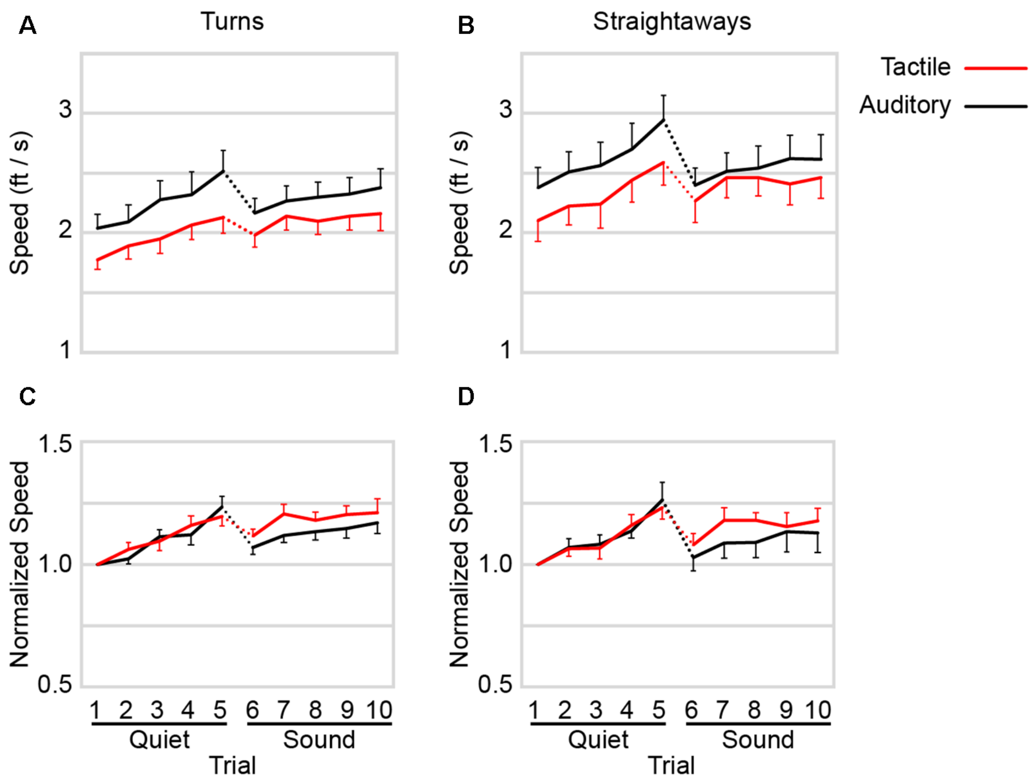

FIGURE 5 | Navigational speed. **(A)** Mean turning speeds as a function of trial by command type. **(B)** Mean straightaway speeds as a function of trial by command type. **(C)** Mean normalized turning speeds. For each participant, turning speed on each trial was divided by the turning speed from trial 1 of the corresponding command type. **(D)** Mean normalized straightaway speeds. For each participant, straightaway speed on each trial was divided by the straightaway speed from trial 1 of the corresponding command type. Dotted lines: background street noise introduced to experiment. Red: tactile navigational commands. Black: auditory navigational commands. Error bars show +1 SE and −1 SE, respectively for the highest and lowest points at each comparison distance.

travel routines, using it in conjunction with my cane, guide dog or human guide; (4) The belt was comfortable to wear; and (5) I'd be better able to attend environmental sounds (traffic, someone talking, etc.) with the belt than with an audio navigation system. As indicated in the table, a clear majority of participants agreed or strongly agreed with these statements. Participants were particularly positive concerning the potential helpfulness of the belt for navigation (nine out of 12 participants—75%—strongly agreed, and the remaining three agreed). Ten of the 12 participants either agreed or strongly agreed that they would be better able to attend to environmental sounds when using the belt than an audio navigation system.

DISCUSSION

The ability to travel independently is crucial to an individual's quality of life but compromised by visual impairment. Several navigational aids have been developed for blind people to address this limitation. These devices typically employ auditory instructions to guide users to desired waypoints (Loomis et al., 2005; Gaunet, 2006). However, the use of auditory navigational commands may interfere with users' awareness of their surroundings, with potentially detrimental consequences. There is an obvious need, then, to explore the use of alternative, under-utilized, sensory modalities to convey information for safe and independent travel. As spatial information can be readily

TABLE 5 | Questionnaire responses.

Statement	End-of-experiment questionnaire responses				
	Response				
	Strongly disagree	Disagree	Neutral	Agree	Strongly agree
1	0	0	0	3 (25%)	9 (75%)
2	0	1 (8%)	2 (17%)	4 (33%)	5 (42%)
3	0	0	0	5 (42%)	7 (58%)
4	0	0	0	8 (67%)	4 (33%)
5	0	0	2 (17%)	4 (33%)	6 (50%)

conveyed to the skin and interpreted by the nervous system, tactile navigational aids would seem to hold particular promise. In the present study, we compared the efficacy of a novel tactile navigational aid and a conventional auditory aid. We predicted that: (1) in the absence of environmental sounds, navigation with the tactile aid would, with sufficient practice, be at least as good as navigation with the auditory aid; and (2) navigation with the tactile aid would be less impaired by concomitant attention to environmental sounds.

The data, while promising, offer a more nuanced view than we had envisaged. To our surprise, we found that, when participants navigated in a silent environment (Quiet conditions), they performed somewhat worse with the tactile belt than the auditory device, taking longer to complete each trial and committing more major errors. When participants navigated in the presence of simulated street noises (Sound conditions), the difference in completion time between auditory and tactile navigation diminished. These results suggest that tactile navigation, although not initially intuitive to the participants, holds promise as an effective navigational method in everyday environments characterized by ambient noise such as street sounds.

Despite the Predicted Superiority of the Tactile Compared to the Auditory Modality for Navigational Processing, Participants Performed Worse When Using the Tactile Belt in the Quiet Conditions

Our findings support previous literature (Ertan et al., 1998; Tsukada and Yasumura, 2004; Van Erp et al., 2005; Pielot and Boll, 2010; Flores et al., 2015; Jimenez and Jimenez, 2017) revealing that tactile displays can successfully guide participants to waypoints. The present study also extends upon this body of literature, by comparing tactile to auditory navigational performance of blind participants in the presence or absence of street noise, and by providing participants with the opportunity to improve performance with repeated testing trials in every condition. Despite these differences in design, like Flores et al. (2015) and Jimenez and Jimenez (2017), we found that participants performed more slowly (in the Quiet conditions) when using the tactile belt than the auditory navigational device. Consistent with Jimenez and Jimenez (2017), we found also that participants made more errors in the tactile condition. These results contradicted our first prediction.

Spatial information is relayed from the skin through the central nervous system *via* topographically organized projections and is represented in somatotopic maps. In light of this spatial fidelity of the somatosensory system, it is not surprising that humans can extract spatial information from the positions of vibrotactile cues (Cholewiak et al., 2004; Van Erp, 2005; Jones and Ray, 2008). Accordingly, it would seem that the somatosensory system is ideally suited to extract navigational directions from vibrotactile stimulus patterns. By contrast, the perception of "turn left" or "turn right" commands requires acoustic, phonological and semantic processing across several separate brain regions, broadly distributed over the right and left hemispheres (Connolly and Phillips, 1994), suggesting that verbal commands require a greater degree of processing before their meaning can be extracted and translated into a spatial direction.

In light of these considerations, it surprised us that our participants generally performed better when using auditory commands than the tactile belt. This difference in performance may be attributed to the novelty of using a tactile device. Novel procedures often induce a cognitive load, resulting in diminished task performance (Sweller, 1994; Brunken et al., 2003; Haji et al., 2015). Participants in this study, who had little or no prior experience navigating with tactile commands, presumably had to process tactile instructions cognitively to a greater extent than an experienced user would have. In this vein, it is worth noting that several participants suggested that future versions of the tactile belt be accompanied by an intensity control to modulate the strength of vibration, as the participants had to expend effort to attend to the vibrations and sometimes missed them.

Improvement With Practice Occurred at the Same Rate for the Two Command Types

Previous navigational studies (Pielot and Boll, 2010; Flores et al., 2015; Jimenez and Jimenez, 2017) did not investigate improvement with repeated testing. We did so by testing participants over five consecutive trials on the same path in each condition. We found that participants improved navigation at the same rate regardless of the command modality. A parsimonious explanation for this finding is that participants did not experience more difficulty acquiring information *via* one command modality than the other, such that the learning rate was constrained, not by the command type, but by the efficiency with which the participants were able to acquire a mental map of the spatial layout of the path.

As the participants repeatedly used the tactile belt, the intuitiveness of the tactile information presumably allowed them to build a spatial representation of the navigated path without excessive cognitive load (Brayda et al., 2013). The similar rates of improvement highlight the efficacy of the tactile belt in providing blind participants a spatial representation of their surroundings. These results suggest that, with practice, blind users could learn to efficiently navigate the real world with the tactile belt system.

The Tactile Belt Benefited Users in the Presence of Background Street Noise

Unlike several previous tactile waypoint studies (but similar to Flores et al., 2015), our study simulated a realistic environment by adding ambient street noise to the navigational task (Sound conditions). Participants were asked to recall events from the street sounds as an incentive to actively attend to those sounds. This procedure simulated scenarios in which, for safety purposes, blind travelers must listen attentively to their immediate surroundings while navigating. We wondered how participants would fare while navigating with either the tactile belt or auditory device in the presence of high auditory load. We predicted that, when navigational instructions are processed through the tactile modality, the consequent mitigation of auditory load would result in two benefits: (1) superior navigational performance; and (2) better recall of the street sounds. Interestingly, although the results from our Sound conditions did not support these predictions, we found trends that strongly suggested the benefit of using tactile commands in an acoustically rich environment.

The primary findings from the Sound conditions would first seem to contradict our predictions, as the auditory performance was still marginally—but non-significantly—superior to tactile performance. However, as previously discussed, the results obtained from the present study were likely skewed to favor auditory commands due to the greater cognitive load associated with the novel tactile device. Hence, we considered it informative to investigate how performance changed from Quiet to Sound (**Figure 4A**). The results suggest that tactile navigational performance was less affected by high environmental auditory load. Specifically, tactile performance was less compromised with the introduction of background street noise as navigation completion times only slightly increased but subsequently improved to their previous levels by the end of the experiment. By contrast, the auditory performance was more compromised with the introduction of background street noise and failed to return to its previous levels. The normalized completion time data (**Figure 4B**) make apparent the extent to which auditory navigation was more compromised by background street noise, as do the normalized speed data (**Figures 5C,D**).

We consider two plausible explanations for why auditory navigation was more compromised by the introduction of background street noise. First, auditory commands and background street noises, when simultaneously present, may have interfered physically at the level of the acoustic waveform presented to the ears, such that in some cases the auditory command signal was physically corrupted or masked by a concomitant street sound. Second, even when physical interference did not occur, task performance may have been compromised in the complex acoustic environment consisting of both auditory commands and street noises, as the concurrent processing of two acoustic inputs may burden shared neural resources. In contrast, distinct sensory modalities may engage independent neural resources as well as shared ones (Wickens, 2008). Consequently, the processing of concurrent inputs—such as navigational commands and street sounds—may be achieved with less difficulty when the inputs are delivered through separate modalities (Duncan et al., 1997; Martens et al., 2010).

Future Directions

The present study provides a proof of concept for a tactile navigational belt for blind individuals. The belt successfully guided users to waypoint destinations, while leaving the auditory modality to attend to environmental sounds. Although participant performance was somewhat better overall with the conventional auditory navigational device than with the novel tactile belt, the data show that performance with the belt improved upon repeated testing and suggest that navigation with the belt was less impaired by the presence of attention-demanding environmental sounds. These findings suggest that tactile navigation systems hold promise and should be further investigated and refined. In particular, future studies should be conducted in order to optimize stimulus timing and intensity, which can exert strong effects on spatial perception (Tong et al., 2016). More sophisticated tactile commands could be implemented, *via* the use of additional actuators, for instance, commands that include turn angles other than 90-degrees. In addition, long-term training should be conducted in order to measure participants' asymptotic performance with the tactile navigational system. Previous research has shown that tactile acuity improves with training (Wong et al., 2013) and that blind individuals have the capacity for enhanced tactile processing (Bhattacharjee et al., 2010; Wong et al., 2011). Accordingly, we expect that, with sufficient practice, blind users would be able to integrate a tactile belt system seamlessly into their daily navigational activities. Ultimately, a tactile belt system may be combined with other advances, such as a technology-enhanced white cane (Khan et al., 2018), to form an integrated navigational assistance system.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by McMaster University Research Ethics Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

SS was part of a team that conceptualized, designed, and built the tactile belt device. AB, SS and DG designed the study and wrote the manuscript. AB and SS conducted the experiments. AB and DG analyzed the data.

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Conflict of Interest: SS was a member of a team that designed and constructed the vibrotactile belt system used in this study. His team has founded a startup company that may market a tactile navigational device; the company is currently at the product verification stage.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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A Novel Training Program to Improve Human Spatial Orientation: Preliminary Findings

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The ability to form a mental representation of the surroundings is a critical skill for spatial navigation and orientation in humans. Such a mental representation is known as a "cognitive map" and is formed as individuals familiarize themselves with the surrounding, providing detailed information about salient environmental landmarks and their spatial relationships. Despite evidence of the malleability and potential for training spatial orientation skills in humans, it remains unknown if the specific ability to form cognitive maps can be improved by an appositely developed training program. Here, we present a newly developed computerized 12-days training program in a virtual environment designed specifically to stimulate the acquisition of this important skill. We asked 15 healthy volunteers to complete the training program and perform a comprehensive spatial behavioral assessment before and after the training. We asked participants to become familiar with the environment by navigating a small area before slowly building them up to navigate within the larger and more complex environment; we asked them to travel back and forth between environmental landmarks until they had built an understanding of where those landmarks resided with respect to one another. This process repeated until participants had visited every landmark in the virtual town and had learned where each landmark resided with respect to the others. The results of this study confirmed the feasibility of the training program and suggested an improvement in the ability of participants to form mental representations of the spatial surrounding. This study provides preliminary findings on the feasibility of a 12-days program in training spatial orientation skills. We discuss the utility and potential impact of this training program in the lives of the many individuals affected by topographical disorientation as a result of an acquired or developmental condition.

Keywords: getting lost, hippocampus, memory, plasticity, rehabilitation, virtual reality

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INTRODUCTION

Human topographical orientation is a complex behavior that serves a critical role in daily functioning: it provides the necessary skills to find the way around in both familiar and novel surroundings and allows individuals to move within the environment with a spatial purpose in mind (Marchette et al., 2011). Such a foundational skill could rely on different orientation strategies involving the use of environmental landmarks, as well as the memorization of given routes independently of landmarks encountered on the way (Siegel and White, 1975; Aguirre and D'Esposito, 1999). Among the many strategies adopted for orientation is the ability to form and make use of cognitive maps (Tolman, 1948). A cognitive map can be formed when an individual has gained configurable knowledge of an environment (understanding the location of landmarks in space with respect to each other) allowing the formation of a mental representation of the surrounding (Farran et al., 2015). This mental representation is formed as individuals become familiar with the surrounding, providing detailed information about salient environmental landmarks and their spatial relationships (Epstein et al., 2017). Cognitive maps are very critical to successful orientation since, once formed, they allow individuals to reach any target location from anywhere within the environment, and even permit generating alternative, unexplored routes if required by environmental circumstances (Arnold et al., 2013).

Cognitive maps are critical for orientation by enabling the planning of routes outside of the visible surroundings (i.e., vista space), which allows individuals to orient from all possible locations within a large-scale environment (i.e., environmental space; Montello, 1993; Wolbers and Wiener, 2014; Epstein et al., 2017). The formation and use of cognitive maps, however, is known to be affected by factors such as age and sex (Liu et al., 2011), as well as unknown factors that create a large amount of variability in the general population (Weisberg and Newcombe, 2018). For instance, in a very recent study, Yamamoto et al. (2019) compared the ability of young and older adults to learn an environment from both an aerial (top-down) perspective and a first-person perspective. They asked participants to watch videos showing a large-scale environment from a first-person perspective moving through the environment, an aerial view changing orientations, and an aerial view in a fixed orientation. The results confirmed that older adults learned an environment better from the fixed aerial perspective as compared to the first-person perspective, and from a fixed aerial perspective as compared to a rotating aerial perspective. This contrasts with young adults who revealed no difference in their ability to learn the environment between the three different perspectives. These results suggest that spatial strategies can differ throughout adulthood, where older adults may start to lose the ability to form new cognitive maps in a first-person perspective (Iaria et al., 2009b), likely due to structural changes in the hippocampus (Iaria et al., 2008), explaining the natural cognitive decline of topographical orientation skills occurring with aging (Wiener et al., 2013).

The critical role of cognitive maps for effective orientation and navigation in daily life is also confirmed by the lack of orientation skills as experienced by individuals affected by Developmental Topographical Disorientation (DTD; Iaria et al., 2009a; Bianchini et al., 2014; Barclay et al., 2016; Conson et al., 2018). DTD refers to a lifelong condition in which individuals get lost in extremely familiar surroundings despite well preserved general cognitive skills, no brain injuries, and no cognitive complaints (Iaria and Barton, 2010). As documented in many cases, the most common cause of topographical disorientation in individuals affected by DTD is the complete inability to form cognitive maps, confirming the important role of this skill for daily effective navigation (Iaria and Burles, 2016).

Despite the fairly well-known behavioral mechanisms underlying the ability to orient by means of cognitive maps, to date, very little is known about the potential effects of training programs that may help to improve such important skills. Although there is evidence that spatial skills are moderately malleable and could be improved by a specifically designed training program (Uttal et al., 2013), these studies have focused solely on improving performance on lower-level spatial functions such as mental rotation, visual selective attention, spatial problem solving, and landmark recognition (Subrahmanyam and Greenfield, 1994; Feng et al., 2007; Cherney, 2008; Lövdén et al., 2012; Nemmi et al., 2017; Xiao et al., 2018; Veurink and Sorby, 2019; Yamamoto et al., 2019). Consequently, this leaves a gap in the literature regarding whether or not it is possible to train the relatively higher-level ability to form cognitive maps. For example, Lövdén et al. (2012) analyzed the effects of a long-term virtual spatial training program on a variety of spatial outcome measures including mental rotation, route memory, and other lower-level spatial skills, but did not administer any tests that were designed to measure the ability to use cognitive maps. Additionally, because their training program utilized a constantly changing environment, it would have been difficult for a participant to develop an accurate, consolidated, cognitive map of the training environment at any given time. The latter highlights a second limitation found in previous literature, where training programs have been designed to train spatial skills other than cognitive map formation. Similarly, Montana et al. (2019) conducted a review on the use of virtual environments for rehabilitating spatial memory in stroke victims. Of the 16 studies analyzed in the review, none used a training program designed appositely to train cognitive map use, focusing instead on skills such as basic day-to-day living (e.g., go to the grocery store, shop for a list of groceries, pay with the right amount of money, and take the groceries home), route-based navigation, or exploration of small environments such as a house or grocery store. Consistent with previous literature, none of the 16 studies used any cognitive map outcome measures.

Here, we present a newly developed training program similar to those described in previous studied (e.g., Lövdén et al., 2012; Uttal et al., 2013; Montana et al., 2019), with the exception that this program was conceived specifically to improve the ability of the individuals to form cognitive maps in a video game-like virtual environment. To evaluate the feasibility of the training program, we asked participants to become familiar with the environment by navigating a small area before slowly building them up to navigate within the larger and more complex environment; we asked them to travel back and forth between environmental landmarks until they had built an understanding of where those landmarks resided with respect to one another. This process repeated until participants had visited every landmark in the virtual town and had learned where each traveled resided with respect to the others. A key component of this training program, described further in the methods section, is the unique task design aimed at mimicking the natural way cognitive map formation is acquired throughout development (Siegel and White, 1975). We expected that participating in the 12-days training program would result in a significant improvement of performance on a test measuring the specific ability to form cognitive maps (i.e., the Spatial Configuration Task, SCT; Burles et al., 2017). Additionally, we hypothesized that this difference would be greater than what would be found as a result of a test-retest effect in a non-training group.

MATERIALS AND METHODS

Participants

We recruited 15 healthy volunteers (10 females; mean age = 34 years, ranging from 19 to 70 years), and referred to them as the "training group." These individuals were recruited through advertisements on the University of Calgary website. After training participants in the experimental group, we recruited 23 participants (21 females, mean age = 20 years, ranging from 18 to 24 years) to be part of a *post hoc*, untrained group to control for any test-retest effects that may explain significant changes in spatial skills detected in the training group. These individuals were recruited through the resident psychology recruitment system at the University of Calgary where students are incentivized to partake in research studies for course credit. This group was only intended to address the testing effect on statistically significant changes detected in the training group. Thus, participants in this group only performed the SCT and did not complete any of the other tasks (i.e., the mental rotation and the Four Mountains Task; see descriptions below). Additionally, it should be noted that this group differs greatly in age range compared to the experimental group. Previous literature has shown that spatial skills are strongest in early adulthood, i.e., from ages 18–30 (Liu et al., 2011). With this in mind, we aimed to recruit individuals within this age range into the testing effect group in order to ensure that they had the highest chance of revealing a testing effect.

Participants in both groups were asked to complete a questionnaire reporting general demographics, neurological conditions or brain damage, anxiety (Marteau and Bekker, 1992) and depression (Kroenke et al., 2001) symptoms. We found that the training group did not vary significantly in the amount of years playing videogames compared to the TE group ($U = 164$, $p = 0.788$). A list of descriptive statistics for both groups can be seen in **Table 1**.

Further, since there is no previous literature aimed at assessing the trainability of cognitive map formation, from a purely exploratory perspective we evaluated whether or not the training program would affect more specific spatial cognitive functions such as mental rotation and perspective-taking; to address this, participants in the experimental group were asked to complete two supplementary spatial tasks (i.e., the mental rotation task and the Four-Mountains Task; see description below) in the pre-training and post-training assessments.

Pre and Post-training Behavioral Assessment

Before and after the training program, participants in the training group were administered a battery of tests. The battery included the following series of tests aimed at assessing a variety of spatial skills that are important for orientation and navigation, including the ability to form cognitive maps.

The Mental Rotation Task is a well-known measure of spatial awareness and the ability to mentally represent and manipulate 3D images in one's mind (Shepard and Metzler, 1971). The task consists of two abstract 3D objects placed side-by-side for the participant to compare and see if they are either the same object or two mirror images of each other (Vandenberg and Kuse, 1978). The objects are presented in varying spatial orientations, so the participant is required to mentally rotate the objects and determine if they are either "the same" or "different." A total of 80 comparisons are made over the course of the task. Participants' performance is scored on accuracy, where a higher amount of correct responses is considered a good performance on the task.

The Four Mountains Task is designed to assess an individual's perspective-taking abilities when presented with simple scenes (Hartley et al., 2007). In a trial, a scene containing a randomly generated set of four mountains is shown for 8 s, during which the participant is asked to memorize the scene to the best of their ability (**Figure 1A**). Following this, in the recall phase, participants are presented with four different pictures, each of four different sets of randomly generated mountains (**Figure 1B**). One of these four sets of mountains is the original set that the participant was been previously shown, however, this new picture was taken from an alternate angle or perspective. In addition, to remove the possibility of using the surroundings to determine the correct answer, both the lighting and texture of the terrain surrounding the mountains are changed. The task consists of 20 trials, each with a different set of randomly generated mountains. Participants' performance is scored by measuring the number of correct responses such that a higher score represents better perspective-taking abilities.

The SCT is designed to assess the ability to generate and use configural (configurable) knowledge (i.e., a mental representation of the relationship between objects in an environment, or a "cognitive map") of geometric objects in an environment (Burles et al., 2017). Participants are presented with a space-like environment containing five abstract objects set in a pentagon (**Figure 2A**). Participants are not presented with the top-down view seen in **Figure 2A**, rather they are situated at one of the objects, facing inwards toward the center of the pentagon in a first-person view. This limits the participant's view so they can only see two of the five objects in the pentagon (**Figure 2B**). They are then presented with the other three objects that are not in view as options to choose from at the bottom of the screen as seen in **Figure 2B**. At each of the 60 trials in this task, participants are required to choose the object they were looking from, or rather, situated on, from the objects presented at the bottom of the screen. Upon responding, the camera moves to

TABLE 1 | Descriptive statistics for both the training and testing effect group.

	Training group	Testing effect group	Group difference	
	M (*SD*)	*M* (*SD*)	*U*	*p*
N	15, 5 males	23, 2 males		
Age in years	34.33 (19.17)	20.00 (1.73)		
Video game experience in years	4.07 (5.47)	2.44 (3.62)	164	0.788
Spatial Configuration Task				
Pre-training accuracy	0.48 (0.20)	0.55 (0.19)		
Post-training accuracy	0.62 (0.24)	0.62 (0.21)		
Four mountains				
Pre-training accuracy	0.62 (0.12)			
Post-training accuracy	0.69 (0.11)			
Mental rotation				
Pre-training accuracy	0.80 (0.10)			
Post-training accuracy	0.86 (0.09)			

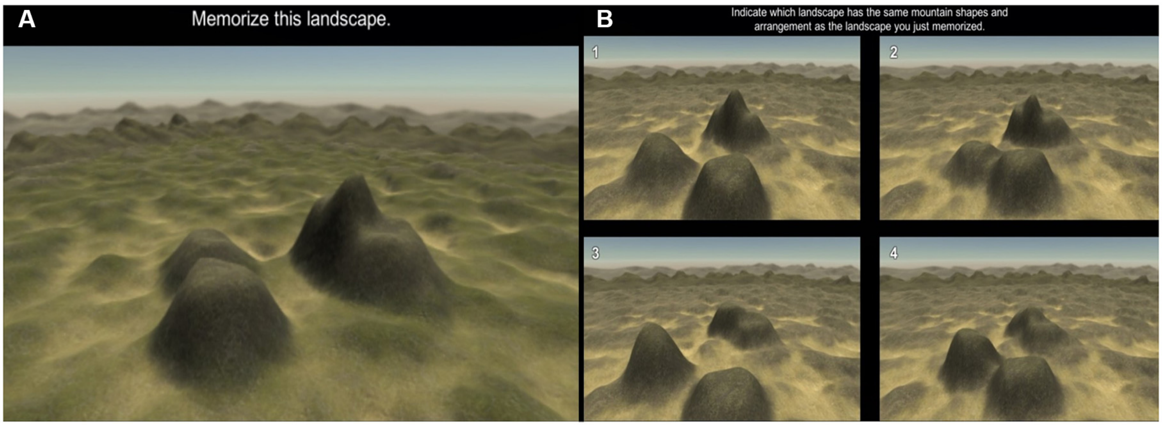

FIGURE 1 | Example of a randomly generated scene from the Four Mountains Task **(A)**, and **(B)** recall phase of the task. In this example the correct answer is option #2.

a new object and the participant is required to repeat the steps described above. Participants are not given any direct feedback, however, while the camera is moving to a new object, it first turns and faces the object they were situated on, providing implicit feedback pertaining to their answer. Participants' performance is scored on accuracy, where having a higher amount of correct responses indicates a better performance. Test-retest reliability conducted on the SCT has shown that it is internally consistent, but only within a single testing session (Burles et al., 2017).

Training Program

Procedure

The training program begins by asking participants to complete the pre-training behavioral assessment, immediately followed by the first 45-min session in the training environment. Participants are then asked to complete eight additional 45-min sessions within the training program over a 10-days consecutive period, with a 2-days break after the first 4 days of training. Participants then complete a final 45-min training session followed by the post-training behavioral assessment. The total amount of training received is 10 45-min sessions across a 12-day period, with a 2-days break between the first five and last five sessions (see **Figure 3**). A 12-days training period was chosen based on the spatial training programs used in the previous literature. For instance, Meneghetti et al. (2016) used the Mental Rotation Task to train generalized spatial abilities where participants were asked to complete six, 45-min training sessions across a 2-week fixed period (three on week 1 and three on week 2) which was sufficient to produce lasting results on an object perspective task. Another study conducted by Wright et al. (2008) also used the mental rotation task to train generalized spatial abilities. In their study, participants were asked to complete 15–20 min of training each day over a 21-days training period, which was enough to improve reaction times on a non-trained spatial task. We chose our training program with the aim of being more intensive and over a shorter period of time than previous studies. Additionally, further support for this training period has been found in a very recent review by Montana et al. (2019) analyzing a variety of training programs using ecological virtual environments to train spatial skills in stroke patients. Results from this review showed beneficial results following 8–15 sessions lasting 40–45 min each, further suggesting that a

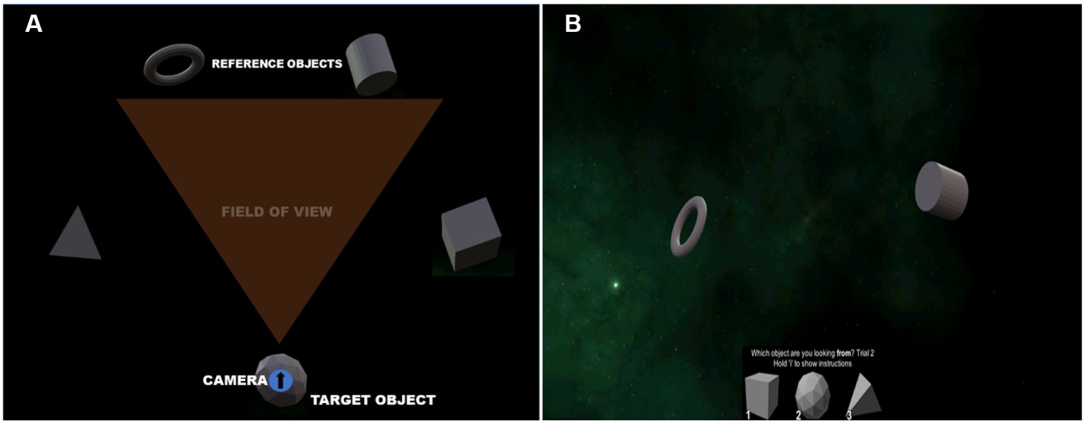

FIGURE 2 | Overhead view of the Spatial Configuration Task (SCT; **A**), and **(B)** a sample trial in which the correct answer is object 2.

FIGURE 3 | Timeframe of the training protocol across a 12-day period.

semi-intensive (5 days per week) 12-days training period would be enough to produce lasting improvements.

Training Environment

The training program takes place in a virtual environment that represents an urban neighborhood that we named Centerville. We designed Centerville using the Unreal Engine 4.15.3 under a Game developer's license. We created the design for Centerville by researching common layouts of popular cities. Centerville was modeled after a curvilinear-loop design that integrates grid-like designs with curved lines. This allows for a combination of easier to navigate sections of the neighborhood in the grid section, with slightly more difficult sections of the environment outside of the grid. Centerville is split into three distinct areas that differ in the architecture and décor of the buildings as well as the types of landmarks present within the area (see **Figure 4A**). Area 1 has a rustic, brick-road look and contains stores, cafés, and restaurants (see **Figure 4B**). Area 2 has a washed-out, industrial look and contains office and government buildings (see **Figure 4C**). Area 3 is more diverse as it is designed to be the city center: it contains a city hall alongside multiple government office buildings (inside of which not available to participants for navigation), a community pool, and a recreational children's park (see **Figure 4D**). Each of the three areas contains one "hub," six major landmarks, and two to three minor landmarks. The hub of an area is the most outstanding landmark within the area and is used as an anchor to give participants a landmark that they can easily remember from each of the three areas. For example, the hub for Area 1 is the character's apartment, the hub for Area 2 is the art gallery, and the hub for Area 3 is the main entrance to the large city hall building. The six major landmarks are stores, office buildings, and other amenities within a given area. The minor landmarks are landmarks that are not buildings, such as statues, billboards, and playground structures.

Each of the three areas is divided into four additional sections: a section that includes the hub, and the three other sections each including two proximal, major landmarks (i.e., section A, B, and C; see **Figure 5A**). For example, referring to Area 1, the two proximal, major landmarks in Section A are the closest landmarks to the hub (e.g., Chelsie's Coffee and Kendra's Korner Store), making them the easiest to find when situated at the hub. The two proximal, major landmarks in Section B are slightly farther away from the hub (Atomic Clothes and City Bank), and generally take more effort to find and navigate to from the hub. Finally, the two proximal, major landmarks in Section C are the furthest away from the hub (e.g., Michael's Bar and Grill and the Liquor Store) and take the most amount of effort to find. Each area also contains two to three minor landmarks: area 1 includes a

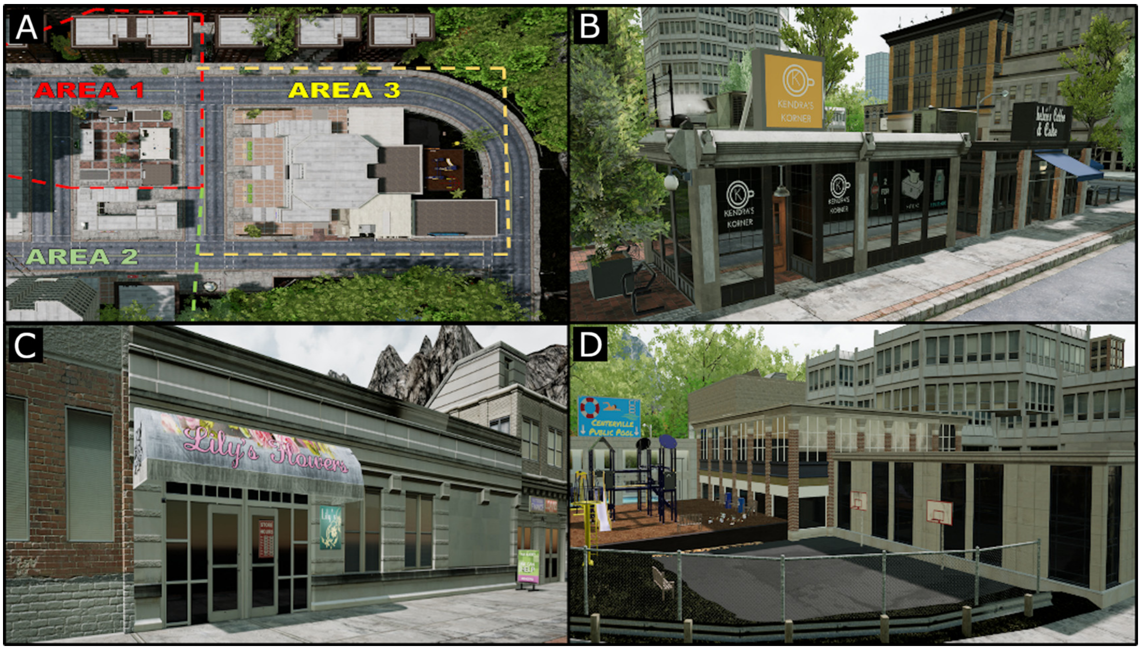

FIGURE 4 | Different views of the virtual town "Centerville." **(A)** Image showing how the training environment is split into three distinct areas. **(B)** Example buildings from Area 1, designed to be rustic with lots of colorful details making the buildings stand out. **(C)** Example buildings from Area 2, designed to be more industrial with gray colors making the buildings blend together. **(D)** Example buildings from Area 3, designed to be more modern and introduce more complex landmarks such as playgrounds, pools, and parking lots.

statue of a man sitting on a bench and the Getting Lost billboard, Area 2 includes a definitive yellow bench and a children's thrift store hidden in an alley, and Area 3 includes the Centerville Sphere statue, a large graffiti art in a tunnel, and a swing-set in the park.

Movement within the environment is achieved using a standard Xbox 360 controller. Participants control a first-person character and only have control of the forward/backward movement and the yaw (left and right turning). The movement is restricted as such to make movement easier for those with less video game experience. This movement is accomplished using the left joystick on the controller. Turning speed is kept at the default setting in the game engine and is smoothed using interpolation to avoid rapid starting and stopping of turning.

In order to practice the motor controls of the task, participants are asked to perform a practice session in a simple maze with one right and one left turn. Participants are instructed to move to the end of the maze without touching any walls and within a designated time period of 17 s. If a wall is touched or the participant takes too long, they are reset back to the beginning of the maze. When participants complete the maze correctly, they are presented with the training instructions and moved onto the training task.

Training Task

Throughout the entire training program, during each training session, participants perform as many trials as possible each asking them to reach a target landmark from a given starting location. Here, we use the term "pathway" to refer to the optimal path between the starting location and the target landmark (see **Figure 5B**). For each pathway, participants are required to perform a series of trials where they attempt to get from the starting location to the target landmark by traveling the shortest distance, as quickly as possible. This pathway is measured by the game engine, providing the optimal distance and time that could be traveled between the starting location and the target landmark, all while accounting for any obstacles (buildings, trees, signs, etc.). Naturally, the distances and times of the pathways vary according to the starting location and the complexity of the pathway to get to the target location. Based on a pilot study that we conducted, we account for a correct performance if participants travel to each target landmark within a 20% overage of the calculated optimal time and distance. Participants are required to perform three correct trials within a given pathway in order to move to a new pathway (i.e., a new starting location and target landmark). These correct trials do not have to be performed consecutively.

Upon reaching the target landmark, a participant receives one of three messages on the screen. If they successfully complete the pathway according to the criteria but have not yet completed three successful trials for the pathway, they are shown a message telling them to go back to the starting landmark (e.g., "Great job, now go to your apartment"). If this trial is their third successfully completed trial for the pathway, they get a message telling them to go find a new landmark (e.g., "Excellent, now go find Fresh Veggies"). If the participant reaches the target landmark but did not satisfy the distance or time criteria, they receive a message telling them to go faster while following

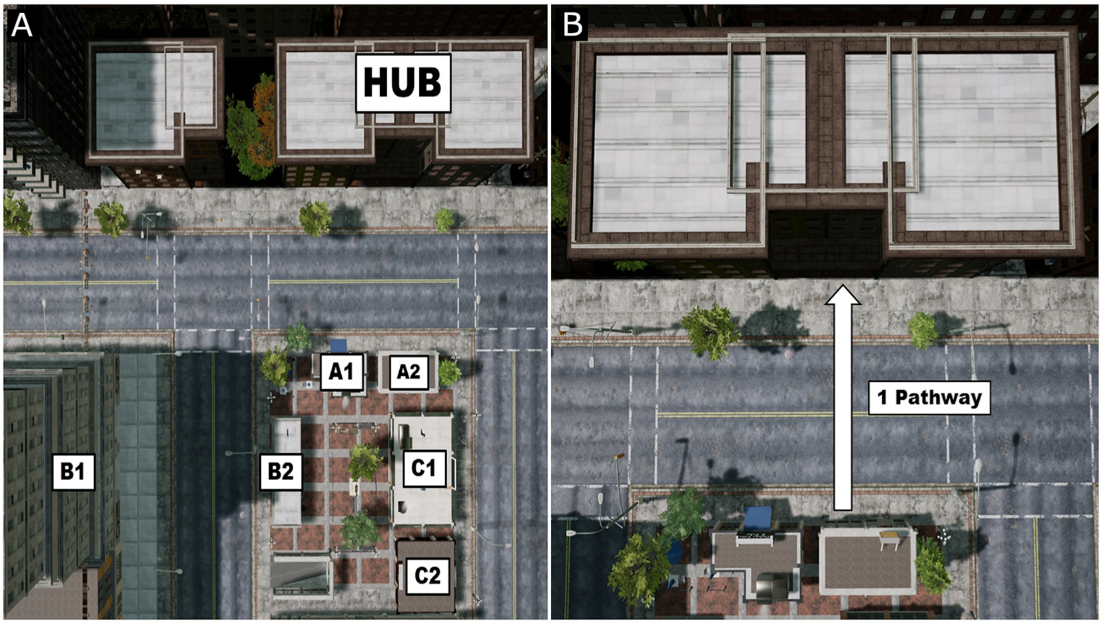

FIGURE 5 | Division of areas in Centerville and example of a "pathway." **(A)** Example of how each area is split into different components. Each area has a hub, two proximal, major landmarks directly near the hub (A1: Chelsie's Coffee and A2: Kendra's Korner Store), two proximal, major landmarks further away from the hub (B1: City Bank and B2: Atomic Clothes), and two proximal, major landmarks that are the furthest away from the hub (C1: Michael's Bar and Grill and C2: The Liquor Store). **(B)** An example of a single pathway. A pathway can be as long or as short as necessary and is defined as the optimal path between the starting location and the target landmark.

the shortest path (i.e., "Good, now try to be faster while following the shortest pathway"); this message is intended to be ambiguous about whether they failed to reach the time or distance criteria so that the participant has to think about how to optimize their route as much as possible. If a participant does not reach the target landmark after a 400% window of the optimal time, they are presented with an arrow at the bottom of the screen pointing in the direction of the landmark (without providing any information of the pathway to be followed in order to get there); this gives the participant a hint of where the landmark is and encourages them to continue exploring on their own.

Participants completed six pathways (e.g., hub to A1, hub to B1, B1 to A1, B1 to C1, C1 to hub, C1 to A1) of major landmarks for each of the three areas. After completing six pathways in each area, they are asked to combine pathways across areas (e.g., hub (Area 1) to B1 (Area 2), B1 (Area 2) to the hub (Area 3), etc). After completing all combinations from these landmarks, they are asked to complete six more pathways between major landmarks that they did not travel the first time around in each of the three areas (e.g., hub to A2, hub to B2, etc). After completing these new pathways in each area, they are asked to combine these new pathways across areas [e.g., hub (Area 1) to B2 (Area 2), B2 (Area 2) to the hub (Area 3), etc]. Upon completing these pathways, they will have traveled every possible pathway between major landmarks in the game (784 pathways) across two full playthroughs of the game. If they reach this point, they are presented with a random pathway chosen from all major landmark pathways they have already completed, as well as the new minor landmarks they have not explored yet. We termed this "infinite mode" as the participant will keep getting a random pathway when they successfully complete a pathway. This will continue as such for the remainder of the training. During this mode, only one successful completion of the pathway is required to be given a new pathway.

Data Analyses

All behavioral data were analyzed while accounting for variability in age, which has been shown to have a significant impact on spatial orientation skills (Iaria et al., 2009b; Uttal et al., 2013; Wolbers et al., 2014; Yuan et al., 2014; Coutrot et al., 2018; Yamamoto et al., 2019). For each behavioral measure collected from each participant in the training group, we conducted a one-way, repeated measure ANCOVA with two levels comparing pre-training and post-training scores using age as a covariate. We then conducted a repeated measures analyses of covariance (RMANCOVAs) comparing overall changes on pre-training and post-training scores in the SCT (measuring the ability to orient by means of cognitive maps), between the training group and the testing effect group using age as a covariate.

RESULTS

We first analyzed the assumptions for repeated measures analysis of covariances. All pre- and post-training measures satisfied the Shapiro–Wilk's test of normality and were within a skewness range of −1 to 1. The covariate (age) was moderately correlated with the dependent variable (difference in SCT scores from pre to post) in the experimental group ($r_{(15)} = -0.536$, $p = 0.04$) but not in the testing effect group ($r_{(23)} = -0.003$, $p = 0.987$) suggesting that age would account for a sufficient portion of the variance in the experimental group. Additionally, the covariate did not violate the assumption of homogeneity of regression slopes between the experimental group and the testing effect group with a change in SCT scores as the dependent variable; $F_{(1,34)} = 0.065$, $p = 0.801$. No significant outliers were present in the data and as a result, all participants were included in the analysis.

One Way Repeated Measure Analysis of Covariances

Three one-way repeated measures analyses of covariance (RMANCOVA) were conducted to determine the effect of assessment (pre-training and post-training) on the scores for each of the behavioral tasks while controlling for age in the training group. The analyses revealed a significant effect of assessment on the SCT accuracy after controlling for age ($F_{(1,13)} = 5.234$, $p = 0.040$, $\eta_p^2 = 0.287$). No significant effect of assessment was found on the Four Mountains Task ($F_{(1,13)} = 0.204$, $p = 0.659$, $\eta_p^2 = 0.015$), and the Mental Rotation Task ($F_{(1,13)} = 0.141$, $p = 0.713$, $\eta_p^2 = 0.011$). An additional one-way RMANCOVA was conducted to determine the effect of assessment (pre-training and post-training) on the calculated scores for the Santa Barbara Sense of Direction Scale while controlling for age in the training group. The analysis revealed that there was no significant effect of assessment on the scale after controlling for age ($F_{(1,13)} = 1.798$, $p = 0.203$, $\eta_p^2 = 0.121$).

A two-way repeated-measures analysis of covariance (RMANCOVA) was conducted to test the hypothesis that the training group would perform better than the untrained testing effect group at the post-training assessment on the SCT while controlling for age. The analysis revealed a significant interaction effect between assessment (pre and post-training) and group (training and testing effect; see **Figure 6**) confirming our hypothesis ($F_{(1,35)} = 6.380$, $p = 0.016$, $\eta_p^2 = 0.154$). With a significant interaction found, follow up comparisons were conducted (Bonferroni corrected) to determine the source of the interaction: the only significant difference that was found was between the pre-training ($M = 0.502$, $SD = 0.345$) and post-training ($M = 0.679$, $SD = 0.357$) scores in the experimental group; $t_{(36)} = 4.784$, $p < 0.001$. All other comparisons were non-significant at an alpha level of 0.05.

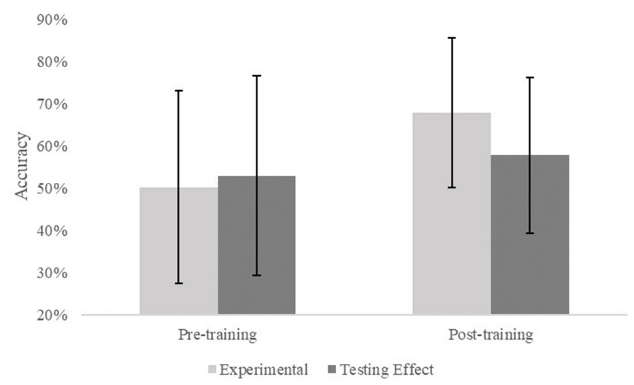

FIGURE 6 | Estimated marginal means of the interaction effect from a repeated-measures analysis of covariance (RMANCOVA) conducted on the SCT comparing pre-training accuracy scores in the training group [$M = 0.502$, 95% CI (0.616, 0.388)] and testing effect group [$M = 0.529$, 95% CI (0.618, 0.440)] to post-training accuracy scores in the training group [$M = 0.679$, 95% CI (0.798, 0.561)] and testing effect group [$M = 0.578$, 95% CI (0.617, 0.486)] after accounting for age as a covariate. Error bars show 95% confidence intervals.

DISCUSSION

This study reports the very preliminary findings of a newly developed training program specifically designed to improve the ability of the individuals to acquire configural knowledge of the spatial surrounding. The preliminary data available in this study confirm the feasibility of such a training program in the healthy population and seem to suggest an increase in the ability to form cognitive maps. This effect is in comparison to the testing effect group, who did not significantly improve in their scores on the SCT following a 12-days of no training period. It should be noted that the testing effect group was not a true control group and was only included in the study to control for the test-retest effect of the SCT across a 12-day period. The same training program did not result in any other significant improvements in task performance across other tests assessing cognitive functions such as mental rotation and perspective-taking. This effect could be explained by the nature of the SCT as it was appositely designed to measure the ability to create a mental representation of elements in space and their spatial relationships (Burles et al., 2017), a crucial, generalized skill in the formation and utilization of cognitive maps for orientation. In the SCT, participants are asked to identify the object (i.e., the location) that the camera is looking from. All objects are geometric abstract objects that are difficult to verbalize, making it much more difficult to remember them without directly visualizing them (e.g., "the round donut looking thing") and consequently easier to simply visualize the objects. This means that to perform effectively and score high on the SCT, an individual must form a mental representation, or "cognitive map," of the relationships between the five objects located in the surrounding. Participants' improvement on the post-training SCT score will have suggested the development of a stronger ability to create and utilize cognitive maps for orientation.

Previous studies have examined the trainability of spatial skills in humans, providing good evidence that some spatial skills can be indeed improved by a training program (Uttal et al., 2013). While this is certainly important evidence, none of the studies reported in the literature have focused on specially training the ability to form cognitive maps. In the meta-analysis by Uttal et al. (2013), the authors examined 217 training studies focused on spatial skills, analyzing in detail the type of training (video game, course-based, and spatial task training) and the type of measure used for evaluating effective trainability of spatial skills. Although video games were a major theme in the meta-analysis, none of the measures reported were indicators of a person's ability to form cognitive maps, which is consistent with more recent studies. For instance, a study by Xiao et al. (2018) examined the potential for a video game platform to train spatial visualization skills without using any measures to determine improvements in cognitive map formation abilities. Similarly, Veurink and Sorby (2019) recently analyzed the results of a longitudinal study aimed at improving spatial skills in engineering students who initially scored low on the Purdue Spatial Visualization Test, a measure of one's mental rotation ability (Bodner and Guay, 1997). In this study, the training program focused on several spatial skills, such as the rotation of objects and cutting planes, but none of their measures focused on cognitive map formation (Veurink and Sorby, 2019). While no studies to date have examined the potential to train the ability to form cognitive maps, it remains difficult to argue that training specific spatial skills (such as mental rotation or landmark recognition) would result in better spatial orientation and navigation. Our present study with a newly developed tool that allows testing the hypothesis that high-cognitive level functions in the spatial domain can be trained, and potentially result in improved spatial orientation skills in people's daily lives.

The evidence that cognitive maps are a trainable skill could have important implications for improving the lives of many individuals. First, individuals affected by DTD have a selective inability to orient in very familiar surroundings due to their inability to form cognitive maps (Iaria and Burles, 2016). Cognitive map training, as described in our study, could directly target this missing skill in people with DTD, leading to an improvement in their navigation abilities and subsequently raising their quality of life. Second, aging has been shown to reduce the effectiveness of navigation in the elderly as it seems to cause a switch from survey-based navigation to less effective route-based navigation strategies (Yamamoto et al., 2019). The ability to train cognitive map skills could keep survey-based navigation as the main strategy in older adults, potentially preserving effective navigation skills in novel surroundings throughout old age. In addition, as shown by Lövdén et al. (2012), navigation training can protect against age-related changes to the hippocampus in both young and older adults. This implies that hippocampal-dependent spatial training, such as training cognitive map formation, may produce neuroprotective benefits leading to healthier brain aging. Finally, recent studies have suggested that spatial navigation quality can be used as an early marker for Alzheimer's disease, a disease that typically starts with degeneration of the hippocampus (Coughlan et al., 2018; Laczó et al., 2018; Parizkova et al., 2018). Alzheimer's patients commonly show orientation issues (Coughlan et al., 2018), suggesting that the early stages of Alzheimer's can be diagnosed with a decline in orientation skills. A training program effective at improving spatial orientation skills such as the one described in our study could have a significant impact on delaying the degenerative process related to the decline of spatial orientation skills in Alzheimer's disease. However, our study does not provide evidence on the efficacy of the training program in any of these populations and thus these speculations should be taken with caution.

Interestingly, we found that scores on the mental rotation and Four Mountain Tasks were not significantly affected by the training program. Although it may seem surprising at first, this is expected given that the mental rotation task is designed to assess the ability to mentally rotate small 3D objects in a scale of space known as *figural space* (Hegarty et al., 2006). Figural space is a scale of space that is external to the individual, can be fully observed from a single viewpoint, and is occupied only by a single object such as a car or a coffee cup (Hegarty et al., 2006). Figural space is smaller than *vista space* which can be fully observed from a single viewpoint but is occupied by multiple environmental elements (objects, landmarks, landscapes, etc.) to create a scene. Figural space also differs from the so-called *environmental space*, which refers to a large environment that can only be fully observed from multiple, different, angles and viewpoints (Hegarty et al., 2006). An individual would need to navigate environmental space to become familiar with it, in comparison to vista and figural space which can be fully observed while standing still. While visualizing and manipulating figural space is important for cognitive map formation and utilization, it is only a small component of the overarching skill. Therefore, one would expect the effects of a training program to be visible on the general ability of cognitive map formation and usage, rather than the individual components of it. The same applies to the lack of effects found in the Four Mountains Task, as this task assesses perspective-taking, another individual component of the ability to form cognitive maps (Hartley et al., 2007).

The main aim of our study was to present a newly developed cognitive training potentially effective in improving the ability of the individuals to form cognitive maps. The preliminary findings reported in this study have some significant limitations that are important to be aware of before making any conclusion on the efficacy of such a program. First, we have collected data on small sample size, and it would be very valuable to test a larger population of individuals to improve the power of the study. Ideally, a future study should be also balanced in sex across the groups, as the current study sample is low on male participants. Sex is known to have a significant effect on spatial abilities, with a specific effect on the ability to form cognitive maps (Moffat et al., 1998; Astur et al., 2004; Iaria et al., 2009b; Liu et al., 2011; Fernandez-Baizan et al., 2019). Thus, in the context of cognitive map formation abilities, keeping the groups balanced can help to remove unexplained variance due to sex from the sample, and investigate the effectiveness of the training program across sex. Similarly, gathering a larger sample size while keeping a broad range of ages will lead to a better understanding of the effect that age may have on the benefits gained from the training program. In addition, a future study should evaluate the long-term effects of the training program on the daily life orientation skills of the individuals; this will require a longitudinal study given that the skills acquired through the training program may require some time and practice to be consolidated in the real-life navigation. Moreover, it is worth noticing that participants in our control group were not asked to complete a non-spatial training program, and the group was included to control only for the passage of time and for repetition effects of the SCT. To overcome this limitation, a future study should include a control group in which participants are asked to perform a daily non-spatial training program with an experimental protocol identical to one followed by the training group. Finally, the lack of follow up measurements does not provide us with information regarding the long-term effects of the training program; without such evidence it would be difficult to prove that the training could have a significant impact on people's daily life. For all reasons described above, the findings reported in our study should be treated as providing preliminary evidence for an effective program aiming to train the ability to form cognitive maps for orientation. Nevertheless, these preliminary findings, together with previous work available in the literature, seem to point towards a very important training opportunity for improving the ability to orient in large-scale surroundings. This has the potential to be beneficial to a variety of individuals suffering from topographical disorientation due to a developmental or acquired condition. However, the limitations in this study cause the results to be too speculative to apply training programs to these populations at this stage. Further testing on healthy subjects using a study design with a true control group will be required before more confident conclusions can be made on the efficacy of this training program.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Conjoint Faculty Research Ethics Board—University of Calgary. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

All Authors contributed significantly to the work presented in this manuscript and approved the final version of the manuscript. MM-G, FB, ID, AU, and GI conceived the training program and developed the experimental protocol. MM-G, KD, and AR performed data collection. MM-G, FB, JH, and GI performed data analyses. MM-G prepared the first draft of the manuscript.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Path Learning From Navigation in Aging: The Role of Cognitive Functioning and Wayfinding Inclinations

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Aging coincides with a decline in navigation and wayfinding abilities, but it is unclear to what extent factors relating to a given individual may contribute to mitigating this decline. The present study aims to analyze how older adults' objective cognitive functioning and self-reported subjective wayfinding inclinations predict their navigation performance. Sixty-four older adults were assessed on their general cognitive functioning (all scoring from 22 to 30 on the Montreal Cognitive Assessment, MoCA), visuospatial working memory (VSWM), and perspective-taking abilities. Their self-assessed wayfinding inclinations (such as their sense of direction, pleasure in exploring places, and spatial anxiety) were also examined. Then participants learned a path in an environment from video navigation and performed a route repetition task (which maintained the same egocentric perspective as the learning phase), and a sketch map task (which involved switching from an egocentric perspective used in the learning phase to an allocentric perspective). The results showed that positive wayfinding inclinations (in terms of pleasure in exploring) related to participants' route repetition accuracy, while their general cognitive performance (MoCA scores) related to their sketch map drawing accuracy. Individual factors such as cognitive functioning and wayfinding inclinations relate differently to older people's navigation performance, depending on the demands of the tasks used to test their environment learning.

Keywords: navigation, route learning, cognitive functioning, MoCA, pleasure in exploring, older adults, aging

INTRODUCTION

Being able to navigate in the environment is fundamental to everyday life, and any impairments in this domain can limit people's independence and safety. Studying populations that have difficulty learning new paths in the environment, such as older adults, is consequently of particular interest. Research on pathological aging has shown that navigation deficits develop in the earliest stages of Alzheimer's disease (AD), and even in cases of Mild Cognitive

Impairment (MCI, Laczó et al., 2011). The navigation issues seen in pathological aging have inspired studies on route learning abilities in normal aging too, in an effort to monitor impairment progression, and identify the first signs of pathological aging (Lithfous et al., 2013).

After we have learned an environment from navigation, several factors can relate to the quality of our mental representation of it, or cognitive map (Tolman, 1948; Wolbers and Hegarty, 2010). Some factors are external to the individual, including the type of task used to assess this learning (Muffato et al., 2019a). Some tasks are based on egocentric knowledge of the environment and involve repeating a previously-taken path (e.g., recalling a series of turns, as in route repetition tasks). Others are based on allocentric knowledge and may involve drawing a map of the environment from memory, or recalling the location of landmarks in an area (as in sketch map tasks). Some studies found that aging affects people's ability to repeat previously-learned routes (e.g., Barrash, 1994; Wiener et al., 2012; Taillade et al., 2016), but others identified no such impairments (e.g., Cushman et al., 2008). As for map drawing, all studies found impairments in older adults (e.g., Cushman et al., 2008; Taillade et al., 2016; Muffato et al., 2019a). These findings suggest that switching from an egocentric point of view (learning from navigation) to an allocentric one (as in map drawing tasks) is particularly problematic, even in normal aging (Lester et al., 2017). In fact, the egocentric component is better preserved than the allocentric one over an individual's lifespan (Ruggiero et al., 2016).

Other individual factors apart from age, including cognitive abilities, and wayfinding inclinations and strategies, can relate to the features of mental representations of a path learned from navigation (Kraemer et al., 2017). In terms of cognitive abilities, studies on navigation in aging have rarely considered the role of an individual's general level of cognition on spatial performance. The most often used measure of general cognition is the Montreal Cognitive Assessment (MoCA), a 30-item screening tool very sensitive to early changes in cognition (Nasreddine et al., 2005). It has been used as an inclusion criterion in various studies on navigation in aging (e.g., Wiener et al., 2012; Bates and Wolbers, 2014; Muffato et al., 2019a,b), but none of these studies related general cognitive performance with spatial recall performance. Only O'Malley et al. (2018) considered MoCA scores as a factor related to spatial navigation performance. They grouped older adults by their higher (26–30) or lower (22–25) MoCA scores and analyzed the two groups' spatial performance after learning a path from navigation. The results showed a superiority of the group with higher MoCA scores in tasks that involved managing spatial information (i.e., recalling directions of landmarks), and required a change of perspective from the learning to the testing phase (i.e., choosing which of three maps depicted the route traveled in the learning phase). No differences emerged between the two groups, however, in a task resembling the format of the learning phase (i.e., recalling the sequence of landmarks). This study provided a first indication that general cognitive level may relate to spatial performance, depending on the type of spatial request. Among the various cognitive abilities, research has focused mainly on visuospatial working memory (VSWM), which retains and processes visuospatial information (Logie, 1995), and perspective-taking, which is a higher-order ability to adopt different views (Hegarty and Waller, 2004). Both are needed for navigation over the adult lifespan and are liable to age-related decline (Muffato et al., 2019b). Self-reported wayfinding inclinations—e.g., an individual's self-rated sense of direction, pleasure in exploring places, and spatial anxiety (finding spatial demands worrying), which are not susceptible to change over time—have also been found positively related to environmental learning across the lifespan (e.g., Meneghetti et al., 2014). They may be important to good navigation performance, even in aging.

To sum up, route learning from navigation is essential for everyone and particularly important in aging. In old age, we become susceptible to a decline in our ability to manage certain types of environment knowledge, when we need to switch the perspective from the learning to the recall phase, for instance. General cognitive functioning (rarely considered in relation to navigation performance), visuospatial abilities, and self-assessed wayfinding inclinations are all factors that contribute to our navigation performance in aging, but studies conducted to date have not analyzed all these factors together.

The present study thus aims to analyze the influence of individual factors—both cognitive skills (i.e., general cognitive level and visuospatial abilities) and self-reported wayfinding inclinations—on route learning from navigation in aging. This learning is measured in terms of the ability to repeat a previously-learned path and to place the landmarks learned on a sketch map. The former is a task retaining much the same format as the learning phase, while the latter requires a change of perspective between the learning and testing phases. This is done with the novel intent to identify factors related to different environmental demands and a view to understanding the first signs of a declining ability to switch perspective that may indicate the increased risk of atypical aging. This will shed more light on the issue of age-related changes in navigation abilities.

Cognitive abilities can be expected to have a role in the task requiring a switch from egocentric to allocentric knowledge (the sketch map task). This aspect is newly explored, however, considering not only VSWM and perspective-taking ability (found to influence navigation, Muffato et al., 2019b), but also general cognitive level. The latter is measured on a continuum from low to high, considering MoCA scores from 22 to 30 (and using the cutoffs adopted by O'Malley et al., 2018) as predictors of navigation performance after accounting for the role of age from young-old to old age. After taking cognitive abilities into account, self-reported wayfinding inclinations are also expected to influence navigation performance (Muffato et al., 2019a), and are explored in relation to the demands of the task.

MATERIALS AND METHODS

Participants

The study involved 64 healthy adults from 60 to 84 years old (34 females; *M* age = 70.55, *SD* = 7.04) who volunteered to take part and were recruited at recreation centers. A MoCA score of at least 22 (Nasreddine et al., 2005; *M* = 26.53, *SD* = 1.96) was needed in order to include only typically-aging individuals (see Bosco et al., 2017, for the Italian normative sample). Participants had attended school from 8 to 13 years old (M = 10.42, SD = 2.20), as is typical in Italy for this cohort (see ISTAT, 2011). None of the participants had a history of psychiatric, neurological or other diseases capable of causing cognitive, visual, auditory or motor impairments (Crook et al., 1986). None of them had ever visited the environment used in the learning phase.

The local ethical committee approved the study, and all participants were informed about its purposes and gave their written informed consent in accordance with the Declaration of Helsinki (World Medical Association, 2013).

Session 1: Individual Measures

Objective Cognitive Measures

MoCA (Nasreddine et al., 2005)

This assesses multiple aspects of executive functioning, attention, working memory, delayed memory, and language. Orientation in time and place is also tested (max score 30).

Jigsaw Puzzle Test (JPT, De Beni et al., 2008)

This VSWM task involves mentally recomposing puzzles of objects (from 2 to 10 pieces, i.e., levels of difficulty). Participants must solve at least two of the three puzzles on a given level in order to proceed to the next. The final score is the sum of the difficulty levels of the last three puzzles solved (max 29).

Short Object Perspective Test (sOPT, Hegarty and Waller, 2004 and De Beni et al., 2014)

Participants have to imagine standing at one object in a layout comprising seven objects, facing another, and pointing towards a third. They indicate the direction by drawing an arrow from the center of a circle to its perimeter (six items; time limit 5 min; score 0–180° mean degrees of error; Cronbach's α= 0.72 in the current sample).

Self-report Measures (From De Beni et al., 2014)

The subjective measures of participants' wayfinding inclinations included: the *Sense of Direction and Spatial Representation scale* (SDSR; 13 items; max score 65; Cronbach's α= 0.85), which measures an individual's self-assessed sense of direction; the *Attitudes Towards Orientation Tasks Scale* (AtOT; 10 items; max 60; Cronbach's α = 0.77) assessing their pleasure in exploring places; and the *Spatial Anxiety Scale* (SA; eight items; max 48; Cronbach's α = 0.87).

Session 2: Environment Learning and Recall Measures

Path Learning

A 6-min video (for details, see Muffato et al., 2019a,b) showing a walk around a botanical garden (in Padova, Italy), with 15 landmarks seen from a walker's perspective, was projected on a screen using a.mp4 file.

Route Repetition Task

This involved watching the same video again, and when it was paused, deciding which way to go along the path (with eight decision-making points; see **Figure 1**). If the wrong way was chosen, the feedback was provided, and the video continued in the right direction (scores: 0–8).

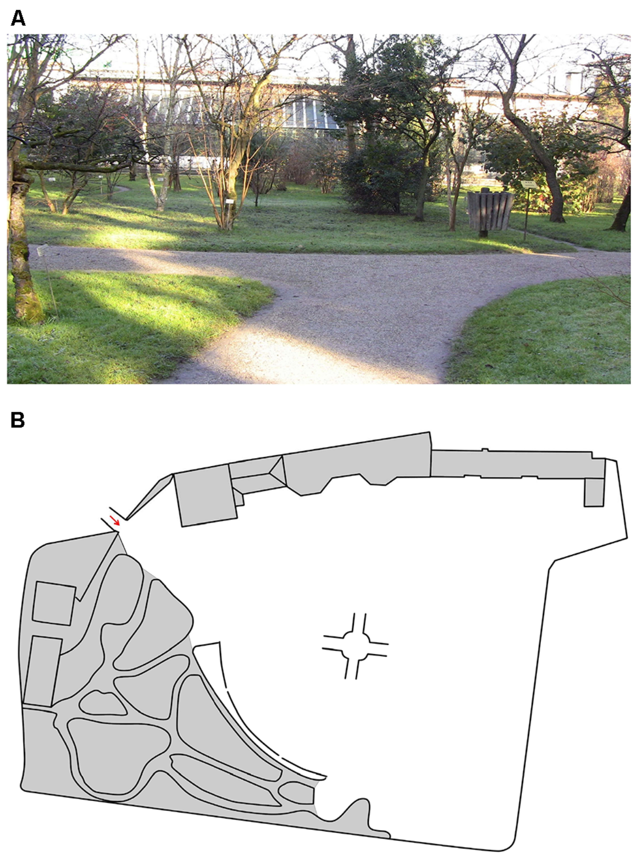

FIGURE 1 | Example of the route repetition task **(A)**, and the sketch used in the sketch map task **(B)**.

Sketch Map Task

This involved placing as many of the landmarks as possible in their right relationship to one another on a sketch map (A4 format; see **Figure 1**). The square root of the canonical organization was considered as a global index of accuracy (scores: 0–1; Gardony et al., 2016).

Procedure

At a first session (lasting 45 min), participants completed a socio-demographic questionnaire, the MoCA, jigsaw puzzle test (JPT) and short object perspective test (sOPT), and the SDSR, AtOT and SA questionnaires in a balanced order. During a second session (lasting 30 min), they learned the route from the video, then performed the route repetition and sketch map tasks, in a balanced order.

RESULTS

The data analysis was conducted with the R software. First, mean and standard deviations of the variables and their correlations

TABLE 1 | Means and standard deviations of the variables and their correlations.

	M (DS)	1	2	3	4	5	6	7	8
1. Age	70.55 (7.04)	-							
2. MoCA score	26.53 (1.96)	−0.12	-						
3. JPT (VSWM)	14.11 (5.51)	**−0.40**	**0.50**	-					
4. sOPT	76.40 (37.11)	**0.27**	−0.13	**−0.50**	-				
5. SDSR	37.11 (8.29)	0.01	0.03	0.24	−0.12	-			
6. AtOT	30.55 (8.23)	−0.15	**0.26**	**0.44**	−0.19	**0.61**	-		
7. SA	22.50 (7.34)	0.24	**−0.38**	**−0.30**	0.22	−0.19	**−0.52**	-	
8. Route repetition accuracy	4.78 (1.92)	−0.15	0.13	**0.26**	−0.16	0.21	**0.45**	**−0.36**	-
9. Sketch map accuracy	0.40 (0.17)	**−0.35**	**0.42**	**0.34**	−0.23	0.18	0.21	**−0.36**	**0.28**

Note. N = 64; significant correlations in bold type: for $|r| \geq 0.25$, $p < 0.05$; for $|r| \geq 0.33$, $p < 0.01$, and for $|r| \geq 0.42$, $p < 0.001$.

were calculated (see **Table 1**). Route repetition accuracy correlated directly with JPT and AtOT scores, and inversely with SA scores. Sketch map drawing accuracy correlated inversely with increasing age, directly with MoCA and JPT scores, and inversely with SA scores.

To shed more light on the effect of the various individual factors, several linear models on route repetition and on sketch map accuracy were run stepwise to see whether the factors added at each step improved the model (changes in R^2 are reported). Age, gender and education were entered in a baseline model (step 0) so as to examine the other factors related to spatial performance after accounting for their role. The objective factors, i.e., the MoCA as a measure of general cognitive functioning, and the JPT and sOPT (measuring VSWM and perspective-taking ability, respectively), were input in a subsequent model (step 1). Then self-reported wayfinding inclinations (SDSR, AtOT, and SA) were added in a further model (step 2) to see whether they still have a role after accounting for all the other factors investigated.

In a preliminary step, MoCA scores were considered as a dichotomous variable, high (26–30) vs. low (22–25; as in O'Malley et al., 2018), and the results were the same as when the MoCA scores were considered on a continuum. We, therefore, opted to report the latter, given the normal distribution of the MoCA scores.

For all models, the variance inflation factors revealed no significant multicollinearity (VIF values ≤ 2.57).

For the route repetition task: step 0 accounted for 11%, and step 1 for another 2% of the variance, but no predictors were significant in these steps; step 2 accounted for 13% of the variance, with pleasure in exploring places (AtOT; $\beta = 0.43$, $p = 0.027$) emerging as a significant predictor. As for the sketch map task: step 0 accounted for 17% of the variance, but no predictors were significant; step 1 accounted for another 14%, with MoCA score a significant predictor ($\beta = 0.39$, $p = 0.005$); step 2 accounted for 4% of the variance, with no factors emerging as significant predictors (see **Supplementary Table S1** for estimates and p-values for all steps in both tasks).

DISCUSSION

The present study aimed to analyze the role of older adults' objective cognitive functioning and subjective (self-reported) wayfinding inclinations on their navigation performance. The general cognitive level of a group of older adults with a broad range of MoCA scores (from 22 to 30) was here newly considered as a potential predictor of their spatial learning from navigation, alongside other cognitive aspects, such as their VSWM and perspective-taking abilities, and their self-reported wayfinding inclinations.

Our results newly showed a different involvement of certain individual factors depending on the type of recall task considered. Objective cognitive factors—and MoCA scores in particular—related to the sketch map task, which demands a switch from egocentric to allocentric knowledge. Subjective wayfinding inclinations—and especially pleasure in exploring places—related to the route repetition task, in which the perspective remains the same as in the learning phase. These results show that cognitive functioning in aging may contribute to better performance in more demanding spatial tasks after learning from navigation (O'Malley et al., 2018). This points to the importance of preserving our cognitive abilities in aging—with the aid of training programs, for instance (Lövdén et al., 2012). Moreover, detecting the very first signs of a decline in the ability to switch perspective could, therefore, be an important indicator of the risk of cognitive impairment and atypical aging (Lester et al., 2017). This issue deserves to be further investigated, administering a comprehensive neuropsychological battery to ensure the exclusion of cases of MCI. This is something not done in the present study, so our participants with lower MoCA scores may have had MCI, which could have affected their performance. Future studies should also disentangle whether the aging effect is triggered by perspective switching *per se*, or due to a generally impaired allocentric knowledge. This could be done by analyzing learning from an allocentric perspective, for instance. The other visuospatial cognitive factors considered in this study seem to not contribute to better learning from navigation, although VSWM correlated with accuracy in both tasks. On the other hand, it is worth emphasizing how self-assessed wayfinding inclinations contributed to environment learning from the navigation. After controlling for individual cognitive abilities, older adults who reported taking pleasure in exploring places were better able to solve the route repetition task. In other words, a positive attitude is helpful when addressing wayfinding tasks after learning from navigation, and an indication of a good spatial profile (Muffato et al., 2019a). In fact, pleasure in exploring correlated positively with a sense of direction and negatively with spatial anxiety, while greater spatial anxiety coincided with a weaker ability to solve spatial tasks. Promoting positive wayfinding inclinations in older people could be another way to help them maintain adequate spatial performance, for less cognitively demanding tasks at least. Finally, it is worth noting that most of the variance in the models remained unexplained. Future studies should analyze the contribution of other factors involved in navigation performance, given the great inter- and intra-individual variability in aging.

In conclusion, these results shed more light on the strengths and weaknesses of old people's navigation abilities. External factors (such as the type of recall task) and internal factors (general cognitive abilities and self-reported wayfinding inclinations) relate differently to the features of old adults' mental representations of environments, depending on the demands of a spatial task.

DATA AVAILABILITY STATEMENT

The data will be made available by the authors to any qualified researcher.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethical Committee for Psychological Research at the University of Padova. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

VM and RB contributed to the design and implementation of the research, to the analysis of the results and to the writing of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2020.00008/full#supplementary-material.

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ORIGINAL RESEARCH
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Difficulties and Problem-Solving Strategies in Wayfinding Among Adults With Cognitive Disabilities: A Look at the Bigger Picture

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Many people with cognitive disabilities avoid outside activities, apparently for fear of getting lost. However, little is known about the nature of the difficulties encountered and the ways in which these individuals deal with them. None of the few studies on wayfinding by people with cognitive disabilities have explored the various specific difficulties they meet in everyday life. Using both a qualitative and quantitative methodology, this study aimed at profiling the types of difficulties encountered in urban mobility and the associated problem-solving strategies. In order to provide more direct evidence from the field, we conducted semi-structured interviews using the critical incident technique (Flanagan, 1954). Among the 66 participants interviewed, 44 had cognitive disabilities and 22 were matched controls. The analysis of the transcripts showed in particular an overall reduced autonomy in problem-solving strategies for people with a cognitive disability. The multiple correspondence analysis highlighted three main types of complex situations, covering a comprehensive range of complex situations that are met in everyday life by these individuals. Results also indicated that people with cognitive disabilities request assistance from another person more frequently when a complex event occurs. These situations are discussed as potential cues for improvements in navigational aids. Conclusions and perspectives are provided to improve wayfinding among people with cognitive disabilities.

Keywords: spatial cognition, mobility, semi-structured interview, critical incident technique, navigational aids

INTRODUCTION

Wayfinding as a Cognitive Process

Getting around the city is the first step in many of our daily activities, whether they are related to work or leisure. This activity is therefore fundamental for autonomy as well as for social integration and community access (Doig et al., 2001; Sohlberg et al., 2007). Still, finding one's way in the environment involves more than just movement (Montello, 2017). Apart from controlled locomotion, spatial navigation relies on a set of cognitive processes referred to as "wayfinding" (Montello, 2005, 2017; Wiener et al., 2009). In his early work on urban architecture, Lynch (1960) coined the term wayfinding to describe the use of environmental cues in order to move toward a destination, considering the physical properties of cities that allow a traveler to find their way. During the following two decades, the scope of research expanded toward a more cognitive perspective, centered on human processes rather than on the environment. Wayfinding has been defined as the cognitive process of finding and following a path that links an origin to a destination (Golledge, 1992). It is considered as a spatial problem solving that depends on the construction of mental models, consisting in internal representations of distinct scales of the environment, from a landmark, first-person perspective, to a comprehensive, "bird's-eye" view (Tolman, 1948; Siegel and White, 1975; Johnson-Laird, 1980). The use of these representations along an itinerary relies heavily on mnesic and executive functions, in order to retain spatial information and perform the adequate actions that govern movement (Vandenberg, 2016; Meneghetti et al., 2017). It involves four main cognitive components (Vandenberg, 2016): decision making, orientation, path integration, and closure. The first step, decision-making, implies that several factors have been taken into account, such as selecting the adequate path between the origin and the destination of the trip (Gärling et al., 1986; Golledge, 1995). Decision making also takes place during the trip: while planning and moving through an itinerary, people use their internal representations of the environment to automatically choose and follow a path (Richter, 2007; Brunyé et al., 2010). A second cognitive resource that supports the use of mental models to find one's way is orientation, the capability of knowing where an individual finds themselves in the environment, in relation to the surroundings (Vandenberg, 2016). A third process deals with updating orientation while moving through the environment, keeping track of the motion and continually acquiring information on the environment to maintain the knowledge of one's location in space (Gärling et al., 1986). Finally, the last step and fourth component is closure (Vandenberg, 2016), i.e., realizing that one has reached the intended destination.

Considering the central role of cognition in wayfinding, any condition that affects either internal spatial representations or cognitive processes can result in difficulties in finding one's way (Postma and van der Ham, 2016). Depending on how challenging the environment is (e.g., noisy or dark), one's current state of health, level of fatigue or stress, nobody is "permanently unimpaired" (Arthur and Passini, 2002). This becomes even truer in the case of permanent cognitive disabilities resulting from strokes or head injuries.

What We Know About Wayfinding in People With Cognitive Disabilities

For a long time, neuropsychology has documented difficulties in spatial representations resulting from cognitive disabilities. These studies make clear the impairments as well as their neuroanatomical correlates, and classify several types of disorientation (for reviews, see Aguirre and D'Esposito, 1999; Claessen and van der Ham, 2017). Studies in cognitive psychology have also highlighted the importance of working memory in spatial representations using interference paradigms (Gyselinck et al., 2009) and have shown how cognitive aging could impair these processes (Meneghetti et al., 2012).

Only a few studies gathered evidence of specific difficulties in wayfinding among people with cognitive disabilities, based on both interviews and experimental settings. Results showed that people with cognitive disabilities appear to lack the ability to link landmarks and paths in a bird's-eye view of their everyday environment (Antonakos, 2004), and that they show little independence when facing a complex situation (Lemoncello et al., 2010). In particular, when observing problem-solving situations, Lemoncello et al. (2010) showed that people without cognitive disabilities mostly resolve spatial problems independently by either guessing or walking a little further to look for a landmark. Conversely, people with cognitive disabilities ask the accompanying experimenter for help, or suggest potential solutions that are generally judged to be vague by the experimenter. These findings seem consistent with these individuals' lifestyle: based on group interviews, Sohlberg et al. (2005) showed that they avoid going outside for fear of getting lost, restricting themselves mostly to routine outside trips.

Up to now, the characteristics of the difficulties encountered by people with cognitive disabilities when traveling outside in their daily activities have remained largely unexplored (Meissonnier, 2016; van der Ham and Claessen, 2016; Nakamura and Ooie, 2017). While "getting lost" appears to be a major factor of avoidance of getting around the city (Sohlberg et al., 2005), one can only conjecture on the nature of this problem, its causes and consequences, and the specificity these characteristics represent for the target population in comparison to the general public. Moreover, not much is known about the problem-solving strategies these people actually implement in their everyday life when they face complex situations, whether these consist in getting lost or not.

The scarcity of research data on this topic seems to be caused by a difficulty in recruiting and categorizing participants with cognitive disabilities, some matters directly discussed by most authors (Dawson and Chipman, 1995; Sohlberg et al., 2005, 2007; Lloyd et al., 2009; Cho et al., 2017). For the last 40 years, very few navigational aids have been designed to meet human spatial cognition needs and functioning (Grison and Gyselinck, 2019). This is even truer for people with cognitive disabilities (Sohlberg et al., 2005, 2007), in part due to the lack of information on the nature of the difficulties encountered by the target population in everyday life.

Objectives of the Study

The present study was designed to explore representative everyday situations in order to develop a broader understanding of the effects that cognitive disabilities can have on all the components involved in completing an itinerary. This means not only taking the right decision at a crossroads but also coping with an unexpected delay in transport, getting along with other pedestrians or simply recognizing a building as the destination of the trip (Vandenberg, 2016). We therefore collected the existing experience of complex wayfinding events among people with and without cognitive disabilities. In particular, we explored the problem-solving strategies used when a complex or an unexpected event occurs. We address the potential specificity of the difficulties met by people with cognitive disabilities by comparing them with a matched control group. Based on these results, we provide some insights that should prove helpful in designing better adapted navigational aids. Our results could also clarify the features of ecological wayfinding situations experienced as complex by people with disabilities, opening up avenues for future research.

Note that the difficulty for people with cognitive disabilities in recalling and articulating specific experiences and feelings (Paterson and Scott-Findlay, 2002) usually prevents the use of semi-directed interviews. Still, the documented benefits, such as avoidance of bias and facilitation in user participation, have led some researchers to advocate such investigations, provided certain precautions are taken (Heal and Sigelman, 1995; Cambridge and Forrester-Jones, 2003; Gilbert, 2004). Moreover, results have shown that interviewing the relatives of these people is not sufficiently reliable when investigating outside activities, suggesting that the target population itself should be included rather than proxies (Cusick et al., 2000).

Thus, to address our research questions, individual semi-directed interviews were conducted based on the "critical incident technique" (Flanagan, 1954; see section "Materials and Methods") in order to perform a step-by-step exploration of representative, detailed everyday-life wayfinding experiences. This technique allows the problematic aspects of complex situations to be rapidly highlighted and offers a way to investigate activities that would otherwise be difficult to observe in laboratory settings. Initially developed to gather data with task experts in order to identify critical competencies for their job, the aim of the technique is to avoid the collection of general thoughts and stereotypes about a theme and to favor verbal reports on specific experienced situations recognized by participants to be significant for the theme under investigation. The features of the critical incident technique make it particularly relevant for the study of complex situations such as getting around a city, and for urban mobility in general (Corneloup and Burkhardt, 2016; Grison et al., 2016). Furthermore, a questionnaire on orientation and spatial abilities (Pazzaglia et al., 2000) was administered to characterize the participants of both groups.

As to our knowledge, this is the first research study on wayfinding to use the critical incident technique with people with cognitive disabilities, we adopted an exploratory perspective. We considered every potentially complex situation that the participants recalled, whether they concerned the action of getting lost or an unpleasant trip in a crowded subway. The aim was to determine the most frequent profiles of complex situations, and whether they were associated with a group of participants or not.

MATERIALS AND METHODS

Participant Recruitment

Two groups of participants were recruited. The experimental group was formed on the basis of the following inclusion criteria: being at least 18 years old at the time of the interview (French legal majority), presenting a legally authenticated cognitive disability, and being able to travel alone in town. Participants had to be stabilized and lived autonomously. An exclusion criterion was the existence of disabilities impacting the visual or motor functions, thereby creating difficulties in mobility possibly unrelated to the cognitive disability itself. Forty-seven volunteers with a cognitive disability and meeting our inclusion criteria came forward to participate in this study. Forty-three came from four partner institutions: 10 participants came from home care services and specialized services for handicapped adults (French "SAMSAH"), and 33 were workers in centers providing care through employment to handicapped adults (French "ESAT"). One participant came from the investigator's indirect network. Participants from the institutions were initially identified and invited by the professionals to volunteer for the study. It was made clear to them that any participant strictly meeting the inclusion criteria could volunteer, whether they had already expressed a mobility complaint or not. With the exception of aphasia that would prevent interviewing, no additional selection criteria were applied by the professionals. Volunteers were then contacted directly by the experimenter for an appointment at their home or within the institution when possible. Three participants in the experimental group were excluded since they expressed difficulties understanding the questions during the interview, and the session was therefore interrupted.

The control group was formed based on the following inclusion criteria: being at least 18 years old at the time of the study, absence of cognitive impairment and absence of daily use of a car as a driver. The latter criterion was applied because no experimental participant declared driving. Also, the partner institutions for the experimental group were located in the outskirts of the cities, and the participants themselves often lived in the suburbs. Therefore, control participants were recruited among companies based outside the city center, and had to use different types of transport (mainly trains and buses) every day, in order to match the environmental context of the experimental group.

Sample Characteristics

The experimental group included 44 participants (28 men, 16 women). The mean age was 38.91 years (Minimum = 21, Maximum = 81, SD = 13.50). Amongst them, five participants suffered from the after-effects of strokes (of whom two had had two strokes), nine from traumatic head injury, one from a brain lesion after surgical tumor removal, two were epileptic, four had developmental cognitive disabilities and 23 suffered from cognitive disabilities of unspecified etiologies.

While cognitive disabilities, like motor disabilities, can refer to a wide variety of difficulties, they cannot be specified in a "standardized" way either by a device (e.g., a wheelchair, crutches) or a functional disability (e.g., blindness). As documented in the neurological literature, there are potentially as many disabilities as lesions. More than half of the participants in our study suffered from cognitive disabilities of unspecified etiologies, suggesting pathogenesis heterogeneity. As we chose to focus on complex events in real life, we selected people who traveled autonomously for their everyday activities. Therefore, our participants were not under medical care. Furthermore, most of them did not have access to their neuropsychological and medical specifics. We therefore used the following two inclusion criteria: the stability of the disability, and the legal authentication of cognitive disabilities according to the 2005 disability policy reform. The authentication procedure consists in successive medical examinations. All partner institutions catered only for people who had strictly complied with this procedure.

The control group included 22 participants (8 men, 14 women) meeting the criteria who volunteered to participate, with a mean age of 37.45 years (Minimum = 21, Maximum = 61, *SD* = 14.39).

Design and Procedure

The experimenter first presented the study to the participant in accordance with the content of the information letter. When all the participant's questions had been addressed, they were asked to sign the consent form. For the participants contacted by telephone, a first call was made to present the study; the information letter and the consent form were then sent by mail. Once the consent form had been read and signed, the interview call was made.

Semi-Structured Interview Using the Critical Incident Technique

The semi-structured interview was based on the critical incident technique (Flanagan, 1954). A "critical incident" is a situation specific in time and geographically localized, resulting in either a satisfactory or an unsatisfactory experience for the participant. The critical incident technique consists in a recall of these events guided step-by-step by the investigator. Each interview was audio recorded and then transcribed. The direction of the interview led the participants to describe each step of the complex situation from the general proposition "*Think of a specific moment that you experienced as complex when you moved around the city during the last few months. It can be a moment that was either pleasant or unpleasant in the end.*" The questions asked by the experimenter to guide the direction of the interview focused successively on the step-by-step process of the event, on the feelings experienced by the participant, on the participant's reactions and strategies, and on whether the participant had learnt anything from the situation (using the question "*If you were in the same situation today, would you do the same?*").

When a description was incomplete, the investigator prompted the participant to elaborate using interview techniques (e.g., summarizations, repetitions of keywords, pauses, nods). To complete a description, the investigator also asked the participant if they would judge the situation overall positively or negatively. When the description of one situation was over, the experimenter asked if the participant could think of another complex situation by reiterating the initial general proposition. Then, a second event was detailed in the same way. When the participant declared that they could not remember any other complex event, the investigator summarized all the situations that had been previously described by the participant in order to make sure they had not forgotten anything. The interview was concluded when the participant stated that they did not remember any other complex event. The average duration of the interview was approximately 20 min (Minimum = 3, Maximum = 58, *SD* = 10).

Questionnaires

Two questions about the age and gender of the participant were asked. When a participant from the experimental group was willing and able to define their disability, the experimenter asked one optional question about their etiology (e.g., stroke, head injury). Then a broader question about the participant's travel behavior was asked to induce them to summarize their daily journeys, recall the types of transportation (i.e., pedestrian, bus, subway, train, tramway, driver or passenger of a car, bike, others) and usual durations of these journeys. This open question served as an ice-breaker for the interview and a confirmation for the investigator that the criterion of autonomous travel was indeed met.

A 16-item questionnaire on orientation and spatial abilities was then completed (Pazzaglia et al., 2000). This questionnaire returns six main scores: "general spatial orientation," "knowledge and use of compass points," "survey representation score," "route representation score," "landmark representation score," and "preference toward survey representation over the rest." The procedure for the completion of this questionnaire was the same for both groups. The questions were formulated orally by the experimenter, who scored the participant's answers on the scales. Since the questionnaire had not previously been tested and adapted to people with cognitive disabilities, when a participant from the experimental group expressed a misunderstanding of certain items, the questions were exemplified or reformulated with synonyms in order to be understood by the participants. The goal of these reformulations was to stay as close as possible to the original question, while adapting to the specific cognitive disabilities of the experimental group. The average duration of this questionnaire was approximately 15 min.

Analyses

Collected Data

The interviews of the experimental group took place between July and November 2017. Nine participants were met at their home, 37 were met within the institutions, and one participant recruited by the indirect network of the experimenter was contacted by phone. The interviews of the control group took place between April and June 2018. Twenty-two participants were met either at their office, at their home or by telephone depending on their availability.

The audio recordings of the 66 semi-structured interviews were entirely transcribed. Two hundred and eighteen critical incidents were obtained from the verbatim, of which 126 were from the experimental group (2.9 incidents per person in average) and 92 from the control group (4.2 incidents per person in average). Situations judged as overall positive (such as an experience deemed satisfactory or a time-saving situation) accounted for 19.9% of the cases in the control group and

5.6% of the cases in the experimental group. It is worth noting that situations triggering positive emotions and overall positive situations do not always match, as situations initially resulting in negative emotions could be evaluated as pleasant by the participants in the end.

Among the 44 questionnaires on orientation and internal spatial representations (Pazzaglia et al., 2000) from the experimental participants, three contained unanswered items. Consequently, these three questionnaires were excluded from this analysis. All 22 questionnaires from the control group were fully completed.

Data Coding

The verbatim of all 218 critical incidents were coded using a grid with six variables to delineate the following dimensions of complex events: cause, type, consequence, emotion, problem-solving strategy, and learning from the situation. The first step in coding the critical incidents remained as close as possible to the story recounted by the participant. The six variables from this first step included between 16 and 48 modalities. In a second step these specific modalities (e.g., "event happened because of rush hour," "event caused by the crowds of people") were combined into broader ones (e.g., "event caused by a punctual environmental difficulty") in order to allow analysis. Finally, we obtained six variables ranging from 7 to 12 modalities detailed in **Table 1**. All the variables (except "consequence") include a modality labeled "other" comprising unique situations that did not fit any other modality.

Among the variables, some could not be coded, resulting in 14.9% of missing data across the modalities. This missing information is labeled "N/A."

Statistical Analyses

Univariate Analyses

For each modality of the six variables, the number of occurrences was compiled into contingency tables, as detailed in **Table 1**. These frequencies were compared between the two groups using the two-sided Fisher's exact test, as some frequencies are lower than five. Benjamini–Hochberg multiple comparison correction was applied to all data with a false discovery rate of 0.05 (Benjamini and Hochberg, 1995). An effect of the gender of the participants was also controlled for each variable, and did not seem to occur across all variables.

For each contingency table, we calculated Cramer's V^2, an estimator of the magnitude of the association between two categorical variables (Corroyer and Rouanet, 1994). Cramer's V^2 lies between 0 and 1. We considered the association as strong when V^2 was greater than 0.16 and as weak when V^2 was less than 0.04 (Wolff and Corroyer, 2004). We therefore analyzed the association when V^2 was greater than 0.04.

In the case of significant statistical difference and V^2 greater than 0.04, we calculated the association between each modality of the contingency table. Relative deviations (RDs) measure the associations and are determined on the basis of a comparison between observed and expected frequencies (i.e., those that would have been obtained if there was no association between the two variables) (Bernard, 2003). There is statistical attraction between two modalities when the RD value is positive, and statistical repulsion when it is negative. By convention, only RD with absolute terms greater than 0.25 are retained. All calculated RD are detailed in **Table 2**. When a modality occurred more than once but less than five times in total across the two groups, we ignored the strength of association between this modality and the groups of participants.

Multiple Correspondence Analysis (MCA)

A multiple correspondence analysis (MCA) was performed in order to obtain a profile of the main types of existing complex situations and the group most associated with each one. We performed the MCA in accordance with the guidelines and recommendations provided by Le Roux and Rouanet (2010). This exploratory analysis determines the most significant associations between modalities across all selected variables by determining factorial axes that contribute to the overall variance.

Consistently with our objective of exploring the relationships between features of the complex situations as well as the projection of the group factor on them, we involved all the variables describing the complex events as active variables, while the "group" variable was added as a supplementary variable. Contrary to active variables, a supplementary variable does not contribute to the construction of the axes.

As positive emotions were not mentioned by the experimental group, no correspondence could be observed between the two groups for this emotion. We therefore did not include the "emotion" variable in this analysis. Also, as learning from the event consisted in a reflection after the situation rather than a factual description of the event itself, we excluded this variable from the analysis. We therefore performed the MCA on four active variables describing the characteristics of the situations (cause, type, consequence, problem-solving strategy).

Among the 218 incidents reported by the participants, incidents containing missing values (N/A) across the four selected active variables were excluded, since a missing value cannot correspond to any modality. Incidents containing "other" modalities were also excluded, since "other" covers heterogeneous unique modalities rather than a specific one. Overall positive incidents (corresponding to pleasant experiences or time saving situations) were excluded as a result of the absence of associated problem-solving strategies. In the end, the MCA was performed on 106 critical incidents (63 out of 126 from the experimental group, 43 out of 92 from the control group). In accordance with the requirements of this analysis, previously determined modalities for each variable were combined to reduce the number of modalities per active variable. These "broader" modalities used for the MCA are detailed in **Table 3**.

The contribution of a modality to a factorial axis determines its coordinate on this axis, therefore allowing for a graphical representation of the MCA. The modalities that frequently appear together in the stories of the participants are graphically close to each other.

The interpretation of an axis is permitted by selecting the categories whose contributions exceeded the "baseline criterion," which is determined by dividing 100 by the total number of

TABLE 1 | Variables and modalities determined from the analysis of the interviews, with examples from the verbatim, and overview of the contingency tables for each variable.

Variable	Number of modalities	Modalities	Examples from the verbatim	Number of incidents in experimental group	Number of incidents in control group	Total
Cause of the event	9	Choosing an unusual route	*"I thought why not take this way today"*	1	3	4
		Environment legibility	*"The road sign was hidden, in the air"*	14	10	24
		Initial interindividual conflict	*"I had an argument with my friend, it stressed me on my way back"*	4	1	5
		Initial request for information	*"We asked and they gave us wrong advice"*	4	0	4
		Internal cause	*"I had forgotten there was a deviation"*	11	5	16
		Not recognizing the environment	*"The place was not like I imagined it would be"*	21	9	30
		Punctual environmental difficulty	*"It was snowy"*	20	21	41
		Transport network	*"They were on strike"*	16	19	35
		Other	*"I met a friend of mine"*	5	2	7
Total				**96**	**70**	**166**
Event type	12	Being in an unwanted or unusual route	*"I had to change and take another line"*	2	8	10
		Being lost	*"I couldn't find the street"*	40	14	54
		Conflict with another person	*"There were drunk people in the station"*	9	1	10
		Disruption of the transport network	*"The subway was not working"*	25	31	56
		Harmful situation	*"I slipped on the wet manhole cover"*	5	0	5
		Missing the transportation	*"I missed the last subway"*	4	2	6
		Mistake by another person	*"The driver did not know the route"*	1	5	6
		Obstacle on the route	*"The exit we wanted to use was closed"*	3	8	11
		Physical or emotional discomfort	*"It was too hot in the train"*	6	5	11
		Route mistake	*"I took the bus in the opposite direction"*	18	11	29
		Transport not on time	*"The bus was late"*	5	1	6
		Other	*"My friend gave me a ride to the station"*	8	6	14
Total				**126**	**92**	**218**
Consequence	9	Delay	*"I arrived late for my music class"*	3	0	3
		Detour	*"I had to go to the terminus and come back"*	18	11	29
		Difficulty reaching the destination	*"I never found the place; I was going round in circles"*	61	38	99
		Exploration	*"I discovered a beautiful district I wouldn't have known otherwise"*	1	6	7
		Missed the desired means of transport	*"I arrived too late at the bus station"*	1	2	3
		Resulting harmful situation	*"I hurt my knee"*	5	0	5
		Resulting physical or emotional discomfort	*"It felt never-ending, I couldn't take it anymore"*	14	11	25
		Simplification	*"I gained 15 min thanks to my friend"*	6	3	9
		Wait	*"We waited for 45 min"*	6	4	10
Total				**115**	**75**	**190**
Expressed emotion	7	Anger	*"It was very upsetting"*	26	25	51
		Fear	*"I was in utter panic"*	23	9	32
		Joy	*"I was very happy"*	0	9	9
		Neutral	*"I felt normal, I wasn't stressed"*	13	12	25
		Sadness	*"I was sad"*	8	4	12

(Continued)

TABLE 1 | Continued

Variable	Number of modalities	Modalities	Examples from the verbatim	Number of incidents in experimental group	Number of incidents in control group	Total
		Stress	*"It was stressful"*	14	20	34
		Other negative emotion	*"I was ashamed"*	13	9	22
Total				**97**	**88**	**185**
Problem-solving strategy	9	Changing to an alternative route	*"Eventually I got off the train earlier and walked"*	14	23	37
		Giving up	*"I thought "to hell with that" and I went home"*	3	1	4
		Going back	*"I walked back to find the right street"*	9	6	15
		Looking for a landmark	*"I looked up to find the bell tower"*	8	4	12
		None	*"I waited until the transit failure ended"*	24	16	40
		Planning	*"I used my app to see what to do"*	5	7	12
		Request for assistance	*"I called for them to pick me up"*	16	3	19
		Request for information	*"I asked some students my way"*	31	14	45
		Other	*"I tried to stay still and sat to avoid the heat"*	5	6	11
Total				**115**	**80**	**195**
Learning from the event	9	Would request assistance	*"I would call a cab"*	7	1	8
		Would request information	*"I would ask a bystander"*	3	1	4
		Would be more attentive	*"I would pay more attention"*	5	0	5
		Would manage by themselves	*"I wouldn't ask for help and I'd look at the screen myself"*	3	1	4
		Would give up	*"I wouldn't try to go there again"*	5	0	5
		Would not change anything	*"I would do the same"*	44	41	85
		Would plan	*"I would look at a map"*	9	21	30
		Would take another route	*"I would take the subway instead"*	4	5	9
		Other	*"I would do something else surely"*	7	6	12
Total				**87**	**76**	**163**

Frequencies for each modality are retrieved from the analysis of the interviews.

TABLE 2 | Overview of the significant relative deviations (RD) between each modality of the critical incident variables and each group.

Variable	Modalities	Experimental group RD	Control group RD
Cause of the event			
Event type	Being in an unwanted or unusual route	−0.65	**0.90**
	Being lost	**0.28**	−0.39
	Conflict with another person	**0.56**	−0.76
	Disruption of the transportation network		**0.31**
	Harmful situation	**0.73**	−1.00
	Missing the transportation		
	Mistake by another person	−0.71	**0.97**
	Obstacle on the route	−0.53	**0.72**
	Physical or emotional discomfort		
	Route mistake		
	Transport not on time	**0.44**	−0.61
	Other		
Consequence			
Expressed emotion	Anger		
	Fear	**0.37**	−0.41
	Joy	−1.00	**1.10**
	Neutral		
	Sadness	**0.27**	−0.30
	Stress		
	Other negative emotion		
Problem-solving strategy	Changing to an alternative route	−0.36	**0.52**
	Giving up		
	Going back		
	Looking for a landmark		
	None		
	Planning	−0.29	**0.42**
	Request for assistance	**0.43**	−0.62
	Request for information		
	Other		**0.33**
Learning from the event	Would request assistance	**0.64**	−0.73
	Would request information		
	Would be more attentive	**0.87**	−0.1.00
	Would manage by themselves		
	Would give up	**0.87**	−1.00
	Would not change anything		
	Would plan	−0.44	**0.50**
	Would take another route		
	Other		

Statistical attractions are in bold and repulsions in regular text. Non-significant RD are left blank. Variables that do not statistically differ between the two groups are left blank.

active modalities included in the MCA. As detailed in **Table 3**, we included the 13 modalities of the incidents, therefore the baseline criterion we used equals 7.69%.

RESULTS

Univariate Analyses: Describing the Complex Situations Step by Step in the Two Groups

Spatial Abilities and Events Recall

The Mann-Whitney test indicated that the control group recalled significantly more complex events (Median = 4) than the experimental group (Median = 3) ($U = 247$, $p < 0.01$). The events in the control group also appear to be more frequently judged positive than in the experimental group when comparing the two groups using a Chi-square test of independence [$\chi^2(1) = 8.95$, $p < 0.01$].

A *t*-test was performed on the six scores provided by the questionnaire on spatial abilities and did not indicate any significant difference between the two groups.

Differences in Event Types and Problem-Solving Strategies Between Groups

The types of events differed significantly between the two groups ($p < 0.001$). As a strong association between variables was found ($V^2 = 0.16$), the associations between modalities (RD) were analyzed. There were four statistical attractions and three statistical repulsions in the experimental group. Compared to the control group, people with cognitive disabilities more frequently encountered situations centered on being lost, in conflict with another person, in a harmful situation or in a situation involving a transport schedule problem, either early or delayed. Less frequently than the control group, they found themselves on an unwanted or unusual route, suffered the consequences of a mistake made by another person, or met an obstacle on their route.

The problem-solving strategies implemented differed significantly between the two groups ($p < 0.05$). As a moderately strong association between variables was found ($V^2 = 0.09$), the associations between modalities (RD) were analyzed. There was one statistical attraction and two statistical repulsions for the experimental group. Compared to the control group, people with cognitive disabilities more frequently chose to request assistance from another person, either a bystander or friend. Less frequently than the control group, they chose to change their current route for an alternative route, or stop to plan the rest of their trip. One statistical attraction for the control group was its relationship to the modality "other."

Comparisons for the causes and consequences of the situations between the two groups returned no statistical significances. V^2 and RD are therefore not discussed here.

Differences in Emotions and Learning Between Groups

The emotions generated by the events differed significantly between the two groups ($p < 0.01$). As the association between variables was moderately strong ($V^2 = 0.10$), the associations between modalities (RD) were analyzed. There were two statistical attractions and one statistical repulsion for the experimental group. Compared to the control group, people with

TABLE 3 | Combined modalities obtained from the preliminary univariate analyses and used for the Multiple Correspondence Analysis.

Variable	Modality used for the MCA	Corresponding modalities combined from the preliminary univariate analyses
Cause of the event	Contextual	Choosing an unusual route
		Initial interindividual conflict
		Initial request for information
		Punctual environmental difficulty
	External	Environment legibility
		Transportation network
	Internal	Internal cause
		Not recognizing the environment
Event type	Mistake	Being lost
		Route mistake
	Obstruction	Mistake by another person
		Obstacle on the route
	Transport problem	Disruption of the transportation network
		Missing the transportation
		Transport not on time
	Unpleasant event	Being in an unwanted or unusual route
		Conflict with another person
		Harmful situation
		Physical or emotional discomfort
Consequence	Discomfort	Resulting harmful situation
		Resulting physical or emotional discomfort
		Wait
	Obstacle to achieving the goal	Difficulty reaching the destination
		Missed the desired means of transport
	Setback	Delay
		Detour
Problem-solving strategy	Autonomous action	Changing to an alternative route
		Going back
		Looking for a landmark
		Planning
	Passivity	Giving up
		None
	Asking someone for help	Request for assistance
		Request for information

cognitive disabilities more frequently experienced emotions of fear and sadness when in a complex or unexpected situation. Less frequently than the control group, they experienced joy.

A comparison of the lessons learnt from the events by the two groups was statistically significant ($p < 0.01$). The association between variables was strong ($V^2 = 0.13$), enabling the associations between modalities (RD) to be analyzed. There were three statistical attractions and one statistical repulsion for the experimental group. Compared to the control group, people with cognitive disabilities anticipated more frequently that if they were to encounter the same situation, they would request assistance, be more attentive or give up and not attempt the journey. Less frequently than the control group, they anticipated planning during the complex situation.

Multivariate Analysis: The Main Profiles of Complex Situations

Based on the decrease in the eigenvalues of the MCA, we considered the first two factorial axes for our analysis. They account for 44.55% of the total variance (axis 1 accounting for 27.34% and axis 2 for 17.21%). The contributions of each active modality are detailed in **Table 4**. The weight of the two modalities and the coordinates for the supplementary "group" variable are presented in **Table 5**. The graphical representation of the MCA is depicted in **Figure 1**.

On the basis of the baseline criterion (7.69%) and the contribution of each modality, we used five modalities for the interpretation of axis 1 ("internal" cause, "mistake" and "unpleasant event" types, "discomfort" consequence, and "passivity" problem-solving strategy). Seven modalities were used for the interpretation of axis 2 ("contextual" cause, "obstruction," "transport problem" and "unpleasant event" types, "discomfort" consequence, "autonomous action" and "asking someone for help" problem-solving strategies). Axis 1 opposes critical incidents relative to the individuals ("internal" cause, "mistake") and critical incidents that arise independently from the individual ("unpleasant event," "discomfort"), leaving a limited margin of maneuver to act on the situation ("passivity"). On the other hand, axis 2 opposes the critical incidents dealt with in an autonomous manner ("autonomous action"), and the critical incidents that require external intervention ("asking someone for help").

To analyze the supplementary group variable, we used the deviation between the categories' coordinates on the axes. A deviation between two categories greater than 0.5 is deemed notable (Le Roux and Rouanet, 2010). Axis 1 does not oppose the two groups, as the deviation between their coordinates on this axis is 0.22, as shown in **Table 5**. Axis 2 however opposes the two groups, as the deviation between their coordinates is 0.74. Thus, the experimental and control groups are mainly distinguished in regard to the way they deal with the complex situations, either by acting autonomously (for the control group) or asking for help (for the experimental group).

Overall, this MCA returned three main profiles of complex situations. The first profile we can identify is a complex situation resulting from an internal cause (e.g., "I did not pay attention"), which results in a mistake. In these situations, achieving the initial goal of the trip becomes uncertain. This situation is mainly encountered by people with a cognitive disability. A second situation deals with contextual problems, emerging because of particular circumstances mainly due to the action of other people (e.g., public works, transport network). This situation triggers autonomous problem-solving actions in order to resolve it. It is mainly encountered by the control group. Finally, a third type of situation is an unpleasant event, which causes discomfort and

TABLE 4 | Contribution of each modality of the Multiple Correspondence Analysis to each axis; the columns "left," "right," or "top," "bottom" refer to their coordinates.

Variable	Modality	Contribution to axis 1 (%)		Contribution to axis 2 (%)	
		Left	Right	Top	Bottom
Cause	Contextual		2.24		**16.15**
	External		3.48	2.15	
	Internal	**13.19**		5.86	
Type	Mistake	**11.68**		6.23	
	Obstruction		1.21		**10.38**
	Transport problem		2.24		**9.07**
	Unpleasant event		**15.1**	**7.83**	
Consequence	Discomfort		**20.17**	**8.25**	
	Obstacle to achieving the goal	5.44			0.07
	Setback		0.01		4.04
Problem-solving strategy	Autonomous action	0.69			**16.9**
	Passivity		**19.55**	3.27	
	Asking someone for help	4.99		**9.8**	
Total		***100%***		***100%***	

The modalities whose contributions to an axis reach the baseline criterion to be used to interpret this axis are in bold.

TABLE 5 | Supplementary "group" variable's weight and coordinates.

Modality	Weight	Coordinate on axis 1	Coordinate on axis 2
Control group	43	0.13	−0.44
Experimental group	63	−0.09	0.30

leaves the individual in a state of passivity (e.g., bad weather, crowded transportation). This situation is met mainly by the experimental group.

DISCUSSION

This study aimed to provide an overview of the difficulties actually experienced by people with cognitive disabilities regarding the complex situations they meet while getting around an urban area in their everyday lives. We used the critical incidents technique to identify the characteristics of the complex situations experienced by people with cognitive disabilities, and compared these characteristics to those of the situations encountered by matched controls. We took into account the situation itself as well as the actions implemented to solve a difficulty. Based on the semi-directed interviews, we divided a complex situation into different components, both factual (cause, event type, consequence and problem-solving strategy) and relative to an evaluation made by the participant (emotion triggered by the event, lessons learnt). We analyzed the potential differences between the two groups across these characteristics and determined a profile of the most frequently encountered complex situations. Based on our results, we propose recommendations for future navigational aids and further research on this matter.

Complex Situations Experienced by People With Cognitive Disabilities Are Specific

While the causes of the encountered events appear to share similarities between the two groups, the types of situations encountered differ between people with and without cognitive disabilities. The complex situations met by control participants are mostly related to external events (unwanted route, disruption of the transportation network, having to deal with a mistake made by someone else or meeting a physical obstacle on the route). Conversely, while they also mention external events, participants with cognitive disabilities mostly describe themselves as the main protagonists of the complex situation. More frequently than the control group, they declare being lost and being in conflict with another person as representative complex events that happened to them.

In our study, being lost designates a situation in which the participant declares not knowing where to go anymore. We distinguished this situation from other events such as taking the wrong direction or being on an unwanted route. This representation of the situation of being lost therefore echoes the results of the study carried out by Sohlberg et al. (2005), which showed that people with cognitive disabilities avoid going outside for fear of getting lost. The present results confirm that being lost is indeed among the complex situations most frequently cited by people with cognitive disabilities. This finding is consistent with the result that taking an unwanted route, therefore running the risk of getting lost, seems to be overall avoided by people with cognitive disabilities in the first place: it is one of the least frequently mentioned events in the experimental group.

Another complex event frequently cited in the experimental group, but not by the control group, is the occurrence of interindividual conflict. These conflicts might thus be seen as a specificity of the experimental group and be related to the disability itself. As many professionals from the partner structures and participants themselves mentioned, the difficulty with cognitive disabilities lies in their absence of visibility. Consequently, other users as well as transport workers may behave with them exactly as with other people without taking their specific situation into account, which causes misunderstandings. Besides, it is well-documented that people with cognitive disabilities tend to experience social difficulties and mood disorders (for reviews, see Morton and Wehman, 1995; Carson et al., 2000), which could also increase the conflictual potential of a situation.

Interestingly, while they apparently mention similar causes of situations, the two groups mention different types of complex situations. A control participant may be more able to detect a potential change and adapt their route accordingly, therefore declaring not the initial precursor but the new route as problematic, whereas participants with a cognitive disability may

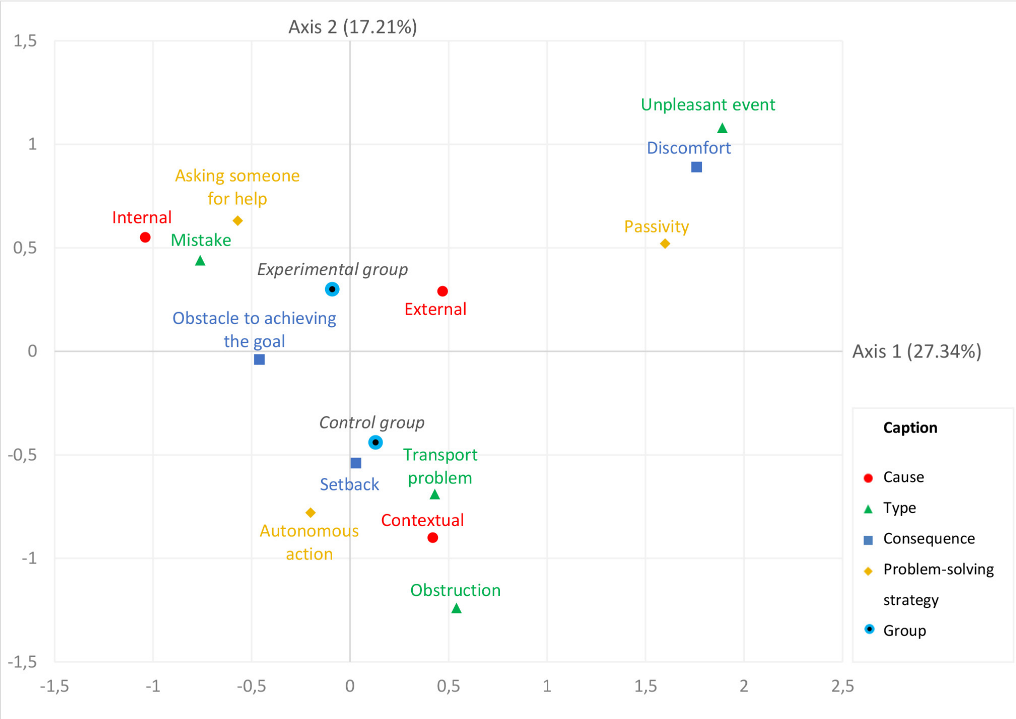

FIGURE 1 | Graphical representation of the Multiple Correspondence Analysis. The coordinates of each modality are determined by its contributions to both axes. The modalities that frequently appear together in the stories of the participants are graphically close to one another. The contribution of each axis to the total variance is indicated in parentheses. Axis 1 (horizontal) opposes events relative to the individuals and events that arise in specific contexts and environments, independently of the individual. Axis 2 (vertical) opposes mainly the experimental and the control groups, with events relative to the control group being dealt with in a more autonomous manner and events relative to the experimental group relying more on the help of another person. The group variable, in italics, is used as a supplementary variable and therefore does not contribute to the overall variance.

experience difficulty in identifying and subsequently adapting their behavior to a new element that is not yet problematic.

Consequences of the Complex Situations: Similarities Across Groups Suggesting Different Traveling Habits

The consequences of the complex situations did not differ between the two groups. Irrespective of the event type, people with and without cognitive disabilities have to make detours, end up being in uncomfortable situations (either physically or emotionally), or have to wait. This is not surprising, as among our sample of 218 critical incidents, 208 deal with situations where people have to go to a specific destination, often at a specific time. Uzan and Wagstaff (2018) proposed five possible categories of motives for an urban journey: physical activity, social activity (e.g., walking around with someone), exploration of an environment, regular route (e.g., going to work), or reaching a place, object or person. The latter two motives comprise more than 95% of the complex situations in our sample. It is worth noting that any consequence of a complex event during this type of journey might therefore disturb the traveler in their activity toward their goal, whatever the event is. These consequences would probably differ for perturbations occurring while doing physical activity outside, for example.

This observation suggests either that participants rarely encounter difficulties when going outside for other reasons than reaching a destination on time, or that they rarely go outside for the other three types of motives. The answer might actually be the latter for the experimental group: as Sohlberg et al. (2005) showed, people with cognitive disabilities avoid outside activities when possible. Therefore, it is not surprising that apart from the journeys that are mandatory (e.g., going to work, to an appointment), they avoid walking around or doing physical or social activity outside. Interestingly, this can be linked to the difference between the two groups regarding the number of complex situations mentioned: participants with cognitive disabilities recall fewer events than control participants. While this may be related to mnesic impairments, it could also be caused by the rarity of their journeys outside, therefore generating fewer complex events.

Different Problems, Different Solutions: People With Cognitive Disabilities Ask for Help While Matched Controls Handle the Situation on Their Own

The problem-solving strategies implemented by the participants reveal an interesting potential for improvements, as they suggest a lesser degree of autonomy for the experimental group.

A major distinction lies in the independence of the action taken: while control participants mostly change their route to an alternative one or plan a solution (either on their phone or on a physical map), the only statistical attraction for people with cognitive disabilities is directed toward the request of assistance from another person. This result therefore provides converging evidence with the findings of Lemoncello et al. (2010) who highlighted the use of the same problem-solving strategy in specifically designed situations where people had to follow incomplete instructions at crossroads. The present study is the first to encompass all the problem-solving strategies reported by people with cognitive disabilities in their everyday life. It is also the first study that compares strategies by people with cognitive disabilities with strategies in the control group when facing complex situations. From this comparison, it can be concluded that the request for assistance is the most frequently used strategy by people with cognitive disabilities. The MCA further strengthens this finding: taking into account all modalities across variables, the experimental group shows statistical ties with requesting help from another person, especially when the event is centered on a mistake (which, in our analysis, includes getting lost and mistaking the route). However, as Sohlberg et al. (2007) and Lemoncello et al. (2010) showed, individuals with cognitive disabilities tend to be vague or inaccurate in their request for help, as judged by experimenters as well as transport workers. This problem-solving strategy therefore appears to be ineffectual for this group.

Another interesting result lies in the statistical attraction of the controls toward the "other" types of problem-solving strategies. Across variables, the "other" modalities group unique events or actions that cannot be combined with any other modality. The attraction toward these "other" modalities in the problem-solving strategies could be interpreted as a form of opportunism, characterized by a greater diversity in the solutions implemented by the controls, as they appear to choose unique, unclassifiable solutions more frequently than people with cognitive disabilities.

Subjective Experience of the Complex Situations: Negative Emotions and Reinforcement of the Strategies Already in Place for People With Cognitive Disabilities

The emotions triggered by complex events are unsurprisingly mostly negative for both groups. However, joy is mentioned in the control group only. In comparison, participants with a cognitive disability express mostly fear and sadness. This is congruent with what has been observed with the causes and event types: the negative emotions might be tied to the difficulty evaluating the origin of the situation and anticipating its evolution. Also, people with cognitive disabilities experience fewer situations that they judge positively overall, compared to the control group. While this evaluation of the event is the object of a separate question and is not necessarily related to the emotion actually triggered during the event, this difference between participants is consistent with the absence of joy observed in the experimental group. This finding is interesting from a wayfinding perspective, as it converges with existing evidence in the literature that emotion structures spatial representations (Storbeck and Maswood, 2016; Ruotolo et al., 2019). In particular, it has been reported that being in a positive mood and feeling positive emotions enhance spatial working memory by favoring a better retention of spatial information, in comparison to being in a negative mood (Storbeck and Maswood, 2016). Emotion has also been shown to affect spatial representations: participants who see landmarks inducing positive emotions while walking along a virtual route are able to locate the landmarks more accurately on a map afterward, as well as drawing the route, in comparison to participants who see landmarks inducing negative emotions (Ruotolo et al., 2019). These findings have led some researchers to advocate the use of positive emotion to improve wayfinding apps in everyday life, for example by computing instructions and routes based on street segments previously evaluated positively by users to allow for an "emotional" wayfinding (Gartner, 2012; Huang et al., 2014). Our results suggest that people with cognitive disabilities, who tend to experience mostly negative emotions when facing an unexpected situation, could also benefit from such a navigational aid proposing routes inducing positive emotions and therefore enhancing spatial working memory and "emotional" wayfinding.

The lessons learnt from the events confirm what is observed with the problem-solving strategies, as a difference emerges between the two groups. People with cognitive disabilities mention more frequently than the control group that they would request assistance, be more attentive or give up and not attempt to make the journey. This strengthens the previous finding that request for assistance seems to be a robust strategy for people with cognitive disabilities. Again, while they mention that they would privilege this action, they cannot plan the action in itself: they do not know in advance at which point of their trip or for what exact reason they would be in need of an outside person. This suggests that for this population, an assistive navigational aid should be available at all times to deal with losing their way at different locations.

The notion of being more attentive is an interesting finding, as it also strengthens the observation that complex situations may indeed emerge from an internal cause. Moreover, this is also congruent with the findings of Lemoncello et al. (2010): this anticipated change seems to be vague, as the participants cannot know in advance what it will be relevant to pay attention to. This also suggests that the directions provided by a navigational aid adapted to this population should tie directions to specific spatial landmarks in order to facilitate the focus of attention on elements that are relevant to the trip.

Giving up, which is also frequently mentioned by people with cognitive disabilities, further confirms the need for an improvement in the mobility of this population. This population, which already avoids most outside activities, contemplates giving up on journeys that are difficult, thereby increasing their difficulty in accessing leisure and social activities.

Overall Remarks on the Complex Situations' Profiles: Not All Events Call for a Wayfinding Improvement

The results of the MCA suggest that not all complex situations can be resolved with a navigational aid, as they do not systematically deal with the act of finding one's way. A frequent profile of complex situations for people with cognitive disabilities concerns an unpleasant event, which causes discomfort, either physical or emotional (e.g., congested transportation, weather conditions). In these situations, the associated problem-solving strategy is mostly passivity, as people wait for the situation to end or simply follow the instructions given by transport workers. This complex situation profile does not seem to hint at a particular solution for people with cognitive disabilities in a wayfinding aid perspective, as they cope with unpleasant conditions rather than spatial cognition. However, while this profile of events does not tie in with decision making, orientation, path integration or closure (Vandenberg, 2016), it can directly impact the following of a path from an origin to a destination (Golledge, 1992). This is particularly true in the case of interindividual conflicts, an event type that occurs especially frequently for people with cognitive disabilities. Our results highlight the multifaceted nature of real-life wayfinding activities, which not only depend on the actual properties of the environment but also on non-spatial properties including the preferences, abilities and beliefs of an individual (Montello, 2017). This emphasizes the interest of taking into account mobility as a whole when discussing wayfinding for specific populations, as an event inducing an actual and recurring difficulty to reach a destination can arise separately from a disability involving the main components of Vandenberg's (2016) model.

CONCLUSION

This study examined the specific difficulties experienced by people with cognitive disabilities during wayfinding. Our perspective was exploratory. Our results show that people with cognitive disabilities encounter specific complex events and especially, that they get lost more frequently. Moreover, they rely more on the help of another person.

Some limitations of our study should be noted. First, as already mentioned, the pathogenesis heterogeneity among our experimental participants might weaken the generalizability of our results, and therefore calls for further studies on this matter. A second limitation directly concerns the cognitive disabilities themselves. Participants with a disability mention significantly fewer events than control participants, a difference that could be linked to the rarity of their urban journeys. Another possible explanation could be that participants with cognitive disabilities indeed suffer from memory impairments. These impairments could thus be a limitation for the validity of the data collected from our interviews.

Still, we can sketch out some recommendations for a navigational aid. As our results tie in with Vandenberg's (2016) cognitive model of wayfinding on several levels, more specific suggestions toward an adapted navigational aid can be proposed. Vandenberg (2016) details four cognitive components of wayfinding: decision making, orientation, path integration, and closure. The analyses of the interviews highlight the relationships between these four components and two variables of complex situations: the event type and the problem-solving strategy implemented by the participant.

The event of "being lost" is among the most frequently encountered by people with cognitive disabilities, and can concern orientation, path integration and closure. One could argue that this situation is already taken into account by existing navigational aids. However, to solve such complex situations, most of the existing solutions provide exclusively bird's-eye views and information (Siegel and White, 1975) which do not meet human needs well as they deal with the most complex levels of spatial representations (Golledge, 1991; Grison and Gyselinck, 2019). These solutions might therefore not be sufficiently helpful for people with cognitive disabilities, since they encounter difficulties mostly based on orientation, path integration or closure when they get lost. The provision of less complex spatial information, such as that based on landmarks, could therefore be considered as more adapted to help this population. Moreover, as "being lost" also refers to situations where individuals do not recognize their destination, obstructing the "closure" part of wayfinding, the analyses of the interviews suggest that an adapted aid should also be able to ease this last step of the journey by describing the destination, either verbally or by showing a picture, in order to make it recognizable by the user.

The problem-solving strategy variable can be directly tied in with the decision-making part of wayfinding. The finding that the most frequently used problem-solving strategy by people with cognitive disabilities is to ask another person for assistance especially hints at a potential improvement for these navigational aids. While people with cognitive disabilities ask someone for help more often than they implement any other strategy, and especially when they are facing an obstacle to the goal of their journey, it has been documented that their requests are often vague, making it difficult for the helper to understand the need and provide sufficient help (Sohlberg et al., 2007; Lemoncello et al., 2010). Still, a possible explanation for the high frequency of use of this strategy despite its flaws lies in the fact that when prompted to describe a route, most people do not use bird's-eye view information such as current aids or maps: they rely on landmarks, which are considered as the key components of route descriptions (Denis, 1997). More specifically, people associate an action to a landmark in order to give directions (Denis et al., 2007). This landmark-based level of spatial information may explain the interest of people with cognitive disabilities in this strategy, also indicated by their intention to ask for help again in the future, as shown by the analysis of the variable "learning from the event." Then again, one could argue that most navigational aids already provide vocal features that could replace information provided by a bystander. However, in current systems, the content of vocal instructions also differs from what a person would actually give. Therefore, an adapted navigational aid should aim at better matching the directions a real person would give and provide instructions linking a landmark to the action to be performed. This would make the aid more relevant to the needs and actual problem-solving strategies of people with cognitive disabilities. Finally, in addition to landmark-based information, considering the negative subjective experience and emotions felt by people with cognitive disabilities, our results are in favor of the use of positive emotions-inducing landmarks in adapted navigational aids, as recommended by several authors, to support a better memorization of spatial information (Gartner, 2012; Huang et al., 2014; Ruotolo et al., 2019).

This study also suggests perspectives for future research. The analysis of the answers to the questionnaire on spatial abilities (Pazzaglia et al., 2000) does not indicate any difference in general spatial orientation, suggesting both people with and without cognitive disabilities have similar spatial skills. Yet, as has been documented, a cognitive disability can be related to several impairments in spatial representations and wayfinding (Lemoncello et al., 2010; Claessen and van der Ham, 2017). Our results provide evidence that people with cognitive disabilities get lost more often than controls. This absence of difference in the questionnaire on spatial abilities therefore does not substantiate the literature, in which most studies have focused only on participants facing prior difficulties in wayfinding. While the present study might indicate an inadequacy of the questionnaire for the target population, or a failure of the questionnaire to detect a difference between people with cognitive disabilities and matched controls, other results suggest a more nuanced picture. Claessen et al. (2017) carried out a study on people with documented cognitive disabilities resulting from strokes. Among their sample of 77 participants, only 33 (43%) actually mentioned difficulties in wayfinding. Moreover, among these 33 people, seven did not show any impairment in internal spatial representations when compared to matched controls over cognitive tests. These data suggest that among the target population, some people do not experience difficulties in wayfinding, and some do not experience difficulties in internal spatial representations. Importantly, these sub-populations may not entirely overlap. Therefore, we cannot rule out that the difficulties observed in wayfinding in the present study do not translate into differences in auto-evaluation of general spatial abilities as measured by the questionnaire. This strengthens the need to supplement quantitative measures by qualitative investigations, allowing deeper understanding of all the dimensions implied in the diverse wayfinding situations encountered by individuals.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

RD designed the experiment, recruited subjects, acquired, analyzed and interpreted the data and drafted the manuscript. J-MB designed the experiment, substantially contributed to the interpretation of the data and critically revised the manuscript. VG designed the experiment, substantially contributed to the interpretation of the data and critically revised the manuscript. All authors gave final approval for publication and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Cognitive Mapping Without Vision: Comparing Wayfinding Performance After Learning From Digital Touchscreen-Based Multimodal Maps vs. Embossed Tactile Overlays

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This article starts by discussing the state of the art in accessible interactive maps for use by blind and visually impaired (BVI) people. It then describes a behavioral experiment investigating the efficacy of a new type of low-cost, touchscreen-based multimodal interface, called a vibro-audio map (VAM), for supporting environmental learning, cognitive map development, and wayfinding behavior on the basis of nonvisual sensing. In the study, eight BVI participants learned two floor-maps of university buildings, one using the VAM and the other using an analogous hardcopy tactile map (HTM) overlaid on the touchscreen. They were asked to freely explore each map, with the task of learning the entire layout and finding three hidden target locations. After meeting a learning criterion, participants performed an environmental transfer test, where they were brought to the corresponding physical layout and were asked to plan/navigate routes between learned target locations from memory, i.e., without access to the map used at learning. The results using Bayesian analyses aimed at assessing equivalence showed highly similar target localization accuracy and route efficiency performance between conditions, suggesting that the VAM supports the same level of environmental learning, cognitive map development, and wayfinding performance as is possible from interactive displays using traditional tactile map overlays. These results demonstrate the efficacy of the VAM for supporting complex spatial tasks without vision using a commercially available, low-cost interface and open the door to a new era of mobile interactive maps for spatial learning and wayfinding by BVI navigators.

Keywords: wayfinding without vision, cognitive mapping, haptic displays, accessible digital maps, blind navigation

INTRODUCTION

Throughout the years, there have been many studies suggesting that people who are blind or visually impaired (BVI), particularly those with early-onset and total blindness, exhibit a range of spatial deficits on spatial learning and wayfinding behaviors as compared to their sighted peers (for reviews, see Golledge, 1993; Millar, 1994; Thinus-Blanc and Gaunet, 1997; Ungar, 2000; Long and Giudice, 2010; Schinazi et al., 2016). Although there is some debate, what can be summarized from this body of literature suggests that BVI individuals tend to perform well on egocentric spatial tasks in small-scale "local" environments, i.e., pointing from their position to another location in a room, or on learning and navigating known routes. However, under-developed spatial skills and performance errors are often cited in the literature with this demographic when learning and navigating larger, unfamiliar environments (e.g., buildings, campuses, neighborhoods, cities, et cetera). The skills needed for successful environmental learning and wayfinding of these large-scale spaces go beyond the perception of one's immediate environment and the following of known routes. Accurate performance requires complex sensorimotor couplings and spatio-cognitive processes that employ allocentric spatial knowledge (understanding spatial relations of the environment independent of one's current position and heading in the space), spatial inference (such as determining shortcuts or detours from known routes), spatial updating (understanding how self-to-landmark and landmark-to-landmark relations change as a function of movement), and developing accurate survey (map-like) knowledge of global environmental relations (Golledge, 1999; Montello, 2005).

Maps represent an excellent tool for supporting many of these critical wayfinding behaviors in large-scale environments that cannot be directly perceived, such as aiding in determining ones current location or planning routes during pre-journey or *in situ* navigation (Montello et al., 2004), and for developing allocentric mental representations of global spatial structure, called cognitive maps (O'Keefe and Nadel, 1978). Although most maps are visual in nature, there is a long history of tactile map use by BVI individuals (Perkins, 2002). Indeed, tactile maps represent an excellent solution for the spatial challenges often ascribed to BVI people as they provide a means of conveying access to off-route landmarks and global spatial structure that is simply not possible to apprehend from nonvisual environmental sensing. We hypothesize that increased availability of tactile maps, accompanied with better formal instruction on how they should be read and used, would greatly improve environmental awareness, spatial inference, and cognitive map development for BVI wayfinders, thereby mitigating many of the spatial deficits that have been ascribed to this demographic in the literature. The two incarnations of accessible maps studied here (a hybrid interactive map and a haptic touchscreen-based digital interactive map) are aimed at meeting this need, each with different pros and cons, as described below.

BACKGROUND

Traditional Tactile Maps

The importance of tactile maps on BVI spatial learning and cognitive map development has strong empirical support in the literature. When comparing tactile map learning and direct experience with blind adults, Espinosa et al. (1998) found that over multiple trials and with delays between training and test, tactile map learners were much better on measures of both route and survey knowledge than participants who learned only from direct environmental experience, showing that access to tactile maps led to improved cognitive map development in terms of accuracy and flexibility. Similar benefits on cognitive map development have been shown with BVI people using tactile maps to learn novel spaces (Golledge, 1991; Ungar et al., 1997a) and for BVI travelers using tactile maps while actively navigating the environment (Blades et al., 1999). Access to maps is especially important for BVI children, as in addition to the benefits found with adults, their use promotes the development of spatial thinking and spatial problem solving (Ungar et al., 1995, 1997b).

Despite the many benefits of tactile maps on spatial learning and navigational performance, they also have several shortcomings that have greatly limited their availability and usage by both BVI children and adults. Most of these issues relate to the map authoring and production process or to limitations of the tangible output. Traditional tactile maps consist of raised elements conveying spatial properties (points, lines, and regions), surface attributes conveying symbolic properties and feature characteristics (dots/dashes, texture variation, and line-height/thickness), and braille labels to convey feature names or semantic information (for reviews, see Edman, 1992; Rowell and Ungar, 2003b). The authoring process for effectively converting visual maps into tactile analogs or developing these materials from scratch involves specialized human expertise, which is expensive in terms of both time and labor costs. Once authored, tactile maps (and other graphical content) are traditionally rendered using specialized, purpose-built, and expensive equipment, such as tactile embossers that produce dots (much like Braille) on hardcopy paper media, raised output produced on thermoform plastic sheets, or tactile output produced on heat-sensitive microcapsule swell paper (Rowell and Ungar, 2003a). Beyond these authoring and production costs, tangible maps/models are limited in that the accessible information provided is static (i.e., presented from a fixed perspective which does not change in register with the observer's movement), cannot be updated if the underlying information changes without re-authoring/production of the map, and the output (often containing many pages of large hardcopy maps) can be cumbersome to carry/use during *in situ* navigation.

Interactive Maps

An alternative to static tactile maps are solutions based on digital displays that convey interactive map information. These displays generally provide context-sensitive information about the navigator's position and orientation on the map through a combination of tactile and auditory feedback. Interactive Digital maps have many benefits and address many of the inherent shortcomings of traditional static tactile maps and models. Some advantages include: (1) they are dynamic vs. static; (2) they can be multimodal vs. just tactile; (3) they can be implemented on portable platforms vs. requiring large-format sheets or map booklets; (4) they are able to be produced on commercial hardware [e.g., the Vibro-audio maps (VAM) rendered using smart tablets studied in this article] vs. expensive, purpose-built hardware; and (5) they support spatial and querying operations that are simply not possible with physical maps (e.g., map panning, zooming, and scrolling operations, where am I queries, and search functionality). Projects developing and evaluating accessible digital maps vary widely in the technology employed and in what nonvisual information is used. A good categorization of this technology, given by Ducasse et al. (2018), distinguishes between "Hybrid Interactive Maps (HIMs)" based on a physical tactile map overlaid on a digital map *via* a touch-sensitive surface and "Digital Interactive Maps (DIMs)" based solely on a digital map.

Hybrid Interactive Maps (HIMs)

One of the earliest incarnations of an accessible digital map was Nomad, a HIM system that incorporated a traditional tactile map overlaid on a digital tablet that provided auditory information about routes and landmarks (Parkes, 1988, 1994). As with most HIM approaches, Nomad worked by registering every x-y location of the tactile map with the corresponding position on the underlying digital map. This allowed for anywhere that the user touched on the tactile map to be augmented with a speech-based label/description (or any auditory information) to be triggered at that location or region. Throughout the years, several other systems have adopted this multimodal audio-tactile HIM approach, with results showing clear benefits for supporting pre-journey route learning, spatial knowledge acquisition, and global environmental understanding by BVI people (Holmes et al., 1996; Jacobson, 1998; Landau and Wells, 2003; Kane et al., 2013). The clear advantage of HIMs is that they afford access to a traditional tactile map but with all the benefits of an underlying digital map. On the one hand, this approach would seem to represent the best of both worlds of accessible map production. On the other hand, the tactile overlays require careful authoring and specialized hardware to produce, the registration process of the overlay to the underlying digital map can be slow and cumbersome, and if the overlay is moved or shifted during use, the correspondence between the physical and digital map is lost. In addition, the use of large touch-sensitive surfaces limits portability and mobile usage. If accessible maps are to be useful tools for BVI navigators *in situ*, as is the case for their sighted peers, then these factors cannot be ignored.

Digital Interactive Maps (DIMs)

An alternative to HIMs are haptic DIMs, representing accessible digital maps with no physical map overlay. These systems generally include some form of haptic information coupled with auditory cues as the user interface (UI) and an underlying digital map. The obvious advantage of the DIM approach is that they are self-contained, with no need for external tactile maps or physical media to be registered with the digital map. Multiple haptic technologies have been used in these interactive maps, including force-feedback devices, refreshable pin-arrays, and vibrotactile displays (see O'Modhrain et al., 2015 for a thorough review). Recent DIMs, as are studied in this article, are even able to be rendered on commercial, mobile hardware, meaning they have significantly lower cost and greater portability than is possible from other interactive mapping solutions.

Force Feedback DIMs

These devices work by providing differential positive/negative forces to the hand or arm to indicate points, edges, and regions of a digital 2D or 3D map/model or by employing differential friction and elasticity information to simulate a material texture or compliance (O'Modhrain et al., 2015). Research with these systems addressing accessible interactive maps has employed either commercial force-feedback devices, such as a haptic mouse or joystick, or a robot arm or manipulandum, such as the PHANTOM, which tends to provide a larger active workspace, more force, and access to 3D renderings but at the cost of greater expense and less portability. Crossan and Brewster demonstrated that the combination of force feedback from a PHANTOM Omni device, with sound feedback and a tactile pin array for specifying the direction, was effective in promoting the learning and navigation of virtual mazes by 10 BVI users (Crossan and Brewster, 2006). In a study comparing learning of maritime environments between traditional tactile maps vs. virtual maps combining auditory sounds/labels and force-feedback using a Phantom device, six totally blind participants showed equivalent accuracy in triangulation of landmark configuration after exposure to both types of maps (Simonnet et al., 2011). Highly similar performance for describing topological relations by five BVI people was also demonstrated after learning an indoor multi-room environment with a traditional tactile map vs. a virtual simulation explored using a commercial Logitech force feedback joystick or mouse, with the most analogous results found with the haptic mouse (Nemec et al., 2004). These studies provide important evidence for the efficacy of multimodal, interactive maps as performance is directly compared with traditional hardcopy tactile maps (HTM), with favorable (i.e., highly similar) results observed after learning from haptic DIMs. These results are in agreement with several other projects utilizing force feedback devices to study haptic learning of maps and virtual environments by BVI users, with results generally being positive in terms of cognitive map accuracy and user preference, e.g., The BATS project (Parente and Bishop, 2003), Open Touch/Sound Maps (Kaklanis et al., 2013), and the Haptic Soundscape project (Jacobson, 2004; Golledge et al., 2005; Rice et al., 2005). Despite these demonstrations, DIMs relying on force-feedback devices are not portable and more importantly, are limited in the information they are able to convey. For instance, they generally rely on a single point of contact (e.g., the Phantom) and while these devices are excellent for conveying surface information (e.g., texture, compliance), force-based interfaces are far less amenable to supporting accurate line tracing and contour following as other DIMs (O'Modhrain et al., 2015), which is problematic as these are critical exploratory procedures for haptic map exploration.

Dynamic Pin Array DIMs

This technology works by individually actuating a matrix of small pins, similar to braille dots, that can be made to move up and down to create dynamic tactile points, lines, regions, and other spatial elements needed for haptic perception of tangible maps (or any visual graphic more generally). Several projects have demonstrated the efficacy of this technology, usually combined with multimodal (audio) cues. Zeng and Weber (2010) showed that audio and haptic information delivered *via* a large, page-sized pin-matrix array (BrailleDis 9000), coupled with an OpenStreetMap (OSM) GIS database, promoted apprehension and pre-journey learning of a university campus by four blind users. The same authors also showed that access to portable audio and tactile-pin matrix system supported accurate understanding of you-are-here symbols during real-time navigation of a north-up OSM street layout by eight BVI participants (Zeng and Weber, 2016). Use of an 8-pin, mouse-like display was also shown to be effective for learning of digital Maps and diagrams in a study with 17 BVI users, especially when combined with an intelligent zooming technique employing intuitive transitions between functional levels of information presentation, rather than traditional stepwise zooming (Rastogi et al., 2013). While dynamically actuated pins have the advantage of providing excellent cutaneous feedback—a hallmark of traditional tactile maps (something that is missing with force-feedback devices and touchscreen-based displays), they involve many moving parts and are expensive to produce and maintain, which has limited their broad deployment. In addition, while there are many types of actuators used in such displays, most are not commercially available and those that are, e.g., the HyperBraille, can cost up to $50,000 for a large pin-matrix display suitable for rendering maps and large graphical content (Russomanno et al., 2015).

Touchscreen-Based DIMs

Touchscreen-based smart devices represent the most recent and fastest growing technology for authoring/rendering digital maps. There are many benefits of these devices for use as an accessible mapping solution. For instance, contrasting with any other technology supporting interactive maps, phones/tablets are built around a portable form factor and computational core that is inherently multi-use, multi-sensory, and incorporates many out-of-the-box universal/inclusive design features in the native interface that benefit BVI users (e.g., screen reader, magnification, and gesture interactions). Perhaps most important, these devices are based on commercial hardware that is inexpensive compared to specialized solutions and is estimated to already be in the hands of 70–80% of BVI cell phone users (WebAIM, 2015; Palani et al., 2016).

With respect to accessible map using, Timbremap was one of the first projects to show that users could learn an indoor layout by exploring a phone's touchscreen with their hand while receiving combinations of speech messages and auditory sound cues to specify the map information, as assessed by the accuracy of subsequent verbal route descriptions (Su et al., 2010). Another system, called Tikisi For Maps, showed that 12 BVI teens could effectively learn and navigate layered map information and perform complex map scaling operations based on audio and speech cues given during exploration of a tablet's touchscreen (Bahram, 2013). Although search performance was varied, Kane et al. (2011) showed that 14 BVI participants could learn the spatial relations of auditory targets *via* bimanual exploration of a large interactive table-top touchscreen.

Touchscreens and Haptic Interactions

A growing number of studies have gone beyond only using auditory/speech information on the phone/tablet by also leveraging the device's built-in vibration motor to provide haptic (vibrotactile) output. While information access is still performed by the user moving their finger around the device's touchscreen, haptic effects are triggered by vibrating the device whenever their finger contacts an onscreen visual element. These vibrations provide robust focal stimulation to the finger, leading to the perception of feeling tactile points, lines, and regions on the touchscreen (Giudice et al., 2012). An obvious shortcoming of this approach is that unlike the traditional reading of tactile maps, there is usually only one point of contact (generally the dominant index finger) when exploring the touchscreen and there is no direct cutaneous stimulation on the finger—the user is just touching a flat glass surface. This means that many of the explicit tactile cues used with traditional embossed tactile maps (or dynamic pin-array systems) are not directly specified using vibrotactile displays, i.e., immediately perceiving a line's orientation, thickness, and elevation (Klatzky et al., 2014). These attributes can be specified *via* vibrotactile cuing, but doing so requires active hand movement and a slower extraction process (Giudice et al., 2012).

We posit that these limitations are more than offset by the positive attributes afforded by the use of vibrotactile information. For instance, tactile information does not mask other potentially useful (or dangerous) environmental cues during real-time navigation, as is often the case when using audio/speech only displays. In addition, in contrast to the other haptic DIM approaches discussed here, the creation of touchscreen-based DIMs allows for the combination of vibrotactile and audio information using portable, inexpensive commodity hardware and does not require the time and effort to produce and register a tactile overlay with the underlying digital map (e.g., HIMs). Finally, the combination of vibrotactile cues, auditory information, and kinesthetic feedback from hand movement on the touchscreen provides more useful information about a map than is possible from touchscreen-based audio-only maps. Indeed, as haptic information (including vibrotactile cuing) is most similar to visual sensing for encoding and perceiving spatial information (as is critical to map using), touch has been argued as the preferred nonvisual analog for conveying spatial data (Giudice, 2018). Support for this claim comes from both behavioral studies and neuroimaging research showing the similarity of spatial representations built up after learning from vision and touch. For instance, functionally equivalent performance on spatial updating tasks has been found after learning haptically or visually encoded route maps (Giudice et al., 2011) and an fMRI study demonstrated that the same brain region, called the Parahippocampal Place Area (PPA), was similarly innervated by spatial computation of scenes apprehended through haptic and visual perception (Wolbers et al., 2011). Explanations for these findings of common performance between modalities, coupled with the same neural basis of action, have been explained by models from several theorists. At their core, all of these models argue that the information learned from separate inputs is stored in unified "amodal" spatial representations in the brain that can be accessed and acted upon in a functionally equivalent manner when supporting subsequent spatial behaviors, e.g., the Spatial Image (Loomis et al., 2013), the metamodal brain (Pascual-

Leone and Hamilton, 2001), or the spatial representation system (Bryant, 1997).

Haptic Touchscreen-Based DIMs

Given these clear advantages, there are a growing number of studies utilizing touchscreen-based smart devices for providing BVI users with audio-tactile access to many types of graphical content (for general reviews, see Grussenmeyer and Folmer, 2017; Gorlewicz et al., 2019). With respect to interactive multimodal maps, TouchOver map was an early project showing that eight blindfolded-sighted users could accurately reproduce an OSM-based road network learned from a touchscreen-based map rendered with vibrational cues indicating roads and auditory labels providing street names (Poppinga et al., 2011). A study using purely vibrotactile cues also showed accurate learning of simple street maps with six BVI users as explored from both a phone and a watch touchscreen interface (Grussenmeyer et al., 2016). In a study using an early version of the VAM touchscreen-based DIM interface evaluated here, nine blindfolded-sighted participants were found to be as accurate in learning an indoor hallway layout with the VAM as with a traditional tactile map, as assessed by both pointing and map reproduction tasks (Raja, 2011). Research investigating rendering of large format maps that extend beyond a single tablet display has also demonstrated that VAM-based DIM interfaces are effective for supporting both nonvisual panning and zooming operations. For instance, accurate learning of simulated indoor maps requiring nonvisual panning techniques to learn the entire spatial extent was shown by 6 BVI users on egocentric pointing and map recreation tasks (Palani and Giudice, 2017), and by performance on similar tasks by 12 blindfolded-sighted participants found after learning VAMs requiring nonvisual map zooming (Palani et al., 2016). Adopting a slightly different approach, the GraVVITAS project demonstrated that floor-plan maps could be accurately understood by six BVI users who explored a touch tablet with multiple vibration motors attached to their fingers (Goncu and Marriott, 2011) and the SpaceSense project showed that 12 BVI users could accurately learn spatial relations of a street network using a 3 × 3 grid of external vibration motors mounted in a case on the back of an iPhone (Yatani et al., 2012). The advantage of these external vibration systems is that haptic cuing could be given to multiple digits and triggered at different regions of the screen when touched, rather than relying on only one finger and a single vibration motor (i.e., the limitation of available commercial hardware). The downside of using external vibration motors is that it requires the purchase of additional hardware and software coordination with that hardware, thereby increasing system cost and design complexity, which will inevitably reduce adoption by BVI end-users compared to systems based on unmodified commercial hardware. There is obviously a trade-off of many factors when designing new technologies but given the specific challenges of the prohibitive cost, limited usability, and low adoption for many assistive technology projects, we argue that it is most important to develop solutions with the highest probability of actually reaching the target end-user for whom it will most benefit. As there is already significant penetration of smart devices by BVI users, we believe it is most fruitful to develop solutions that can leverage all the existing advantages of this technology without requiring any additional hardware. We also feel strongly in the principle of utilizing as much multimodal information as possible from auditory, haptic, and kinesthetic channels, even if redundantly specified, as both empirical and user preference results from multiple touchscreen-based mapping studies support the benefit of multimodal vs. unimodal interfaces. For instance, the development of cognitive maps was more accurate (and less effortful) when the digital maps were learned by 12 BVI users with a combination of vibration and audio feedback vs. only audio information (Yatani et al., 2012). Similar empirical/preference benefits for combined haptic and audio multimodal touchscreen interfaces vs. their unimodal analogs have been shown for map learning with 14 BVI participants on comprehension of indoor layouts (Adams et al., 2015), with 6 BVI and six blindfolded-sighted user's on map recreation tasks after learning the relation between three landmarks on a tablet-based digital map (Simonnet et al., 2019) and with map learning by 12 BVI users *via* a small touchscreen-based watch interface (Bardot et al., 2016). In aggregate, these studies demonstrate the value of using multimodal information for learning maps *via* the touchscreen, and germane to the current study, show that the use of vibrotactile information is particularly important for supporting cognitive map development and is most preferred as a mapping interface by users.

EXPERIMENT AND METHODS

The current study addresses environmental learning, cognitive mapping, and wayfinding performance by blind and visually impaired (BVI) participants in unfamiliar indoor layouts (floors of university buildings). The study was designed to directly address two key gaps in the extant literature on accessible digital maps.

(1) Comparison between DIMs, vs. hybrid interactive maps (HIMs): most research with interactive tactile maps has used one or the other of these techniques but has not directly compared them using a within-subjects design and testing procedure that explicitly probes cognitive map development and wayfinding performance. This study evaluates learning with a VAM, which is a DIM that is rendered using vibrotactile and auditory information and is explored on the touchscreen of a commercial tablet vs. learning by exploring a HIM comprised of a traditional HTM overlaid on the same touchscreen and augmented with the same auditory cues. This comparison is important as the VAM-based DIM is implemented on commercial touchscreen-based smart devices and does not require the cost/effort associated with production and registration of additional tactile maps, i.e., the HIMs approach. Results showing that learning from the VAM supports similar performance as is found with the HTM-based HIM would open the door for a new class of DIMs that

can be easily rendered and broadly deployed on what has become the fastest-growing computational platform used by BVI individuals.

(2) Perceptual vs. cognitive focus: Most of the research discussed above, irrespective of the technology used to render the digital maps, has focused on perceptual factors relating to map reading (e.g., information extraction/encoding). When cognitive factors were addressed, they related to how well the map information was learned and represented in memory. Common performance metrics included map reconstruction, distance and direction estimates, route following, and answering spatial questions. Most studies assessed map learning through non-ambulatory spatial tasks (but see Zeng and Weber, 2016) and did not probe the relation of how map-learning impacted cognitive map development. While cognitive maps were assessed through map reproduction tasks, this approach does not speak to how well those cognitive maps can be subsequently accessed and used during *in situ* navigation, i.e., the gold standard metric for determining efficacy for actual usage. Here, we adopted an environmental transfer technique that allows us to directly evaluate cognitive map accuracy by comparing wayfinding performance after learning maps rendered using a DIM vs. a HIM on a common testing protocol done from memory (i.e., without access to the map). With this approach, virtual representations of the two physical environments are first learned through free exploration using the two accessible mapping conditions. After the learning phase, participants are brought to the corresponding physical environment and asked to perform wayfinding tasks, such as target localization, route finding, or spatial inference. Since no explicit routes were specified during map exploration, and the map used during learning is not present at the test, successful *in situ* wayfinding behavior requires accessing an accurate cognitive map built up during the learning process. Similar transfer paradigms have proven effective for supporting accurate navigation in large physical indoor layouts after learning in the corresponding virtual space using verbal and auditory interfaces (Giudice and Tietz, 2008; Giudice et al., 2010; Connors et al., 2014). To our knowledge, the transfer paradigm has never been used to study cognitive mapping and wayfinding behavior after learning large-scale environments using touchscreen-based DIMs. However, a study by Brayda et al. (2018) has investigated small-scale environmental transfer using a refreshable pin-based dynamic tactile display. In their study, 10 BVI participants learned a map conveying a scaled representation of a physical 6 × 4.5 m room and a virtual target either *via* a static tactile map or from a dynamic 12 × 16 pin array display. After haptic learning, participants first recreated the map and then were brought to the physical room and asked to walk to the target location. Results showed that access to the dynamic map at learning led to lower errors on map recreation and faster, more accurate, and greater confidence during subsequent physical room navigation. While this study did not compare a haptic DIM with a HIM, i.e., the tactile map control was static, and the environment was much smaller than is studied here, we believe that its findings, in conjunction with results from the transfer studies in large-scale environments with auditory displays, suggest that learning from our interactive map displays will lead to accurate wayfinding performance at test. Further, building on the success of previous studies evaluating touchscreen-based haptic DIMs for map learning, discussed above, we predict that the use of the VAM-based DIM at learning will be as effective as learning from the tactile map overlay (HIM) in cognitive map development and subsequent wayfinding behavior.

Participants

Eight blind participants, four females and four males (ages 18–55, *SD* = 13.9), were recruited for the study (see **Table 1** for participant demographics). All participants were daily iPhone users and had received at least 10 h of formal orientation and mobility training. The study was approved by the University of Maine's IRB and all participants were given informed consent and were paid for their time.

Environments

Three virtual indoor map layouts were created based on partial floor plans within two buildings at the University of Maine. The practice map included a section of the first floor of Boardman Hall and the two experimental maps included sections of the third floor of Little Hall. The experimental maps/building layouts varied in overall topology but were matched in terms of their complexity. The experimental maps (and associated physical layouts) were similar in size, e.g., the overall corridor length of the navigable space was 398 and 411 ft (121.3 and 125.3 m) and both layouts consisted of 3 two-way intersections, 2 three-way intersections, two dead ends, and a loop (see **Figure 1**). All junctions were 90° and all participants were unfamiliar with the testing environments prior to the experiment. Each map contained a home location and three unique targets (map 1: doll, cat, knife; map 2: duck, carrot, shoe). Target names were selected from an index of highly visualizable and readily memorable words (Snodgrass and Yuditsky, 1996). The target name was spoken *via* synthesized speech triggered whenever its x-y location on the map was touched during the learning period. The target and home locations were selected to ensure they were spread evenly throughout each layout as well as to provide multiple walking paths between each object, thus allowing us to assess optimal and sub-optimal route-finding performance at test.

Apparatus

Maps were presented *via* a Samsung Galaxy Tab (GT-P2610) with a 7.6″ × 4.8″ (19.3 × 12.2 cm) screen running Android 3.2 Honeycomb. Following earlier work in our lab, each map consisted of lines [0.35 in (0.9 cm)] on the tablet which represented walkable hallways (see **Figure 2**) and squares [0.35″ × 0.35″ (0.9 × 0.9 cm)] which represented objects and hallway junctions (Giudice et al., 2012). When a user's finger touched a map element, the interface provided information *via* text-to-speech audio labels. Spoken elements included key

TABLE 1 | Descriptive information for blind participants is provided for the following factors: etiology of blindness, age of blindness onset, residual vision (if any), level of education (high school = HS, undergraduate degree = UG, and graduate degree = G), primary mobility aid, and days per week of independent navigation.

Etiology of blindness	Onset age	Residual vision	Education	Mobility aid	Ind. Nav. Days
Diabetic retinopathy	29	N	UG	Cane	2
Microphthalmia	Birth	N	UG	Dog	7
Retinitis pigmentosa and glaucoma	Birth	Avoid obstacles	UG	Cane	7
Posterior polymorphs dystrophy, cataracts and glaucoma	Birth	N	UG	Dog	2
Lebers congenital amaurosis	Birth	Light/dark, blurry objects, avoid obstacles	UG	Dog	7
Genetic neurofibromatosis (optic pathway tumor)	5	Blurry objects, fingers at arm's length	HS	Cane	3
Lebers congenital amaurosis	Birth	Light/dark, blurry objects, avoid obstacles	G	Dog	7
Juvenile glaucoma	Birth	Light/dark, blurry objects, avoid obstacles, fingers at arm's length	UG	Cane	7

features of the map such as hallway junctions: "corner," "three-way," or "dead-end," or target objects: "cat." Users could tap on any part of a hallway to hear the total length of the corridor between junctions (in feet). The spoken target labels and home location could also be repeated by tapping on its x-y position on the map. Volume was user selected, and the speech played at a rate of approximately 150 words per minute. The edges of the screen were framed using cardboard (see **Figure 3**) to provide a haptic border and to eliminate accidental contact with any "soft" buttons of the device that could interfere with the map presentation software. Maps were presented *via* either the VAM interface (DIM) or a HTM that was mounted on the same tablet (HIM), allowing for identical auditory cues between conditions and equivalent logging of finger movement behavior.

The VAM interface, representing a haptic DIM (see **Figure 3**) provided user feedback in the form of a continuous vibration when the user's finger touched a walkable hallway and a pulsed vibration when the user's finger touched an object or the home location. Contact with a walkable hallway produced a steady, 250 Hz vibration. Contact with objects and hallway junctions produced a pulsed vibration consisting of a 100 ms pulse (50% duty cycle). Use of pulsing to differentiate map/graphical elements and to draw the user's attention to that location has been found to be important for information extraction and interpretation using vibrotactile stimuli in other studies using similar non-visual touchscreen interfaces (Giudice et al., 2012; Klatzky et al., 2014).

The HTM overlay, representing a haptic HIM (see **Figure 4**), provided embossed tactile lines of the corridors instead of vibrotactile lines. These lines were produced using a View Plus Tiger Emspot embosser with 20 DPI resolution. The audio cues and functions (e.g., tap for hallway length and repeating of audio labels) with the HIM were identical to those in the VAM condition. As traditional tactile maps do not generally use (or need) additional cues to indicate vertices or intersections, as has been found for VAMs (Giudice et al., 2012), they were not included on the HTM-based HIM. However, to provide a redundant cue for targets, both map modes employed an alert tone that was triggered at that location prior to the speech message. As such, although the maps were not strictly identical, they were functionally matched in terms of all relevant information based on the rendering modality. Participants were blindfolded during map learning in both conditions to eliminate the chance of unintended effects of vision for people with any residual sight.

Design and Procedure

A 2 (VAM-based DIM vs. HTM-based HIM) × 2 (two experimental map layouts) mixed factorial design was employed. Each participant ran in two conditions, including learning both layouts, using both map interfaces (one for each layout), and performing wayfinding tests in both physical environments. We intended for interface and map layout to be counterbalanced but due to a balancing error, five participants started with the DIM and three started with the HIM (thus the design was not fully counterbalanced). The experiment took between 75 and 120 min for each participant.

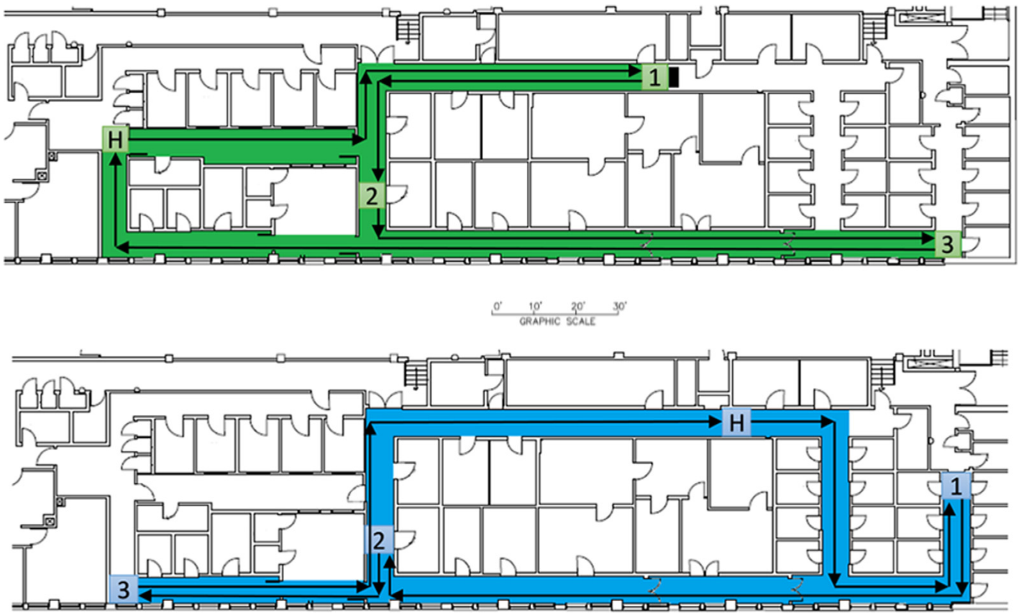


FIGURE 1 | Building floorplan and maps for the two experimental conditions. The highlighted region of the floorplan was selected for the map. The four, square regions in each map represent the three objects and home locations of the maps. The arrows illustrate an example of optimal route efficiency for successful wayfinding (start and end at H).

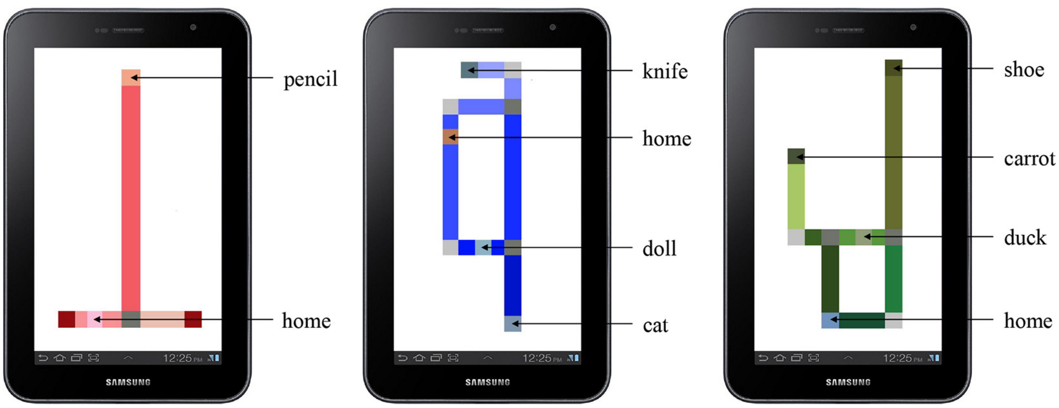


FIGURE 2 | The practice map (left) and experimental maps (center and right) as rendered on the tablet. The home and target object locations are labeled.

Practice Phase

Each participant began with a practice phase which started with an explanation of each map interface. They were first given examples of the stimuli to familiarize themselves with how each would feel. The sample HTM (which would be familiar to most BVI people) was given and then a sample practice map was provided using the DIM. They were also encouraged to practice haptic scanning strategies using the DIM, with several exploratory procedures demonstrated based on evidence of effective strategies found in earlier studies from our lab (Giudice et al., 2012; Palani and Giudice, 2017). One exploratory procedure involves back and forth sweeping of the finger across the vibrating line. Another strategy was to trace a circle around an encountered junction to determine how many legs (i.e., hallways) were intersecting at that decision point.

During the formal practice session, participants were given up to 5 min to freely explore the practice map, which consisted of a single object and two hallways (see **Figure 2**). Participants were instructed to inform the experimenter when they felt confident in their understanding of the spatial layout of the map as well as the location of the object within the environment. After initial exploration, the practice map was removed and a spatial memory task was administered as a learning criterion test. For this test, they were instructed to indicate the target location using a blank map. To perform the task, participants were provided a sheet of cardstock on which the borders of the tablet screen were represented by an embossed line. An embossed square

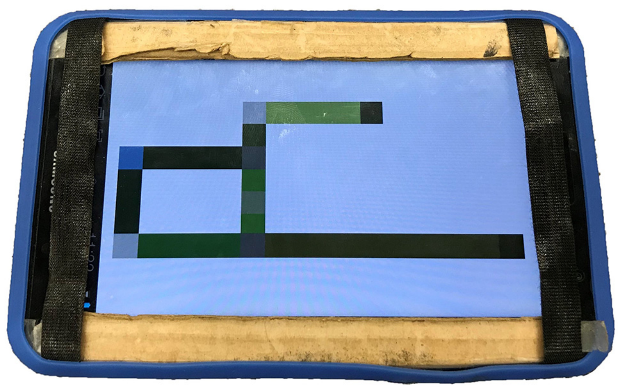


FIGURE 3 | Photograph of the vibro-audio map (VAM)-based Digital Interactive Map (DIM) as used by participants.

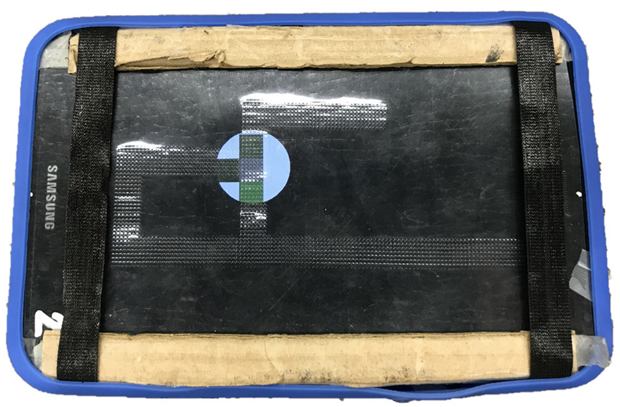


FIGURE 4 | Photograph of the Hybrid Interactive Map (HIM) tactile overlay as used by participants.

[0.35″ × 0.35″ (0.9 × 0.9 cm)] was provided that corresponds to the home position on the map. They were then instructed to indicate the approximate location of the target object within the map by placing a finger at that location (which was then marked on the paper by an experimenter). Participants were considered to have passed the criterion test if they were able to (1) correctly identify the target object (or multiple targets for the experimental conditions) by name; and (2) place the named target(s) on the paper such that the relative positions between each target and the start location maintained the correct global spatial relation (topology) of the original map. If participants did not pass this spatial memory criterion test, they were to be allotted an additional 5 min of learning time before reattempting the test (no participants required more than one learning period to pass).

Participants were then led (blindfolded) to the real-world location they had studied using the practice map and were positioned at the location corresponding to the "home" location from the map. The blindfold was removed, and they were then instructed to walk to the location where they believed the target object was located (the object was not physically present). The trial concluded when the participant informed the experimenter that they were standing at the physical location where they believed the target object had been located on the map.

Note that the practice phase was designed to clarify the experimental procedure, and to ensure any procedural based issues were not present in the experimental trials. Due to time constraints, the full practice session only occurred using the DIM, which was a completely novel interface as compared to traditional tactile maps. If problems were to arise with the procedure, they were more likely to manifest with the unfamiliar VAM-based DIM and we wanted to make sure that such issues were fully resolved before the experimental trials. As such, the practice was done with the DIM to ensure participants understood the task and could effectively use (and were comfortable with) the novel interface. It is possible this may have introduced a slight advantage for the DIM compared to the HIM. However, considering that our overall goal was to evaluate the effectiveness of the DIM to support wayfinding behavior, this possibility was deemed acceptable for the present research. Upon accurate completion of the practice phase, participants were guided (blindfolded) to the third floor of Little Hall to begin the two experimental conditions.

Experimental Phase

Learning Task

Participants completed the map learning task while seated, with the tablet placed in front of them. They were allowed to adjust the position of the tablets based on personal preference. Prior to the learning tasks, all participants were blindfolded. The learning phase began immediately after the experimenter placed the participant's dominant index finger on the "home" position on the map.

Learning was self-paced, with participants given up to 10 min to freely explore the experimental maps. This time limit was based on the average exploration time established during pilot testing. They were instructed to find the three objects (in addition to the home location), learn the global spatial layout of the map, and to remember where the three target objects were located within the map. Although they knew that they would be required to find routes between targets at test, explicit route information was not given during the learning phase. The learning period ended either when participants informed the experimenter they felt confident in their learning, or when 10 min had elapsed.

The learning criterion test was then administered requiring correct placement of the three targets (as described in the "Practice Phase" section). All of the participants met the criterion after the first learning period.

Environmental Transfer Phase

After meeting criterion, participants were led (blindfolded) to the corresponding real-world floor layout, and positioned at the same "home" location, in the same orientation, as was used during map learning. They were then asked to lift their blindfold and were given a target name to find in the environment. The targets were not physically present in the environment. During this wayfinding task, they were instructed to walk at their normal speed to the given object location using the most efficient route possible and to stop and verbally name the target once they believed they had reached its location (e.g., "I am now at the carrot"). Access to the digital map or any indication of the targets in the environment was not provided during testing, meaning that correct wayfinding performance required accessing an accurate cognitive map in memory built up from previous map learning. Testing occurred without blindfold as we were interested in the ability to access the learned cognitive map to perform wayfinding tasks, not in participant's mobility skills for using their cane or dog guide to detect environmental features (which are very different skills). As such, to ensure that all BVI wayfinders had access to the same layout information, and to avoid any potentially confounding biases from individual differences in mobility performance, the experimenter verbalized basic corridor intersection information when encountered during the route-finding task. Thus, whenever the participant entered an intersection, they were provided a consistent verbal prompt structured to match the information provided during learning (i.e., "three-way," "corner," or "dead-end"). This information was provided so participants could focus on the navigation task of interest, rather than mobility challenges of identifying the intersection geometry. The experimenter did not give any information about where to go at the intersection or other cues about the current location or target position, meaning that users had to independently plan/execute their walking trajectory. After a successful navigation attempt, participants were then asked to navigate to the next object. In this manner, the participant navigated to the first object, a second object, third object, and then a return to the home location. If the participant incorrectly localized the target, they were informed they made an error and were walked (blindfolded) to the correct target location, where they once again lifted the blindfold and proceeded with the next navigation trial. This corrective procedure ensured that errors did not accumulate between route trials. Two experimenters accompanied the participant through this *in situ* navigation phase. One supervised the participant, prepared the route by opening doors and blocking unused halls, and recorded navigation time *via* a stopwatch. The other experimenter recorded the participants' route and their response for each target location on a printed floorplan.

Statistical Analysis

Our goal in this research was to evaluate the use of a new commercial DIM, called a VAM as a robust alternative to traditional HTM overlays on digital maps (e.g., HIMs). Based on the efficacy of previous research with similar Vibro-audio interfaces, our hypothesis was that learning with the VAM-based DIM used here would demonstrate functionally equivalent wayfinding performance as was observed after learning from the information-matched HIMs. When the *a priori* goal of hypothesis testing is to not find an effect between conditions, i.e., not reject the null hypothesis, the use of traditional frequentist based null hypothesis significance testing (NHST) is less meaningful, as these procedures allow for the determination that data are unlikely given that the null hypothesis is true (Raftery, 1995), but they do not provide evidence in support of the null hypothesis (which is the goal of the present research). An advantage of the Bayesian approach is that it provides an opportunity to analytically determine whether the null hypothesis is more likely than the alternative(s) given the observed data (e.g., Raftery, 1995; Gallistel, 2009). As such, the current data was analyzed using Bayesian methods. Although these procedures require more effort, we believe they are better suited for our purposes and that this is the first use of this rigorous (equivalence) analysis with wayfinding data in the BVI literature.

RESULTS

The effect of the user interface [i.e., learning a building layout *via* a DIM (touchscreen-based VAM) vs. a HIM (touchscreen-based HTM)] was evaluated using three dependent variables (DVs): (1) wayfinding accuracy; (2) route efficiency; and (3) learning time. Wayfinding accuracy was recorded as a binary variable (0 or 1) indicating failure/success for each target localization trial during the wayfinding task. Each route navigation trial was recorded as correct if the participant stopped within a 12 feet (3.7 m) radius of the target location. This accuracy threshold was selected because it is similar to the spatial extent of a single finger width on the map. Thus, there was a natural correspondence between felt location during learning and the error allowed during navigation in the physical space when localizing the target. Route efficiency was also recorded as a binary variable indicating if, during successful wayfinding, the route navigated was either the shortest/most direct route or a longer/suboptimal route. Each of the eight navigation trials were designed so that there were two possible direct routes to the target. The most efficient route was the shortest and required the least amount of turns. The direct (but inefficient routes) included traveling a greater distance, more turns, or both. Additionally, any successful navigation trial not following any of these defined routes was scored as inefficient. This included situations in which participants changed their route mid navigation (e.g., backtracking). These types of accuracy data are often analyzed by submitting participant averages (e.g., proportion correct) to an ANOVA or *t*-test; however, accuracy data (like that in the present study) are often based on a series of binary (correct/incorrect) outcomes. As a result, there is a growing body of literature that argues it is more appropriate to use generalized linear mixed-effects probit/logit models to analyze these types of accuracy data (Dixon, 2008; Jaeger, 2008; Quené and van den Bergh, 2008; Song et al., 2017). Therefore, these data were evaluated using separate mixed-effects probit regression models to estimate the effect of the interface (DIMs vs. HIMs) on the DVs of wayfinding accuracy and route efficiency. Each model included random effects (varying intercepts) for subjects and the target during wayfinding. The effect of the interface was included as a fixed effect in each model. Initial considerations of the raw data (see **Table 2**), suggests that the DIM is effective as a navigation aid. These data revealed similar wayfinding accuracy using both the HIM (78%) and DIM (75%). Additionally, the route efficiency data suggest that participants were using accurate cognitive maps during wayfinding after learning with both the HIM (79%) and the DIM (67%) conditions. The primary disadvantage of the DIM

TABLE 2 | Mean participant data (±1 SD) for accuracy, route efficiency, and learning time.

Interface	Accuracy (%)	Route efficiency (%)	Learning time (s)
HIM/HTM	78 (±25)	79 (±19)	194 (±111)
DIM/VAM	75 (±23)	67 (±32)	364 (±180)

Means for accuracy and route efficiency were calculated as the mean of each participant's average wayfinding accuracy or route efficiency.

over the HIM is revealed in the learning time data in which the mean learning time was 194 s for the HIM and 364 s for the DIM. This disadvantage is not surprising as the DIM has less explicit cues which may slow the learning time, albeit not the overall wayfinding performance. In the following sections, we consider the effect of interface on the DVs of wayfinding accuracy and Route Efficiency. First, we define the statistical models used in this analysis ("Bayesian Model Description for Wayfinding Accuracy and Route Efficiency" section). Then, we describe how we calculated and evaluated the posterior distribution ("MCMC Sampling" section). Next, we present our conclusions based on the posterior distribution ("Evaluation of the null hypothesis *via* HDI and ROPE" section). Finally, separate analyses for the DV of learning time are presented ("Learning time" section).

Bayesian Model Description for Wayfinding Accuracy and Route Efficiency

For both wayfinding accuracy and route efficiency, multilevel models (see **Figure 5**) considered participants' responses using a Bernoulli distribution with a probit link function. Prior distributions for the intercept and the fixed effect were assigned as Cauchy distributions using parameters recommended for weakly informative priors in logistic/probit regression (Gelman et al., 2008, 2013). Prior distributions for the random effects were assigned as normal distributions with weakly informative inverse-gamma distributions as hyper-priors for the variance parameters.

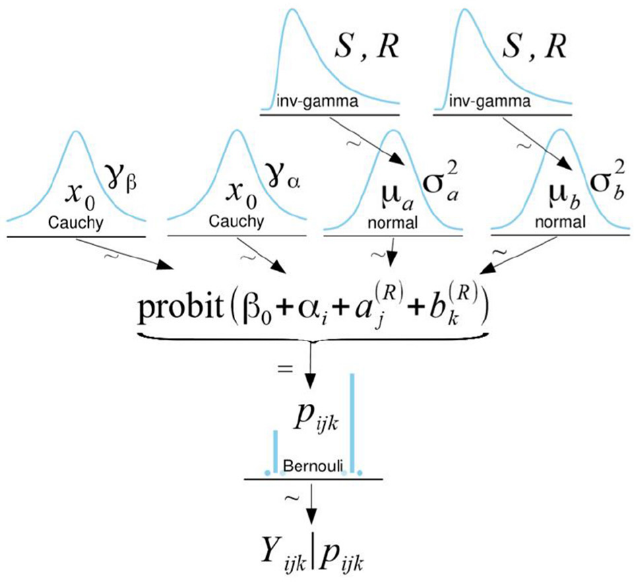
MCMC Sampling

Bayesian analyses on the above models were conducted using the BinBayes.R function (Song et al., 2017), which uses the coda (Plummer et al., 2006) and rjags (Plummer, 2018) packages in R (R. Development Core Team, 2018) to run Markov chain Monte Carlo (MCMC) sampling *via* JAGS (Plummer, 2003) to compute the posterior distribution of the model parameters. After MCMC sampling, model convergence was first verified through visual checks of trace plots and then *via* the Gelman-Rubin convergence diagnostic (Gelman and Rubin, 1992) for each parameter using a 95% confidence interval. For all parameters, $\hat{R} < 1.01$ indicating MCMC chain convergence. To ensure the MCMC sample was sufficiently large, the effective sample size (ESS) was calculated (Kass et al., 1998). To ensure stable estimates of the Highest Density Interval, it is recommended that MCMC sampling should be run, for parameters of interest, until the ESS for the posterior distribution exceeds 10,000 (Kruschke, 2014). The primary parameter of interest was the fixed effect of the map interface (α_2 in the model) on both wayfinding accuracy and route efficiency. To ensure that the ESS for this parameter exceeded 10,000, the BinBayes.R function was modified in the following ways. The number of MCMC burn-in iterations was set to 5,000 and then MCMC sampling was conducted using three chains for 30,000 iterations, with a thinning interval of 5, leaving 6,000 samples per chain (total samples = 18,000). ESS exceeded the recommended value for the fixed effect both on wayfinding performance (ESS = 11,777.6) and route efficiency (ESS = 11,964.4), indicating the MCMC sample should be large enough to produce stable estimates of the HDI. The extent to which the prior informed the posterior distribution was assessed *via* the prior posterior overlap (PPO) calculated using the MCMCvis package (Youngflesh, 2018). The overlap for the fixed effect (α_2) and intercept (β_0) was 32.6% and 17.5% for wayfinding accuracy and 34.6% and 17.2% for route efficiency, indicating the priors did not excessively influence the posterior distribution.

Evaluation of the Null Hypothesis *via* HDI and ROPE

Using mean values from the posterior distribution, we first calculated the marginal effect at the mean, to predict future wayfinding accuracy depending on the interface. This revealed, for an average participant, navigating to an average target, the predicted probability of successful wayfinding is nearly identical for both the HIM (82%) and the DIM (83%) conditions. This is consistent with the observed data (see **Table 2**) which also shows highly similar wayfinding accuracy after learning *via* the HIM (78%) and DIM (75%).

To test for equivalence, the effect of the interface (HIM vs. DIM) was assessed *via* Bayesian parameter estimation using the Highest Density Interval (HDI) plus a Region of Practical Equivalence (ROPE) decision rule (Kruschke, 2011; Kruschke and Liddell, 2018; Kruschke and Meredith, 2018). In the present research, we used a 95% HDI, which describes the range in which a measure should fall 95% of the time. The purpose of the present research is to evaluate the feasibility of the DIM-based VAM to support wayfinding ability among BVI users. Thus, we were primarily interested in testing for the noninferiority of the DIM (when compared to the HIM-based HTM). For noninferiority testing, only one side of the ROPE is emphasized (Kruschke, 2018). These accuracy data were only collected in 25% intervals (wayfinding accuracy was assessed four times for each interface); thus, the lower boundary of the ROPE was set to reflect −25% probability of accurate navigation when using the DIM compared to the HIM. As the mean predicted wayfinding accuracy for the HIM was 82%, the lower boundary of the ROPE (−0.741) corresponded to 57% accuracy. If the most credible values, from the posterior distribution of the effect of interface, predict greater than 57% accuracy (i.e., the entire HDI is greater than −0.741) when using the DIM, we could then conclude that the DIM is not inferior to the HIM (i.e., wayfinding performance is at least as good after using the DIM as after using the HIM). 97.8% of the HDI (see **Figure 6**) for the effect of the user interface (−0.747 to 0.78) falls above the decision criteria. These values correspond to a predicted wayfinding accuracy between 57–96% when using the DIM; however, 2.2% of the most credible values for the parameter fell below the decision criteria. Thus, we are unable to definitively confirm that the DIM is not inferior to the HIM. However,



$$Y_{ijk} | p_{ijk} \sim \text{Bernoulli}(p_{ijk})$$

$$g(p_{ijk}) = \beta_0 + \alpha_i + a_j^{(R)} + b_k^{(R)}$$

$$\beta_0 \sim \text{Cauchy}(0, 10)$$

$$\alpha_1 = 0$$

$$\alpha_2 \sim \text{Cauchy}(0, 2.5)$$

$$a_j^{(R)} \sim N(0, \sigma_a^2)$$

$$b_k^{(R)} \sim N(0, \sigma_b^2)$$

$$\sigma_a^2 = \text{inv} - \Gamma(0.01, 0.01)$$

$$\sigma_b^2 = \text{inv} - \Gamma(0.01, 0.01)$$

FIGURE 5 | On the left is a diagram illustrating the model, priors, and hyperpriors used in the analysis. Specific model parameters are listed on the right. α_1 is set to zero as a constraint for model identification such that α_1 reflects the baseline condition (HIM). Therefore, $\alpha_2$represents the effect of the DIM relative to the HIM (Song et al., 2017). In the model, the subscripts i, j, and k refer to the interface, target, and participant, respectively.

when considered alongside the HDI for the HIM (0.053–1.89), which corresponds to a predicted wayfinding accuracy between 52–97%, and given the small sample size here, these data suggest both the DIM and HIM are likely to support equivalent wayfinding performance. Another point to consider is, while the mean of posterior density for the effect of the interface (α_2 = 0.025) is close to zero, the range of the HDI is nearly ±0.8. This is similar in magnitude to the mean of the posterior density for the intercept (β_0 = 0.922). This point is emphasized because, although these data suggest a similarity between the DIM and HIM, there is still considerable variability in these estimates.

The ROPE used to evaluate the effect of map interface on Route Efficiency (see **Figure 7**) was also set such that the lower bound (−0.716) reflected a reduced performance by 25% when using the DIM compared to the HIM. Only 73% of the HDI (−1.29 to 0.315) for the effect of interface on route efficiency was greater than the lower limit (−0.716) of the ROPE. Thus, with 27% of the HDI below the lower limit of the ROPE, these data are inconclusive regarding the effect of interface on route efficiency. Using mean values from the posterior distribution, we also calculated the marginal effect at the mean, to predict route efficiency depending on the interface. For an average participant, navigating to an average target, the predicted probability of navigating along the most efficient route is higher for the HIM (80%) than the DIM (65%). This is consistent with the observed data (see **Table 2**). Even though these results were inconclusive, overall route efficiency (observed data) after learning in both HIM (79%) and DIM (67%) conditions suggests participants were still using accurate cognitive maps during wayfinding. It is important to again note the variability in the parameter estimates. For route efficiency, the upper and lower values of the HDI for the effect of the interface (α_2) deviate from the mean by approximately ±0.8. This is again similar in magnitude to the mean of the posterior density for the intercept (β_0 = 0.843).

Learning Time

Learning time was defined as the time (in seconds) participants spent using each interface (HIM or DIM) to learn the map. The effect of interface on learning time (see **Table 2**) was also assessed *via* Bayesian parameter estimation using the HDI plus a ROPE decision rule. The posterior distribution was generated *via* MCMC sampling using the default options (Kruschke, 2013) in the BEST package (Kruschke and Meredith, 2018). MCMC chain convergence was indicated by an $R < 1.01$ for all parameters. To ensure stable estimates of the parameter of interest (mean), the ESS was confirmed to be greater than 10,000 (ESS = 45,918. **Figure 8** shows the posterior distribution for the effect of interface on learning time. The positive value for the mean estimate indicates 166 s greater learning time when using the DIM compared to using the HIM. The ROPE was set such that a difference in learning time reflecting ±60 s would be considered equivalent performance. With 8% of the HDI in the ROPE, we are unable to determine whether the map interface has a reliable effect on learning time or not. However, the posterior probability that the difference in learning time is greater than zero (i.e., that it takes longer to learn using the DIM than using the HIM) was 97.4%. Thus, it is likely that the DIM requires increased learning time as compared to the HIM, an outcome that is consistent with findings

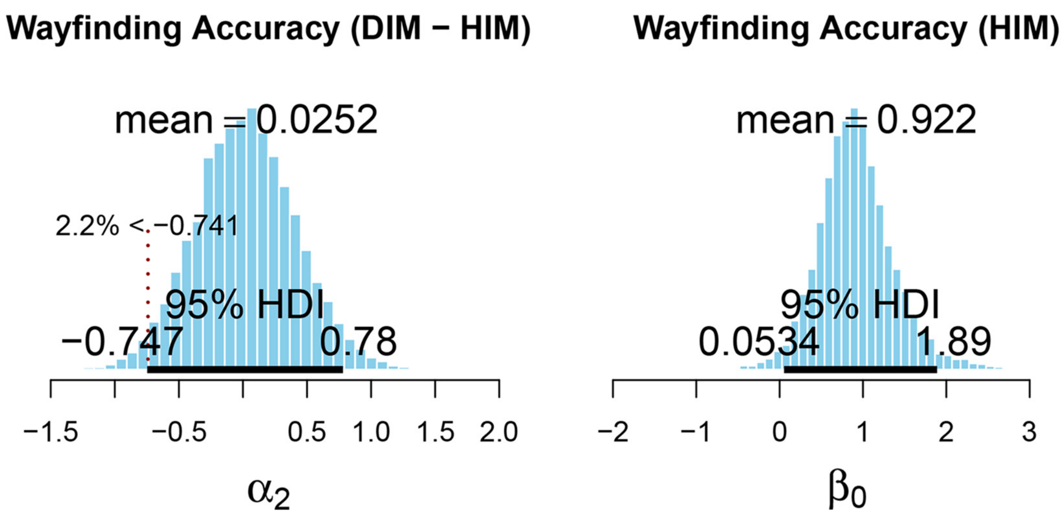


FIGURE 6 | Posterior distribution for the fixed effect (left) and intercept (right) of the model for wayfinding accuracy. The lower limit of the Region of Practical Equivalence (ROPE) is indicated by the dashed vertical line and was set to correspond to −25% probability (based on the mean of the posterior for β_0) of successful wayfinding when using the DIM compared to the HIM. This figure was created using the code provided in *DBDA2E-utilities.R* (Kruschke, 2014).

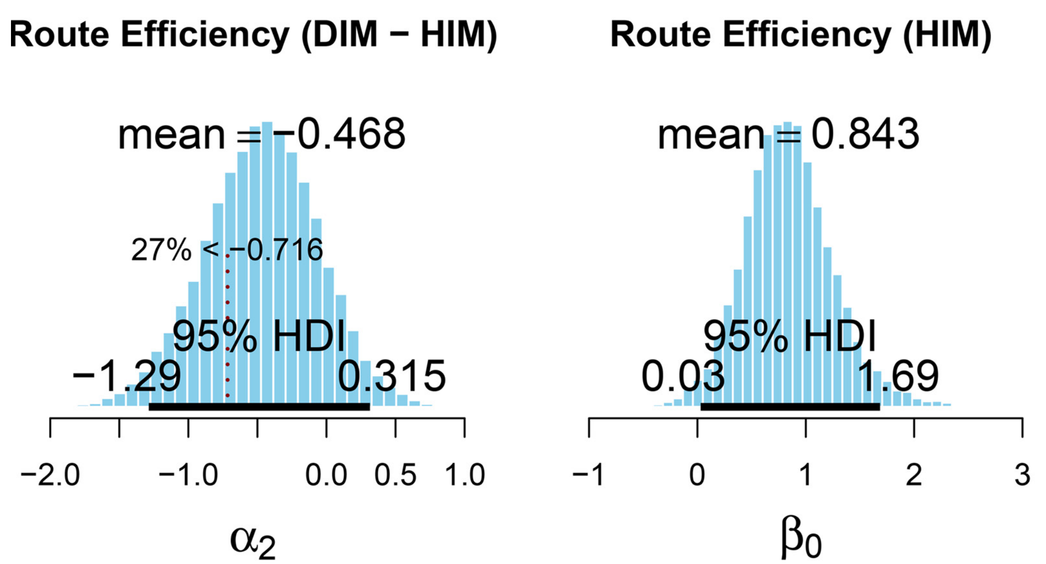


FIGURE 7 | Posterior distribution for the fixed effect (left) and intercept (right) of the test model for route efficiency. The lower region of the ROPE is indicated by the dashed vertical line and corresponds to a value of −25% route efficiency (based on the mean of the posterior for β_0) when using the DIM compared to the HIM. This figure was created using the code provided in *DBDA2E-utilities.R* (Kruschke, 2014).

comparing similar interface conditions for learning graphs and shapes (Giudice et al., 2012).

DISCUSSION

The goal of this experiment was to evaluate map learning and cognitive map development using a wayfinding task with BVI participants. Comparisons were made between learning with hybrid interactive maps (HIMs) consisting of traditional HTM overlays that were mounted on a touchscreen and augmented with audio information and a new class of digital-only interactive maps (DIMs) that conveyed functionally-matched information using vibratory and auditory cues *via* the same touchscreen interface. The literature has unequivocally shown that access to tactile maps greatly benefits the accuracy of environmental learning, cognitive map development, and wayfinding performance by both children and adult BVI users (Golledge, 1991; Espinosa et al., 1998;

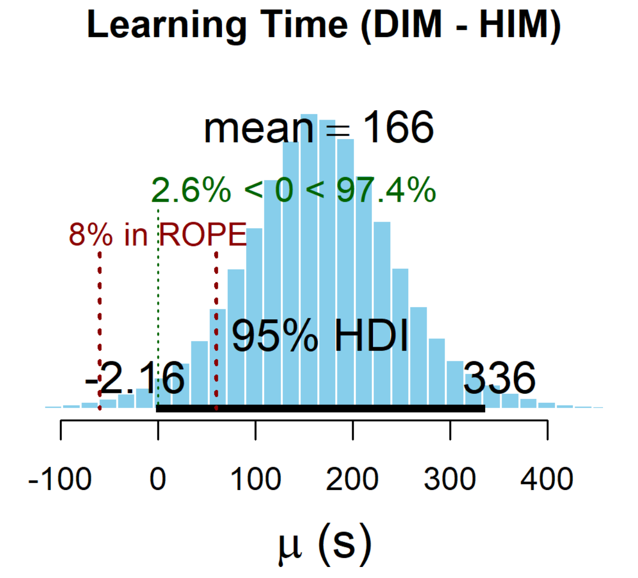


FIGURE 8 | Posterior distribution for the effect of interface on learning. The ROPE is indicated in red and was set to correspond to a ±60 s of learning time. This figure was created using the BEST package (Kruschke and Meredith, 2018).

Blades et al., 1999). These advantages have also been demonstrated with accessible interactive maps, using a host of technologies (Ducasse et al., 2018). As discussed previously, there are many benefits of digital maps, e.g., they are interactive, dynamic, and multimodal as compared to traditional static, tactile-only maps but there are also challenges. For instance, most digital maps require the use of technology that is expensive, highly specialized, and non-portable. In addition, the HIMs approach still requires the use of tactile map overlays, which are costly to produce and necessitate careful registration with the underlying digital map.

The DIM we evaluated here adopted a new approach to multimodal interactive mapping, called a VAM, which is based on commercial touchscreen-based smart devices. Filling a gap in the literature, this study set forth to directly compare environmental learning with the VAM-based DIM with traditional HIMs using a within-subjects design. Rather than focusing on perceptual factors related to map reading or route following, as has been the emphasis of previous research, we adopted an experimental paradigm that evaluated map learning by measuring cognitive map development on a series of *in situ* wayfinding tasks. Our interest here was in assessing similarity between the two map learning conditions (e.g., supporting the null hypothesis), rather than the traditional hypothesis testing approach of detecting differences (e.g., rejecting the null). As such, we used Bayesian analyses aimed specifically at determining whether or not performance on a battery of wayfinding tasks between conditions was functionally equivalent. Although definitive equivalence was not observed using the strict parameters of our analyses, the consistent wayfinding data found between the two map learning conditions are indicative of a highly similar performance. This is an important outcome as it demonstrates that traditional (raised line) tactile stimuli are neither necessary for developing accurate cognitive maps nor required for supporting efficient wayfinding behaviors. While the observed data does suggest a difference in the time needed to learn the maps, with the HTM-based HIM being faster than the VAM-based DIM, these findings are not all that surprising given the nature of tactual encoding from the hardcopy stimuli, i.e., faster and more direct perception of edges and intersections compared to extraction of the same attributes using vibrotactile cues (Klatzky et al., 2014). Indeed, these results are in-line with previous literature comparing learning time between vibrotactile and traditional tactile modes across a range of spatial patterns (Giudice et al., 2012). The role of expertise is also an unknown factor that could impact learning time. While most BVI individuals have interacted with some form of traditional embossed tactile renderings during orientation and mobility training, none of our participants had previously used our VAM interface. Additional studies are needed to assess whether increased experience with the VAM leads to corresponding improvements in learning speed. The results from the navigation test are far more relevant to our interest in cognitive map development, with the current findings suggesting that once the maps are learned, the ensuing cognitive maps are similarly robust in supporting a high level of wayfinding behavior, irrespective of map learning condition.

Our study of haptic map learning between the encoding of traditional embossed tactile stimuli and vibrotactile stimuli is more than a comparison of two map presentation modes. The VAM-based DIM and HTM-based HIM also utilize different types of sensory receptors and physiological channels of haptic information processing. That is, converging results from psychophysical studies and direct physiological recordings from the glabrous skin of the human hand have identified four types of mechanoreceptors that have different spatial and temporal response properties (Bolanowski et al., 1988). While tactual perception likely involves multiple types of these mechanoreceptors and overlap of the neural substrates/channels mediating this information depending on what combinations of spatial, temporal, and thermal parameters are present, the embossed tactile maps and vibrotactile maps at the heart of our comparison utilize different fundamental receptor types. For instance, the HIM, relying on traditional mechanical stimulation from skin deformations and displacements during movement would have prioritized activation of the slowly adapting type I (SA I) and slowly adapting type II (SA II) receptors, which are most sensitive to this type of stimulation (Loomis and Lederman, 1986). By contrast, the DIM, which was based primarily on vibrotactile stimulation, would have involved the "P channel," with Pacinian corpuscles as the primary receptor inputs. The Pacinian are sensitive to changes in thermal properties and stimulus duration but are most associated with vibration and vibrotactile stimulation between the broad range of 40 and 800 Hz (Bolanowski et al., 1988). Given that the Pacinians are most sensitive at around 250 Hz (Loomis and Lederman, 1986) and that the majority of vibration motors and actuators used in commercial smart devices operate around 200 Hz

(Choi and Kuchenbecker, 2013), it is likely that our VAM-based DIM primarily activated these receptors. While more psychophysical studies are needed to formally compare the similarities and differences of haptic perception between traditional embossed tactile stimuli vs. the vibrotactile perception elicited from movement of a flat touchscreen display *via* a vibrating motor/actuator (as was the case for our VAM-based DIM), the current results provide a compelling story for how these different haptic presentation modes support real-world spatial learning. Indeed, we interpret the similarity of test performance after learning with both DIM and HIM maps observed here as supporting the notion that different encoding sources, as long as they convey functionally relevant information, can lead to a common spatial representation in the brain that functions equivalently in the service of spatial behaviors (Loomis et al., 2013). Although additional research is needed to further probe the structure of the underlying neural representation between these approaches, the current results clearly support the efficacy of the VAM as a new type of DIM interface that is comparable to traditional HIM solutions. In addition, the similarity we observed between our low-cost DIM and the traditional HIM suggests that the trade-off of additional expense and increased design complexity in producing HIMs is not justified by a corresponding offset in improved behavioral performance.

The high level of wayfinding performance observed after learning with both map conditions also provides evidence demonstrating that the use of one-finger for encoding environmental information during map exploration is sufficient for supporting accurate spatial learning and cognitive map development. These findings speak to a longstanding debate in the literature about the relevance of the use of one or more fingers in tactile perception. Clarification of this issue is particularly relevant to touchscreen-based haptic DIMs, which are generally explored by moving only one point of contact on the display, usually the dominant index finger. This exploration strategy contrasts with traditional tactile map reading, which is often done with unrestricted movement of both hands over the map. Studies using vibrotactile patterns (Craig, 1985) or embossed tactile objects (Klatzky et al., 1993) have argued for the benefit of using multiple fingers/hands. However, other research investigating the exploration of raised line drawings found no consistent benefit of using multiple fingers (Loomis et al., 1991). In two systematic studies investigating reading of simple tactile maps with multi-finger/hand use by both blindfolded-sighted and BVI participants, Morash and colleagues found that while performance tended to improve with multiple fingers, it was not a simple "more is better" scenario. For instance, while Line-tracing tasks were found to be fastest when using two hands, performance benefits were not found by using more than one finger per hand. By contrast, tasks requiring a search of both local and global structures were faster with multiple fingers, but not with both hands. Finally, BVI users tended to perform better than their sighted peers when using both hands (Morash et al., 2013, 2014). In studies employing more complex map-reading tasks, two hands were found to be beneficial but only one was employed for map exploration, while the other hand (or finger) was used as a fixed "anchor" on the map or its edge (Perkins and Gardiner, 2003). This 2-hand anchoring strategy has also been found to be useful with touchscreen-based haptic DIMs, similar to the VAM studied here, in supporting map panning operations (Palani and Giudice, 2017). Although the one vs. multiple finger issue has not been extensively studied with interactive maps, research with 14 BVI participants on learning indoor layouts *via* a touchscreen-based tablet interface using haptic cues delivered to one finger from the devices embedded vibration motor vs. stereo haptic cues delivered by vibrating rings worn on two fingers, showed that one-finger exploration was usually more accurate and actually preferred (Adams et al., 2015). The combination of these findings, in conjunction with the current results, provide compelling evidence for the efficacy of one-finger search strategies with touchscreen-based DIMS for supporting accurate information extraction, map learning, and cognitive mapping enabling efficient wayfinding behavior.

The current findings also speak to the issue of under-developed spatial abilities of BVI navigators that are often described in the literature, e.g., deficits in building up and accessing accurate cognitive maps (for review, see Thinus-Blanc and Gaunet, 1997). The consistently high wayfinding performance observed here would not have been possible without map learning leading to accurate cognitive map development. These results argue against the standard explanation of lack of vision or visual experience as being the root cause of spatial deficits by BVI people (see Schinazi et al., 2016 for discussion). We interpret our findings as supporting the information-access hypothesis of blind spatial cognition. According to this perspective, the spatial differences (if manifest) found in studies with BVI individuals compared to their sighted peers are less about the role of visual experience or a necessary outcome of vision loss but occur as a result of insufficient access to environmental information from nonvisual sensing and under-developed teaching of key spatial skills underlying complex spatial behaviors (Giudice, 2018). Maps are an excellent tool for conveying normally inaccessible environmental information; as such, they represent an important solution for leveling the "spatial" playing field for BVI wayfinders.

Finally, while visual maps are often used during *in situ* navigation, the physical limitations of traditional tactile maps (i.e., their size and cumbersome nature) and the lack of portability of most interactive mapping technologies greatly constrain analogous real-time map usage. While the small form factor of VAM-based DIM interfaces makes them particularly amenable to *in situ* use scenarios, the success of this interface in the transfer task evaluated here (i.e., map learning followed by subsequent wayfinding in the physical space) suggests that they would also be excellent pre-journey learning tools. This procedure involves exploring maps in an offline learning mode, where the map is used to learn routes, configurational information, and convey spatial relations before going to space. This strategy has been extremely effective for teaching spatial concepts to BVI navigators in a safe and low-stress scenario (Holmes et al., 1996; Zeng and Weber, 2010; Ivanchev et al., 2014). As the haptic DIMs tested here can be used for both pre-journey learning and during real-time wayfinding, they represent an important advancement in accessible mapping technology. Note that although our current findings are limited to indoor building layouts, we are confident in the VAM's efficacy in also supporting outdoor travel, where there are far more complementary tools, technologies, and environmental cues. This prediction will be tested in a future environmental transfer study with wayfinding at test occurring in an outdoor environment.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of Maine Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

NG designed the experimental tasks and contributed to the implementation of the research and to the writing of the manuscript. BG performed the data analysis and interpretation and also wrote the results sections. KH assisted with conducting the study and initial data organization and analysis. NJ assisted with piloting the research and conducting the experimental trials.

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The Effectiveness of Home-Based Training Software Designed to Influence Strategic Navigation Preferences in Healthy Subjects

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One approach to the rehabilitation of navigation impairments is to train the use of compensatory egocentric or allocentric navigation strategies. Yet, it is unknown whether and to what degree training programs can influence strategic navigation preferences. In validating this approach, the key assumption that strategic preference can be changed by using a navigation training was assessed in a group of healthy participants (n = 82). The training program consisted of a psychoeducation session and a software package that included either allocentric or egocentric navigation exercises in virtual environments. Strategic navigation preference, objective and self-reported spatial abilities were assessed in pre- and post-training sessions. Based on their pre-training strategic preference, participants received either the egocentric training (n = 19) or the allocentric training (n = 21) version of the training. These participants engaged in four training sessions over a period of 2–3 weeks. The second group of participants did not use the training software (n = 43) and served as a control group. The results show that 50% of participants that received the egocentric training shifted from an allocentric to and an egocentric strategic preference. The proportion of participants that switched their strategic preference as a result of the allocentric training was identical to this proportion in the control group (19%). The training did not affect objective and self-reported navigation abilities as measured in the pre- and post-training sessions. We conclude that strategic navigation preferences can be influenced by using home-based training in healthy participants. However, using the current approach, only a preference shift from an allocentric to an egocentric navigation strategy could be achieved. The effectiveness of this navigation strategy training should next be assessed in relevant patient populations.

Keywords: spatial navigation, navigation strategy, cognitive training, strategy training, serious game

INTRODUCTION

Spatial navigation is a complex cognitive ability that is essential to our daily functioning. On a daily basis, humans traverse a range of environments (e.g., a crowded city or an open rural environment), with different navigational goals (e.g., exploration, finding one's way home). In order to adapt to the variety of spatial challenges we are faced with regularly, evolution favored a complex and flexible navigation system in the human brain (Cashdan and Gaulin, 2016). Neuroimaging and lesion studies have identified a large neural network associated with spatial navigation, including the hippocampal formation, parahippocampal gyrus, retrosplenial cortex, medial temporal lobe, prefrontal cortex, precuneus and regions of the parietal lobe (Maguire et al., 1999; Chrastil, 2013; Boccia et al., 2014; Spiers and Barry, 2015). This widespread recruitment of the brain renders the navigation ability highly vulnerable to brain damage. Disruption of neural networks involved in navigation often results in navigation impairments (also known as topological disorientation) as observed in patients with acquired brain injury (Claessen and van der Ham, 2017), neurodegenerative diseases (Kalová et al., 2005) and developmental (Lind et al., 2013) and mental disorders (Hanlon et al., 2006). Navigation impairments are known to have a debilitating effect on the daily life activities of patients (Aguirre and D'Esposito, 1999). As such, navigation impairments have been associated with lowered quality of life, heightened levels of spatial anxiety and reduced autonomy (van der Ham et al., 2013).

Developing a standardized treatment for navigation impairments has proven to be a challenge due to the multifaceted nature of spatial navigation (Maguire et al., 1999; Wolbers and Hegarty, 2010; Claessen and van der Ham, 2017). Problems reported by navigation impaired patients are diverse and deficits are often specific. This is illustrated by a wealth of rapports of patients displaying specific spatial impairments: difficulty encoding novel landmarks (Herdman et al., 2015), recognizing famous landmarks (Rainville et al., 2005), understanding the order in which landmarks are encountered (van der Ham et al., 2010), remembering what actions to take at a landmark to follow a route (van der Ham et al., 2010), utilizing maps (Suzuki et al., 1998), forming a topological understanding of an environment (Ino et al., 2007) or switching between spatial reference frames (Ruggiero et al., 2014).

Over the past years, training programs have been developed with the goal of improving navigation ability in healthy subjects and patients. Most training programs for healthy subjects have been directed towards knowledge acquisition of specific environments. Examples of these include training for firefighters (Bliss et al., 1997), evacuation scenarios (Burigat and Chittaro, 2016) and astronauts learning to orient themselves in a space station (Aoki et al., 2008). One notable training program that has been developed for healthy participants has been reported in a study in which pre-school children were trained for 12 weeks to enhance their spatial orientation skills. After engaging in a variety of spatial exercises, children were able to encode and utilize map-like knowledge of an environment, a spatial skill that normally arises years later in development (Boccia et al., 2014). Several training programs have been reported that were specifically tailored to the impairments of a patient (Brooks, 1999; Incoccia et al., 2009; Bouwmeester et al., 2015; Claessen et al., 2016a). Some rehabilitation programs have focused on learning how to navigate a specific route through the environment (errorless learning; Lloyd et al., 2009) while other programs aimed to strengthen general spatial abilities by developing route learning (Kober et al., 2013). Generally, patients do benefit from navigation rehabilitation training. However, previous training programs have been either specifically designed for an individual patient or were directed at training navigation in a specific, spatially limited environment. Furthermore, the programs involve intensive supervision of experts as training programs required repeated sessions.

There is a need for a standardized navigation training that can be used to treat a broad range of navigation impairments. To account for the diversity in navigation impairments, the training should include exercises for navigational abilities in different spatial domains. Becoming acquainted with different navigation abilities should allow for the development of a more beneficial, compensatory navigation strategy, which can be used in real life. In order for this standardized training to be feasible in today's healthcare system, the training should include both face-to-face therapy and repeated (unsupervised) training sessions (Wentzel et al., 2016). To this end, we propose a home-based navigation rehabilitation training that can be installed on and used from a patient's home computer. Training exercises provided by the software should be modeled after experimental paradigms described in the field of spatial cognition.

When interacting with an environment, humans encode, update and process spatial information using distinct representations of space, referred to as reference frames (Klatzky, 1998). Spatial information about objects in the environment, in relation to the navigator's own body is encoded into a body-centered, egocentric reference frame. Spatial relations between objects in the environment, irrespective of the navigators own position, are encoded into a world-centered, allocentric reference frame. The type of spatial information that is encoded and used during navigating reflects the employed navigation strategy. Remembering sequences of bodily turns (Iglói et al., 2009), landmark-direction associations at intersections (Wiener et al., 2013) and path integration (Wang et al., 2006) are all spatial abilities that rely on egocentric reference frames. As such, spatial behavior that relies on these abilities can be regarded as an egocentric navigation strategy. Conversely, spatial abilities such as place finding (Parslow et al., 2004), utilizing configurational knowledge of landmarks (Iglói et al., 2009) and the use of maps during navigation (Palermo et al., 2012), makes use of a world-oriented, allocentric reference frame. Spatial behavior that focusses on external cues during navigation can be classified as an allocentric navigation strategy. It is well established that (partially) distinct neural subsystems underlie navigation based on egocentric and allocentric reference frames (Jordan et al., 2004; Zaehle et al., 2007; Boccia et al., 2014; Colombo et al., 2017). This distinction between navigation strategies and their underlying neural correlates, suggests that a compensatory rehabilitation approach might be an effective approach to rehabilitation of navigation impaired patients.

Compensatory and metacognitive strategy training programs are practice standards in the rehabilitation of cognitive functions after brain injury (Cicerone et al., 2000, 2005, 2011, 2019). Such training programs start with the construction of a strengths and weaknesses profile in which a patient's impairments and intact cognitive abilities determined. Then, training is constructed that focusses on the improvement of the intact abilities and the development of strategies that are beneficial to a patient. In terms of navigation impairment, participants with intact egocentric abilities, but difficulties in the allocentric domain, should be trained to adopt an egocentric navigation strategy and vice versa.

It is currently unknown whether navigational strategies can be influenced by training interventions. The aim of the current study was to test the key assumption that strategic navigation preference can be influenced by using home-based navigation training. By validating the concepts of the training in healthy subjects, we will provide the basis for a randomized control trial with navigation impaired acquired brain injury patients. To demonstrate a change in strategic navigation preference, we will train participants to adopt a navigation strategy other than their naive strategic preference. To this end, a home-based navigation training was developed in the form of a serious game. Two versions of the game were constructed: a version designed to train allocentric navigation strategies and a version designed to train egocentric navigation strategies. In order to provide evidence that strategic shifts were the result of the training intervention, a control group was used that did not receive the intervention. In addition, we aim to provide insight into the mechanisms by which a shift in strategic preference might occur. We will explore to what degree individual differences in objective and subjective navigation abilities determine naive strategic preference. Furthermore, we aim to examine individual characteristics that could potentially predict training success.

We hypothesized that participants who used the training program would display a preference for the navigation strategy trained in a situation where using both strategies can be deployed. As we expected the training to induce the strategic preference shifts, we expected a higher proportion of strategy shifts in the training group compared to the control group. Second, we hypothesized that using the training will lead to increased performance on spatial abilities associated with the trained domain. Specifically, egocentric spatial abilities (e.g., route continuation) will improve after the egocentric training, and allocentric spatial abilities (e.g., location on map) will improve after allocentric training. No performance changes were expected in the control group. Third, we hypothesized that subjective navigation ability will increase after using the training, whereas no change in subjective navigation ability was expected in the control group.

MATERIALS AND METHODS

Design

A pre-test–post-test design was employed in this study including a control group, consisting of a "control" and "control + psychoeducation" subgroup and an experimental group consisting out of an "allocentric training" subgroup and an "egocentric training" subgroup. Measurements took place during two sessions: pre- and post-training. These measuring phases were separated by a 2 week intervention period. During the pre-training session, participants completed the screening/general questionnaire, the strategy assessment task, the Virtual Tübingen testing battery, which measured objective navigational ability, wayfinding questionnaire, which measured self-reported navigation and four neuropsychological assessments. During the post-training session, participants again completed the strategy assessment task, the Virtual Tübingen testing battery, and the wayfinding questionnaire. Participants in the experimental condition would engage in either the allocentric or egocentric training software in the period between pre- and post-training sessions.

Participants

Participants were recruited from the university campus using posters, the university's recruitment website, and social media. The inclusion and exclusion criteria for the study were: (1) between 18 and 35 years old; (2) Dutch-speaking; (3) access to personal computer and internet; (4) willingness and capability to complete the training program; and (5) no history of neurological or psychiatric disorders. All participants were required to sign an informed consent form in order to participate and were compensated for participation in participant hour credits or with a monetary reward of 6 € per hour. The study was performed in concordance with the Declaration of Helsinki (2013) and was approved by Leiden University's local ethics committee for psychological research.

Materials

Tasks

Screening/General Questionnaires

All participants completed a screening questionnaire in which they filled in demographic characteristics such as age, gender, handedness, level of education and gaming experience. Furthermore, screening information about psychiatric or neurological disorders was obtained.

Navigation Strategy Assessment

Strategic navigation preference was assessed during the pre- and post-training sessions using an adapted version of the Starmaze (Iglói et al., 2009). Two variants of the Starmaze were used: the original environment described by Iglói et al. (2009) and a mirrored environment. The Starmaze consisted out of five alleys that formed a pentagon and five alleys that radiated from this pentagon. The alleys were surrounded by a small wall that could not be traversed. Surrounding the environment were two distinct mountains, two distinct forests, and two radio towers, which were visible throughout the maze. Participants were instructed to explore the environment to find the goal location, which was located in one of the arms. Upon finding the goal location, the text "Bravo" would be displayed on-screen and the next trial was started. Over the course of the first five trials (training trials), participants would start in the same arm of the maze and learn to find the goal location. In the 6th trial (probe trial) participants started in a different arm of the maze. Participants could navigate using either the sequence of left-right turns that was learned during the training trials or by determining their location based on the configuration of landmarks in the environment. Participants utilizing the turn sequence approach would end in an alley that was different from the goal location in the training trials. Participants that utilized the configuration of cues would end in the original ending alley.

The ending location and the travel path measured in the probe trial were used to identify egocentric, allocentric or mixed navigation strategies. Participants who ended at the different goal location, and thus utilized a sequential egocentric navigation strategy, were classified as egocentric navigators. Participants who traveled directly (using the shortest route) to the original goal location, and thus utilized the configuration of landmarks to orient themselves, were classified as allocentric navigators. Participants that initially followed the turn sequence strategy, but changed direction and headed for the original goal location, were classified as mixed navigators.

Subjective Navigation Ability

Self-reported navigation ability was assessed during the pre- and post-training sessions using the Wayfinding Questionnaire (de Rooij et al., 2017). The Wayfinding Questionnaire contains 22 items in three subscales: navigation and orientation (11 items), distance estimation (three items) and spatial anxiety (eight items). All items were rated on a seven-point Likert scale.

Objective Navigation Ability

Objective navigation ability was assessed during the pre- and post-training sessions using an adapted version of the Virtual Tübingen testing battery (Van Veen et al., 1998; Claessen et al., 2016b). Four routes through the city were selected that were comparable in terms of distance and number of intersections. Participants watched a video of a route through a virtual replication of the city of Tübingen. Participants were instructed to memorize as much as possible about the spatial characteristics of the route and the environment. Afterward, participants completed 6 tasks in which navigation abilities were assessed.

Participants completed two variations of the task at each measuring phase. In the first variation of the tasks, participants saw the route from a first-person perspective. In the second variation, participants observed a red arrow icon moving along a route from a birds-eye view, the map perspective. The camera was placed at a height of 38 m and was focused on the red arrow. The camera did not rotate with the arrow and thus, was always aligned in the same direction.

After viewing the video, a Route Sequence task was conducted. Participants had to indicate what action was taken sequentially at each intersection point along the route. Options were left-turn, right-turn or straight. No images of the related decision points were shown. Numbers 1–8 were listed and participants selected the arrow icon indicating the response options for each number. Scoring was based on the number of correct responses. A participant's score was the sum of correct responses (ranging from 1 to 8).

Then the Route Continuation task was performed. Participants were presented with eight images of the intersection points in random order. Participants had to indicate whether they turned left, right or went straight ahead at each decision point by pressing the arrow keys left, right or up arrow, respectively. Scoring was based on the number of correct responses. A participant's score was the sum of correct responses (ranging from 1 to 8).

Participants then performed the Point to Start and Point to End tasks. Participants were shown eight scenes taken along the route in random order. Participants were asked to indicate where the start/end location of the route was using a rotational device. In the first-person perspective version, the rotational device was placed horizontally on the desk in front of the participants. Participants were asked to point from the perspective shown in the image. In the dynamic map perspective version, the rotational device was placed vertically on the desk next to the monitor. Participants had to indicate the start/end location on the map, relative to the red arrow icon the camera was following. Scoring was based on the mean pointing deviation angle for each trial, ranging from 0 to 180 degrees deviation.

In the Distance Comparison task, participants were shown a target image and two response images. In the first-person perspective version, the images corresponded to locations visited along the route. In the dynamic map perspective version, the images were landmarks encountered along the route. Participants had to indicate which of the two response locations was closest to the target location (direct path distance). A participant's score was the sum of correct responses (ranging from 1 to 8).

Finally, participants performed the Locations on Map task. Participants were shown a schematic map of the city including icons indicating starting and ending locations. In the first-person perspective version, participants were shown images of eight locations along the route in random order. Participants had to indicate the correct location on the city map using the mouse. In the dynamic map perspective version, participants had to indicate where landmarks were located on the city map. Scoring was based on the amount of pixels deviation from the correct location.

Neuropsychological Assessment

Four neuropsychological tests were performed to assess general cognitive ability. The Corsi Block tapping tasks, both forward and backward, were used to asses visuospatial working memory (Kessels et al., 2000). The WAIS VI Digit span test, both forward and backward, was used to assess verbal working memory (Wechsler, 1987). A digital 46-item adaptation of the Mental Rotation test was used to assess object-based transformation ability (Shepard and Metzler, 1971; Vandenberg and Kuse, 1978). An adaptation of the 12-item Santa Barabara perspective-taking test was used to assess egocentric transformation ability (Hegarty and Waller, 2004).

Training Intervention

The training intervention consisted of a short psychoeducation session and home-based navigation training software that was used over the course of 2–3 weeks.

Psychoeducation

The psychoeducation session took 20–30 min. The experimenter placed a document with illustrations on the table and read an educational text for the participants. After reading the text aloud, the experimenter discusses the illustrations on the document to clarify the content. The educational text addressed the following topics: the formation of egocentric and allocentric reference frames and the use of egocentric and allocentric navigation strategies. It was explained that people are capable of using both strategies and that certain strategies are more effective in specific situations. To verify whether participants understood the concepts, participants were asked to give examples of both egocentric and allocentric navigation strategies they have used. Participants were told that they would engage in a training program designed to train egocentric or allocentric navigation strategies. Importantly, participants were not informed about their performance or strategy preference in the Starmaze and Virtual Tübingen tasks.

Home-Based Training Software

Two versions of the training were constructed. Participants would receive either the egocentric navigation training or the allocentric navigation training. Each training consisted of 3 modules that were designed to train spatial abilities that are central to either an egocentric or allocentric navigation strategy. The egocentric training was composed of the modules: "landmark-action association," "turn-sequence" and "egocentric updating." The allocentric training was composed of the modules: "place-finding: distal landmarks," "place-finding: local landmarks" and "effective map-use." Each module resembled a simple game, set in the theme of ancient Greece. A comprehensive description of the training modules can be found in the Supplementary Material (**Supplementary Figures S1–S10**).

The navigation training software was installed on the participants' home computer. Participants received a personal account, which allowed for data transfer with an online server. *Via* the server, progress during the training could be stored and tracked. Furthermore, training adherence was recorded by storing training time and the number of trials started and completed. Participants were instructed to engage in at least four separate training sessions, in which all three training modules should be used. Mails reminding the participant to train were automatically sent two times per week.

During a single training session, participants were instructed to perform at least one attempt to increase their level in all three training modules that were available to them. Each training module contained four difficulty blocks. Each difficulty block was composed of three levels of increasing difficulty levels. All participants started on difficulty block 1. When engaging in a training session, participants completed three levels within a difficulty block. If participants scored 75% or more of the points obtainable over the levels, participants would advance to a higher difficulty block. If participants failed to obtain 75% of the points, participants would remain on the same difficulty block. Depending on the participant's skill level and progress, a training session was estimated to take 10–15 min.

Procedure

All participants were invited to the laboratory at the Faculty of Social Science at the Leiden University, where participants read the information letter and signed the informed consent form in accordance with the Declaration of Helsinki (2013). Participants filled in the screening/general questionnaire followed by the Wayfinding Questionnaire and completed the Starmaze task.

Participant was assigned to the control or training condition based on participation order. The first half of the participants were assigned to the control groups. The second half of the participants were allocated to the training condition. Participants allocated to the training condition were assigned to the egocentric or allocentric training depending on the navigation strategy displayed in the Starmaze. Participants ending in the allocentric ending location, thus displaying a mixed or allocentric navigation strategy, received the egocentric training program. Participants ending in the egocentric ending location received the allocentric training program.

Following the Starmaze task, participants would complete the Virtual Tübingen testing battery. Route and order of the perspective (first-person or map perspective) were counterbalanced between conditions. A 10-min break was introduced following the Virtual Tübingen test. After the break, the four neuropsychological tests were completed.

For participants in the control condition, the first session ended here. Participants in the experimental condition would continue to receive psycho-education and were instructed on how to use the home-training software. During the training period, participants in the experimental condition would practice with the navigation training software during four occasions. During a training session, participants were instructed to perform all three training modules at least once. A periodically repeating mail was sent to the participants, reminding them to use the training application.

After 2 weeks, participants were invited back to the lab to perform the post-intervention measurement. The Starmaze, Virtual Tübingen and Wayfinding Questionnaire were conducted. The session ended with a debriefing.

Analysis

Demographics, Neuropsychological and Visuospatial Measures

MANOVA analysis was performed to assess potential differences between participants in the conditions. Demographic, neuropsychological and visuospatial scores were compared between conditions.

Navigation Strategy

A Fishers' exact test was used to compare the proportions of participants who changed strategy between the pre- and post-training sessions. To assess the effect of psychoeducation, the proportion of strategy shifts in the control conditions was analyzed. Then, proportional analysis was performed on the control condition and the egocentric and allocentric training conditions. In order to assess whether factors other than condition determined strategy change, the proportional analysis was performed for gender, gaming experience and education between strategy shifters and those who did not shift. Binary logistic regression was performed to investigate the relationship between training adherence and strategic shift.

Objective Navigation Ability

The effect of condition on performance in the Virtual Tübingen tasks was analyzed using a differences score analysis. A difference score was calculated for each navigation task by subtracting the pre-training score from the post-training score. A MANOVA was used to assess the effect of condition (control, egocentric training or allocentric training) on performance change. Three participants had an extreme score ($Z > 3$) on the map perspective point to start task and were removed from the analysis.

Subjective Navigation Ability

The effect of condition on self-reported navigational ability, measured using the Wayfinding questionnaire, was analyzed using a differences score analysis. A difference scores for each of the subscales (Spatial Anxiety, Navigation and Orientation and Distance estimation) was calculated by subtracting the pre-training score from the post-training score. A MANOVA was used to assess the effect of condition (control, egocentric training or allocentric training) on wayfinding questionnaire change scores.

Interaction Between Strategic Preference, Preference Shift, and Navigation Abilities

To explore the interaction between strategic navigation preference and navigation abilities, a MANOVA was conducted with strategic preference at T1 as between-subject factor (egocentric, allocentric or mixed strategy) and performance on egocentric (composite score of route sequence, route continuation and point to start) and allocentric (composite score of point to end, distance estimation and location on map) tasks as dependent variables. Separate composite scores were calculated for the egocentric and allocentric tasks for the first-person and map-perspective tasks. A similar analysis was conducted with the self-reported navigational scores (spatial anxiety, navigation and orientation, and distance estimation) as dependent variables.

MANOVAs were conducted to assess differences in objective and self-reported navigation abilities between participants that shifted their strategic preferences between T1 and T2 and participant that did not shift strategic preference.

A binary logistical regression was conducted to assess whether performance on objective egocentric (composite score of route sequence, route continuation and point to start) and allocentric (composite score of point to the end, distance estimation and location on map) predicted strategic preference shifts. A similar analysis was performed with self-reported navigational abilities (spatial anxiety, navigation and orientation and distance estimation) as predictors.

RESULTS

Participants and Demographics

One-hundred and twenty-nine participants were recruited into the screening procedure. To maintain a gender balance in the egocentric training condition, the sessions of 29 females and one male were terminated during screening as they displayed an egocentric navigation strategy in the Starmaze, while this condition was already filled. Revealing a clear gender effect for strategy preference (22.97% females vs. 42.85% males displayed an allocentric navigation strategy during the first Starmaze task). Seven participants were screened on the basis of exclusion criteria as they reported psychological or neurological disorders, two participants did not perform the training at home (or trained for less than 5 min), three participants were lost to attrition, two participants were wrongly classified into the allocentric training condition. As a result, 82 participants successfully completed the experiment.

Participant characteristics for each condition are presented in **Table 1**. A MANOVA revealed that were no differences in scores on visuospatial and neuropsychological assessments between conditions, $F_{(12,148)} = 0.40$, $p > 0.05$; Wilk's $\Lambda = 0.94$, partial $\eta^2 = 0.03$, nor were there differences between age, education and gaming experience between conditions, $F_{(6,154)} = 0.77$, $p > 0.05$; Wilk's $\Lambda = 0.94$, partial $\eta^2 = 0.03$. Independent t-tests did reveal that training time significantly differed between the egocentric and allocentric strategy training groups, $t_{(37)} = 4.05$, $p < 0.01$, and the number of trials completed in the allocentric strategy training group was significantly higher than in the egocentric strategy training group, $t_{(37)} = -7.21$, $p < 0.01$.

Strategy Change

A Fisher's Exact test revealed a significant effect of condition on the proportion of strategic preference changers ($p < 0.05$; *FET*, **Figure 1**). *Post hoc* analysis, using Bonferroni corrected Chi-squared tests, revealed that a higher proportion of participants changed strategy in the egocentric training condition compared to the control condition[1] (50% vs. 19%), $\chi^2_{(1)} = 5.95$, $p = 0.015$. *Post hoc* analysis did not reveal a significant difference between the proportion of participants that changed strategic preference after the "egocentric training condition" compared to the proportion of participants that changed strategic preference after the "allocentric training condition" (50% vs. 19%), $\chi^2_{(1)} = 4.18$, $p = 0.041$ (not passing the Bonferonni correction). No significant differences were found between the allocentric training condition and the control condition in the proportion of participants that changed strategic preference (19% vs. 19%), $\chi^2_{(1)} = 0.0$ $p = 1$. Overall, this analysis revealed that strategic preference shifts between pre- and post-training were present in all groups. However, the proportion of the participants who shifted strategic preference after receiving the egocentric training was significantly larger compared to the control group.

[1]Fisher's exact test did not reveal a significant difference in the proportion of strategy changers between the "control" and (14.3%) "control + psycho-education" conditions (23.8%; $p = 0.69$; FET). In the remainder of the analysis, the control groups were combined to enhance the power of the analyses.

TABLE 1 | Overview of demographics data, neuropsychological scores and training adherence.

	Control (*n* = 43)	Experimental (*n* = 39)	
		Egocentric training (*n* = 18)	Allocentric training (*n* = 21)
Demographics			
Age in years, *M (SD)*	22.42 (2.85)	22.44 (3.11)	21.48 (2.14)
Gender, *% female*	62.79	55.56	57.14
Education, *M (SD)*†	6.77 (0.43)	6.80 (0.43)	6.76 (0.44)
Gaming experience, *M (SD)*‡	1.51 (0.94)	1.72 (1.18)	1.67 (1.02)
Neuropsychological test scores at T1			
Corsi block tapping task forward span, *M* (*SD*)	6.51 (0.94)	6.33 (1.03)	6.43 (0.87)
Corsi block tapping task forward product score, *M* (*SD*)	66.3 (20.16)	62.11 (20.91)	63.76 (16.68)
Corsi block tapping task backward span, *M* (*SD*)	6.74 (0.82)	6.61 (0.92)	6.52 (0.81)
Corsi block tapping task backward product score, *M* (*SD*)	71.14 (17.98)	69.11 (18.76)	66.86 (16.93)
Digit span forward span, *M* (*SD*)	6.14 (1.21)	6.78 (1.39)	6.38 (1.43)
Digit span forward product score, *M* (*SD*)	60.67 (24.19)	69.56 (25.67)	64.52 (30.44)
Digit span backward span, *M* (*SD*)	5.35 (1.15)	5.83 (1.15)	5.38 (1.12)
Digit span backward product score, *M* (*SD*)	52.98 (22.13)	57.89 (21.96)	53.04 (22.34)
Santa Barbara perspective taking test, deviation, *M* (*SD*)	14.99 (9.15)	15.04 (9.05)	16.88 (9.48)
Mental rotation slope, accuracy, *M (SD)*	76.98 (12.09)	76.67 (12.97)	75.29 (11.93)
Mental rotation slope, reaction time, *M (SD)*	4, 992.63 (2, 822.06)	5,520.32 (2,047.83)	5,047.36 (2,765.06)
Mental rotation slope, ms/degree, *M (SD)*	19.11 (11.69)	24.06 (19.65)	20.2 (15.11)
Training adherence			
Training time in minutes, *M* (*SD*)	-	62.31 (31.95)*	30.70 (15.00)*
Training Trials completes, *M (SD)*	-	27.94 (8.29)*	78.90 (28.94)*

†*Level of Education measured on the Verhage scale, a Dutch scale of education level ranging from 1 (low) to 7 (high; Verhage, 1964).* ‡*Gaming experience was measured on a five point scale, represented indicating 1 = 0–2 h/week, 2 = 2–4 h/week, 3 = 4–8 h/week, 4 = 8–12 h/week, 5 = 12+ h/week.* **T-tests indicate significant differences between groups (p < 0.05).*

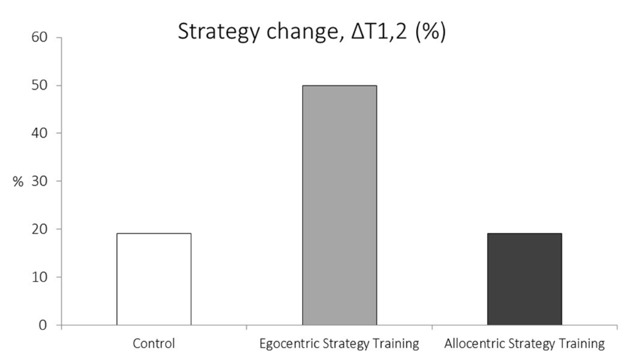

FIGURE 1 | Proportion of participants that changed navigation strategy between the pre- and post-training sessions.

TABLE 2 | Direction of change in participant that changed navigation strategies between the pre- and post-training sessions.

Strategy T1	Strategy T2	Control (*n* = 42)	Egocentric training (*n* = 18)	Allocentric training (*n* = 21)
Egocentric	Allocentric	0	-	2
Egocentric	Mixed	1	-	2
Allocentric	Egocentric	2	5	-
Allocentric	Mixed	1	1	-
Mixed	Egocentric	3	2	-
Mixed	Allocentric	1	1	-

The values in the table indicate the number of participants that changed strategic preference from T1 to T2. Participants that received the allocentric training, always displayed an egocentric strategy at T1 and could only shift towards a mixed or allocentric strategy. Vice versa, participants that received the egocentric training, always displayed a mixed or allocentric strategy at T1 and could shift towards any other strategy, not displayed at T1.

Additional proportional analyses were performed to determine whether strategic preference change could be attributed to other factors that are known to influence navigation strategy or learning processes. No effect of gender, $\chi^2_{(1)} = 0.65$, $p > 0.05$, education, $p > 0.05$; *FET*, or gaming experience, $p > 0.05$; *FET*, was found. Training time and number of trials completed differed significantly between the egocentric and allocentric training groups (**Table 1**). Exploratory binary logistical regression analyses were conducted to explore whether strategy change could be attributed to these differences. Binary logistic regression revealed that there was no effect of training time on strategy change, $\chi^2_{(1)} = 1.07$, $p = 0.74$. However, a significant relationship between the number of trials completed and strategy change was found, $\chi^2_{(1)} = 4.8$, $p < 0.028$), with fewer trials completed leading to higher training success.

Inspection of the strategic preference changes shows that the direction of the change in the control condition was not uniform. Participants in the control group changed from egocentric to allocentric strategic preference and vice versa (**Table 2**).

Objective Navigation Ability Assessment

MANOVAs were performed to test the hypothesis that navigation training leads to an increase in performance on the objective navigation tasks compared to the control group. Specifically, we expected that participants in the egocentric training condition had a higher, positive differences score on egocentric navigation tasks (route sequence, route continuation, point to start), whereas allocentric training would lead to higher, positive differences scores on allocentric navigation tasks (distance comparison, location on map, point to end). First, the analysis was run for the dynamic map perspective condition. A MANOVA on the difference scores (post-training—pre-training) of six navigation tasks as independent variables and conditions as a between-subject factor was performed (**Table 3**). A trend effect of condition was found on the differences scores $F_{(12,140)} = 1.65$, $p = 0.07$; Wilk's $\Lambda = 0.77$, partial $\eta^2 = 0.13$. Second, the analysis was run for the first-person learning condition. A MANOVA with the difference scores of six navigation tasks as independent variables and conditions as a between-subject factor was performed. No significant effect of condition was found on the differences scores $F_{(12,148)} = 2.083$, $p > 0.05$; Wilk's $\Lambda = 0.94$, partial $\eta^2 = 0.03$.

Subjective Navigation Ability

MANOVAs were performed to test the hypothesis that navigation training leads to an increased rating of subjective navigation ability on the "Navigation and Orientation" and "Distance Estimation" scales and decreased score on the "Spatial Anxiety" subscale, in the experimental groups compared to the control group (**Table 4**). No main effect of condition on difference scores was found, $F_{(6,148)} = 1.29$, $p > 0.05$; Wilk's $\Lambda = 0.90$, partial $\eta^2 = 0.05$.

Interaction Between Strategic Preference, Preference Shifts, and Navigation Abilities

MANOVAs were performed to explore the relation between strategic preferences at T1 an objective and self-reported navigational abilities. Performance on egocentric and allocentric spatial tasks did not differ between participants with allocentric, egocentric or mixed strategic preference, $F_{(8,148)} = 1.51$, $p > 0.05$; Wilk's $\Lambda = 0.85$, partial $\eta^2 = 0.08$. Similarly, self-reported navigation abilities did not differ between subjects with different strategic preferences $F_{(6,152)} = 0.26$, $p > 0.05$; Wilk's $\Lambda = 0.98$, partial $\eta^2 = 0.01$.

To explore differences in egocentric and allocentric spatial abilities between participants that shifted strategy after the intervention and those who maintained the same strategic preference, a MANOVA was performed. Performance on egocentric and allocentric tasks did not differ between strategy shifters and non-shifters, $F_{(4,75)} = 0.82$, $p > 0.05$; Wilk's $\Lambda = 0.96$, partial $\eta^2 = 0.04$. Similarly, self-reported navigation abilities did not differ between strategy shifters and non-shifters, $F_{(3,77)} = 0.26$, $p > 0.05$; Wilk's $\Lambda = 0.99$, partial $\eta^2 = 0.01$.

Binary logistic regression analysis was performed to determine whether objective navigation abilities would predict shifts in strategic preference. Shifts in strategic preference were not predicted by objective navigation abilities, $\chi^2_{(4)} = 2.2$, $p = 0.69$, or self-reported navigation abilities $\chi^2_{(3)} = 0.54$, $p = 0.91$ at T1.

TABLE 3 | Mean and standard deviation of performance on navigation tasks on the pre- and post-training sessions and differences scores.

Subtask in virtual Tübingen battery	Control			Egocentric training			Allocentric training		
	T1	T2	ΔT1, 2	T1	T2	ΔT1, 2	T1	T2	ΔT1, 2
Map Perspective									
Route sequence, % correct	61.01 (25.78)	70.35 (22.50)	8.93 (28.19)	76.39 (25.69)	61.81 (31.35)	−14.58 (38.41)	67.26 (23.54)	68.45 (25.50)	1.19 (36.64)
Route continuation, % correct	82.56 (17.71)	84.01 (14.52)	1.45 (19.33)	88.89 (9.48)	82.64 (16.12)	−6.25 (20.22)	79.17 (17.38)	80.95 (18.38)	1.79 (24.78)
Distance estimation, % correct	63.08 (15.66)	70.35 (20.05)	7.27 (23.82)	67.36 (17.22)	75.00 (16.61)	7.64 (18.26)	72.02 (20.88)	75.00 (15.81)	2.98 (26.49)
Point to start, average deviation	22.04 (13.61)	23.71 (21.60)	1.67 (19.23)	32.77 (39.31)	33.35 (38.88)	0.58 (23.90)	24.64 (33.84)	24.76 (14.23)	0.13 (37.56)
Point to end, average deviation	28.44 (14.59)	34.78 (26.83)	6.34 (29.19)	22.42 (9.28)	35.11 (25.02)	12.69 (22.25)	27.17 (12.38)	35.63 (16.14)	8.46 (14.77)
Location on map, average pixels	128.40 (68.54)	121.26 (83.95)	−7.14 (85.90)	110.91 (79.53)	80.19 (57.32)	−30.72 (72.83)	114.70 (56.18)	110.72 (63.06)	−3.98 (75.01)
First-Person Perspective									
Route sequence, % correct	59.59 (28.33)	61.34 (28.58)	1.74 (32.23)	64.58 (36.19)	75.00 (26.78)	10.42 (49.31)	60.71 (25.40)	72.62 (24.24)	11.90 (26.36)
Route continuation, % correct	70.93 (18.44)	70.93 (18.84)	0.00 (22.66)	74.31 (18.92)	76.39 (22.23)	2.08 (21.54)	65.48 (19.73)	66.67 (17.38)	1.19 (27.64)
Distance estimation, % correct	63.66 (21.27)	62.50 (17.25)	−1.16 (26.70)	70.83 (16.04)	64.58 (20.22)	−6.25 (26.52)	68.45 (21.51)	63.10 (19.56)	−5.36 (28.66)
Point to start, average deviation	47.64 (19.67)	47.63 (20.44)	0.00 (27.94)	48.85 (28.14)	44.74 (24.00)	−4.11 (17.71)	47.75 (14.05)	42.31 (17.47)	−5.43 (23.74)
Point to end, average deviation	48.07 (22.53)	60.75 (29.84)	12.68 (31.99)	46.92 (21.64)	48.14 (21.67)	1.22 (25.68)	58.20 (24.10)	61.19 (23.71)	3.00 (32.56)
Location on map, average pixels	139.96 (73.62)	163.24 (89.51)	23.28 (87.74)	135.69 (69.65)	139.62 (72.10)	3.93 (56.38)	149.54 (69.87)	152.72 (68.59)	3.18 (68.65)

TABLE 4 | Scores on the Wayfinding Questionnaire pre- and post-training.

Wayfinding questionnaire subscales	Control			Egocentric training			Allocentric training		
	T1	T2	ΔT1.2	T1	T2	ΔT1.2	T1	T2	ΔT1.2
Navigation and orientation	50.16 (9.96)	50.47 (9.79)	0.30 (3.91)	49.44 (8.54)	50.56 (7.37)	1.11 (3.80)	49.62 (9.35)	48.90 (10.11)	−0.71 (4.79)
Distance estimation	11.44 (3.19)	11.42 (3.49)	−0.02 (2.23)	11.00 (2.83)	10.61 (2.91)	−0.39 (1.24)	11.19 (3.71)	11.57 (3.80)	0.38 (1.99)
Spatial anxiety*	21.42 (8.34)	22.19 (8.25)	0.77 (3.89)	21.78 (5.48)	22.06 (6.91)	0.28 (3.77)	24.24 (8.19)	23.76 (8.70)	−0.48 (3.93)

*Displayed values are mean scores, and standard deviations in brackets. *A higher score represents a higher level of spatial anxiety.*

DISCUSSION

There is a strong need to develop rehabilitation programs for acquired brain injury patients with navigation impairments. A core approach to cognitive rehabilitation is the application of compensatory strategies. In the current study, we assessed the effectiveness of a home-based rehabilitation software designed to train and develop alternative navigation strategies in healthy participants.

The current study shows that strategic navigation preference can be influenced by using a navigation training program. A large portion of the participants that received the egocentric navigation training shifted from an allocentric or mixed navigation strategy preference before training, to an egocentric navigation strategy preference after training. This shift in strategic preference was the result of the training intervention as the proportion of shifters observed in the control group was significantly lower. Exploration of the individual characteristics of participants indicated that strategy shift was not predicted by a demographic factor such as gender, education or gaming experience. Furthermore, objective and self-reported navigation abilities did not predict strategic preference shifts. While an earlier study has shown that navigation strategy can be influenced by the use of intensive therapy sessions (Claessen et al., 2016a), these findings provide support for the hypothesis that strategy training can be achieved by the use of a standardized home-training program in combination with psychoeducation.

Important to note, however, is that the increase in strategy shifts was only demonstrated for the egocentric strategy training program. Participants who engaged in the allocentric training did not change strategy more often than the control groups. These results suggest that the current home training program was ineffective in inducing an allocentric navigation strategy. There are several factors that might explain why the allocentric training seemed to be ineffective in altering strategy preference.

First, the training time was significantly higher in the egocentric training condition compared to the allocentric training condition. This difference was the result of inherent differences between the training modules that were used in both programs. The duration of the allocentric modules was mostly dependent on the skill of the participant, as the goal of the modules was to find the shortest path to a location. Conversely, the turn sequence and landmark-action modules in the egocentric training required participants to traverse lengthy routes through an environment regardless of a participant's skill level. While a higher training time was observed in the egocentric training condition, a significantly higher number of trials were attempted and completed in the allocentric training. Exploratory analysis revealed that within the experimental groups, training time did not predict the strategic preference shift. Conversely, a lower number of trials completed predicted a higher chance of preference shifts. Clearly, exposure time and the number of exercises were not the most prominent factors that predict training success. Rather, the content and presentation of the training exercises in the allocentric training modules should be improved. A small number of lengthy trials seemed to be preferable over many short trials for the development of navigation strategies.

A second explanation for the lack of strategy shifts observed after allocentric training regards the difficulty of switching between allocentric and egocentric reference frames during navigation. Egocentric navigation entails a focus on landmark-response associations, sequences, and spatial updating rather than forming relational representations (Bullens et al., 2010). Conversely, the formation and utilization of map-like representation of space are central to allocentric navigation. Constructing such allocentric representations is cognitively demanding (Wen et al., 2011; Nemmi et al., 2017; Ruggiero et al., 2018). Furthermore, a considerable processing cost is involved in switching between egocentric and allocentric reference frames (Lee and Tversky, 2001). As such, shifting from an allocentric to an egocentric navigation strategy reflects a shift towards a strategy that is cognitively less demanding, whereas a switch from an egocentric to an allocentric navigation strategy, can be regarded as a switch to a more demanding strategy. The environment used to assess the navigation strategy in this study was developed to facilitate both allocentric and egocentric strategies (Iglói et al., 2009). It is, therefore, possible, that participants who received the allocentric training, were not prompted by the environment to adopt the trained strategy and instead reverted to their default strategy.

Related this explanation are the results reported by Pazzaglia and Taylor (2007), who examined the cognitive style of spatial processing in participants with high and low survey abilities. In this study, participants with high survey abilities were less depended on learning perspective and were able to shift more efficiently from one representation to another compared to participants with low survey abilities. A similar effect was found when regarding the participants with a naïve allocentric preference as the high survey participants, as participants with an allocentric strategic preference were more responsive to the training. One important difference with this study however, is that naïve strategic preference did not correspond performance in objective navigation tasks in this study.

In addition to a shift in strategic navigation preference, we expected that exposure to the training programs would lead to an increase in objective navigation ability and self-reported navigation ability. Contrary to expectations, no effect of the training was found on both objective and subjective navigation ability. This result indicates that the strategy training did not strengthen specific navigational abilities, but rather, affected meta-cognition and behavioral selection. Additionally, we did not find differences in objective navigational abilities between the groups before the training. Preferred strategy during the pre-training session, did not correspond to higher performance on allocentric or egocentric objective navigation abilities. This finding supports a study that has shown that strategic navigation preference does not correspond to navigation ability (Prestopnik and Roskos-Ewoldsen, 2000). The relation between strategy preference and navigation skills has yet to be studied thoroughly, but might be of particular importance to the rehabilitation of navigation impairments. It appears that someone's preferred navigation strategy is not grounded in their spatial strengths and weaknesses. When developing compensatory strategy therapies for navigation impaired patients, care should be taken to make patients aware of their strengths and focus their efforts to maximizing the use strategies that utilize these abilities.

An important distinction between this study and the intended clinical application should be noted. In order to assess whether strategy use can be changed, participants were trained to adopt a navigation strategy that was contrary to their initial preferences. Patients however, will be trained to focus on and expand upon their intact navigation abilities. Ineffective strategies and abilities will be recognized and discouraged, while effective a strategies and abilities will be expanded upon. As the training is tailored to their strengths, rather than to their weakness, we expect that it will be easier for patients to utilize the training and transfer this information to real life situations.

Furthermore, the rehabilitation training that was investigated here focused on promoting the use of allocentric and egocentric navigation strategies. Both strategies rely on the use of landmarks. There have been rapports of patients with specific impairments in landmark recognition, encoding and processing (Rainville et al., 2005). Therefore, future therapies should be developed that train navigation strategies that include a minimal focus on landmarks.

Using the current iteration of the navigation training, participants with an egocentric navigation strategy preference did not adopt an allocentric navigation strategies. While it might not be possible to train allocentric navigation strategies, we expect that improvements to the training program will lead to training success. Based on the findings of this experiment, we propose to following improvements. First, fewer but lengthier training modules in the allocentric training. One explanation for the training success of the egocentric strategy training is the longer training time compared to the allocentric training. Second, as the "distal landmarks" and "local landmarks" place learning modules might have been too similar in terms of what navigation techniques were taught. A larger variety of training modules in the allocentric might be beneficial to strategy development. Third, an extended discussion of an individual's strengths and weaknesses during the psychoeducation phase of the training. The results suggest that people display navigation strategies that are not necessarily in line with their spatial abilities. Making people aware of their strengths and weaknesses might lead to higher adherence to beneficial navigation strategies. More research should be performed to determine whether a change towards an allocentric strategy preferences can be achieved when these novel features are implemented.

Over the past years, there has been a growth in software applications that combine game-like features with health related goals such as diagnosis of cognitive impairments. Spatial cognition in particular, lends itself well to serious-gaming adaptations as illustrated by applications such as "Sea Hero Quest" (Coutrot et al., 2018), "Navigeren kun je leren" and "Squirrel away" (Prpic et al., 2019). While substantial progress is being made in regards to the diagnosis of spatial impairments using these tools, the validity of treatment applications has yet to be explored. In context of this emerging field, the current study provides the encouraging results for a compensatory approach to the rehabilitation of navigation impairments using a game-like application.

In conclusion, we have developed a home-based rehabilitation training designed to treat navigation impairments that are often reported in acquired brain injury patients. A key assumption of this training is that strategic navigation preferences can be influenced by using a training. This study demonstrates that strategic navigation preference can indeed be influenced in healthy participants. Allocentric navigators could be trained to adopt an egocentric strategic preference. The current version of the training, did not induce a change in strategic preference in egocentric navigators. This may be due to factors inherent to the allocentric training such as its focus on multiple short exercises or a lack of diversity between exercises. Alternatively, switching from an egocentric to an allocentric navigation strategy, requires a switch towards a strategy that is cognitively more demanding. Egocentric navigators might not have been prompted to rely on the trained strategy in an environment, which was ambiguous regarding navigation strategies. Future research should be conducted to optimize the training for acquired brain injury patients with navigation impairments. The feasibility and effectiveness of the current approach should next be assessed in a patient population.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by CEP FSW LEIDEN. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

MK, AE, JV-M, and IH conceptualized, prepared the original draft and wrote the manuscript. MK and IH contributed to the methodology. MK was responsible for the formal analysis and investigation. IH was responsible for the funding acquisition. AE, JV-M, and IH supervised the study.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2020.00076/full#supplementary-material.

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Egocentric and Allocentric Spatial Memory in Korsakoff's Amnesia

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The goal of the present study was to investigate spatial memory in a group of patients with amnesia due to Korsakoff's syndrome (KS). We used a virtual spatial memory task that allowed us to separate the use of egocentric and allocentric spatial reference frames to determine object locations. Research investigating the ability of patients with Korsakoff's amnesia to use different reference frames is scarce and it remains unclear whether these patients are impaired in using ego- and allocentric reference frames to the same extent. Twenty Korsakoff patients and 24 matched controls watched an animation of a bird flying in one of three trees standing in a virtual environment. After the bird disappeared, the camera turned around, by which the trees were briefly out of sight and then turned back to the center of the environment. Participants were asked in which tree the bird was hiding. In half of the trials, a landmark was shown. Half of the trials required an immediate response whereas in the other half a delay of 10 s was present. Patients performed significantly worse than controls. For all participants trials with a landmark were easier than without a landmark and trials without a delay were easier than with a delay. While controls were above chance on all trials patients were at chance in allocentric trials without a landmark present and with a memory delay. Patients showed no difference in the ego- and the allocentric condition. Together the findings suggest that despite the amnesia, spatial memory and especially the use of ego- and allocentric reference frames in Korsakoff patients are spared.

Keywords: Korsakoff's syndrome, spatial memory, landmarks, delay, egocentric, allocentric, amnesia

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INTRODUCTION

Spatial memory is extremely important for successful navigation through our environment. Therefore, information about landmarks, spatial locations and routes have to be processed efficiently. Locations of landmarks can be determined in two fundamental ways to allow successful navigation and orientation; by egocentric and allocentric reference frames (e.g., O'Keefe and Nadel, 1978; Klatzky, 1998; van den Brink and Janzen, 2013). Egocentric coding involves the representation of positions of objects in relation to the observer's body (subject-to-object). This system can be used when the observer is not moving or when he/she can track his/her movements based on optic flow, vestibular and proprioceptive cues. The second system, allocentric coding, involves an externally referenced spatial coding based on inter-object relations to determine the location of an object

(object-to-object). Allocentric coding is independent of the observer's current position. In adults, there is growing evidence for a parallel spatial-representational system of these two different coding types (Simons and Wang, 1998; Wang and Simons, 1999; Committeri et al., 2004; Mou et al., 2004; Nadel and Hardt, 2004; Burgess, 2006; Waller and Hodgson, 2006, see Ekstrom et al., 2014 for a critical review on the neural correlates of allocentric spatial representations).

Several disorders and syndromes are known to have impaired episodic memory functions, including spatial memory dysfunction, as one of the symptoms. Despite their memory deficits, patients with amnesia can also have spared spatial memory aspects (Kessels et al., 2011; Oudman et al., 2011; see Rosenbaum et al., 2015 for a case study with a developmental amnesia patient showing preserved as well as impaired spatial memory). Previous research findings have shown for example impaired allocentric spatial memory functions in patients with Alzheimer's dementia or mild cognitive impairment, while egocentric spatial memory seems to be spared (Hort et al., 2007; Iachini et al., 2009). Difficulties in allocentric processing have also been observed in normal aging (Moffat and Resnick, 2002; Iaria et al., 2009; Wiener et al., 2012, 2013). Harris et al. (2012) observed a more specific decline showing that aging impairs switching from an egocentric to an allocentric strategy while switching to an egocentric strategy remained unaffected. Together these findings show that especially the allocentric reference frame is challenging not only for patients with memory deficits but also for healthy elderly (see Lester et al., 2017 for a review on spatial cognition in normal and impaired aging).

Another group of patients who are known to suffer from memory deficits, but also have spared memory capacities, are patients with Korsakoff's syndrome (KS); patients with this syndrome all display excessive memory disorders, lack of binding abilities, attention deficits and disorientation in time and place due to excessive alcohol abuse in combination with vitamin B1 deficiency (Kopelman, 2002; Tielemans et al., 2012; Arts et al., 2017; see Heirene et al., 2018 for a systematic review on the assessment of alcohol-related cognitive impairment including KS). Spatial memory performance in patients with this syndrome is characterized by a deficit in explicitly remembering spatial information (Holdstock et al., 1999; Kessels et al., 2000; van Asselen et al., 2005; Postma et al., 2006, 2018; Kessels and Kopelman, 2012).

To date, reports exploring the ability of amnesic patients with KS to use the different object-location-framing types are scarce, but a previous study by Holdstock et al. (1999) focused on this process. They designed a spatial memory task in which participants should recall the position of single spot LED lights after various delays. Three task conditions were used, a short-delay condition (0, 3 or 8 s), as well as an allocentric and egocentric condition (both with delays of 5, 20 or 60 s). In the short-delay condition, the participant was instructed to look away from the board, without changing position. In the allocentric condition, the participant had to move around the light-board during the delay. Due to the enlightened room during this procedure, participants could make use of external stimuli to re-orientate. The egocentric condition took place in a dark room where participants could not make use of external stimuli and could only rely on his/her body position. Participants were encouraged not to move during this condition. The results showed that patients with amnesia due to KS were impaired in both the ego- and allocentric condition to the same extent. In both conditions, patients' performance declined to a greater extent due to the extension of the delay as compared to the performance of the controls. Due to this accelerated forgetting, it was concluded that the KS patients have impaired memory for both allocentric and egocentric information.

The present study aimed to investigate the ability of patients with KS to use egocentric and allocentric frames of reference to determine object locations. While previous studies have shown an impairment particularly in allocentric processing with spared egocentric memory function in healthy aging (Moffat and Resnick, 2002; Iaria et al., 2009; Wiener et al., 2012, 2013) as well as in patients with memory deficits (Hort et al., 2007; Iachini et al., 2009), Holdstock et al. (1999) observed impairment in both allocentric as well as egocentric processing in KS patients. The present study aimed to shed light on these diverse findings.

Here, we extended the previous work by Holdstock et al. (1999) by using an ecologically valid paradigm. Furthermore, we examined the effect of adding landmarks, as this may facilitate allocentric representations and contributes to re-orientation (Learmonth et al., 2002). To further clarify the ability of these patients to use egocentric and allocentric strategies, the current study applied a paradigm previously designed by van den Brink and Janzen (2013). In their study, a group of 30 to 35-month-old toddlers watched an animation of a bird flying in one of two trees standing in a virtual environment. After the bird disappeared, the camera turned around, by which the trees were briefly out of sight and then turned again to the center of the environment in which the trees were located. Participants were asked in which tree the bird was hiding. In half of the trials, a landmark was shown. Comparable to Holdstock et al. (1999), and in addition to the paradigm of van den Brink and Janzen (2013), the present study made use of a direct condition and of a delay condition in which the camera turn is delayed by 10 s.

In addition to the studies of Holdstock et al. (1999) and van den Brink and Janzen (2013), the current study explored if the performance on this virtual spatial memory task (the "bird task" of van den Brink and Janzen, 2013) was related to the performance on an everyday memory test (the Global Memory Index of the Rivermead Behavioral Memory Index—Third Edition; RBMT-3). Since mental rotation is of great importance in the present paradigm, we also studied whether the participants were able to mentally rotate spatial information and to which degree the performance on this paper-and-pencil mental rotation task was related to the performance on the bird task.

Concerning the underlying neural correlates, the hippocampus is crucial in using allocentric frames of reference (e.g., O'Keefe and Nadel, 1978; Maguire et al., 1998; Holdstock et al., 2000). Although KS is primarily characterized by diencephalic lesions (Aggleton and Saunders, 1997; Arts et al., 2017), hippocampal atrophy has been reported in these patients as well (e.g., Sullivan and Pfefferbaum, 2009). Furthermore, damage to diencephalic structures that are connected to the hippocampus may also result in impaired allocentric representations (Holdstock et al., 1999). Holdstock et al. (1999) not only observed an impairment in the allocentric condition, but also in egocentric processing. Similarly, we expect that KS patients perform worse in all conditions as compared to healthy controls. We furthermore hypothesize that KS patients have a preference for an egocentric strategy and will perform worse in conditions where only an allocentric strategy will be successful (Neave et al., 1997; Kopelman, 2002). Since, an egocentric representation alone is not providing enough information to correctly perform the task, we hypothesize that the amnesic patients will make more egocentric errors (i.e., selecting the position of the tree the bird was hiding in before the turn) in comparison to the controls. Although landmarks facilitate the use of an allocentric object-location strategy, we expect the patients not to benefit from these to the same extent as controls. Concerning the findings of Holdstock et al. (1999) and the memory deficits KS patients have (Kopelman, 2002; Kessels and Kopelman, 2012), we expect the performance of the patients to decline more than the control performance after a delay.

MATERIALS AND METHODS

Participants

Twenty patients with severe anterograde amnesia, diagnosed with KS and 24 healthy age- and intelligence matched controls (see **Table 1**) successfully participated in the present study and were included in the analyses. Four more patients did not want to complete the task and two patients did meet the exclusion criteria; their data was not included in the further analyses. General exclusion criteria for both groups were a stroke in history, other neurological disorders, like alcohol-related dementia, premorbid intelligence level below 65 and not being able to communicate in Dutch. All patients were abstinent for at least 6 weeks before being tested. All patients were recruited from the Centre of Excellence for Korsakoff and Alcohol-Related Cognitive Disorders of Vincent van Gogh Institute for Psychiatry, Venray, The Netherlands. Inclusion criteria for the patients were a DSM-5 diagnosis of a Major Neurocognitive Disorder due to Alcohol, Amnestic/Confabulatory Type (confirmed by neuropsychological assessment, neurological examination, and neuroradiological findings) and meeting the criteria for the KS (Kopelman, 2002; Arts et al., 2017). Available MRI scans were visually rated to exclude other diseases by an experienced researcher, focusing on global cortical (GCA) and medial temporal lobe atrophy (MTA; see Wahlund et al., 2000) as well as white-matter hyperintensities (WMH; Fazekas et al., 1993). MRI data were available for fifteen patients; five patients did not undergo an MRI-scan. The matched healthy volunteers were recruited from the staff of the clinic or through relatives of one of the researchers.

In all participants, premorbid intelligence was estimated using the Dutch version of the National Adult Reading Task (NART; Schmand et al., 1992). Additionally, the Mental Rotation Task (MRT; Shepard and Metzler, 1971; Aleman et al., 2004) was administered. In this paper-and-pencil task, consisting of seven items, the participant is asked which two of four turned

TABLE 1 | Demographical and other characteristics for both groups.

Characteristic	Healthy controls	Korsakoff's amnesia	Statistic	*p*-value
Number of participants	24	20	-	-
Sex (men:women)	19:5	16:4	$\chi^2_{(1)} = 0.005$	$p = 0.946$
Age (*M, SD*)	57.79 (6.90)	60.20 (7.70)	$t_{(42)} = 1.095$	$p = 0.280$
Educational level (*Mode, range*)	5 (2–7)	3 (2–7)	*Mann–Whitney U* = 164.500	$p = 0.066$
Intelligence level estimation (NART IQ; *M, SD*)	89.96 (11.51)	89.65 (15.84)	$t_{(42)} = -0.075$	$p = 0.941$
Mental Rotation Task (*M, SD*)	9.42 (2.62)	7.10 (1.68)	$t_{(42)} = -3.46$	$p = 0.001$
RBMT-3 Global Memory Index (*M, SD*)	-	59.79 (5.86)	-	-
MTA		0: *N* = 8		
		1: *N* = 3		
		2: *N* = 3		
		3: *N* = 1		
		4: *N* = 0		
		NA: *N* = 5		
GCA		0: *N* = 2		
		1: *N* = 9		
		2: *N* = 3		
		3: *N* = 1		
		NA: *N* = 5		
WMH		0: *N* = 2		
		1: *N* = 11		
		2: *N* = 2		
		3: *N* = 0		
		NA: *N* = 5		

Notes. NART, National Adult Reading Test; RBMT-3, Rivermead Behavioral Memory Test—Third Edition; MTA, Medial Temporal Lobe Atrophy Rating: 0 = no atrophy, 1 = widening of choroid fissure, 2 = widening of choroid fissure and temporal horn of lateral ventricle, 3 = moderate hippocampal volume loss, 4 = severe hippocampal volume loss; GCA = Global Cortical Atrophy Rating: 0 = no atrophy, 1 = opening of sulci and mild ventricular enlargement, 2 = volume loss of gyri and moderate ventricular enlargement, 3 = knife-blade atrophy and severe ventricular enlargement; WMH = Fazekas White-Matter Hyperintensities Rating: 0 = no WMH, 1 = pencil-thin periventricular or punctuate focal deep WMH, 2 = smooth halo periventricular or early confluence of focal deep WMH, 3 = irregular periventricular WMH extending into the deep white matter or large confluent regions of deep WMH.

block-patterns is the same as the target pattern. The other two patterns are a mirrored version of the target. During the explanation of this task, we made use of two 3D demonstration pieces to clarify the rotation of the patterns.

Besides, the KS patients completed the Rivermead Behavioral Memory Task—Third edition (RBMT-3; Wilson et al., 2008), an ecologically valid episodic memory test battery. Education level was classified on a 7-point-scale based on the Dutch educational system (Verhage, 1964). **Table 1** shows the demographical and behavioral characteristics of both groups. Note that the healthy controls had a slightly higher education level compared to the patients, but no group differences were found for estimated premorbid intelligence (see **Table 1**).

Spatial Memory Task

A computerized paradigm, developed by van den Brink and Janzen (2013), was adopted and used to study spatial memory. Commercially available animation suited software Blender[1] was used to construct 48 movies, which were shown by the software Presentation. Each movie lasted 30 s in the direct condition and 40 s in the 10-s delay condition. The movies showed an animated bird, appearing in front of the camera, turning around and flying into one of three identical trees. Other than in the original experiment designed by van den Brink and Janzen (2013) in which two trees were shown, we here used environments in which three trees were positioned to increase the difficulty of the task. Each tree was positioned at different distances within an open 3D environment, forming an equilateral triangle. After the disappearance of the bird, the camera perspective followed a path that resulted in a perspective change; 90° to the left or 90° to the right of the center of the environment. This change in perspective led to the illusion of self-motion by the participant. During this turn, the trees and all other objects were out of sight for a while, preventing tracking of the bird's hiding place. While the camera turned away the empty landscape without trees and landmark was shown. In the delay condition, the empty landscape was shown for 10 s and in the direct condition, the camera turned back to the center of the environment directly after turning away. At the end of the turn path, the camera again turned to the center of the environment which led to the reappearance of all objects. The total duration of the turn was 4 s. In 24 movies, the turn that led to the perspective change, was delayed by 10 s. The distance to the center of the environment (before and after the spatial transformation) was six Blender units (6 m). The distance of the spatial transformation was 8.5 m. After the reappearance of all objects in all conditions, the participants should point to the tree in which they believe the bird was hiding.

The trees were positioned in four 3D environments: snow, autumn, mud and a grass landscape. In the environments of autumn and grass, a landmark was added; respectively a bench and a slide. The landmark was positioned inside the cluster of the three trees, but closer to the front tree (see **Figure 1**). The presence of a landmark possibly facilitated the use of an allocentric strategy (object-to-object relation use). To survey subject-to-object relation use, in one-third of the trials the starting position of the correct tree, in which the bird was hidden, corresponded to the position of that tree relative to the participant's body after the camera angle change (for example before and after the turn, the tree in which the bird was hidden, was at the most right side relative to the participant). These trials that allow the use of an egocentric strategy as well as the use of an allocentric strategy were called position-congruent trials. In all the other trials, the position of the correct tree before the turn did not match the position of that tree, relative to the participant's body, after the turn (position-incongruent trials). These trials only allow the use of an allocentric strategy to be successful (see **Figure 2**). In this position-incongruent trials, an egocentric choice resulted in an incorrect response; only an allocentric representation led to the correct response in these trials.

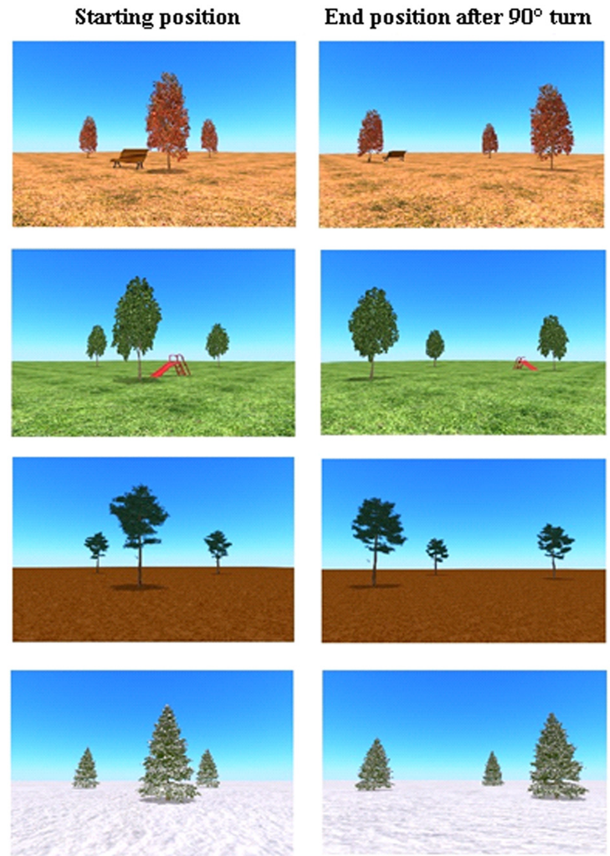

FIGURE 1 | Overview of the four environments, with and without landmark and a reproduction of the spatial transformation in the virtual spatial memory task. The direction of the turn is respectively right, left, left, right.

Consequently, all factors led to the distribution of the movies/trials into eight conditions: (1) position congruent, delay, landmark; (2) position congruent, delay, no landmark; (3) position congruent, direct, landmark; (4) position congruent, direct, no landmark; (5) position incongruent, delay, landmark; (6) position incongruent, delay, no landmark; (7) position incongruent, direct, landmark; and (8) position incongruent,

[1] www.blender.org

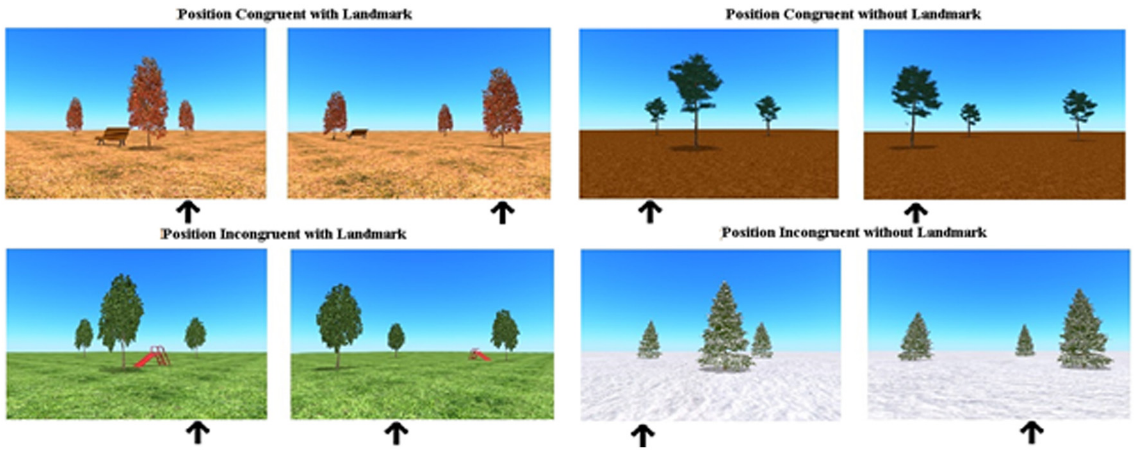

FIGURE 2 | Overview of position congruent (ego- as well as allocentric strategies are successful) and position incongruent trials (only allocentric strategy is successful), with and without a landmark in the virtual spatial memory task.

direct, no landmark. The position congruent conditions each consisted of four movies; position incongruent conditions each consisted of eight movies. Alternately, a block of 12 trials with a delay and 12 trials with a direct recall was shown. Whether the participant began with a block with or without delay was counterbalanced.

Procedure

The total duration of the procedure was 45–60 min and it took place in a quiet room in the Korsakoff Clinic in Venray or the participant's environment. The procedure started with the paper-and-pencil MRT, which took 10 min. After this, the participants were seated in front of a 16.7-inch laptop, which was within arm reach of the participant, for the start of the bird-task. Participants were told that they were going to see an animation of a bird flying away and hiding in one of three trees and that they should watch carefully in which tree the bird flew and to remember this place during a spatial transformation of the camera. They were also told that in half of the movies, this turn of the camera was delayed so that they should remember the position of the bird for a longer time. After the spatial transformation, the participant was required to indicate where the bird was hiding; they could respond verbally or they could point to the tree in which they thought the bird was hiding. After the administration of this response by the researcher, the bird reappeared from the correct tree, giving the participant feedback. Participants were given two practice trials in advance: one with a direct recall and one with a delay. The study was approved by the Institutional Review Board of Vincent van Gogh Institute for Psychiatry (CWOP27/1/2014) and written informed consents were obtained, as per the Declaration of Helsinki.

Analyses

The total percentage of correct responses for each condition was measured. As each trial has three response options, the chance level performance was indicated at 33.3% correct answers. An alpha of 0.05 was used in all analyses. Effects with an alpha between 0.05 and 0.10 were judged marginally significant. First, we compared both groups using a 2 (Group: KS vs. controls) × 2 (Position: incongruent vs. congruent) × 2 (Delay: direct vs. delay) × 2 (Landmark: yes vs. no) repeated measures ANOVA. Participants could respond in an allocentric way (which leads to the correct answer), in an egocentric way (which leads to incorrect responses in position incongruent trials) or in a random way. *T*-tests were used to compare the amount of egocentric errors in both groups.

Furthermore, it was calculated if both groups differ in their performance on the MRT and if they performed above chance level of seven correct answers (max = 14). Also, we correlated the spatial memory performance with the performance on the RBMT-3 and the MRT and the visually rated MTA, and GCA.

RESULTS

Figures 3, **4** show the performance on the spatial memory task. Controls performed above chance level on all eight conditions (all p-values < 0.05). Patients performed at chance in the position incongruent condition without a landmark and with a delay ($p = 0.16$). Performance on all other conditions were above chance ($p < 0.05$, **Figure 4**). This analysis revealed that, in general, patients performed significantly worse ($M = 60.0$, $SE = 4.33$) than controls ($M = 84.95$, $SE = 3.95$; $F_{(1,42)} = 18.15$, $p < 0.005$, $\eta_p^2 = 0.30$, **Figure 3**). In addition, a significant performance difference between trials with and without a landmark was found ($F_{(1,42)} = 27.24$, $p < 0.005$, $\eta_p^2 = 0.39$), with participants scoring higher on the trials with a landmark ($M = 82.97$, $SE = 3.05$) than on the trials without landmark ($M = 61.98$, $SE = 3.99$). Furthermore, a significant main effect of Delay was found ($F_{(1,42)} = 6.36$, $p = 0.02$, $\eta_p^2 = 0.13$), with participants scoring higher on trials with a direct recall ($M = 75.25$, $SE = 3.01$), than on trials with a delay ($M = 69.70$, $SE = 3.24$). The main

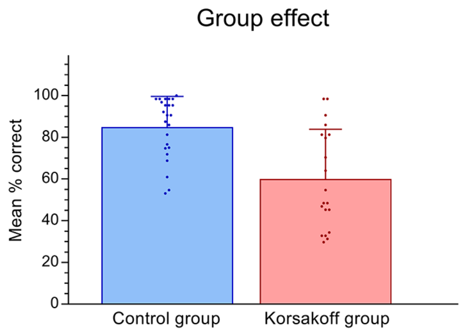

FIGURE 3 | Means and standard deviations for Korsakoff's patients vs. controls. Scatter dots show individual mean performance for all conditions.

effect of Position type was not significant ($F_{(1,42)} = 3.36$, $p = 0.07$, $\eta^2_p = 0.07$). There the performance on position-congruent trials ($M = 74.71$, $SE = 3.12$) was only slightly higher compared to position-incongruent trials ($M = 70.23$, $SE = 3.20$). The Landmark × Delay interaction effect was not significant ($F_{(1,42)} = 2.92$, $p = 0.095$, $\eta^2_p = 0.07$). Furthermore, the 3-way interaction of Landmark × Delay × Group was not significant ($F_{(1,42)} = 2.92$, $p = 0.095$, $\eta^2_p = 0.07$).

Neither the interaction between Group and Landmark nor the interaction between Group and Delay were significant ($F_{(1,42)} = 0.40$, $p = 0.53$ and $F_{(1,42)} = 0.92$, $p = 0.34$, respectively), nor was the Group by Position interaction ($F_{(1,42)} = 1.53$, $p = 0.22$), the Position by Landmark interaction ($F_{(1,42)} = 1.19$, $p = 0.28$) or the Position by Delay interaction ($F_{(1,42)} = 0.26$, $p = 0.61$). Furthermore, none of the other 3- or 4-way interactions were statistically significant (all p-values > 0.27).

Additional analyses for the separate groups were performed, showing a strong interaction effect of Landmark and Delay in the control group ($F_{(1,23)} = 8.35$, $p = 0.008$, $\eta^2_p = 0.27$), which was absent in the KS group ($F_{(1,19)} = 0.00$, $p = 1.00$), indicating that in controls a delay resulted in a worse performance on the no-landmark trials as compared to the landmark trials. In both groups, a significant main effect for Landmark was found (Patients: $F_{(1,19)} = 9.78$, $p = 0.006$, $\eta^2_p = 0.34$; Controls: $F_{(1,23)} = 18.63$, $p = 0.001$, $\eta^2_p = 0.45$). A significant main effect of Delay was observed in the controls only ($F_{(1,23)} = 7.84$, $p = 0.01$, $\eta^2_p = 0.25$; KS: $F_{(1,19)} = 0.95$, $p = 0.34$). In neither of the two groups did we find a significant main effect for Position type or any significant 2- or 3-way interactions with Position type (all $p > 0.10$).

A t-test to compare the percentage egocentric errors in both groups did not reveal a group difference ($p = 0.16$). Correlational analyses showed that better performance on the bird task was strongly correlated with better performance on the MRT ($r = 0.41$, $p = 0.005$). Separate correlations for both participant groups were not significant. A Spearman rank correlation on the MRI measures revealed a not significant correlation between the amount of MTA and the performance on the bird task ($r = -0.492$, $p = 0.062$) as well as between the amount of GCA and performance on the bird task ($r = -0.216$, $p = 0.440$). The correlation between RBMT-3 performance and spatial memory performance was not significant ($p > 0.19$).

DISCUSSION

The present study aimed to investigate the ability of KS patients to use egocentric and allocentric frames of reference to remember an object's location. In a virtual reality environment, amnesic patients and healthy controls should determine the hiding place of an animated bird with and without a delay of 10 s as well as with and without a landmark present. Position incongruent trials needed to be processed in an allocentric frame of reference while position congruent trials could be solved with the use of an egocentric frame of reference. Results showed that patients generally performed significantly worse than controls. While controls performed above chance level on all trials, patients were at chance in position-incongruent trials without a landmark present and with a memory delay. As expected for all participants trials with a landmark were easier than without a landmark and trials without a delay were easier than with a delay. A trend was observed with position congruent trials being easier than position incongruent trials. An interaction effect with the factor group involved showed that controls benefited more from a landmark and the no-delay condition than patients.

Results did not show any differences between cognitively unimpaired controls and Korsakoff amnesics in their performance on position incongruent trials in comparison to position congruent trials. Furthermore, the performance of the patients on position-congruent and position-incongruent trails did not differ. This shows that the performance of patients with Korsakoff's amnesia does not decline more in a task in which an allocentric frame of reference is needed relative to a task in which an egocentric strategy is sufficient. Besides, adding a landmark improved the performance of the patients, which indicates their use of an allocentric strategy. Furthermore, the amount of egocentric errors did not differ between healthy controls and Korsakoff amnesics. Together these results indicate, in comparison to our hypotheses, the efficient use of an allocentric frame of reference to determine object locations by patients with Korsakoff amnesia. This finding is in line without results from Holdstock et al. (1999) that showed that patients with KS had a comparable impairment on both ego- and allocentric conditions.

In general, all participants' achievements declined due to the addition of a delay. However, the controls only showed a delay effect in the condition without a landmark; for an optimal delay performance, a landmark was helpful for them. The patient group alone did not show a delay effect. This might be because trials without an extra delay of 10 s involved a memory component. After all, the hiding place of the bird needed to be remembered for the short period in which all objects were out of sight. Therefore, possibly the difference between both conditions was too small to detect a memory decline in the patient group.

All participants benefited from a landmark being present. Landmarks are helpful in navigation because they may facilitate the use of an allocentric reference frame and contribute to re-orientation in an environment (Learmonth et al., 2002; Janzen and Jansen, 2010; Eppstein and Vass, 2013). However,

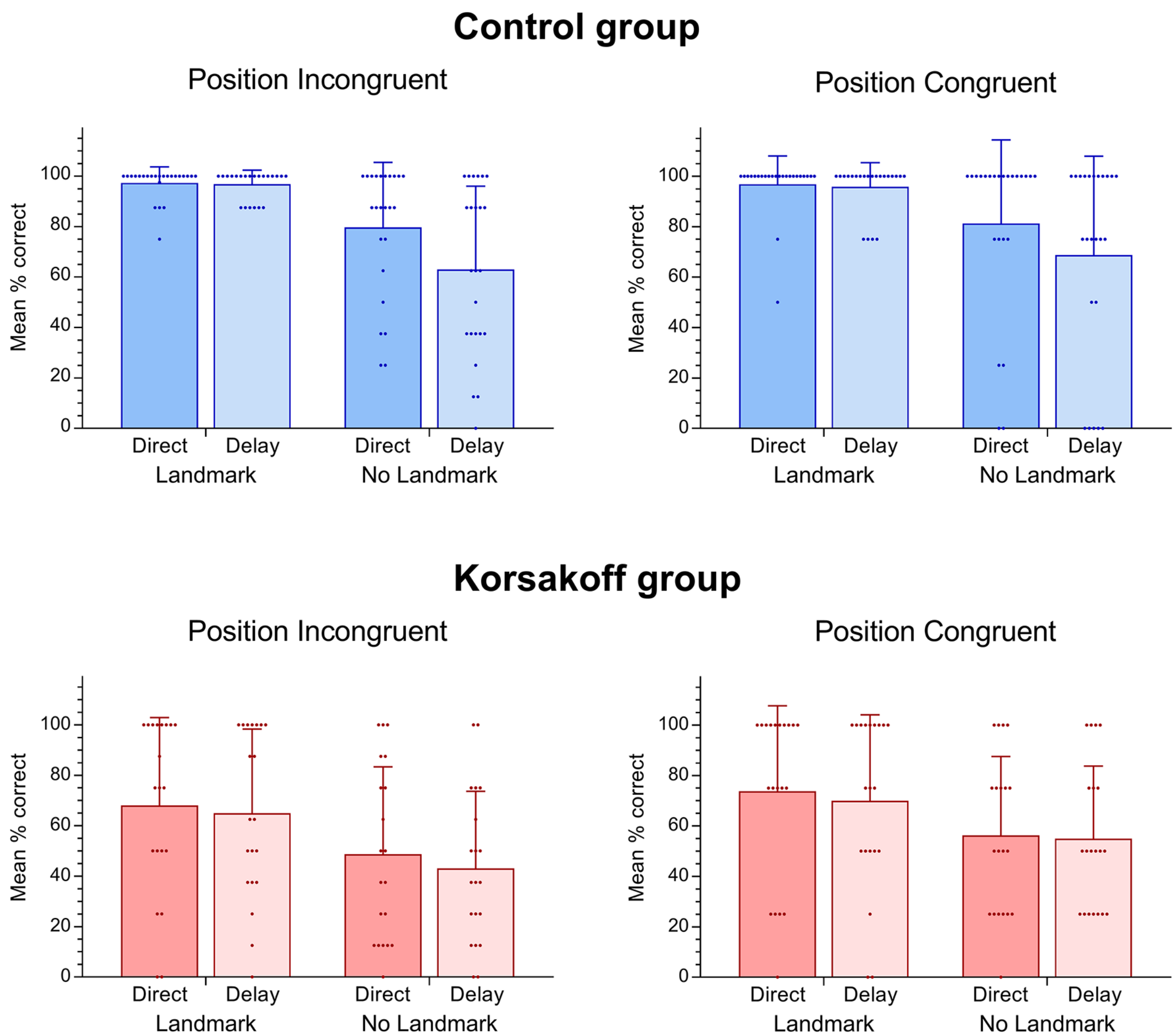

FIGURE 4 | Means and standard deviations as well as scatter dots of individual performance in the virtual spatial memory task for both controls and patients with Korsakoff's syndrome (KS).

findings in toddlers with the "bird task" developed by van den Brink and Janzen (2013) showed that young children did not benefit from the presence of landmarks. On the contrary, children were distracted and even performed worse in trials with landmarks that were of interest to children. Young children seem to rely on optic flow cues only, whereas adults prefer a landmark strategy which might be beneficial in trials that require an allocentric strategy. Other than the patients and controls in the present study who did not show a difference between position-congruent and incongruent trials, 30-month old children in the study by van den Brink and Janzen (2013) demonstrated a strong decline in position incongruent trials. Most likely successful landmark use also requires the ability to use an allocentric strategy.

Further analyses show that better performance on the bird task strongly correlates with better performance on the paper-and-pencil mental rotation task (Shepard and Metzler, 1971). Both participants groups showed similar relationships which were not significant for the separate groups, most likely due to the low number of participants. This finding shows that the ability to mentally rotate objects also links to reorientation skills in larger environments. Furthermore, although patients with more hippocampal atrophy performed worse on the bird task, the results on the RBMT-3 and bird task were not correlated. This—in combination with the correlation between the bird task and mental rotation—supports the notion that the effects we have found with our paradigm do not only reflect the effect of the overall amnesia but may be the result of a specific deficit in the processing of spatial orientation related information.

It is worthwhile to notice that the patient group with 20 Korsakoff patients could be seen as relatively small. However, studies with larger groups are sparse as described in the review on implicit memory by Hayes et al. (2012). The number of patients in the present study, in addition, does not differ from previous studies examining spatial memory (e.g., Oudman et al., 2016). Nevertheless, a replication in a larger sample would be valuable. A further limitation is that the present study includes a visual rating of the MRI scans for a subset of 15 patients only. Future research in a larger sample including a more precise method to measure hippocampal volume, such as voxel-based morphometry, would allow for concrete conclusions about the relationship between behavioral performance and brain regions. A future study could additionally consider to include more trials per condition. A larger number of trials might reduce the variability in the data. However, a new study design should balance between more trials and possible fatigue and motivation problems in the patient group, since Korsakoff's amnestic have severe cognitive impairments.

Since KS is more common in men than in women our participant sample has a corresponding unequal distribution which makes analyses including sex not informative and beyond the scope of our manuscript. However, sex-related differences in spatial navigation tasks are a matter of debate, with often men outperforming women in navigation tasks (see e.g., Coutrot et al., 2018), whereas women often have an advantage in object location tasks (Murphy et al., 2009; Bocchi et al., 2020; but see also Postma et al., 2004). Note, however, that the effect sizes for sex difference on cognitive tasks are typically small, and that the effects of the amnesia itself overshadow these subtle sex differences.

In sum, our study confirms a spatial memory deficit in KS patients with the patient group performing at chance in the most difficult condition (position incongruent trials, without landmark and with memory delay). In contrast to previous findings in young children both participant groups benefited from a landmark present. In line with findings by Holdstock et al. (1999), KS patients showed no difference in the ego- and the allocentric condition, suggesting that patients can efficiently use an allocentric frame of reference to maintain orientation in a spatial environment. Our study suggests that despite the amnesia and in line with findings by Oudman et al. (2016), spatial memory in KS patients can be spared.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The study was approved by the Institutional Review Board of the Vincent van Gogh Institute for Psychiatry (CWOP #27-01-2014) and written informed consents were obtained in accordance with the Declaration of Helsinki.

AUTHOR CONTRIBUTIONS

GJ and RK planned and designed the study, with input from CR. CR collected and coded the data with input from JO. GJ, RK, and CR analyzed the data. GJ, CR, and RK drafted and wrote the manuscript. JO provided input on the drafted version of the manuscript.

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[2]https://mensenlinq.nl/overlijdensberichten/arie-wester-6373680/

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CASE REPORT
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Travel Planning Ability in Right Brain-Damaged Patients: Two Case Reports

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Planning ability is fundamental for goal-directed spatial navigation. Preliminary findings from patients and healthy individuals suggest that travel planning (TP)—namely, navigational planning—can be considered a distinct process from visuospatial planning (VP) ability. To shed light on this distinction, two right brain-damaged patients without hemineglect were compared with a control group on two tasks aimed at testing VP (i.e., Tower of London-16, ToL-16) and TP (i.e., Minefield Task, MFT). The former requires planning the moves to reach the right configuration of three colored beads on three pegs, whereas the latter was opportunely developed to assess TP in the navigational environment when obstacles are present. Specifically, the MFT requires participants to plan a route on a large carpet avoiding some hidden obstacles previously observed. Patient 1 showed lesions encompassing the temporoparietal region and the insula; she performed poorer than the control group on the ToL-16 but showed no deficit on the MFT. Conversely, Patient 2 showed lesions mainly located in the occipitoparietal network of spatial navigation; she performed worse than the control group on the MFT but not on the ToL-16. In both cases performances satisfied the criteria for a classical dissociation, meeting criteria for a double dissociation. These results support the idea that TP is a distinct ability and that it is dissociated from VP skills.

Keywords: planning, spatial navigation, navigational planning, navigational impairments, travel planning, right brain lesions, topographical orientation

BACKGROUND

Planning ability is fundamental for ensuring efficient spatial navigation and it can explain the wide individual differences frequently reported in spatial navigation, both in healthy (Wolbers and Hegarty, 2010; Sharma et al., 2016; Bocchi et al., 2017, 2019) and in clinical populations (Passini et al., 1995; Ciaramelli, 2008). Passini et al. (1995) reported that patients with mild-to-moderate Alzheimer's Disease failed in reaching a destination in the hospital and this was mainly due to their impaired planning ability. They struggled to build structured plans, binding each sub-goal to the next one (take the elevator to reach the 5th floor) employing a "trial-and-error" strategy. The Prefrontal Cortex (PFC) seems to support the planning ability involved in navigational skills (Ciaramelli, 2008). Indeed, PFC is crucial for several functions of spatial navigation, such as processing of context representations following the navigational goal (Martinet et al., 2011), keeping some spatial information in working memory (Ciaramelli, 2008; Lithfous et al., 2013), planning a path (Spiers, 2008), applying strategies (Dahmani and Bohbot, 2015) and modifying the ongoing plan when detours are required (Boccia et al., 2014a; Spiers and Gilbert, 2015).

Several studies suggested the existence of a specific planning ability devoted to solving navigational tasks (Basso et al., 2006; Cazzato et al., 2010; Martinet et al., 2011; Boccia et al., 2014a; Schacter et al., 2017). This ability has been called in several ways: visuospatial planning (VP; Basso et al., 2006; Cazzato et al., 2010), spatial navigational planning (Martinet et al., 2011; Schacter et al., 2017; Carrieri et al., 2018: Bocchi et al., 2017) or, even, travel planning (TP; Bocchi et al., 2019) that is the term we will adopt thereafter. Martinet et al. (2011) defined TP as the mental evaluation of alternative action-sequences to infer optimal trajectories for reaching a goal, suggesting the dynamic nature of this kind of planning. In this vein, TP could be differentiated from visuospatial planning. Direct evidence of the dissociation between travel and visuospatial planning is not yet available, but some indirect evidence could help in disentangling this issue.

A distinction between the processing of visuospatial and navigational information is detectable in different cognitive domains, being visuospatial information, at least in part, acquired and processed differently from navigational information (Piccardi et al., 2008; Nemmi et al., 2013; Bianchini et al., 2014a). Indeed, the perception of navigational stimuli involved specific cortical areas that are not involved in the perception of other types of visuospatial inputs (Epstein and Kanwisher, 1998). Another distinction refers to memory processing. While visuospatial working memory requires remembering positions in the environment, navigational working memory also requires the continuous updating of our perspective every time a new orientation is provided (e.g., a turn); this would constitute an additional load for the navigational working memory (Nori et al., 2009). Some studies have provided evidence for double dissociations in visuospatial and navigational working memory (Piccardi et al., 2010, 2011). Also, in the mental imagery domain, different kinds of mental images have been described (Guariglia and Pizzamiglio, 2006; Palmiero et al., 2019): topological images are mental representations of stimuli in which the subject can navigate (i.e., rooms, squares, cities, maps, etc.,) and that can be transformed into (or correspond to) cognitive maps of the environment. Non-topological images are mental representations of stimuli, such as a desktop, the interior of a car (Ortigue et al., 2003), single objects or arrays of objects, which can be manipulated but never navigated. Clinical evidence demonstrated the existence of double dissociations between topological and non-topological mental images in brain-damaged patients (Palermo et al., 2010a; Guariglia et al., 2013) as well as differences in mental generation process across the life span (Piccardi et al., 2015); further supports derives from behavioral (Boccia et al., 2014b) and neuroimaging studies (Boccia et al., 2015). Indeed, navigational stimuli are processed differently from common objects (i.e., the clock hands or the map of Italy; Boccia et al., 2014b) in healthy participants; also, these differences are more evident in older individuals (Piccardi et al., 2015). Furthermore, different brain networks support mental imagery of familiar environmental space, geographical space and non-spatial categories, such as faces or clock hands (Boccia et al., 2015, 2019). Neuroimaging evidence also points towards a functional segregation between the processing of navigational information. Indeed, pictures of navigational stimuli (buildings and/or landmarks) activate specific brain regions (i.e., retrosplenial complex, parahippocampal place area, and occipital place area; Aguirre et al., 1998; Epstein and Kanwisher, 1998; Ishai et al., 1999; Dilks et al., 2013) when compared with other categories of objects, such as faces (Ishai et al., 2000; O'Craven and Kanwisher, 2000; Gorno-Tempini and Price, 2001).

Several neural models tried to disentangle the key nodes of TP in the brain. Based on the idea that TP is a complex and multifaceted ability, neural models highlighted the interplay between different brain regions. For example, Martinet et al. (2011) proposed that the interaction between the hippocampus and the PFC yields to the encoding of manifold information pertinent to TP, including prospective coding and distance-to-goal correlates. The hippocampal formation would send the representations of the spatial context to the PFC, which in turn would process such representations according to the current situation. Similarly, Ekstrom et al. (2014) suggested that hippocampal and extra-hippocampal areas (i.e., parahippocampal, retrosplenial, prefrontal and parietal cortices), characterize the neural basis of spatial representations during navigation. According to Spiers and Gilbert (2015), the hippocampal-prefrontal reciprocal interactions would be fundamental when detours require to revise a travel plan.

Finally, the view that travel planning is a distinct ability from visuospatial planning is also supported by the evidence that individuals with developmental topographical disorientation (DTD; Iaria et al., 2009; Bianchini et al., 2010, 2014b; Nemmi et al., 2015; Conson et al., 2018) show impairments in travel planning but not in planning *per se* (Bianchini et al., 2010). Indeed, patients with DTD and good visuospatial planning skills may show a selective deficit in planning a route to reach a destination. In other words, this study suggested that travel planning could be selectively impaired.

Overall, these studies lead to hypothesize that travel and visuospatial planning may be distinct abilities. Despite studies directly testing these differences, especially in brain-damaged patients, are still lacking, it is important to approach such an investigation for both theoretical and clinical implications. It can disclose not only if these processes are dissociated, but also it can be useful for disentangling subtle deficits in travel planning in brain-damaged patients who usually show motor impairments (Mohr and Binder, 2011), and thus may need to set out alternatives ways to blocked-routes. Therefore, in this study we describe patients who performed worse than controls in only one of the two tests tapping TP and VP, showing also greater differences between the two tasks than those expected for the controls, thus meeting the criteria for classical dissociation. The opposite pattern of performance we detected in the two patients provides evidence for a double dissociation between TP and VP skills, for the very first time.

CASE REPORT

This study was designed following the principles of the Declaration of Helsinki and was approved by the ethical committee of the IRCCS Fondazione Santa Lucia (Protocol number: CE/PROG.670; date: 18th April 2018). Written informed consent was obtained from all participants for participation in the study and the publication of this case report.

Patients underwent an extensive neuropsychological assessment aimed at excluding deficit in general cognitive functioning and visuospatial disorders. After that, they underwent the Mine Field Task (MFT) and the Tower of London (ToL-16) task, to test TP and VP, respectively. For easiness of exposition, neuropsychological assessment and planning evaluation will be divided into subheadings.

Neuropsychological Assessment and Lesion Description

The neuropsychological assessment included tasks of orientation in time and space (Spinnler and Tognoni, 1987); abstract and/or verbal reasoning (Raven, 1938; Spinnler and Tognoni, 1987); language functions (Ciurli et al., 1996); visuospatial and verbal working memory (Spinnler and Tognoni, 1987), as well as verbal long-term memory, constructive apraxia and attention (Spinnler and Tognoni, 1987; **Table 1**).

The Walking Corsi Test (WalCT; Piccardi et al., 2008, 2013; De Nigris et al., 2013) was administered to assess topographic short-term memory in a vista navigational space, namely "the space that can be visually apprehended from a single location or with only little exploratory movements" (Wolbers and Wiener, 2014, p. 3).

Patients also performed a standard battery for evaluating the presence of hemineglect (Pizzamiglio et al., 1989; **Table 1**). The battery includes: Letter Cancellation Test; Line Cancellation Test; Wundt-Jastrow Area Illusion Test; Sentence reading.

Finally, patients were assessed for perceptual and representational neglect through the Familiar Squares Description Test (Bisiach and Luzzatti, 1978) and the O'Clock Test (Grossi et al., 1989).

Patient 1 was a 65-year-old right-handed woman with 13 years of education, employed as a teacher. Sixty-one days before our examination, she suffered from a stroke involving the right temporal and parietal lobes, extending also to the insula and the subcortical structures (**Figure 1**). Naming and comprehension abilities were within the normal range. She had neither difficulty in verbal and visuospatial memory tests nor deficits in abstract reasoning. She showed no signs of perceptual or representational neglect.

Patient 2 was a right-handed 51-year-old woman with 13 years of education, employed as a healthcare worker. Fifty-five days before our examination, she suffered from a stroke, involving the right parietal and occipital lobes and the thalamus (**Figure 1**). Her speech was fluent and informative; her naming and comprehension abilities were intact. The patient did not show difficulty in either verbal or spatial short and long-term memory. She had no deficit in abstract reasoning and did not show signs of perceptual or representational neglect.

Details about lesions (**Supplementary Tables S1**, **S2**) and disconnections (**Supplementary Table S3**) are reported in **Supplementary Material**, along with the procedure used to derive them. In brief, patient 1 showed a lesion mainly located in the frontal lobe, extending to the parietal and the temporal lobe, as well as disconnection in a wide number of frontal and frontoparietal tracts. Instead, patient 2 showed a lesion of the occipital lobe (including the posterior cingulate/retrosplenial cortex), extending only marginally to the temporal lobe, the basal ganglia, and the cerebellum; she also showed disconnection of these posterior areas and fronto-temporal tracts. Lesion reconstructions are also depicted **Supplementary Figure S1**.

Assessment of Planning Abilities

Participants were tested individually in a quiet room. The administration order of the MFT and ToL-16 was counterbalanced across the participants.

The Minefield Task (MFT)—TP Task

The MFT aims to assess the ability to plan a route on a matrix, avoiding some invisible obstacles (false mines) previously seen for a few seconds. It consisted of a walkable white/black chessboard (8 × 8 matrix, 2.5 × 2.5 m) placed on an empty room (**Figure 2**). An additional tile was placed out of the matrix (1 meter below the chessboard) to indicate the starting position. Two circles with a 10 cm diameter (one red, one green) were used to indicate the starting and the ending positions of each route. Some "mines" of 15 cm diameter made with red and white felt were placed during the observation phase on the chessboard. The number of mines that could be placed on the matrix ranged from two to nine depending on the trail difficulty. In the first trial, two mines were placed on the chessboard, with the number of mines progressively increasing by one in the successive trials (three mines in the second trial, four in the third and so on). Each trial included two items; therefore, the total number of possible trials was 16.

TABLE 1 | Neuropsychological Assessment: Spatial orientation, Temporal orientation, Raven's progressive matrices; Digit span forward, Digit span backward, Rey 15 item memory test immediate recall, delayed recall and recognizing (Rey 1- Rey 2-Rey 3), Story recall test; ca, Constructive apraxia; Corsi Block Tapping Task (Corsi, 1972; Walking Corsi Test, Piccardi et al., 2008; De Nigris et al., 2013), Visual search test.

No.	Spatial orientation	Temporal orientation	Raven	Digits f	Digits b	Rey 1	Rey 2	Rey 3	Story	Ca	CBT	WalCT	Visual Search
1	+++	+++	2	n.a.	n.a.	n.a	n.a.	n.a	3	4	5	6	4
2	+	+	4	4	4	4	4	0	4	4	6	6	3

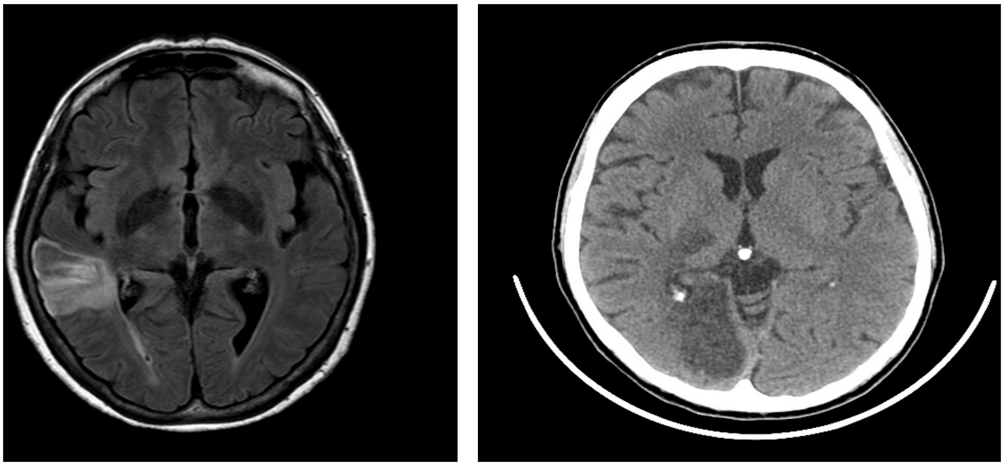

FIGURE 1 | Lesion of Patient 1 (on the left) and Patient 2 (on the right). Patient 1 that showed a lesion in temporoparietal regions with subcortical structures and insula, performed poorly on the visuospatial planning (VP) task (ToL-16) but showed no deficit on the travel planning (TP) task (MFT). Patient 2 that showed a lesion on parieto-occipital areas with the involvement of thalamus performed poorly on the TP task (MFT) but not on the visuospatial task (ToL-16).

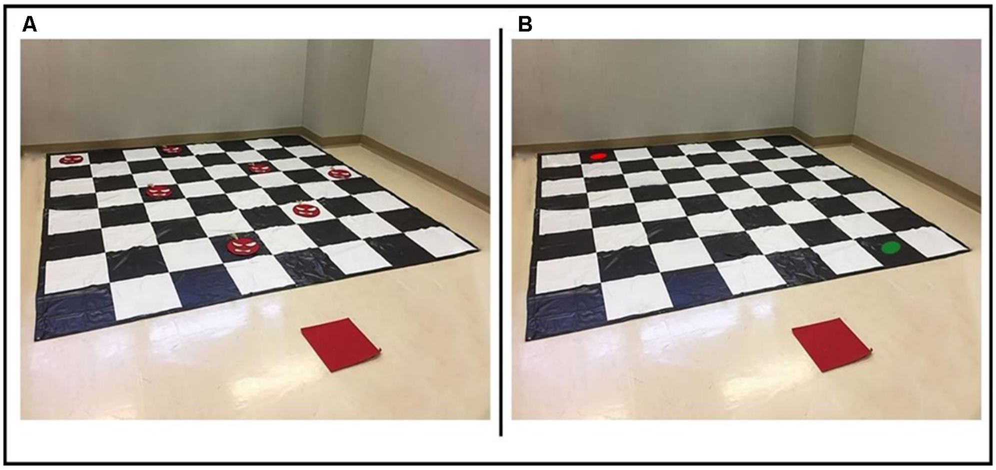

FIGURE 2 | An item from the MFT. **(A)** The chessboard in the acquisition phase; participants were allowed to see the mine locations. **(B)** The chessboard without the mines on it; participants were allowed to see it after being unblindfolded. Green and Red circles indicated the start and the end of the route to plan.

Participants were placed on the starting position outside the matrix and blindfolded. Two experimenters placed the mines on the matrix. Then, in the observation phase, the eye patch was removed, and participants were required to carefully study the position of the mine on the chessboard. After 10 s, participants were blindfolded again, and the experimenters removed the mines on the chessboard and put the starting and ending point on the chessboard within 10 s. After that, in the testing phase, the blindfold was removed, and the participants had to perform the planned route. Participants moved on the chessboard only vertically and horizontally but not diagonally. Between the starting and ending points, many routes were possible; to avoid that they chose a long, peripherical ride to avoid all the mines, participants were instructed to use the shortest one. To allow testing patients with motor disorders, similarly to the WalCT adopted with patients (De Nigris et al., 2013), participants performed the route from the green circle to the red circle by using a pointer, being careful to avoid the positions in which they had seen the mines in the observation phase. The task began with only two mines on the matrix. If the participant succeeded to avoid them, the second item of the same trial was not presented, and the next trial was administered. On the contrary, if the participant failed, the second item of the same trial was administered. The task was stopped when participants failed to reproduce both items of a given trial. The MFT allowed obtaining a score that corresponded to the number of mines in the longest sequence correctly performed. The maximum score was 9, the minimum was 0.

TABLE 2 | Patients' score at the standard battery for evaluating the neglect syndrome (Pizzamiglio et al., 1989).

No.	Left H	Right H	Left-lines	Right-lines	W-J test (unattended responses)	Sentence reading	O'Clock test (LQ)	Square description test (LQ)
1	53/53	51/51	11/11	10/10	0	6/6	−10.34	12.5
2	52/53	51/51	11/11	10/10	0	6/6	0	0

Left h/Right h: Letter Cancellation Test, left and right (Pizzamiglio et al., 1989). L-Lines: Line Cancellation Test, left and right (Pizzamiglio et al., 1989). W-J Test: Wundt-Jastrow Area Illusion Test (Pizzamiglio et al., 1989). Sentence reading (Pizzamiglio et al., 1989); O'Clock test- Laterality Quotient (Grossi et al., 1989); Familiar Square Description- laterality quotient (Bisiach and Luzzatti, 1978).

ToL-16 (Shallice, 1982; Boccia et al., 2017a)—VP Task

The ToL-16 was aimed at assessing visuospatial planning ability. Although ToL has long been considered a measure of general planning ability, recent studies have disclosed a visuospatial component in this task (Unterrainer et al., 2004; Franceschi et al., 2007; Cheetham et al., 2012).

The version used in the present study included 16 trials (Boccia et al., 2017a) of increasing difficulty with a maximum number of allowed moves that vary from 2–7. Following Krikorian et al. (1994), the accuracy corresponded to the number of solved problems according to the number of attempts needed to achieve the solution (i.e., 3 = solved at first attempt; 2 = solved at second attempt; 1 = solved at the third attempt; 0 = not solved). Here, the sum of the accuracy at each trial (maximum accuracy = 48) was considered.

Statistical Analyses and Results

Neuropsychological assessment of the two patients is summarized in **Tables 1**, **2**. Performances on the experimental tasks of patients and controls are reported in **Table 3**.

We analyzed patients' performances on the ToL-16 and MFT tasks considering a group of control (**Table 1**) which included participants with no signs of neurological or psychiatric disorders (10 participants, 3 males; mean age = 57.2 years; SD = 9.1; mean education = 9.1; SD = 3.5). We expected that the difference between the cases' standardized scores on the two tests was greater than the difference between the same two tests obtained from a control group. The Crawford's analysis for single cases (Crawford and Howell, 1998; Crawford and Garthwaite, 2002) was applied using the computer program DISSOCSBayes_ES.EXE (Crawford and Garthwaite, 2007), which first tests whether the case's scores meet the criterion for a deficit on Tasks X and Y (Crawford and Howell, 1998) and then applies the Bayesian Standardized Difference Test (BSDT) to the standardized difference between the case's scores on Tasks X and Y.

Patient 1 showed a deficit in the ToL-16 task ($t_{(1, 9)} = -1.8$; one-tailed $p = 0.05$) but not in the MFT ($t_{(1, 9)} = 2.5$; one-tailed $p = 0.01$); actually patient 1 scored better than the control group in the MFT. She fulfilled the criteria for a dissociation, putatively a classical dissociation. Patient 2, instead, showed a deficit in the MFT ($t_{(1, 9)} = -2.0$; one-tailed $p = 0.03$) but not in the ToL-16 task ($t_{(1, 9)} = 1.4$; one-tailed $p = 0.09$). She fulfilled the criteria for a dissociation, putatively a classical dissociation.

The BSDT on the difference between the case's standardized scores obtained on the ToL-16 task and MFT showed a probability that the standardized difference for a member of the control population would be greater than that of the case showed for patient 1 of 0.00226 (one-tailed), being the effect size (Z-DCC) for the difference between the case and controls (plus 95% Credible Interval): Z-DCC = −3.715 (95% CI = −5.704 to −2.042). The BSDT on the difference between the case's standardized scores obtained on the ToL-16 task and MFT showed a probability that the standardized difference for a member of the control population would be greater than that of the case shown for patient 2 of 0.00851 (one-tailed), being the Z-DCC for the difference between case and controls (plus 95% Credible Interval): Z-DCC = 2.899 (95% CI = 1.538–4.497).

TABLE 3 | Scores of the patients 1 and 2 and the mean scores obtained by the Control Group on the experimental tasks (MFT: Minefield Task and on the ToL-16).

	MFT SCORE	ToL-16 SCORE
CONTROL GROUP	5.1	36.5
PATIENT 1	9	24
PATIENT 2	2	46

DISCUSSION

Dissociations play a key role in building and testing theories in cognitive neuroscience, for instance providing critical support for several models in the field (Dunn and Kirsner, 2003). A classical dissociation (Shallice, 1988) requires that a patient obtained an "impaired" performance on task X, but his/her performance is "not impaired" on task Y (see also Ellis and Young, 1996). Furthermore, Crawford et al. (2003) argued that a further criterion is needed for a classical dissociation: "a comparison of the difference between a patient's scores on the two tasks of interest to the differences on these tasks observed in the control sample" (Crawford and Garthwaite, 2007, p. 349). This also allows avoiding incorrectly classified cases (Crawford and Garthwaite, 2005, 2006). Following this criterion, this study compared the performances on VP and TP of two right brain-damaged patients without neglect. We provided evidence for a double dissociation between the two types of planning, supporting the idea that they could be considered distinct abilities, involving different cognitive processes that are subtended by, at least partially, different neural bases.

Patient 1, whose performance was good on the MFT, performed poorly on the ToL-16 task, showing impairment in VP and an intact TP. Patient 2 performed poorly on the MFT but not on the ToL-16 task, showing a normal VP and an impairment in TP. These results suggest that only the lesion of Patient 2, which involved the right occipitoparietal lobes, impaired TP. This result allows drawing some conclusions. First of all, Patient

1 showed an adequate level of cognitive functioning. Her lesion and disconnections involved the temporoparietal regions and the insula, but not the PFC; nevertheless, she showed an impairment in the ToL-16 task, in the absence of deficits in TP. This performance can be explained considering that brain regions compromised in Patient 1 contribute to VP (e.g., insular cortices Owen, 1997; Robbins, 1998; van den Heuvel et al., 2003).

Patient 2 showed a globally preserved cognitive profile with performances adequate in all the cognitive domains. She performed worse than the control group on the TP but not on the VP. Interestingly, her lesion encompassed areas of the occipital and the parietal lobe involved in learning positions within navigational vista space (Nemmi et al., 2013) during the WalCT, which is the same space of the MFT. In light of Wolbers and Wiener's definition (2014; p. 3) of the vista space, Patient 2's performance could be explained considering the common features of WalCT and MFT and the navigational nature of the MFT. Both the WalCT and MFT require to remember positions in the navigational vista space, to implement a route and to process information available only for a short time. Importantly, during the MFT, differently from the WalCT, this information should be further manipulated and used to perform the task (i.e., avoiding the mines). Furthermore, the MFT requires to decide which route to perform to reach the goal of choosing the shortest one among several alternatives, while the WalCT requires to remember a given route. Thus, a more active involvement of the PFC should be present in the MFT, since planning is less involved in the WalCT. Accordingly, patients performed well within the normal range on the WalCT, supporting the idea that the two tasks tested different aspects of navigation. In other words, it is possible to explain Patient 2's performance considering that the lesioned areas are important to navigation, for example when positions in the environment should be remembered. These areas could be important either during the WalCT and the MFT considering that both of them take place in the navigational vista space. However, Patient 2's performance on the WalCT was good, suggesting that these areas could specifically be involved during TP that, unlike the WalCT, requires to further manipulate the spatial information (position in the environment) to plan the right route to the goal. In sum, Patient 1, who showed lesions and disconnections involving more anterior areas (e.g., insular cortex and frontoparietal tracts), was spared on TP but performed worse than controls on VP. Instead, Patient 2, who showed lesions and disconnections involving the occipitoparietal network of navigation, was spared on VP but performed worse than controls on TP, likely due to an impairment in using spatial information to plan the "right" route to the goal.

At date, TP and VP have been considered two aspects of the same planning, sub-served by the same neurocognitive processes. Indeed, both of them may require the correct functioning of PFC (Martinet et al., 2011; Carrieri et al., 2018; Choi et al., 2018) to put together the right sequence of actions to reach a goal; however, TP could be considered a specific planning that shares with VP common processes but also differences. Indeed, Patient 2 who showed a deficit of TP but intact capabilities of VP did not show any lesion of PFC, suggesting that TP involves a network that mostly relies on other brain areas.

The ToL-16 task and the MFT share many processes: working memory useful for maintaining online the final configuration in the ToL-16 task and the position of mines in the MFT; the visual mental imagery necessary to plan the sequence of beads movements in the ToL-16 task and that of steps in the MFT; the planning process itself which put in sequence a series of hand actions in the ToL-16 task and a series of displacements in the MFT; the monitoring process which compares the result of the planning with the desired outcome (namely, the right configuration in the ToL-16 task and the reaching of the goal avoiding mines in the MFT). However, at least two of these processes, i.e., working memory and visual imagery, do not rely on the same brain networks. Indeed, several studies showed that memory in navigational space is subserved by a specific network which, at least partially, differs from that involved in visuospatial memory in no-navigational space (Piccardi et al., 2010; Nemmi et al., 2013). Also, for what attains visual imagery, TP would rely more on topological images (mental representations of environmental stimuli, i.e., rooms, squares, etc., corresponding to cognitive maps of the environment—Guariglia and Pizzamiglio, 2006) rather than on non-topological images. The existence of two different systems processing these two types of mental images has been demonstrated by the observation of double dissociation in right brain-damaged patients (Guariglia et al., 2013) and by neuroimaging studies in healthy participants (Boccia et al., 2015).

Thus, the present double dissociation may be due to the derangement of navigational working memory or to a deficit in the topological imagery deficit which does not affect in any way the VP. This interpretation is consistent with previous lesion locations and disconnections observed in patients with representational neglect restricted to topological mental images (Committeri et al., 2015; Boccia et al., 2018). It is also consistent with findings from DTD patients, who seem to struggle to build a navigational plan even though they still perform well within the normal range on the ToL-16 task (Bianchini et al., 2010).

The double dissociation reported here also suggests that the proper functioning of PFC, although fundamental for planning, it is not sufficient to ensure TP. Indeed, TP likely relies on the cooperation of several areas instead of a specific region, in line with recent neural models including travel planning (Martinet et al., 2011; Ekstrom et al., 2014; Spiers and Gilbert, 2015; Schacter et al., 2017). This is in line with the idea that TP is a complex ability requiring other cognitive processes, such as memory and mental imagery (Byrne et al., 2007; Schacter et al., 2007; Wolbers and Hegarty, 2010; Bocchi et al., 2017, 2019).

To sum up, three key findings emerged from the present case reports. First, lesions in the right occipitoparietal lobes impair the ability to plan a route in the navigational space, even in the absence of lesions in the PFC. Second, TP is likely associated with the parieto-medial temporal lobe network of spatial navigation (Kravitz et al., 2011; Boccia et al., 2014a, 2017b; Sulpizio et al., 2016). Third, and most importantly, a double dissociation exists between VP and TP, suggesting that they involve different brain areas, even sharing some processes.

Despite the importance to describe such a dissociation, the present study has some limitations. For instance, the control group should be increased additional indexes for MFT could be derived, such as planning and execution time. Also, memory for mines position could be investigated in future studies, to disentangle the contribution of memory for positions to a deficit in TP. Finally, even if our patients did not show hemineglect, future studies should investigate the possible association between mental imagery deficits in patients with representational neglect (Palermo et al., 2010b; Guariglia et al., 2013) and TP, to definitively disentangle if the deficit in planning a route is due to an impairment of topological mental images.

Notwithstanding, the present findings are important from both a theoretical and a clinical point of view. On the one hand, they provide the first evidence for a double dissociation between TP and VP skills. On the other hand, knowing that TP can be selectively impaired may be useful for improving rehabilitation programs in brain patients who often show motor impairments (Mohr and Binder, 2011).

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The study, which involved human participants, was reviewed and approved by Comitato Etico Indipendente della Fondazione Santa Lucia-Santa Lucia Foudation, Via Ardeatina, 306, 00179 Roma. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

AB, LP, MB, AD, and CG conceived and designed the experiment. AB the collected data. AB and MB analyzed the data and made the lesion mapping. All authors contributed to writing the article.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2020.00117/full#supplementary-material.

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Path Learning in Individuals With Down Syndrome: The Floor Matrix Task and the Role of Individual Visuo-Spatial Measures

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Environment learning is essential in everyday life. In individuals with Down syndrome (DS), this skill has begun to be examined using virtual exploration. Previous studies showed that individuals with DS can learn and remember paths in terms of sequences of turns and straight stretches, albeit with some difficulty, and this learning is supported by their cognitive abilities. This study further investigates environment learning in the DS population, newly examining their ability to learn a path from actual movements, and to learn increasingly long paths, and how their performance relates to their visuo-spatial abilities and everyday spatial activities. A group of 30 individuals with DS and 30 typically-developing (TD) children matched for receptive vocabulary performed a 4 × 4 Floor Matrix task in a grid comprising 16 squares (total area 2.3 × 2.3 meters). The task involved repeating increasingly long sequences of steps by actually moving in the grid. The sequences were presented in two learning conditions, called Observation (when participants watched the experimenter's moves), or Map (when they were shown a map reproducing the path). Several visuo-spatial measures were also administered. The results showed a clear difference between the two groups' performance in the individual visuo-spatial measures. In the Floor Matrix task, after controlling for visuo-spatial reasoning ability, both groups benefited to the same degree from the Observation condition vis-à-vis the Map condition, and no group differences emerged. In the group with DS, visuo-spatial abilities were more predictive of performance in the Floor Matrix task in the Observation condition than in the Map condition. The same was true of the TD group, but this difference was much less clear-cut. The visuo-spatial working memory and visualization tasks were the strongest predictors of Floor Matrix task performance. Finally, the group with DS showed a significant relation between Floor Matrix task performance in the Observation condition and everyday spatial activity. These results enlarge on what we know about path learning in individuals with DS and its relation to their visuo-spatial abilities. These findings are discussed within the frame of spatial cognition and the atypical development domain.

Keywords: down syndrome, route learning, floor Matrix, working memory, visuo-spatial abilities, environment measures

INTRODUCTION

Path Learning in Individuals With Down Syndrome

Knowing how to find your way through an environment is essential in everyday life. When people experience a new environment, they form an internal mental representation of it, showing elements (such as landmarks) and their relations, called cognitive maps (Tolman, 1948). This spatial information can be acquired using different modalities, such as from looking at maps or from navigation. Maps depict a whole area, showing landmarks and paths connecting them based on an aerial view of the layout, so they present the information in an allocentric way. Route learning by navigation involves memorizing a particular sequence of movements and changes of direction, and a set of place-action associations in order to reach a destination. In navigation the environment is experienced from an egocentric point of view, based on sensorimotor (e.g., vestibular and kinesthetic) information identifying an individual's positions in space and self-to-object distances (Montello, 2005). The related representations and their features can be assessed using various tasks, such as map drawing or retracing a previously-explored route.

Learners' personal characteristics are a source of variability in how successfully they retain environmental information (Meneghetti et al., 2019). When investigating the features of mental maps and the conditions that favor their formation, individuals with Down syndrome (DS) seem an interesting population to study because of the constraints imposed by the syndrome. DS is a genetic syndrome caused by chromosome 21 trisomy. Individuals with DS generally have an intelligence quotient between 25 and 70, and a mental age of between 5 and 6 years (Dykens et al., 2000; Kittler et al., 2008). It is worth further analyzing the visuo-spatial skills of individuals with DS because, although they are generally considered a relative strength (Dykens et al., 2000; Silverman, 2007), they have yet to be extensively explored. For example, these individuals' environment learning ability (which is one type of visuo-spatial ability) has been little investigated compared with other visuo-spatial skills (see below). There is growing interest in how individuals with DS gain confidence with moving around and reaching places outside their home (a workplace, supermarket, gym, or other people's homes), and returning home by various means (walking, taking public transport, or asking someone for directions; Yang et al., 2018). Broadening our knowledge of the DS population's environment learning can shed light on their adaptability and capacity for autonomy, which are strongly related to their quality of life. There is reason to believe that individuals with DS can encounter difficulties with environmental learning, based on neuropsychological evidence. We know that hippocampal structures (and other related brain regions, like the parahippocampal cortex) are involved in navigation (e.g., Burgess et al., 2002). We also know that hippocampal volumes have been found smaller in individuals with DS than in matched typically-developing (TD) individuals (Pinter et al., 2001; Contestabile et al., 2010), and that the former perform less well than the latter in cognitive tasks measuring hippocampal functions (Pennington et al., 2003).

Visuo-spatial abilities have commonly been found relatively stronger than verbal abilities in individuals with DS (Dykens et al., 2000; Silverman, 2007). It would be wrong to generalize from this observation, however, since visuo-spatial abilities include a whole set of different skills. Any individual may have different strengths and weaknesses relating to a given construct, and different tasks may be used to assess these various subsets of skills. Most studies examined visuo-spatial abilities in terms of small-scale abilities, when the spatial information needing to be handled concerned objects, figures, etc., in small spaces (i.e., smaller than the individual's body), as in paper-and-pencil tasks. A few studies examined large-scale abilities, i.e., for managing information about, or moving in larger spaces, as in the case of learning a path (Yang et al., 2014; Meneghetti et al., 2019). For instance, Yang et al. (2014)'s review showed that most of the small-scale abilities examined refer to: recalling locations (placing previously-seen objects in their appropriate positions in an empty layout, e.g., Vicari et al., 2005); closure (combining different pieces of information into larger wholes, and breaking larger wholes down into smaller parts; e.g., Vicari et al., 2006); mental rotation (mentally turning 2D and 3D objects; Hinnell and Virji-Babul, 2004; Vicari et al., 2006; Meneghetti et al., 2018); and visuo-spatial construction (reconstructing a whole object from a number of parts, as typically assessed with the WISC block design test; Cornish et al., 1999; Wechsler, 2003; Kittler et al., 2004). The results of such studies vary. For instance, they usually show that individuals with DS perform less well than TD children -matched (or controlled) for general cognitive functioning – in recalling locations and closure, while the results for mental rotation and visuo-spatial construction are less consistent. Another ability amply investigated in individuals with DS is visuo-spatial working memory (WM), which is concerned with retaining and processing visuo-spatial information, and can be distinguished as sequential or simultaneous (remembering increasingly long sequences or locating increasingly large numbers of elements, respectively, Cornoldi and Vecchi, 2003). There is evidence of individuals with DS performing as well as matched TD children in sequential WM tasks that involved remembering in the right order positions presented one at a time on a matrix. In contrast, individuals with DS performed comparatively less well in simultaneous WM tasks, which involved recalling the position of colored cells displayed simultaneously on a matrix (Lanfranchi et al., 2004, 2009; Carretti and Lanfranchi, 2010).

Only a few studies conducted to date examined large-scale visuo-spatial abilities, such as path learning, in individuals with DS, however, and most of them did so using the exploration of virtual environments (VE).

Path Learning in Virtual Environments

To our knowledge, there have been five studies on path learning in individuals with DS (Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015; Purser et al., 2015; Toffalini et al., 2018). They all support the impression that individuals with DS are more confident when forming environment representations with egocentric features (as seen from the person's point of view), while they have more difficulty with forming allocentric representations (based on relations between landmarks). In most of the studies, participants were asked to learn a virtual indoor or outdoor path presented from the person's point of view. Then they were asked to reproduce the previously-seen path by trial and error until they completed it without making any mistakes, with a maximum of attempts allowable (the criterion condition). This was conceived as an egocentric task as it retained the person's own point of view. Afterward, participants were asked to find a shortcut to their destination. This was conceived as an allocentric task as it involved linking landmarks located in the layout and taking new paths (not previously covered). It should be noted that the paths to learn generally consisted of about four segments (based on the number of turns and straight stretches), i.e., in a regular environment (a grid of 3 × 3 streets), participants needed to learn two segments for each of two paths (Courbois et al., 2013; Farran et al., 2015), or in a square-shaped environment they had to cover all four sides (Toffalini et al., 2018), or in an irregular environment they had to take a path with around four choice points (Davis et al., 2014; Purser et al., 2015).

The results of these studies show that individuals with DS were able to recall (Davis et al., 2014) or recognize landmarks irrespective of their position (Courbois et al., 2013; Toffalini et al., 2018), though their performance was not as good as that of matched TD controls. They could also trace previously-learnt paths, but it took them more attempts to do so, and they made more mistakes (Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015; Purser et al., 2015), or took longer (Toffalini et al., 2018). When asked to find a shortcut, they had a clearly worse performance than TD controls (Courbois et al., 2013; Farran et al., 2015), and the few individuals with DS who succeeded in producing representations with allocentric feature still preferred to use a strategy based on their personal point of view in their moves (Farran et al., 2015). The evidence that individuals with DS can form environment representations with egocentric, but not with allocentric features supports the assumption that large-scale environment knowledge is first acquired egocentrically, and it is only afterward that individuals can form allocentric mental representations of an environment (Siegel and White, 1975).

Given that individuals with DS seem able to handle egocentric-sequential information (as expressed by their ability to repeat a previously-explored path) better than allocentric-simultaneous information, it seemed worthwhile to examine in the present study how much sequential information presented from a personal point of view they are able to learn and recall in the right order.

Path Learning With Actual Moves

Any actual moves are necessarily egocentric because of the body's involvement (Montello, 2005), and they are an interesting condition to consider when examining path learning in individuals with DS.

It has been demonstrated that individuals with DS placed in a controlled setting, such as a platform about 4 m square (drawing inspiration from the Water maze task; Morris, 1984), are just as capable of making moves using distinctive local cues as matched TD children, while they perform less well when they need to use environmental information such as the locations of other elements in the layout, or the edge of a platform (Mangan, 1992; Pennington et al., 2003; Lavenex et al., 2015). Studies on the typically-developing population, designed mainly to shed light on the development of egocentric and allocentric representations, used a similarly controlled setting (like a platform) to examine children's actual moves to reach a target (such as a hidden toy) that could be identified from local cues (e.g., colored elements) or environmental features (other elements on or off the platform). The studies showed that TD children learn to form egocentric representations by 2–5 years old, and can even use environmental information, such as the shape of a room (Hermer and Spelke, 1994; Newcombe et al., 1998). At around 6 years of age their mental representations can become view-independent, showing the children's ability to use the structural features of an environment to infer target locations (e.g., Nardini et al., 2008, 2009; Ruggiero et al., 2016).

In the case of individuals with DS, findings obtained with tasks that involved moves in controlled settings showed that they were able to reach places using information learned from a personal point of view, and the same was true of studies using virtual exploration (Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015; Purser et al., 2015). However, they had difficulty using environmental information related to a whole layout, and organizing elements in relation to each other (i.e., allocentrically), even when they should have been able to do so according to their mental age (given evidence of TD children definitely being able to using environmental information by around 6 years old).

Using a controlled real-life setting could be an interesting way to investigate to what extent individuals with DS are able to learn sequences of actual moves. This condition represents a large-scale setting in "vista space" (where the spatial setting and the movements inside it are all visible to participants; Montello, 1998). Given the current paucity of evidence concerning the ability of individuals with DS to learn sequences from actual moves in controlled settings (Mangan, 1992; Pennington et al., 2003; Lavenex et al., 2015), it is worth considering some inspiring studies conducted in the TD domain. In an effort to distinguish between small-scale and large-scale processing abilities (as used for navigation), Piccardi et al. (2008) suggested a 1:10 scaled-up version of the classical Corsi Block-Tapping Test (CBT; Corsi, 1972; a board with nine blocks irregularly placed on it), called the Walking Corsi Test (WalCT; Piccardi et al., 2008), i.e., a space marked on the floor (3 × 2.5 meters in size) with squares irregularly placed inside it. The task involved actually walking around the floor and repeating the same sequence of moves so as to pass through the same squares as an examiner had previously done (stopping in each square for 2 s). Since this task assesses the ability to learn increasingly long sequences of moves (generating a measure of span), it is conceived as a navigational WM task. TD children proved able to perform this task: from 5.4 to 6.7 years of age, which largely corresponds to the mental age of individuals with DS, they were able to reproduce sequences of up to about 3 squares (from M = 1.90, SD = 1.18 up to M = 2.43, SD = 0.84; Piccardi et al., 2014). There was evidence of their performance in the WalCT gradually improving, and becoming stable by 10 years of age, with virtually no gender-related differences (Piccardi et al., 2014). It should be noted that, when asked to learn a sequence of 4 squares, 6 years old were already able to do so, though they had more difficulty than older children, around 11 years old (Piccardi et al., 2015). The WalCT seems specifically to capture the processing ability involved in moving within a vista space setting, which differs from the processing ability involved in small-scale WM tasks. In fact, TD 5–6 years old performed less well in the WalCT than in the CBT or a verbal WM task that involved repeating increasingly long series of digits (Piccardi et al., 2014). In other words, the WalCT is a task capable of providing a key to elucidating the type of strength or weakness in participants' navigation ability (e.g., Bianchini et al., 2010; Palermo et al., 2014).

Overall, this kind of vista space task (which is a way to reproduce a large-scale environment in a controlled setting) holds promise for assessing the path learning ability of individuals with DS. The actual moves necessarily involve adopting the egocentric view, enabling us to examine participants' ability to learn a path based on information gained from a personal point of view.

It is important to note individuals with DS benefit from being given a regular visuo-spatial context, such as a uniform grid layout (Carretti et al., 2013), so a vista space setting with regular squares placed within a grid (as reproduced virtually by Courbois et al., 2013; Farran et al., 2015) could represent a favorable condition for assessing path learning with actual moves in the DS population.

The first aim of the present study was therefore to examine the ability of individuals with DS to learn a path from increasingly long sequences of actual moves.

The Role of Cognitive Abilities in Supporting Path Learning

It is generally assumed that large-scale abilities (like path learning) are related to small-scale abilities (Hegarty et al., 2006), which include a large set of skills used in basic processing, such as WM, and higher-level functions like mental rotation (Hegarty and Waller, 2005). The relation between small-scale (spatial) abilities and environment learning performance has been demonstrated in adults (Hegarty et al., 2006; Weisberg et al., 2014) as well as in developmental age, in 5–6 years old children (e.g., Fenner et al., 2000; Purser et al., 2012, 2015; Merrill et al., 2016; Thomas et al., 2016), albeit with some inconsistencies in the findings. In a recent study, Merrill et al. (2016) found that the visuo-spatial abilities – i.e., mental rotation, spatial visualization (the ability to arrange spatial stimuli), and visuo-spatial working memory (in a task resembling those used to test simultaneous WM) – of children 6–12 years old were related to their path learning accuracy (after exploring VE), but it was only in females that verbal WM was also a significant predictor of their performance. Fenner et al. (2000) also found visuo-spatial abilities (including visualization, mental rotation and tasks resembling those used to test sequential WM) related to path learning (after exploring VE) in 5–6 years old, but not in 8–9 year old children. Other studies found additional cognitive abilities involved in path learning (again after exploring VE), including attention, perception, memory and executive functions (Purser et al., 2012; Nys et al., 2015). There is also evidence of 6 year old performance in the WalCT being related to individual visuo-spatial factors, such as field-independent cognitive style (Boccia et al., 2019), and even verbal abilities (such as grammar comprehension, when the squares used in the WalCT are identified with images reproducing landmarks; Piccardi et al., 2015).

It is useful to analyze the contribution of the individual (small-scale) abilities involved in environment learning (a large-scale ability) because this helps us to pinpoint factors that can explain variability in people's performance. Analyzing these issues can be particularly important in the atypical development domain, given the more variable spatial performance of the individuals concerned (Yang et al., 2014). The few studies on individuals with DS (Davis et al., 2014; Farran et al., 2015; Purser et al., 2015; see also Lavenex et al., 2015) found a role for both visuo-spatial reasoning [measured with Raven's colored progressive matrixes (CPM); Raven et al., 1998] and other cognitive abilities like executive control, attention and memory in participants' environment learning (especially in path reproduction). Visuo-spatial reasoning seems to have a fundamental role in path learning, in both TD and DS groups (Farran et al., 2015), but possibly even more so in the latter (Purser et al., 2015). These findings should be considered with caution because of the small sample sizes considered, but they do seem to suggest that several cognitive abilities – and visuo-spatial reasoning in particular – are involved in supporting path learning (Farran et al., 2015; Purser et al., 2015). Other visuo-spatial abilities, such as visuo-spatial WM, visualization and mental rotation, should be considered in individuals with DS too, as studies on TD populations have found them involved in environment learning (Fenner et al., 2000; Merrill et al., 2016).

Hence the second aim of the present study, which was to examine the role of a set of visuo-spatial cognitive abilities in supporting path learning by individual with DS.

Rationale and Aims of the Study

On the basis of the literature reviewed, the aims of the present study were to examine: a) the ability of individuals with DS to learn actual paths (in a vista space setting) of increasing length (in terms of the number of steps involved in a sequence), by comparison with matched TD children; and b) how path learning performance relates to visuo-spatial cognitive abilities and everyday spatial activities.

To address these aims, groups of individual with DS and TD children were presented with a large-scale (vista) task (inspired by Piccardi et al., 2014), in which cells were arranged in a uniform grid (a facilitating feature for individuals with DS; Carretti et al., 2013). The task was based on a span-like procedure, as used by Piccardi et al. (2014), to identify the longest sequence participants could learn (i.e., the range of their performance).

Since the way in which spatial information is presented can influence our mental representation of it (e.g., Picucci et al., 2013), two path learning conditions were presented: one involved learning from observation (Observation condition), in which participants watched a person move through a sequence of squares (as in Piccardi et al., 2014); in the other participants learned from a map (Map condition), showing the cells in the grid involved in the sequence of steps. This solution was chosen based on the evidence that children aged 3–5 years understand the representative function of maps in showing a correspondence with a larger space (e.g., Frick and Newcombe, 2012), although they have trouble with handling it flexibly (rotating it, for example; Vosmik and Presson, 2004). Preschoolers are able to use a map illustrating spatial information in a layout to address related spatial tasks involving a room (Bluestein and Acredolo, 1979; Shusterman et al., 2011) or larger spaces (Uttal and Wellman, 1989; Sandberg and Huttenlocher, 2001; Uttal et al., 2006). Uttal and Wellman (1989) found that 4 years old children shown a map indicating a path through the layout of a playhouse were better able to reproduce the path with actual moves than children not shown the map. Six years old children also proved capable of using a map at the same time as they actually moved in an environment, such as a hallway (Sandberg and Huttenlocher, 2001). These results suggest that preschoolers can integrate information shown on a map (allocentric view) while completing navigation tasks (egocentric view), in line with studies showing their ability to integrate allocentric and egocentric information (Nardini et al., 2008, 2009). This matter has been poorly investigated in individuals with DS, however, and the few studies on the effect of seeing a sketch map before learning a path through an environment found that individuals with DS did not benefit from the map in the same way as matched TD children (Toffalini et al., 2018; see also Meneghetti et al., 2017).

To address our second aim, the two groups were administered a series of visuo-spatial measures. Some assess basic processing ability, such as tasks measuring sequential and simultaneous aspects of visuo-spatial WM (i.e., the ability to manage increasingly long sequences and configurations of elements, respectively). The (small-scale) sequential WM task could be particularly relevant because the (large-scale) Floor Matrix task is based on sequences to learn, and the two types of task are related (Piccardi et al., 2014). Other measures were used to assess higher-level abilities, such as visualization and mental rotation (i.e., the ability to arrange and to rotate stimuli, respectively), that have been shown to influence path learning in TD children (Fenner et al., 2000; Merrill et al., 2016).

An everyday spatial activity questionnaire was also administered to assess the extent to which participants' Floor Matrix task performance was associated with their performance in everyday spatial activities.

We expected to find:

(a) that both the TD and the DS group would be able to complete the Floor Matrix task with sequences involving from 2 to 4 steps. Our assumption was based on: previous evidence of TD children of an age comparable with the mental age of DS individuals being able to reproduce sequences comprising 1.90–2.44 steps correctly in an irregular vista space setting (Piccardi et al., 2014); and on VE studies on individuals with DS showing that they were able to learn paths involving 2–4 segments (including turns and straight stretches; Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015; Purser et al., 2015; Toffalini et al., 2018). Differences between the groups in relation to learning condition were explored. If individuals with DS preferred the person's point of view for acquiring spatial information (as suggested by VE studies; e.g., Courbois et al., 2013; Farran et al., 2015), and had more difficulty learning the same information presented on a map (Meneghetti et al., 2017; Toffalini et al., 2018), we might expect the group with DS to perform better in the Observation than in the Map condition. On the other hand, if TD children are able to transfer information from a map to their personal point of view (expressed by their moves in an environment; e.g., Uttal and Wellman, 1989; Sandberg and Huttenlocher, 2001), they might perform the Floor Matrix tasks just as well in the Map condition as in the Observation condition. It might be that the TD children's performance is better in the Observation condition than in the Map condition, however. This would be due to their retaining a preference for egocentric representations (Siegel and White, 1975; Ruggiero et al., 2016);

(b) an involvement of (small-scale) visuo-spatial cognitive abilities in the performance of the Floor Matrix task in both groups, TD children and individuals with DS (Farran et al., 2015; Purser et al., 2015; Merrill et al., 2016). Differences in their involvement as a function of learning condition (Observation vs. Map) were explored in relation to the type of visuo-spatial tasks administered. Among several possibilities, visuo-spatial WM, and especially sequential WM could be involved in the Floor Matrix task, given that both rely on learning a sequence (Piccardi et al., 2015), an ability that individuals with DS also develop (Lanfranchi et al., 2004; Lanfranchi et al., 2009).

We also expected Floor Matrix task performance to be related with participants' everyday behavior, as suggested by the evidence of a link between vista space tasks and individuals' attitudes to moving in the environment (e.g., their self-assessed sense of direction, Mitolo et al., 2015), and their environment navigation difficulties (Palermo et al., 2014). Differences emerging between learning conditions and groups were examined in this respect.

MATERIALS AND METHODS

Participants

A group of 30 individuals with DS (11 females; M_{age} = 12.72 years; *SD* = 3.44; age range = 7.75–17.92 years), and a group of 30 TD children (11 females; M_{age} = 5.49 years; *SD* = 0.23; age range = 5.17–6.00 years) participated in the study. The two groups were similarly distributed by gender. As the measure for matching the two groups we chose to use the Peabody Picture Vocabulary Test-Revised (PPVT-R; Dunn and Dunn, 1981; Italian adaptation by Stella et al., 2000), a measure of receptive vocabulary (aware of the complexity of the issue concerning how to match groups for age equivalence in the presence of a population characterized by peaks and troughs, e.g., Jarrold and Brock, 2004). The TD group was selected from a larger group of 90 TD children aged 5–6 years old, which can be considered as the mean equivalent age, in terms of intellectual functioning, of individuals with DS from adolescence onward (Dykens et al., 2000). This age range is also considered adequate for a child to fully understand the necessary verbal instructions and perform the Floor Matrix task. The PPVT-R consists of a series of 175 pictorial stimuli of increasing difficulty, each comprising 4 black-and-white drawings. The respondent is asked to indicate which of the four drawings best represents the word an experimenter speaks aloud when presenting each stimulus. The task is terminated when the respondent makes six mistakes in eight consecutive responses. The final score is the total number of correctly chosen drawings. The DS group had an average PPVT-R score of $M = 67.2$ ($SD = 25.91$), corresponding to a mean equivalent age of 5 years, 9 months (Stella et al., 2000). The TD group had an average score of $M = 69.13$ ($SD = 15.62$), corresponding to a mean equivalent age of 5 years, 11 months. The between-group difference was negligible, Cohen's $d = 0.09$.

Material

Floor Matrix task (Adapted From Mitolo et al., 2015)

This task assesses path learning from actual movements in a controlled vista space setting. It consists of a 4 × 4 matrix on the floor comprising 16 squares (of stiff cardboard) 50 × 50 cm in size, with a 10 cm gap between them, forming a whole square layout about covering 2.30 × 2.30 meters. The layout of the Floor Matrix task is shown in **Figure 1A**. The Floor Matrix is aligned with the walls of the room to avoid a mismatch between the matrix (local) and room (global) spaces, which would affect performance (Shelton and McNamara, 2001; Lavenex et al., 2015). The task involves looking at sequences of positions presented consecutively, one square at a time, on the matrix and then reproducing them in the same order. The starting position is in one of the 4 squares in the bottom row of the matrix marked with an "X" (**Figure 1B**). There were 14 trials (two for each number of steps in a path, which ranged from 1 to 7).

Two learning conditions were considered: in one participants watched the examiner make a series of moves to complete the sequence (Observation), while in the other they looked at a map (Map condition). In the Observation condition the experimenter stood on the square ("X" marked), then completed a sequence of steps, stopping for 3 s in each square, while a participant stood outside the matrix and watched the experimenter's moves. The time spent on observing the sequence ranged from 3 to 21 s (for paths from 1 to 7 steps), plus the time taken to move from one square to the next (up to 1 s for each step). In the Map condition a participant was given 8 s to look at the matrix layout (a pilot study had suggested this was enough to memorize the path without participants' attention being distracted) on a sheet of paper (16 × 16 cm) reproducing the layout on the floor, with the starting square marked with an "X" and the sequence of steps indicated by squares linked together and highlighted with thicker edges (**Figure 1B**). During the learning phase, in both conditions, participants stood outside the matrix in front of the square marked with an "X," either observing the experimenter's movements or looking at a map.

Then they were asked immediately afterward to reproduce exactly the same sequence of steps, covering the same path by walking in the matrix. To ensure that participants understood what was required of them, the experimenter first explained the task by means of verbal instructions, then provided a direct demonstration. Two trials (with 2-step sequences) were used for each condition for familiarization purposes: if participants made a mistake, the experimenter demonstrated the right move and asked them to repeat it until they could complete the sequence correctly.

The paths involved consecutive squares that could go in one of three directions (forward or to right or left). All paths were randomly chosen so as not to feature any particular regular pattern. The task was terminated when a participant failed to reproduce both trials with the same number of steps correctly. It should be noted that the sequences in the Floor Matrix task involved moving from one square to another adjacent square, whereas the sequences to recall were not adjacent in the other visuo-spatial WM tasks (e.g., the Corsi Blocks task and the sequential WM task, see below). The final score for each condition was the participant's memory span, i.e., the maximum number of steps correctly reproduced by the participant in at least one of a pair of trials, and it ranged from 0 to 7.

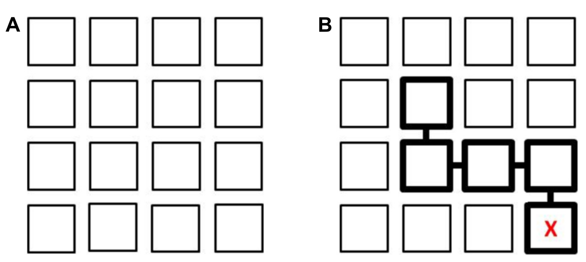

FIGURE 1 | Layout of the 4 × 4 Floor Matrix task **(A)**. Example of a sequence of 4 steps with the starting point identified by a "X" **(B)**. In the Map learning condition, this is shown to the participant; in the Observation learning condition, the experimenter stands in the square with the "X" and then moves to each square in the sequence. After each learning phase, participants stand on the square marked with an "X" and reproduce the four steps in the same order.

Visuo-Spatial Reasoning

Raven's Colored Progressive Matrices (CPM; Raven et al., 1998; Italian adaptation by Belacchi et al., 2008). This is a measure of fluid reasoning that uses items of a visuo-spatial nature. It consists of 36 increasingly complex colored matrices, and each matrix has a piece missing: the respondent is asked to choose the best fit for the missing piece from among six options. The reliability is good: the test-retest stability and convergent validity with other intelligence tests is strong in all international versions of the CPM, with r in 0.60–0.90 (Belacchi et al., 2008). The final score is the number of matrices correctly completed (and ranges between 0 and 36).

Visuo-Spatial Individual Measures

Ghost Picture Test (GPT; adapted from Frick et al., 2013). This is a measure of mental rotation ability. It consists of 21 items, each depicting a target silhouette of a ghost inside a circle on the top of the page, and two similar silhouettes underneath. The respondent has to choose which one is identical to the target figure in a rotated position (the alternative figure is a mirror image). The items require different degrees of rotation to match the target figure, i.e., 0° (3 items), 45° (3 items), 90° (7 items), 135° (4 items), and 180° (4 items). The internal consistency is good: Cronbach's alpha calculated on the matrix of the tetrachoric correlations (because the responses are of binomial type) on the current sample was 0.83. The final score is the total number of correct answers, and ranges from 0 to 21.

Primary Mental Abilities – Spatial relations – K1 (PMA-K1; Thurstone and Thurstone, 1949; Italian adaptation by Thurstone et al., 1981). This is a measure of spatial visualization ability. It consists of 12 incomplete target figures, each with four different pieces beneath it from which the respondent is asked to choose the one that completes the target figure. The internal consistency is good, the Italian adaptation of the test reportedly achieving an adjusted split-half correlation of $r = 0.81$ in preschoolers (Thurstone et al., 1981). The final score is the total number of correctly chosen pieces, and ranges between 0 and 12.

Working Memory Matrices – sequential and simultaneous – (Lanfranchi et al., 2004, 2015). These are two tasks respectively measuring the sequential and simultaneous aspects of visuo-spatial WM. Both tasks consist of a series of matrices presented on a sheet of paper. The matrices comprise cells measuring 3 cm each. Two trials are presented for each level of difficulty (i.e., the length of the sequence or the positions to learn range from 1 to 4).

In the sequential WM version, matrices of 3 × 3 and 4 × 4 square cells were used. The experimenter showed a path covered on the matrix by a small frog, which jumped onto cells in the matrix, stopping at each cell for 1 s showing sequences moving throughout the matrix, not necessarily in adjacent cells. Participants had to reproduce the sequences of jumps in the right order. In the simultaneous WM version, matrices comprising from 2 × 2 to 4 × 4 square cells were used. The experimenter showed participants the matrix with some cells colored in green and others left blank for 8 s, then showed them an all-blank matrix and asked them to remember the position of the green cells. In both conditions, participants had to respond immediately, and the task was terminated when they failed both trials on the same level of difficulty. In both tasks the final score corresponds to the number of trials completed correctly, and ranges from 0 to 8. The internal consistency is moderately good (0.59 for sequential WM and 0.89 for simultaneous WM, Lanfranchi et al., 2004).

Everyday Spatial Activity Questionnaire (ESAQ; Meneghetti et al., 2018)

This is a 6-item questionnaire examining an individual's ability to move around and reach locations out of doors (e.g., a school, a care center, a public park; e.g., "Can he/she move around the neighborhood unassisted?"), and indoors (e.g., in a classroom, a supermarket; e.g., "At the grocery store, can he/she go and get a product by moving along the aisles?"). It is completed by adults (the parents for the TD children, parents or educators for the individuals with DS), and scored on a 3-point Likert scale (from 1 = very poorly to 3 = very well). If the respondent feels the child shows no evidence of being able to do something, a score of 0 is also allowable. One participant in the DS group had to be excluded from the analysis concerning the ESAQ because some values were missing from the questionnaire. The internal consistency was acceptable: Cronbach's alpha was.77 (Meneghetti et al., 2018).

Procedure

Participants were tested individually during two sessions on two different days in the same week (for the participants' convenience). The first session (lasting around 30 min) was used to administer the Floor Matrix task. The matrix was set up on the floor of a room made available at the day center or school attended by participants. The rooms were similar in size (ranging 4–6 meters in length and width), and enabled the matrix to be aligned with the walls (doors and windows remained visible). The order of presentation of the learning conditions (Observation and Map) was balanced across participants. Each version of the task started with a familiarization phase (two trials) and the instructions emphasized the need for participants to pay careful attention to the sequence of steps shown by the experimenter's moves or on the map, and then reproduce the same sequence in the right order as best they could.

The second session (lasting around 40 min) was used to administer the measures of individual differences, which were counterbalanced across participants. The tasks were performed in a quiet room (different from the one used for the first session) at the day center or school, where a table and chairs were available. Specific instructions were given for each measure, making sure participants understood the task by practicing with examples before approaching it.

The Everyday Spatial Activity Questionnaire was delivered to parents or guardians after they consented to their child's participation in the research, and was completed and returned within 2 weeks.

RESULTS

A Bayesian approach was used for estimations and inferences, mainly because it enables evidence to be quantified taking the uncertainty due to factors not considered into account, including evidence in favor of the null hypothesis, where relevant (McElreath, 2016). The "BayesFactor" (Morey and Rouder, 2015) package and the "brms" package (Bürkner, 2018) of the R software were used for statistical estimation and model fitting.

Descriptive Statistics

The descriptive statistics (means and standard deviations) of all measures of interest are listed in **Table 1**, distinguishing between the two groups (DS vs. TD).

The standardized difference (Cohen's d) was used as a measure of the effect size of the between-group comparisons for all measures of interest. Cohen's d was calculated for each variable of interest using MCMC resampling with the "lmBF" function of the "BayesFactor" package in R. As a measure of uncertainty, 95% Bayesian credible intervals (BCI) were estimated using the percentile method on posterior distributions. In the Bayesian framework, a posterior distribution represents the probability distribution of an effect of interest (e.g., model

TABLE 1 | Descriptive statistics of individual measures for the two groups.

	Range of possible values	DS group (*N* = 30)		TD group (*N* = 30)	
		M	***SD***	***M***	***SD***
Peabody Picture Vocabulary task	0–167	67.20	25.91	69.13	15.62
Raven's Colored Progressive Matrices	0–36	14.17	5.00	18.80	4.12
Ghost Picture Test	0–21	11.40	3.86	15.87	1.98
Primary Mental Ability, Spatial – K1	0–12	5.30	2.52	7.77	1.85
Sequential working memory task	0–8	3.60	2.27	5.23	1.17
Simultaneous working memory task	0–8	2.77	2.18	4.97	1.35
Floor Matrix task, Map condition	0–7	3.00	1.70	3.53	0.82
Floor Matrix task, Observation condition	0–7	3.50	1.31	4.23	1.19
Everyday Spatial Activity Questionnaire†	0–18	9.10	3.92	12.23	2.64

DS, Down syndrome; TD, typically developing. †For the Everyday Spatial Activity Questionnaire, the DS group has N = 29.

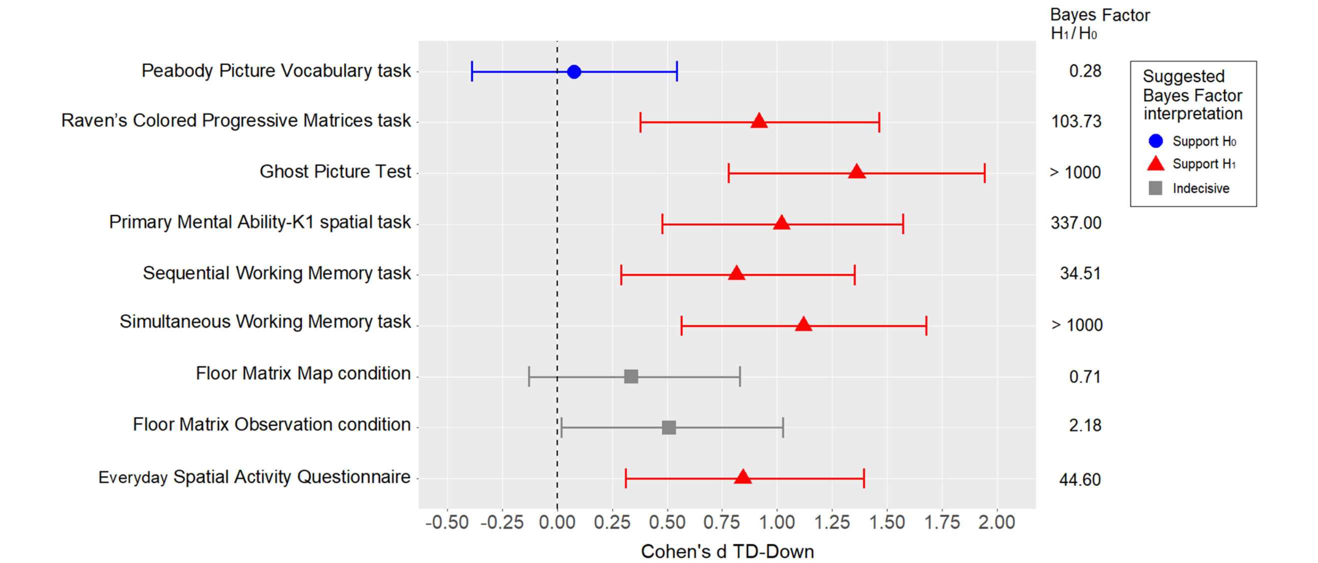

FIGURE 2 | Between-group comparisons using standardized differences (Cohen's d), with error bars representing 95% BCIs. The Bayes factor refers to *t*-tests.

parameter, standardized difference) after the data has been taken into account, and considering the *a priori* (prior) probability distribution. When objective default priors are used, as in the present case, the posterior distribution is determined solely by the data, and the 95% BCI tends to coincide with the 95% confidence interval reported using the frequentist framework. The standardized differences are shown in **Figure 2**. Apart from the PPVT-R matching measure, which obviously supports means equality, all the other measures were weaker in the group with DS than in the TD group, with medium to large standardized differences. Interestingly, the differences were smaller in the Floor Matrix task (in both conditions) than in the other visuo-spatial measures.

Given the relatively large number of variables and the relatively small sample size, statistical inference was not a relevant goal at this point. Nonetheless, to obtain an indication of the level of evidence, a t-test Bayes factor (BF) was calculated for each comparison (using the "BayesFactor" package in R). A weakly informed Cauchy prior with rscale = $\sqrt{2}/2$ was used for H_1 (set by default by the "ttestBF" function). Referring to Schönbrodt and Wagenmakers (2018), we interpreted a BF > 3 as at least "moderate" evidence of H_1 (i.e., the hypothesis that the two means are not equal at the population level), and a BF < 1/3 as "moderate" evidence of H_0 (i.e., the hypothesis that the two means are equal at the population level). Any BF coming between these two cutoffs was regarded as "indecisive" evidence. The BFs and their suggested interpretations are also shown in **Figure 2**.

Floor Matrix Task

Linear models were fitted on the Floor Matrix task scores, considered as the dependent variable, to examine the simultaneous roles of group (TD vs. DS), learning condition (Map vs. Observation), and their possible interactions. Because the data consisted of repeated measurements (in the two learning conditions) by participant, mixed-effects linear models were fitted, with random intercepts for the participants. The models were fitted using the "lmBF" function of the Bayes Factor package in R (Morey and Rouder, 2015), which allows for BFs

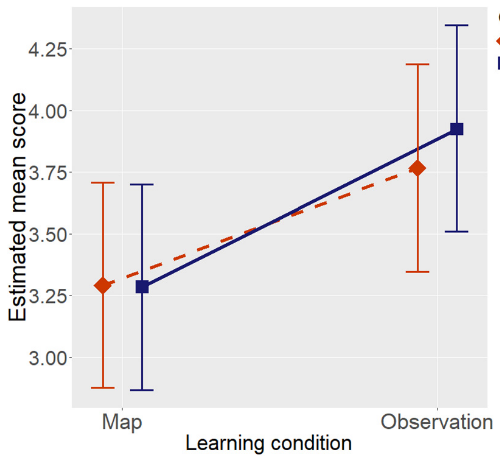

FIGURE 3 | Estimated mean scores in the Floor Matrix task by Group (DS, individuals with Down syndrome; TD, typically-developing children), and by Learning condition, after controlling for Raven's CPM score. Error bars represent 95% BCIs of the estimated means.

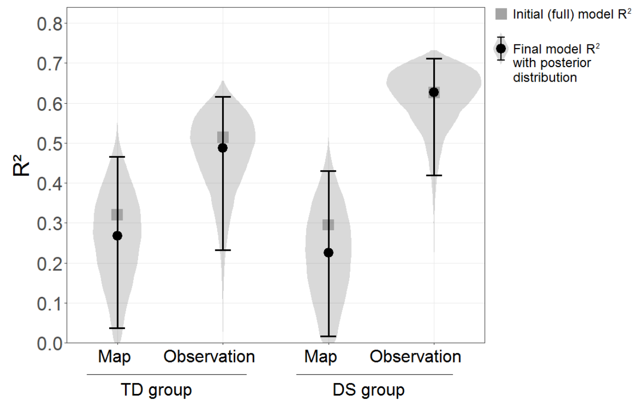

FIGURE 4 | R^2 of the initial models (squares) and final models (circles) with the Floor Matrix task scores as the dependent variables and visuo-spatial abilities as the predictors, estimated separately by Learning condition and Group. Error bars represent 95% BCIs. Violin plots represent posterior distributions of R^2 (i.e., probability distributions of the R^2s having certain values across Learning conditions and Groups).

to be computed by comparing the models with vs. without a given effect of interest. Default non-informed priors were used in all models.

Group showed a main effect, supported by weak evidence, BF = 2.11, when Raven's CPM was not entered as a covariate in the model; the effect size estimated from the mixed model was medium, with lower scores in the DS group than in the TD group (Cohen's d = -0.55). We opted to insert the effect of Raven's CPM in the model as a control variable, given that it is a general fluid measure capable of influencing environment learning in individuals with DS as well (Farran et al., 2015; Purser et al., 2015). After controlling for Raven's CPM the evidence supported no effect of group, BF = 0.30 (H_0 was suggested). There was a strong effect of the covariate Raven's CPM on the Floor Matrix task score, BF > 1000.00, so the two groups could be considered as not differing in terms of their scores in the Floor Matrix task once the role of non-verbal fluid reasoning had been taken into account. The learning condition had a main effect supported by fairly strong evidence, BF = 30.60, such that scores were higher in the Observation condition than in the Map condition (see **Figure 3**; Cohen's d = 0.58). There was evidence against an interaction between group and learning condition, BF = 0.31. **Figure 3** shows the estimated score in the Floor Matrix task as a function of group and learning condition: after controlling for Raven's CPM, the two groups' performance was much the same, and they both benefited equally from the Observation condition vis-à-vis the Map condition.

Relations Between Floor Matrix Task Performance and Individual Visuo-Spatial Measures

Additional analyses were run to quantify the relation between visuo-spatial abilities and everyday spatial activity using the Floor Matrix task. All correlations can be found in the online Supporting information (**Supplementary Table S1**).

Visuo-Spatial Abilities

The Floor Matrix task was treated as the dependent variable in the linear model, and scores in the GPT, PMA-K1, and sequential and simultaneous WM tasks were entered as independent variables. For ease of interpretation, the same model was computed separately for each learning condition and group. The "brm" function of the "brms" package in R was used, which fits Bayesian regression models using the MCMC algorithm implemented in the STAN programming language. Default non-informed priors were adopted for all models, using 4 Markov chains, with 10,000 iterations each, in each model. The outcome of interest was the explained variance, estimated as the model R^2. As shown in **Figure 4**, the R^2 was clearly higher for the DS group in the Observation condition, R^2 = 0.63, than in the Map condition, R^2 = 0.30. A similar pattern was seen for the TD group, R^2 = 0.51 in the Observation condition vs. R^2 = 0.32 in the Map condition. An evidence ratio was used for comparisons between R^2 in different conditions, calculated as the probability of the R^2 in one condition being superior to the R^2 in the other (this follows the logic underlying the "hypothesis" function of the "brms" package in R). Although there is no conventional cut-off for the evidence ratio, a value exceeding 39 could be interpreted as roughly equivalent to the amount of evidence given by $p < 0.05$ (two-tailed) in a frequentist framework, as it implies that more than 97.5% of the probability distribution is on one side of a given threshold, and less than 2.5% on the other (0.975/0.025 = 39). The evidence ratio was 134.14 in favor of the R^2 being higher in the Observation condition than in the Map condition in the group with DS, while in the TD group the evidence ratio for the same comparison was only 10.29.

In a second step, a selection procedure was adopted to avoid inflated R^2-values due to irrelevant predictors in the models. The same models as before were fitted, but removing one predictor at a time from each model for as long as this improved its fit. Model fit was assessed using the WAIC and the LOO indexes (Vehtari et al., 2017). The final models included the combinations of predictors that maximized model fit (WAIC and LOO led to the same final models). The final models included as predictors: only PMA-K1 for the TD group in the Map condition; PMA-K1 and simultaneous WM for the TD group in the Observation condition; only simultaneous WM for the DS group in the Map condition; and PMA-K1, and both sequential and simultaneous WM for the DS group in the Observation condition. The R^2 estimated were very similar to those reported above, and the difference between the Observation and Map conditions in the group with DS emerged even more clearly, with an evidence ratio = 306.69.

Figure 4 shows the R^2 of both the initial (full) models and the selected final models, along with the posterior distributions and the 95% BCIs of the R^2 for the final models.

Everyday Spatial Activity Questionnaire (Hetero-Assessment)

To better qualify the role of the Floor Matrix task as a measure capturing at least some aspects of everyday spatial activity, the Floor Matrix task scores in the two conditions (Observation and Map) were correlated with the results of the Everyday Spatial Activity Questionnaire (ESAQ).

In the group with DS, performance in the Floor Matrix task correlated strongly with the ESAQ in the Observation condition, $r = 0.43$, while the correlation was negligible in the Map condition, $r = 0.07$. In the TD group, the correlations were negligible in both conditions, $r = 0.15$ (Map) and $r = 0.00$ (Observation). Full details of the correlations between the ESAQ scores and all other variables considered in the present study can be found in **Supplementary Table S1**.

DISCUSSION OF THE RESULTS AND CONCLUSION

The aims of this study were to compare individuals with DS with matched TD children in terms of: (a) their ability to learn increasingly long sequences of steps from actual moves; and (b) how much this learning is supported by their visuo-spatial cognitive abilities and related to their everyday spatial activities.

Concerning the first aim, our results show that – in a vista space setting (with a 4 × 4 matrix of cells placed on the floor of a room) – individuals with DS could learn a path and reproduce it with a sequence of actual moves (turns and straight stretches) in the right order. The two learning conditions considered had a different impact on their performance, however, with the Observation condition proving easier than the Map condition. Intriguingly, this pattern was much the same in the group of TD controls. In fact, after controlling for visuo-spatial reasoning (given its impact on path learning; Farran et al., 2015; Purser et al., 2015), the most relevant result is the difference made by learning condition (in favor of Observation), whereas no group difference emerged.

In particular, in the Observation condition the mean number of steps in the sequences successfully reproduced was around 3–4 (the DS group learnt a mean 3.5 steps, the TD children a mean 4.23). This points to the number of steps learnt in a 4 × 4 floor matrix being higher (descriptively, at least) than when TD children of comparable mental age were administered the WalCT (when they learnt an average of 3 steps). This difference may be attributable to the fact that the squares in the matrix used in the WalCT are placed irregularly on the floor (Piccardi et al., 2014), whereas in our Floor Matrix task they formed a uniform 4 × 4 grid. The number of steps in the sequences learnt by our participants seem more similar to the findings in VE studies in which individuals with DS proved capable of learning and reproducing paths 4 segments long (Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015; Purser et al., 2015; Toffalini et al., 2018), although some of these studies envisaged repeatedly tracing the path until all or most of the segments had been reproduced correctly, not just once as in the present study.

In the Map condition, on the other hand, both of our groups were less successful in reproducing the path: the group with DS learnt a mean 3 steps, children with TD a mean 3.53 steps. As hypothesized for individuals with DS, these results indicate a greater difficulty of using map-based information (simultaneously presenting the whole grid layout on which the path is marked, on a sheet of paper 16 × 16 cm in size) to learn sequences and reproduce them with actual moves in a corresponding grid on the floor (2.3 × 2.3 meters in size). This is consistent with earlier evidence of individuals with DS not benefiting from seeing a map before exploring an environment from a personal point of view (Meneghetti et al., 2017; Toffalini et al., 2018), and their difficulty with applying allocentric information to their actual movements (Lavenex et al., 2015). This difficulty was surprisingly found to apply to TD children too, whereas they might have been expected to benefit from seeing a map before exploring an environment – in the light of previous evidence obtained in preschoolers – (Uttal and Wellman, 1989; Sandberg and Huttenlocher, 2001). Studies on navigation in TD children have differed in some ways, however. For instance, the space tested was limited in the present study (2.30 × 2.30 m in all), whereas previous studies tested children navigating in larger spaces (such as a series of rooms in Uttal and Wellman, 1989; or hallways in Sandberg and Huttenlocher, 2001). Larger spaces can be more useful for detecting the integration of allocentric information (such as room layouts, or walls) with egocentric information (experienced during navigation). In this sense, how TD children benefit from preserving the person's point of view in learning sequences (indicating the prevalence of egocentric representations) warrants further investigation, because there is evidence in the literature of children 5–6 years old being able to use allocentric information to manage their movements (Nardini et al., 2008, 2009; Ruggiero et al., 2016).

Although these results for Floor Matrix task performance are encouraging, there are some limitations to consider relating to the method used. For a start, the rooms where the matrix was set up contained elements outside the matrix that remained visible to participants, such as doors and windows. This ensured that the task was performed in a "natural" setting, but also gave participants the chance to rely on external reference points as part of their spatial representation, and this would have influenced its final features (e.g., Pennington et al., 2003; Purser et al., 2015). Second, the time of presentation varied in the Observation condition, increasing with the length of the sequence to be remembered, whereas it remained the same in the Map condition (8 s). This difference (involving a generally longer time of presentation in the former condition, for sequences of more than 3 steps at least) could affect performance, and may explain why it was generally better in the Observation condition. It is worth adding that, in a preliminary pilot study, a time of presentation longer than 8 s in the Map condition did not seem beneficial, as it only led to participants' attention wandering. These methodological aspects need to be carefully considered in further studies.

As for the second aim of our study, to clarify the involvement of visuo-spatial factors in Floor Matrix task performance, our results show how the contribution of individual abilities changed as a function of learning condition (Observation or Map) and group (TD or DS). It is important to note that the individuals with DS were matched with TD children on a verbal measure (receptive vocabulary), but were still weaker than the latter on a series of visual-spatial tasks, both basic sequential and simultaneous WM tasks, and higher-level mental rotation and visualization tasks. These results are not in contrast with the findings of the review by Yang et al. (2014). Individuals with DS performed less well than TD children matched for cognitive functioning (where studies in the review also reported matching them on the PPVT-R) in tasks measuring closure, like our Primary Mental Ability (Spatial – K1) task, which involved identifying the part of a figure needed to complete it. Yang et al. (2014) also reported inconsistent evidence regarding mental rotation, and our results are in line with studies showing a poor performance using a task based on the detection of rotated figures (as in the Ghost Picture Test; in Meneghetti et al., 2018). We also confirmed the poor performance of individuals with DS in simultaneous WM tasks (Carretti and Lanfranchi, 2010), and found that they had difficulty with a sequential WM task as well. This latter result differs from the findings of previous studies (e.g., Lanfranchi et al., 2004), and will need to be confirmed or refuted in future. Overall, the present findings support the assumption that visuo-spatial abilities generally are not a relative strength in individuals with DS (Yang et al., 2014), but this depends on the type of ability tested and the type of measure used. They certainly warrant further investigation in this population.

That said, visuo-spatial abilities influenced Floor Matrix task accuracy in both the individuals with DS and the TD children, to a different degree in the two learning conditions, from Observation or a Map. Judging from our results, the DS group's visual-spatial abilities (particularly visualization, and sequential and simultaneous WM) were more heavily involved when they learnt a path from direct observation than when they saw a map (only simultaneous WM is involved in the latter case). The same pattern was seen in TD children, but it was weaker (less variance was explained by the model in the Observation condition): visualization and simultaneous WM were especially involved in the Observation condition; and visualization in the Map condition. These results must be considered with caution, however, due to the relatively small sample size of both groups. More specifically, the model selection procedure used to define the "final" best-fitting models or set of predictors should be considered only as an exploratory analysis.

These results prompt some considerations. In the easier learning condition (Observation), visuo-spatial abilities clearly emerged to ensure success in recalling the path, particularly in the group with DS. The contribution of visuo-spatial abilities in the Map condition was less relevant in this group. The same trend was probably at work in the TD group, but with a weaker contribution of their visuo-spatial abilities. Considering the contribution of specific visual-spatial abilities, it seems that a role for visuo-spatial WM (a basic ability) is more detectable in individuals with DS, while the role of visualization (a higher-level ability) seems more apparent in TD children. It is worth noting that the role of sequential WM emerged in the Observation condition for individuals with DS, as expected, but in combination with simultaneous WM (probably due to the sharing of WM processing resources). In the DS population, visuo-spatial WM seems to be a core process in their execution of such a complex cognitive task as path learning. While a higher-level spatial ability like visualization (i.e., the ability to arrange and manage the shapes of objects) seemed relevant in the TD children, this was not the case for mental rotation (unlike previous findings in TD children; Merrill et al., 2016). Other researchers found egocentrically-based abilities (such as the one needed to imagine yourself in different positions in space) related to path learning in TD children (Nazareth et al., 2018).

This seemingly stronger contribution of visuo-spatial abilities in DS than in TD individuals can be explained by the fact that, despite generally weaker visuo-spatial abilities in the DS group than in the TD group (as reported above), some individuals with DS have well-developed visuo-spatial skills. In fact, the DS group showed a greater heterogeneity in its performance, also as regards visuo-spatial skills (see **Table 1**). There is therefore more room for some individuals with DS – those whose visuo-spatial abilities were relatively strong – to dedicate these resources to underpinning their performance in the Floor Matrix task (especially in the Observation condition, which is generally more manageable for them), whereas those whose underlying abilities are more severely impaired would be unable to do so. This has to do with the question of cognitive profile variability within the same population. There are studies suggesting that the classical profile of individuals with DS does not always apply, and that individual differences in this population can be even twice as great as in the TD population (e.g., Tsao and Kindelberger, 2009; Karmiloff-Smith et al., 2016).

These results confirm the importance of taking the role of individual cognitive abilities into account when examining environment learning in individuals with DS as well (Farran et al., 2015; Purser et al., 2015). At the same time, they offer insight on how to explore the role of visuo-spatial abilities in relation to the variability of task performance in a given population to gain a better picture before drawing any definitive conclusions.

Finally, examining the relations between our participants' path learning and hetero-assessed everyday life spatial activity (ESAQ) suggested quite a strong association, particularly in the group with DS. This applied especially to this group's path learning from Observation ($r = 0.43$), rather than from a Map. In the TD group the correlations between Floor Matrix task performance (in both the Map and Observation learning conditions) and the EASQ were negligible. This result supports the use of the Floor Matrix task in individuals with DS to capture aspects of their everyday navigation ability, such as outdoor movements to reach places (as previously suggested by Mitolo et al., 2015). The absence of any relation between Floor Matrix task performance and everyday life spatial activity in the TD group is plausible because 5 and 6 years old children (like those in our TD group) are not required or allowed to go around in the outside world alone (to go to school or visit other parts of their neighborhood). Their parents' ratings were probably higher than for the DS group because the activities mentioned in the ESAQ were judged as something the children were capable of doing (rather than something they actually did), and there was little or no association between these ratings and the TD children's Floor Matrix task performance. The adults' ratings of the individuals with DS are more likely to have captured their real abilities because these individuals were older (from 7.75 to 17.92 years of age), and the older ones would have actual experience of the movements considered. This type of result offers insight on the relationship between everyday experiences of navigation (when hetero-assessed, at least) and an actual navigation task in a controlled setting (as in the Floor Matrix task) in individuals with DS, a relationship that deserves to be better explored. Although these results support the use of the Floor Matrix task to assess large-scale navigation ability with actual moves in a vista space, it would be even better to employ more ecological navigation conditions (such as actual movements in the neighborhood, or to reach a given room in a building) in this population (Yang et al., 2018).

Though further research is certainly needed on the role of small-scale (spatial cognitive) abilities in successful path learning, our results support the spatial cognition model postulating a relationship between small- and large-scale abilities not only in young adults (Hegarty et al., 2006), and TD children (Merrill et al., 2016), but also in cases of atypical development. This relationship can be demonstrated using VE (Farran et al., 2015), and also – as our study newly showed – using actual movements in the environment. Such findings are important not only for the purpose of extending the theoretical framework to cover different populations but also for their various implications. One such implication may be particularly relevant to individuals with DS, for the purpose of training their cognitive abilities (such as visuo-spatial WM) in order to improve other, related cognitive skills (such as spatial learning), or directly practicing with learning from navigation in controlled settings (using the Floor Matrix task, for instance), and analyzing its impact on everyday navigation ability. This issue has yet to be approached directly, but promising evidence has emerged of individuals with DS benefiting from visuo-spatial WM training (Lanfranchi et al., 2017), and future studies can be designed to examine more closely how their navigation abilities might be improved.

Overall, although the results of the present study need to be confirmed, they shed new light on the path learning ability of individuals with DS. They show that: (a) individuals with DS can learn increasingly long sequences of steps in a vista space setting (as in the Floor Matrix task) almost as well as matched TD children, though it seems easier for them to learn from watching a person actually make the moves rather than from looking at a map; and (b) visuo-spatial cognitive abilities are important in supporting path learning accuracy, especially when learning from observing other people's moves, with visuo-spatial WM seeming particularly relevant in individuals with DS, and visualization ability in TD children. In short, our findings show that individuals with DS are able to learn sequences of steps forming a path from actual moves, and their accuracy in reproducing the path is supported by their individual visuo-spatial abilities.

DATA AVAILABILITY STATEMENT

The dataset analyzed for this study can be found on Figshare, doi: 10.6084/m9.figshare.10055438.

ETHICS STATEMENT

The study was approved by the Ethics Committee for Research in Psychology (University of Padua), Number: 2EDFB5F0133B6675FE88D0E3F714B17C. Prior written consent for the children's participation was obtained from both their parents (or the legal representatives of the individuals with DS).

AUTHOR CONTRIBUTIONS

ET organized the database and performed the statistical analysis. CM wrote the first draft of the manuscript. BC, SL, and ET wrote sections of the manuscript. All authors contributed conception and design of the study, manuscript revision, read and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2020.00107/full#supplementary-material

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BRIEF RESEARCH REPORT
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Path Integration Changes as a Cognitive Marker for Vascular Cognitive Impairment?—A Pilot Study

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Path integration spatial navigation processes are emerging as promising cognitive markers for prodromal and clinical Alzheimer's disease (AD). However, such path integration changes have been less explored in Vascular Cognitive Impairment (VCI), despite neurovascular change being a major contributing factor to dementia and potentially AD. In particular, the sensitivity and specificity of path integration impairments in VCI compared to AD is unclear. In the current pilot study, we explore path integration performance in early-stage AD and VCI patient groups and hypothesize that: (i) medial parietal mediated egocentric processes will be more affected in VCI; and (ii) medial temporal mediated allocentric processes will be more affected in AD. This cross-sectional study included early-stage VCI patients (n = 9), AD patients (n = 10) and healthy age-matched controls (n = 20). All participants underwent extensive neuropsychological testing, as well as spatial navigation testing. The spatial navigation tests included the virtual reality "Supermarket" task assessing egocentric (body-based) and allocentric (map-based) navigation as well as the "Clock Orientation" test assessing egocentric and path integration processes. Results showed that egocentric integration processes are only impaired in VCI, potentially distinguishing it from AD. However, in contrast to our prediction, allocentric integration was not more impaired in AD compared to VCI. These preliminary findings suggest limited specificity of allocentric integration deficits between VCI and AD. By contrast, egocentric path integration deficits emerge as more specific to VCI, potentially allowing for more specific diagnostic and treatment outcome measures for vascular impairment in dementia.

Keywords: navigation, egocentric, virtual-reality, dementia, VCI, vascular cognitive impairment, vascular-dementia

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INTRODUCTION

Vascular cognitive impairment (VCI) is the second most prevalent cause of cognitive decline after Alzheimer's disease (AD) and is thought to account for ∼20% of all dementias (Goodman et al., 2017; van der Flier et al., 2018). Although, individuals with mixed (AD and VCI) pathology are estimated to account for up to 70% of all dementia cases (Toledo et al., 2013). Despite the high prevalence of vascular impairment, its cognitive correlates are still being explored. Clinically, VCI is considered to involve a decline in executive function and

Abbreviations: VCI, vascular cognitive impairment; AD, Alzheimer's disease.

higher-order cognition such as information processing, planning, set-shifting and working memory (Hachinski et al., 2006; Sachdev et al., 2014). These changes are mostly attributed to micro and macro infarcts in subcortical and cortical regions, as well as their connecting white matter tracts (Beason-Held et al., 2012; van der Flier et al., 2018), in particular affecting frontoparietal networks. Nevertheless, attributing such executive changes to VCI specifically has remained challenging, as deficits in executive function can also present as part of AD or related pathophysiology (Neufang et al., 2011; Girard et al., 2013; Guarino et al., 2018). However, the recent development of novel spatial navigation cognitive markers for AD show promise in being more specific to underlying disease pathophysiology (Coughlan et al., 2018b) and may help to identify cognitive decline specific to VCI. A clear distinction between VCI and AD is essential to both clinicians and patients as with appropriate intervention VCI can be slowed or halted, whereas AD has a fixed and terminal prognosis.

Spatial navigation is a fundamental cognitive skill that requires the integration of egocentric (body-based) and allocentric (map-based) frames of orientation. Both frames are required for everyday navigation with egocentric and allocentric processes shifting as a function of navigational demands (McNaughton et al., 2006). Path integration is integral to spatial navigation as it allows an individual to keep track of and return to their starting location based on visual, self-motion, vestibular and proprioceptive feedback which represent the current position and heading direction in references to a permanent location (Etienne and Jeffery, 2004: McNaughton et al., 2006; Knierim et al., 2014). This process involves translating distance traveled with changes in direction of movement either relative to our allocentric or egocentric orientation (Burgess, 2006). Multisensory (visual, self-motion, vestibular and proprioceptive) feedback combine egocentric and allocentric frames of reference, allowing path integration to continuously update this information, allowing one to keep track of one's position in space (Rieser, 1989; Coughlan et al., 2018b).

Egocentric orientation relies more on the prefrontal and parietal cortex to localize the position of objects relative to the body (Goodale and Milner, 1992; Arnold et al., 2014), the precuneus then uses these location cues to form the basis of an egocentric representation of the surrounding space, integrating self-motion cues with the egocentric reference frame (Wolbers and Wiener, 2014). While allocentric orientation is reliant on the formation of maps using place, grid and boundary vector cells situated mainly in the medial temporal lobe (Lester et al., 2017; Coughlan et al., 2018b). The integration of egocentric and allocentric frames occurs in the retrosplenial cortex (RSC), which is a critical interface between the medial temporal and medial parietal regions (Alexander and Nitz, 2015). Dorsal-medial regions of the RSC are thought to be implicated in orientating and recalling unseen locations from a current position in space, whilst ventrolateral portions were more linked to updating and integrating scene information (Burles et al., 2017).

Tasks that tap into path integration, therefore, provide a promising ecological, cognitive framework to detect medial temporal and medial parietal pathophysiology. Not surprisingly, path integration has been already explored in AD (Morganti et al., 2013; Serino et al., 2014; Vlček and Laczó, 2014; Ritchie et al., 2018) and the advent of VR based testing has allowed such tests to be clinically available (Plancher et al., 2012; Morganti et al., 2013; Parizkova et al., 2018). We have developed previously such a test, the Virtual Supermarket task, which is now used across many large cohorts and drug trials as it can reliably detect path integration differences in preclinical and clinical dementia populations (Tu et al., 2015, 2017). The VR task reliably measures spatial processes of: (i) egocentric self-reference navigation; (ii) allocentric map-based navigation; and (iii) heading direction. For example, we have previously shown that the test allows the distinction of behavioral variant fronto-temporal dementia (bvFTD) from AD, with AD showing particularly problems in switching between egocentric and allocentric frames during path integration (Tu et al., 2017). Importantly, these switching problems in AD were associated with grey matter atrophy in the RSC (Tu et al., 2015).

In contrast to the exciting findings in AD, less is known about path integration in VCI, despite path integration potentially allowing as well to tap into parietal deficits in VCI (Maguire et al., 1998; Wolbers et al., 2004; Papma et al., 2012; Haight et al., 2015). A previous case study by our group explored path integration in a 65-year-old male with VCI. The findings showed that the vascular patient had normal performance on allocentric orientation but a clear and isolated deficit in egocentric and heading direction sub-components of the path integration tasks (Coughlan et al., 2018a). These findings are consistent with frontoparietal network disruptions typically seen in vascular dementia patients (Beason-Held et al., 2012; Sachdev et al., 2014; van der Flier et al., 2018) and may suggest medial parietal changes imped the egocentric frame of reference and subsequent path integration.

The current study leads on from this case study by exploring path integration in a group of VCI patients and importantly comparing them against a group of AD patients and controls. Navigation will be tested using the Virtual Supermarket task where participants move through the virtual environment to a series of locations and are tested on their egocentric, allocentric and heading direction response. We hypothesize that: (i) medial parietal mediated egocentric processes will be more affected in VCI; and (ii) medial temporal mediated allocentric processes will be more affected in AD.

MATERIALS AND METHODS

Participants

Nine early-stage vascular cognitive impairment and 10 early-stage Alzheimer's disease patients along with 20 healthy controls were recruited from the community using "Join Dementia Research" to participate in the study at the University of East Anglia as part of the wider The Dementia Research and Care Clinic (TRACC) study. The research was approved by the Faculty of Medicine and Health Sciences Ethics Committee at the University of East Anglia (reference 16/LO/1366) and written informed consent was obtained from all participants. Clinical diagnosis (VCI or AD) was classified by a consultant at the Norfolk and Suffolk Foundation Trust by interviewing the patient, examining neuropsychological assessment scores, structural clinical MRI scans, and the patient's medical history which met the diagnostic criteria for VCI (see Sachdev et al., 2014) or AD (see Dubois et al., 2007). For clarity, the structural MRI profile of VCI was indicated by subcortical infarcts and white matter hyperintensities, whilst volume loss focused on medial temporal lobes was associated with AD pathology. Disease duration was reported by the person's study partner (a spouse or relative). Participants had no history of psychiatric or neurological disease, substance dependence disorder or traumatic brain injury and had normal or corrected-to-normal vision. None of the patient's study partners in this experiment reported problems with spatial orientation before dementia onset or a history of developmental topographical disorientation (Iaria et al., 2009). All participants underwent neuropsychological screening, including cognitive screening, episodic memory and spatial memory tasks, Addenbrooke's cognitive examination (ACE-III; Hsieh et al., 2013), Rey–Osterrieth Complex Figure Test (RCFT) copy and with 3-min delayed recall (Lezak, 1983), Cube Analysis, Dot Counting and Position Discrimination from the Visual Object and Space Perception Battery (VOSP; Warrington and James, 1991), Free and Cued Selective Reminding Test (FCSRT; Buschke, 1984).

Virtual Supermarket Task

The Virtual Supermarket Task has been developed by our group previously and used in symptomatic mild cognitive impairment (MCI), AD, frontotemporal dementia (FTD) and VCI patients (Tu et al., 2015, 2017; Coughlan et al., 2018a). The VR task is an ecological test of spatial navigation abilities designed to simulate navigating through a real-world supermarket. An iPad 9.7 (Apple Inc.,) was used to show participants 20–40 s video clips of a moving shopping trolley in the virtual supermarket (**Figures 1A–C**). Videos were presented in a first-person perspective and participants are provided with optic flow cues from the moving shopping trolley and changing scenery as they followed different routes to reach a different endpoint in each trial. The task avoids the use of landmarks or salient features within the environment and limits the demand on episodic memory, reflecting similar tasks in the literature (see Cushman et al., 2008; Morganti et al., 2013; Wolbers et al., 2007) and taps into path integration processes *via* three core spatial processes: (i) egocentric self-reference navigation; (ii) allocentric map-based navigation; and (iii) heading direction. Once the video clip stops, participants indicate in real-life the direction of their starting point (egocentric orientation; **Figure 1D**). In a second step, participants indicate their finishing location on a birds-eye view map of the supermarket (allocentric orientation; **Figure 1E**), performance is calculated using the distance error (mm) between this and the coordinates of the actual finishing location. This map-based component provides an assessment of the geocentric encoding of the virtual environment. The participant then indicates their heading direction at the finishing point, which determines the ability to which heading direction was encoded and updated throughout the task. The tasks consist of 14 trials and takes approximately 15 min to complete.

Clock Orientation Test

The Clock Orientation test has also been developed by our lab (Coughlan et al., 2018a) as a bedside clinical test for egocentric orientation. It requires participants to imagine they are standing in the center of a large clock, facing a particular number, e.g., the number 3. Participants are then asked, "Which number is directly behind you?" (Answer: number 9). Next participants are asked to point, in real life, to the positions of different numbers on the clock face in relation to the number that they are currently facing. For example, "You are facing number 12, can you point to the number 3?" (Answer: pointing right). The questions increase in complexity across the test and require medial parietal mediated mental imagery, rotation, and egocentric processes, with no episodic memory demand. The test consists of 12 trials and takes 5–10 min to complete.

Procedure

Participants completed a battery of neuropsychological assessments at their homes (see **Table 1** for a list of tasks). In a second session held at the Norfolk and Suffolk Foundation Trust, participants undertook cognitive experimental tests (including the Virtual Supermarket task and Clock Orientation test) and completed a clinical interview with the Chief Investigator of the study.

Statistical Analysis

Statistical analysis was performed using IBM SPSS (Version 25). Chi-square and two-tailed one-way univariate analysis of covariance (ANCOVA) with age and sex as covariates were used to test the significance of any demographic or neuropsychological differences between the clinical groups. When quantifying group differences, partial eta squared (η_p^2) was used as a measure of effect size. The Virtual Supermarket task has three measures - specifically egocentric response, allocentric response and heading direction. Each outcome measure was individually entered into a one-way ANCOVA with group as the independent variable and age and sex as covariates. Although groups were well matched for age and sex, these covariates were decided as evidence suggests they can affect navigational behavior (Coutrot et al., 2018). The Clock Orientation test was also analyzed using a one-way ANCOVA with group as the independent variable and age and sex as covariates. *Post hoc* pairwise comparisons were conducted using Bonferroni adjustment for multiple comparisons. The sensitivity and specificity of the egocentric supermarket task and clock orientation test performance in VCI and AD were compared using logistic regression and ROC curve analysis. A Z-score of AD performance was computed for seven missing values for one AD patient in the Virtual Supermarket task.

RESULTS

Demographics and Neuropsychology

Participant groups were well matched and no significant differences in demographic measures were observed between the VCI, AD and control groups (all p-values > 0.1). ANOVA of participant groups showed both VCI and AD patients performed significantly lower on a general cognitive screening

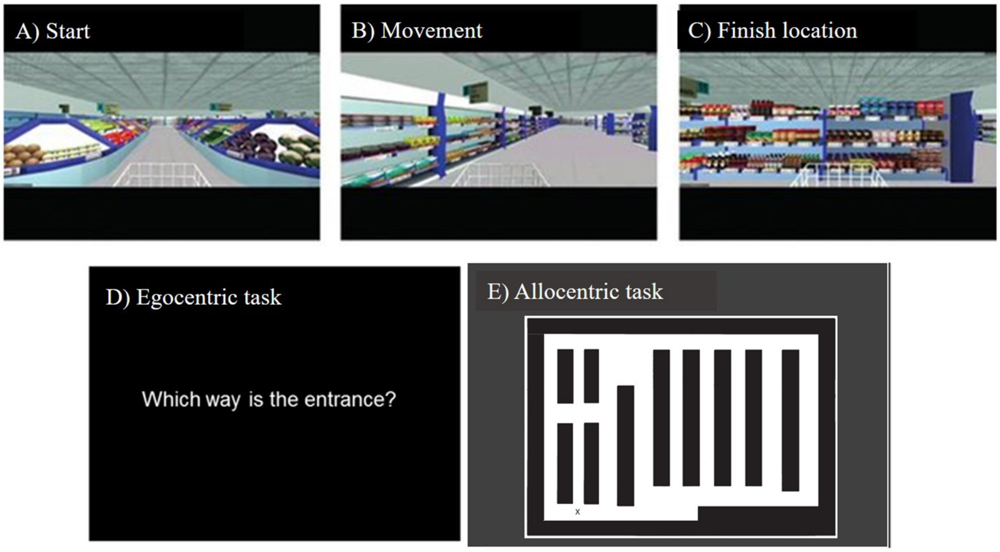

FIGURE 1 | Screenshots from the Virtual Supermarket task, showing **(A)** starting viewpoint, **(B)** movement during the example video clip, **(C)** end location of an example video clip, **(D)** onscreen instructions prompting the participant to indicate the direction of their starting point, **(E)** the supermarket map participants use to indicate their finishing location and their heading direction when the video clip ends.

TABLE 1 | Demographic characteristics and neuropsychological performance.

	VCI *Mean (SD)*	AD *Mean (SD)*	Control *Mean (SD)*	*Sig post hoc VCI vs. AD comparisons*
n	9	10	20	
Sex (F/M)	3/6	2/8	9/11	ns
Age	70.22 (4.57)	69.91 (7.7)	69.6 (6.45)	ns
Disease duration	3.13 (2.64)	2.81 (2.21)	n/a	ns
General cognition				
Total ACE-III	69.44 (12.9)	72.1 (22.41)	95.1 (3.13)	ns
ACE: attention	13.5 (0.72)	15.75 (0.72)	17.6 (0.45)	ns
ACE: memory	13.5 (1.73)	17.13 (1.17)	24.3 (0.74)	ns
ACE: fluency	7.13 (0.59)	8.12 (0.59)	11.7 (0.37)	ns
ACE: language	21.77 (2.44)	22.33 (3.04)	25.6 (0.61)	ns
ACE: visuospatial	11.5 (1.19)	16.67 (1.12)	15.8 (0.75)	*
Visuospatial ability				
RCFT: copy	22.1 (7.17)	28.4 (8.92)	32.72 (3.23)	*
RCFT: recall	7 (5.65)	11.8 (8.12)	17.55 (5.43)	ns
Dot counting	9.5 (0.71)	9.8 (0.42)	10 (0)	ns
Position discrim	18.87 (1.27)	19.7 (0.67)	19.85 (0.37)	*
Cube analysis	8.11 (2.62)	8.7 (1.88)	9.8 (0.52)	ns
Memory ability				
Total FCSRT	29.21 (2.84)	42.91 (2.63)	47.92 (2.01)	**
FCSRT: free recall	8.83 (7.94)	17.14 (8.83)	26.83 (4.17)	ns
FCSRT: cued recall	25.7 (4.94)	20.5 (7.2)	23.35 (4.87)	ns
Supermarket task				
Egocentric	3.44 (3.24)	9.4 (2.27)	8.1 (3.7)	**
Allocentric	69.1 (38.11)	48.41 (12.17)	30.2 (14.13)	ns
Head direction	4.8 (1.33)	5 (3.41)	7.1 (0.9)	ns
Clock test	5.43 (0.81)	10.1 (1.2)	10.1 (0.51)	***

**Significant group differences between VCI and AD patients. *p < 0.1, **p < 0.01, ***p < 0.001, ns = non significant. ACE-III = Addenbrooke's cognitive examination. RCFT: Copy = Rey-Osterrieth Complex Figure Task, copy condition. RCFT: Recall = Rey-Osterrieth Complex Figure Task, recall 3 min after copy. Dot Counting, Position Discrimination, and Cube Analysis = sub-sets from Visual Object and Space Perception Battery (VOSP). FCSRT: free recall = Free and Cued Selective Reminding Test, free recall Test condition, FCSRT: free recall = Cued and Cued Selective Reminding Test, cued condition.*

test (ACE-III) and the memory recall domain of RCFT compared to controls (all p-values < 0.01). Results showed no significant neuropsychological differences between the VCI and AD patients for the ACE-III, RCFT recall condition, VOSP dot counting, and cube analysis sub-sets (all p-values > 0.1. However, VCI patients were significantly more impaired than AD patients in

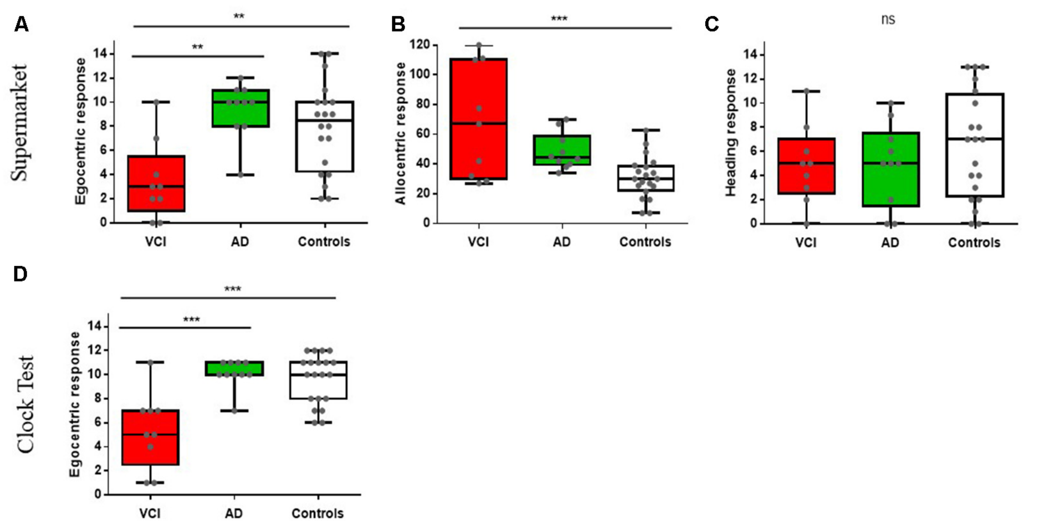

FIGURE 2 | Spatial orientation performance between vascular cognitive impairment (VCI), Alzheimer's disease (AD), and controls. $^{**}p < 0.01$, $^{***}p < 0.001$, ns = non significant. Figures **(A–C)** show The Virtual Supermarket task performance; **(A)** egocentric response (correct), **(B)** allocentric response (error in mm) and **(C)** heading response (correct). Figure **(D)** displays The Clock Orientation test egocentric response (correct).

the RCFT copy condition, FCSRT free recall condition and the VOSP position discrimination (all *p*-values < 0.1; see **Table 1**).

Virtual Supermarket Task

An ANCOVA with age and sex as covariates revealed a significant differences between egocentric responses on the supermarket task, $F_{(2,34)} = 8.14$, $p < 0.001$, $\eta_p^2 = 0.32$. *Post hoc* comparisons revealed significantly greater egocentric impairment in VCI ($M = 3.5$, SD = 3.24) compared to AD ($M = 10.01$, SE = 1.11), $p < 0.002$, 95% CI (−10, −2.1) and control groups ($M = 8.1$, SD = 3.7), $p < 0.009$, 95% CI (−7.95, −1.1). No other significant group differences were observed ($p > 0.1$; see **Figure 2A**).

Allocentric responses showed a significance difference between groups, controlled for age and sex $F_{(2,34)} = 10.1$, $p < 0.001$, $\eta_p^2 = 0.37$. *Post hoc* comparisons showed significantly greater impairments in VCI patients ($M = 68.33$, SD = 38.1) compared to controls ($M = 30.85$, SD = 14.13), $p < 0.001$, 95% CI (16.02, 61.1) but impairments did not reach statistical significance in AD patients ($M = 50.1$, SD = 7), $p = 0.09$, 95% CI (−41.11, 2.1) compared to controls. However, there were no significant groups differences between VCI and AD ($p > 0.1$; see **Figure 2B**).

Heading direction (correct judgement of facing direction after travel period) did not reveal significant group differences when controlling for age and sex $F_{(2,34)} = 1.11$, $p > 0.1$, $\eta_p^2 = 0.06$ (see **Figure 2C**).

Clock Orientation Test

An ANCOVA with age and sex as covariates revealed a significant difference between egocentric responses on the Clock Orientation task $F_{(2,34)} = 13.4$, $p < 0.001$, $\eta_p^2 = 0.44$. *Post hoc* comparisons showed significantly greater egocentric deficits in VCI patients ($M = 5.42$, SD = 3.16) compared to AD ($M = 10.1$, SD = 1.21), $p < 0.001$, 95% CI (−7.2, −2) and control groups ($M = 9.65$, SD = 2.06), $p < 0.001$, 95% CI (−6.56, −7.1). No other significant group differences were observed ($p > 0.1$; see **Figure 2D**).

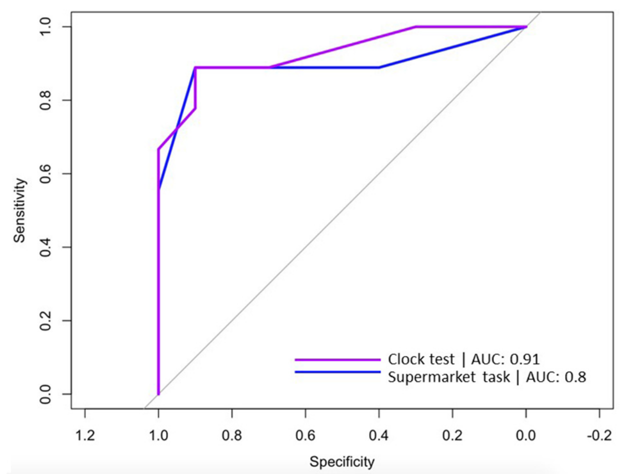

FIGURE 3 | ROC curves for Virtual Supermarket task (blue line) and Clock Orientation test (purple line) predicting correct diagnosis (VCI or AD).

Sensitivity and Specificity

The sensitivity and specificity of egocentric Virtual Supermarket and Clock Orientation test performance in VCI and AD were explored using logistic regression and ROC curves. Logistic regression indicated that the regression model based on egocentric scores of Supermarket and Clock Orientation predictors was statistically significant, $X^2_{(2)} = 16.36$, $p < 0.001$.

The model explained 77% (Nagelkerke R^2) of variance in VCI and AD patients and correctly classified 84% of patients (7 out of 9 VCI; 9 out of 10 AD) into their respective cohorts. ROC curves were computed for the supermarket and clock test predictors in discerning VCI from AD patients. Similarly, Area Under the Curve (AUC) values indicated that egocentric orientation in the Supermarket [AUC = 0.8, SE = 0.12; 95% CI (0.56, 1)] and Clock test [AUC = 0.91, SE = 0.06, 95% CI (0.8, 1)] had strong diagnostic accuracy in distinguishing VCI from AD patients (see **Figure 3**).

DISCUSSION

Overall, our results indicate that medial parietal mediated egocentric path integration processes are a sensitive and specific cognitive marker selective for VCI. By contrast, allocentric orientation deficits were less sensitive, and not specific to distinguish between the underlying pathologies.

In more detail, the egocentric path integration measures of the Virtual Supermarket task and Clock Orientation test successfully detect vascular changes in patient populations. More importantly, the measures allowed to reliably distinguish vascular from AD pathophysiology in the patient populations. Notably, egocentric orientation was impaired in VCI, but relatively intact in AD patient groups when controlling for age and sex. This supports findings from our vascular patient case study (Coughlan et al., 2018a) and suggests egocentric impairments indicate a more medial parietal focused change (Weniger et al., 2009) in VCI. Furthermore, the AD patient's egocentric ability remained intact which supports suggestions that MCI and earlier stage AD groups show an undisturbed egocentric orientation (Coughlan et al., 2019), which is consistent with our early-stage AD patient population (see total ACE-III score of 72.1). It would be interesting to explore whether more moderate to advanced AD patients might show problems using both allocentric and egocentric orientation, as it is known that medial parietal structures might be affected only later in the disease course (Braak and Del Tredici, 2015).

The egocentric demands in the virtual Supermarket requires the individual to form an accurate representation of the starting point by integrating virtual self-motion with heading direction to reach their end destination. Path integration plays an important role in updating spatial orientation during self-motion but this process is accumulative, therefore it can be liable to directional errors with respect to the original starting position (McNaughton et al., 2006), which may be responsible for problems observed across both egocentric tasks. The Clock Orientation test also demands path integration to configure the position of numbers on a clock face relative to the individual's current position. Both tasks rely on accessing scene construction, mental rotation and imagery translated from an egocentric orientation. At the neural level, translation of these egocentric processes depend mainly on the medial parietal cortex (Goodale and Milner, 1992; Galati et al., 2000; Zaehle et al., 2007; Coughlan et al., 2018b) as well as the prefrontal cortex (Spiers, 2008; Bird et al., 2012; Spiers and Barry, 2015), indicating potential disruptions in frontoparietal structures typically seen in vascular patients (Beason-Held et al., 2012; Heiss et al., 2016; Vipin et al., 2018; van der Flier et al., 2018).

Medial parietal mediated egocentric deficits appear to characterize VCI patients. This is consistent with emerging evidence suggesting the earliest signs of dysfunction appear in medial frontal and anterior cingulate regions in at VCI-risk individuals (Papma et al., 2012; Haight et al., 2015), which is accompanied by a more typical vascular profile of reduced integrity of white matter in the bilateral superior longitudinal fasciculus (Beason-Held et al., 2012). Since egocentric orientation does not deteriorate in healthy aging and early-stage AD, compared to medial temporal based cognitive functions (for review, see Colombo et al., 2017) it emerges as a potential powerful cognitive marker to identify early vascular-related pathology. Given the prevalence of vascular-related dementia, it is surprising that investigation to isolate cognitive deficits unique to this pathology is so sparse. However, based on our findings, it appears that egocentric orientation may be a useful diagnostic tool to discriminate VCI from other neurodegenerative conditions.

Our study suggests allocentric orientation deficits were not statistically present in AD, only VCI showed significant impairments compared to healthy controls. This does not support our prediction that allocentric deficits would be more profound in AD. The literature suggests allocentric deficits are more prominent in preclinical AD (Coughlan et al., 2019) with a loss in selectivity as the disease stage progresses and deficits become more widespread (Braak and Del Tredici, 2015). Yet, for the early-stage AD patients in our study results were not significant. A *post hoc* power analysis was employed using G*Power3 (Faul et al., 2007) and results indicate power at Cohen's $d = 0.32$ would have been sufficient to yield significant results between AD and VCI allocentric performance. The actual power yielded between groups was reported at Cohen's $d = 0.71$. Therefore, group sizes should have been large enough to yield significant effects. Indeed, as evident from **Figure 2**, it is clear that AD patients perform differently from controls but this did not reach statistical significance.

One potential explanation for the results observed may be provided by the large range in allocentric scores across the VCI group (see **Table 1**). VCI is a highly heterogeneous disorder in terms of disease pathology and subsequent cognitive impairments which may account for this variation, compared to AD pathology and symptoms that are more uniform. As VCI patients revealed both egocentric and allocentric orientation problems this is likely to represent a disruption to translational and integration processes where both frames are combined to produce effective navigation. This view also explains the reduced visuospatial performance exhibited by the VCI patients during neuropsychological testing across RCFT copy and position discrimination tasks.

It is also important to consider the domain of memory when interpreting our findings. Results from the FCSRT suggest VCI patients had significantly worse memory than the AD and control groups, sub-score results indicate this is driven by reduced performance during free recall. This is likely due to the retrieval demands on prefrontal and parietal structures

(Staresina and Davachi, 2006) which are typically disrupted in VCI. However, when cued VCI patients outperform AD patients. This finding is consistent with evidence that suggests providing a cue has little bearing on improved memory recall in AD (Sarazin et al., 2007; Wagner et al., 2012). This finding may be relevant to the poor allocentric results observed for VCI patients, as reduced retrieval mechanisms may have disrupted their task performance as opposed to pure allocentric (medial temporal) mapping problems, which we would expect to see in the AD patients.

Despite these exciting findings, our study is not without limitations. First and foremost, replication in larger patient cohorts is important. Further, clinical characterization of VCI subtypes (Skrobot et al., 2017) would help to better classify vascular pathology and determine accompanying cognitive symptoms, this may also help inform the variation of results seen in allocentric performance for the VCI patients. Future studies may also wish to examine the relationship between spatial navigation performance and the patient's perceived navigational abilities. Findings suggest perceived spatial ability assessed by the self-report Santa Barbara Sense of Direction Scale (Hegarty et al., 2002) is correlated with spatial accuracy and hippocampal volume (Burte et al., 2018). Therefore, the assessment of perceived spatial abilities may help inform spatial navigation as a marker of pathological aging. Finally, as the study did not access the patient's clinical MRI scans, confirmation of vascular lesions and their locations, as well as AD specific biomarkers would be important in the future to corroborate our cognitive findings.

Nevertheless, to our knowledge this in the first study to isolate a selective navigational deficit in VCI. This showcases the important role of virtual navigation and spatial tests in the future development of sensitive and specific diagnostic tests for VCI. Further investigation into the cognitive symptoms selective to VCI as well as longitudinal cohort studies in at VCI-risk individuals is critical to identify the emergence of the disease and intervene with therapeutic strategies as early as possible.

In conclusion, our findings show a distinct egocentric orientation deficit that is specific for VCI relative to AD. This is critical given the lack of specificity in current diagnostic tests and the indistinct diagnostic criteria for cognitive symptoms in VCI. In turn, this will inform diagnostic work-ups and aid personalized treatment pathways to treat underlying vascular changes in patients.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Faculty of Medicine and Health Sciences Research Ethics Committee, University of East Anglia. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

EL and MH contributed to the conception and design of the study, statistical analysis and intellectual contribution to the writing of the manuscript. VP, GC, and SJ contributed to the data collection and intellectual contribution to the manuscript.

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Spatial Perspective-Taking in Children With Autism Spectrum Disorders: The Predictive Role of Visuospatial and Motor Abilities

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Despite its impact on everyday functioning, spatial perspective-taking has rarely been investigated in autism spectrum disorders (ASD), and previous findings are surprisingly sparse and inconsistent. In the present study, we aimed to investigate spatial perspective-taking abilities in children and adolescents with ASD without intellectual disabilities, comparing them with a group of typically developing (TD) peers. Our objectives were: (i) to test similarities and differences between these groups in a spatial perspective-taking task; and (ii) to see whether similar or different underlying processes (i.e., fine and gross motor skills, and visuospatial abilities) might account for the groups' performance in the spatial perspective-taking task. A group of children with ASD (N = 36) was compared with a TD group (N = 39), aged from 8 to 16 years. Participants were administered tasks assessing spatial perspective-taking, fine and gross motor skills, visuo-constructive abilities, visuospatial working memory, visual imagery, and mental rotation. Our results revealed that the ASD group had more difficulty with the spatial perspective-taking task than the TD group. The two groups also had some shared and some different processes that predicted their perspective-taking performance: a significant predictive effect of fine motor skills and visuospatial working memory emerged for both groups, while gross motor skills (i.e., walking heel-to-toe) and visuospatial imagery only revealed a role in the TD group. These findings suggest that different abilities might account for the two groups' performance in the spatial perspective-taking task. Gross motor skills and complex visuospatial abilities seem to be more important in sustaining spatial perspective-taking ability in typical development than in the event of ASD. Some of the clinical and educational implications of these findings are discussed.

Keywords: spatial perspective-taking, neurodevelopmental disorders, autism spectrum disorder, visuospatial abilities, motor abilities

INTRODUCTION

Autism spectrum disorders (ASD) are characterized by deficits in social communication, social interaction, and obsessive/stereotyped patterns of behavior, interests or activities (American Psychiatric Association, 2013). Other, non-social factors also have an important role in the cognitive profiles of children with ASD (Cardillo, 2018), even for those with no intellectual disabilities (ID).

One of the features of the cognitive phenotype of this disorder is an atypical perceptual processing, particularly for complex visual stimuli (Caron et al., 2006; Cardillo et al., 2018). A vast amount of research on the role of these processing peculiarities in the visuospatial domain in individuals with ASD has revealed a heterogeneous profile of strengths and weaknesses, depending on the type and complexity of the tasks administered (e.g., Edgin and Pennington, 2005; Happé and Frith, 2006; Kuschner et al., 2007; Mammarella et al., 2019; Cardillo et al., 2020). The crucial role of visuospatial functioning in ASD emerges clearly from its possible consequences on everyday life and adaptive behaviors. Visuospatial abilities are essential to interaction with the environment (Hegarty and Waller, 2005; Jansen and Heil, 2010) and involved in many daily activities, from navigating in the environment to recognizing and manipulating objects, to recalling locations (Tzuriel and Egozi, 2010; Cardillo, 2018). From the academic standpoint, visuospatial skills predict success in science, technology, engineering, and math (Humphreys et al., 1993; Uttal and Cohen, 2012; Andersen, 2014; Khine, 2017; Mammarella et al., 2018). Visuospatial abilities can be trained (Uttal et al., 2013a,b; Meneghetti et al., 2017), so it is fundamentally important to understand the factors that influence performance on measures of these skills (Schmidt et al., 2013; Tarampi et al., 2016).

One of the crucial components of the multi-faceted construct of visuospatial ability is spatial perspective-taking (Eilam and Alon, 2019), which involves a higher-level, conscious, and deliberate mental transformation that corresponds to the spatial orientation factor (Thurstone, 1950; Huttenlocher and Presson, 1973; Lohman, 1988; Kessler and Rutherford, 2010). Spatial perspective-taking consists in seeing a space from a different perspective, adopting new imaginary orientations, mentally viewing a scene from an external viewpoint (Pearson et al., 2013). This spatial transformation process occupies a crucial place at the convergence of perception and mental imagery (Kessler and Rutherford, 2010). It is particularly important in "large-scale" spatial activities, when individuals can imagine "being part of" or "move through" a space (Münzer et al., 2018). In fact, tasks investigating spatial perspective-taking abilities have revealed an important role in predicting people's environment-learning (Allen et al., 1996; Pazzaglia and De Beni, 2006), navigating and wayfinding abilities (Kozhevnikov et al., 2006).

One task that enables spatial perspective-taking abilities to be investigated is the Object Perspective-Taking Test (OPT) developed by Kozhevnikov and Hegarty (2001) and Hegarty and Waller (2004). This test assesses an individual's ability to mentally adopt new imaginary positions within a configuration of objects. It was developed to better explore the distinction between spatial orientation and spatial visualization performance, or the ability to make egocentric and object-based spatial transformations, respectively (Meneghetti et al., 2012). Hegarty and Waller (2004) confirmed that a distinction could be drawn between these two spatial factors using a confirmatory factor analysis in which the perspective-taking factor was dissociated from mental rotation. Despite this dissociation, these two factors proved to be strictly related (Kozhevnikov and Hegarty, 2001; Hegarty and Waller, 2004). Specifically, Hegarty and Waller (2004) using different measures of perspective-taking and mental rotation abilities, showed that these two spatial factors were highly correlated ($r = 0.80$), indicating that they have a consistent portion of shared variance. In order to account for this shared variance, authors suggested different hypotheses. First, perspective taking and mental rotation may rely on common processes (i.e., encoding and memory of spatial images). Second, participants might use the same strategy to perform both perspective taking and mental rotation tasks. Third, similar innate or environmental factors might influence one's ability to solve the two types of spatial transformations (Hegarty and Waller, 2004).

Given the complex nature of spatial perspective-taking, some published studies investigated the role of different factors underlying people's performance. Meneghetti et al. (2012) showed that OPT performance is sustained by specific spatial abilities and by the use of different strategies. The authors administered the OPT and several visuospatial tasks and self-report measures to undergraduate students to investigate whether different spatial abilities and strategies sustained their OPT performance. The results showed that OPT performance was positively associated with spatial visualization ability and a preference for spatial imagery strategies, while it was negatively associated with the use of a mental rotation strategy.

Visuospatial working memory and motor abilities have also been found to have an important influence on spatial perspective-taking performance of children and adults (Kaiser et al., 2008; Eilam and Alon, 2019). Neuroimaging studies, conducted with adults, showed activation of areas involved in general cognitive control processes (such as working memory) and the supplementary motor area during the execution of a mental rotation task or a spatial perspective-taking task (Johnston et al., 2004; Kaiser et al., 2008). In particular, Kaiser et al. (2008), found the activation of the supplementary motor area in healthy adults during the execution of a spatial perspective-taking task. Authors highlighted that the activation of this brain region can relate to the encoding of the stimuli in relation to the observer, as well as to the cognitive processes involved in the perspective transformation. Only few studies have explored the relationship between perspective taking and motor abilities in children. Newcombe and Frick (2010) suggested that the developmental progress of perspective taking abilities is strictly related to motor development, and motor activity has been found to facilitate children's performance in this kind of tasks. According to the authors, it would seem that children's mental spatial transformation abilities can profit from active movements, by allowing them to draw into consolidated links between action and cognition (Newcombe and Frick, 2010). In addition, children's perspective taking skills were found to be related with their spatial drawing abilities, which involve visuo-motor skills (Ebersbach et al., 2011). However, to the best of our knowledge, no research has explored the relationship between spatial perspective taking and motor abilities in children with neurodevelopmental disorders.

In addition, despite its impact on everyday functioning and strong association with various visuospatial abilities, spatial perspective-taking in ASD has been investigated only rarely (David et al., 2010), and with inconsistent results (Pearson et al., 2013).

Some studies involving adults or/and children, found spatial perspective-taking performance intact in participants with ASD, and concluded that any deficits in this area were not crucial in the ASD profile (Hobson, 1984; Reed and Peterson, 1990; Tan and Harris, 1991; David et al., 2010). Others reported evidence of poor spatial perspective-taking abilities in children with this clinical diagnosis (Yirmiya et al., 1994; Warreyn et al., 2005). Some authors argued that a possible explanation for the discrepant findings across studies lies in the different tasks administered (i.e., items vs. appearance questions) and the way the instructions were presented (i.e., viewer vs. object-rotation instructions) (Langdon and Coltheart, 2001; David et al., 2010). Considering the tasks, item questions ask to judge which object in an array of features occupies a specific position relative to another viewpoint, while appearance questions ask how an array would appear from another perspective (Langdon and Coltheart, 2001). Concerning the instructions, in the viewer rotation the examinee is asked to imagine moving himself relative to a fixed array, while in the object rotation is asked to imagine rotating an array relative to the viewer fixed position (Langdon and Coltheart, 2001). According to David et al. (2010), adults with ASD seem to perform better on item questions, particularly when they have to manage with viewer rotation instructions (i.e., "Which object would be to your right if you were in that position?") while, employing object rotation instructions (i.e., "Which object would be to your right if we turned the stand so that side over there were in front of you?") would be disadvantageous for this clinical group. Thus, the use of different tasks and instructions could explain discrepant findings across studies.

As for motor skills, to our knowledge no research has investigated their role in predicting the spatial perspective-taking performance of participants with ASD. However, previous studies involving adults or/and children extensively reported poor fine and gross motor skills in individuals with ASD (Ming et al., 2007; Fournier et al., 2010; Staples and Reid, 2010; Whyatt and Craig, 2012). Differences in these underlying processes should therefore be taken into account when considering the variability in spatial perspective-taking performance of individuals with ASD.

The findings described thus far highlight the need to analyze the spatial perspective-taking abilities of individuals with ASD in more depth. Only a handful of studies have explored these spatial skills in such individuals, and none investigated the concurrent role of both visuospatial and fine and gross motor skills. The present study aimed to investigate spatial perspective-taking abilities in children and adolescents with ASD without ID comparing them with a group of typically developing (TD) peers. Participants were administered tests to measure their fine and gross motor skills, visuo-constructive abilities, visuospatial working memory, visual imagery and mental rotation.

To clarify the similarities and differences between the two groups' spatial perspective-taking performance, our first aim was to seek possible differences in terms of angular disparity. To establish whether similar or different underlying processes might account for the groups' performance in the spatial perspective-taking task, we also used two separate models: one for the role of fine and gross motor skills in predicting spatial perspective-taking performance; the other for the involvement of visuospatial abilities (i.e., visuo-constructive abilities, visuospatial working memory, visuospatial processing, and mental rotation).

Although we expect some differences between groups, the inconsistency of previous reports on the spatial perspective-taking abilities of individuals with ASD (Pearson et al., 2013) prevented us from making any specific predictions regarding our groups' performance. Given the role of motor abilities and visuospatial factors underlying spatial perspective-taking, we might expect to find a significant effect of visuospatial imagery (Meneghetti et al., 2012), visuospatial working memory, and motor abilities (Johnston et al., 2004; Kaiser et al., 2008; Meneghetti et al., 2016) in sustaining our participants' perspective-taking performance.

MATERIALS AND METHODS

Participants

The study involved 75 participants aged between 8 and 16 years old: 36 (34 M) children with ASD but no ID, and 39 (36 M) matched TD controls. The two groups did not statistically differ in chronological age [$F(1, 73) = 0.34$, $p = 0.563$; $R^2_{adj} = 0.009$], gender distribution [$\chi^2(df = 1) = 0.008$, $p = 0.926$], or total IQ [$F(1, 73) = 1.34$, $p = 0.250$; $R^2_{adj} = 0.018$]. A summary of the participants' characteristics is shown in **Table 1**.

All participants were recruited via local community contacts, at specialized centers (for children with ASD), or schools (for the TD children).

Children in the ASD group had all received an independent clinical diagnosis according to the DSM-IV-TR (American Psychiatric Association, 2000) or ICD-10 (World Health Organization, 1992) criteria. They also scored above the cut-off for ASD in the Autism Diagnostic Interview – Revised (ADI-R; Rutter et al., 2005). Children with ASD were only included in this study if they achieved a standard score of 85 or more for total IQ on the Wechsler Intelligence Scales (WISC IV: Wechsler, 2003).

The TD group consisted of healthy children of normal intelligence with no history of psychiatric, neurodevelopmental or neurological disorders. In addition, having a family member with a neurodevelopmental disorder was an exclusion criterion for this group. They were tested individually at school.

All participants spoke Italian as their native language and had no neurological, visual or hearing impairments. The study was approved by the research ethics committee at the University of Padua, Italy, and all parents had given prior written consent to their children's participation by signing an informed consent form.

Materials

Spatial Perspective-Taking

The *Short Object Perspective-Taking* (sOPT) task (adapted from Kozhevnikov and Hegarty, 2001; Hegarty and Waller, 2004) is a paper-and-pencil task comprising six items, each containing a configuration of seven objects drawn on the top half of an A4 piece of paper and a circle for the answer placed at the bottom half of the same page (see **Figure 1**). On each item, participants were asked to imagine being at one object in the layout (the station

TABLE 1 | Characteristics of the two groups: children with autism spectrum disorders but no intellectual disability (ASD); and typically developing (TD) peers.

Measures	ASD (*n* = 36)	[Min–Max]	TD (*n* = 39)	[Min–Max]	*F*(1, 73)	*P*	Cohen's *d*
Gender (M:F)	34:2		36:3				
Age (year; month)							
Mean (*SD*)	10;10 (2;8)	[8;0–16;10]	11;3 (2;10)	[8;00–16;8]	0.337	0.563	0.13
IQ[a]							
Mean (*SD*)	98.30 (12.75)	[80–135]	101.62 (11.97)	[83–132]	1.34	0.250	0.27
ADI-R: A							
Mean (*SD*)	15.77 (7.26)	[10–29]	4.05 (3.64)	[0–9]	80.03	<0.001	2.04
ADI-R: B							
Mean (*SD*)	11.64 (5.07)	[8–23]	2.85 (2.12)	[0–7]	98.61	<0.001	2.27
ADI-R: C							
Mean (*SD*)	6.61 (3.05)	[3–14]	1.41 (0.75)	[0–2]	87.59	<0.001	2.36

[a] *Standard scores on the Wechsler Intelligence Scale for Children – Fourth Edition. IQ, Intelligence Quotient; ADI-R, Autism Diagnostic Interview (Rutter et al., 2005); ADI-R: A, Reciprocal Social Interaction; ADI-R: B, Language/Communication; ADI-R: C, Repetitive Behaviors/Interests. Higher scores on the ADI-R reflect more severe autistic symptoms.*

point), facing another (imagined heading), and pointing to a third (target object). Participants were asked to give their answers using the circle provided at the bottom half of the page, which displayed the station point (e.g., the flower) in the center of the figure, and the imagined heading (e.g., the tree) drawn as an arrow pointing vertically up. Participants were asked to draw an arrow from the center toward the edge of the circle, indicating the direction to the target object (e.g., the cat). An item example is reported in **Figure 1**; the dashed arrow indicates the correct response to the item. The time limit for completing the task was 5 min. The six items were divided into three categories, depending on the angular disparity with respect to the respondent's point of view (0–60°, 60–120°, 120–180° in the right or left half-disk). The answers of two of the items fell in each of the three categories. The score corresponded to the deviation in degrees between the participant's response and the correct direction to the target, for each item (degrees of error or angular disparity). The higher the degrees of error the worse the performance.

Fine and Gross Motor Abilities

Fine and gross motor abilities were analyzed using four subtests of the Movement ABC-2 (Henderson et al., 2007), two each from the Manual dexterity and Balance domains, respectively. Manual dexterity refers to the fine motor control of hands and fingers needed to manipulate objects. Dynamic balance involves gross motor skills that are specific goal-directed movement patterns. The following tasks were administered, and according to the manual, the version for the younger children (7–10 years old), and for the older ones (11–16 years old) was used:

Manual Dexterity 1 (MD 1)

Participants were asked to insert (younger children) or rotate (older children) 12 pegs in a pegboard. Children were asked to take the pegs one at time and to put them in the pegboard as soon as possible. The task was performed first with the dominant hand and then with the non-dominant hand. The task was timed and two trials were given for each hand; the best trial for each hand was used to rate the task. Response times were considered for scoring purposes.

Manual Dexterity 3 (MD 3)

Participants had to draw a trail between the two lines of a path of variable size (wider for the younger children, narrower for the older ones). Only the dominant hand was considered.

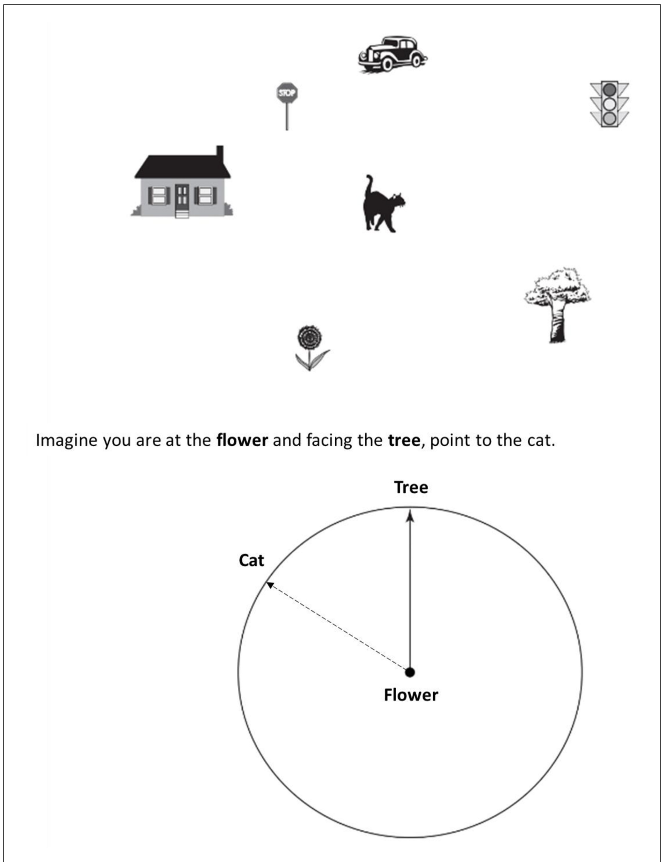

FIGURE 1 | Example of an item in the Short Object Perspective-Taking task (sOPT). The dashed arrow indicates the correct response to the item (direction to the cat).

A maximum of two trials were given, and the best trial was used to rate the task. If the child completed the first trial without errors, the second trial was not required. The number of errors was considered.

Dynamic Balance 1 (BAL 1)

Participants were asked to walk forward heel-to-toe (the younger children) or backward toe-to-heel (the older ones) on a 4.5 m long strip of adhesive tape placed on the floor. A maximum of two trials were given, children had to walk up to 15 steps or to the end of the line, and the best trial was used to rate the task. If the child completed the first trial without errors, the second trial was not required. The number of correctly completed steps was recorded.

Dynamic Balance 2 (BAL 2)

Participants were asked to hop on one-foot straight forward (the younger children) or zig-zagging from side to side (the older ones). Participants were required to jump five consecutive jumps on mats first with the dominant leg and then with the non-dominant leg. A maximum of two trials were given for each leg. If the child completed the first trial without errors, the second trial was not required. The best trial for each leg was used to rate the task and the number of hops completed was recorded.

For each task of the Movement ABC-2, raw scores (accuracy, errors or response times, depending on the task) were compared with normative values, and Scaled scores ($M = 10$, $SD = 3$) were computed. A scaled score from 1 to 7 is described as a below average score, a scaled score from 8 to 12 is described as an average score, finally a scaled score from 13 to 19 is described as an above average score.

Visuo-Constructive Abilities

The Rey-Osterrieth Complex Figure Test (ROCFT; Rey, 1941, 1968) is a neuropsychological test measuring visuo-constructive skills. Participants were asked to copy from the original figure a complex geometrical figure. To perform the copy condition, the stimulus figure was placed in front of the examinee, with the request to copy the figure as accurately as possible. The standard scoring system (Rey, 1968) was used to measure the accuracy of their drawing, awarding different scores (from 0 to 2) to each of the 18 elements comprising the figure depending on their presence or absence, and/or correct location in a participant's drawing. There were not time limits for drawing the figure. The raw scores were considered for each participant. The higher the score the better the performance.

Visuospatial Working Memory

Two computerized tasks, adapted from Mammarella et al. (2018), were used to measure simultaneous and sequential spatial working memory. Each task consisted of a maximum of 21 items administered with a self-terminating procedure. Participants were shown a 5 × 5 grid and asked to memorize a number of cells presented simultaneously or sequentially. After 3 s, the initial stimulus was removed, and participants were shown a blank grid in which they had to reproduce the previously seen pattern of cells. In the spatial-simultaneous matrices (SSM), participants were asked to recall the position of the stimuli, while in the spatial-sequential matrices (SSQM), they needed to recall the stimuli in their order of presentation. In both tasks, the number of cells presented in each grid ranged from 2 to 8. The accuracy was calculated as a proportion i.e., the number of correct responses out of the total number of items performed. The higher the score the better the performance.

Visuospatial Processing

The Arrows task is a subtest of the Nepsy-II battery (Korkman et al., 2007), which assesses the ability to create and manipulate a mental representation of an object, and the ability to judge line orientation. The task consisted of 21 items. For each item participants looked at an array of arrows placed around a target and indicate the arrows that were pointing to the center of the target. The number of correct responses were considered and one point was awarded for each correct arrow detected. The scores obtained by each participant were compared with the normative values and expressed as scaled scores.

Mental Rotation

The Animal Rotation task derived from Kaltner and Jansen (2014) is a paper-and-pencil task used to assess mental rotation abilities. Participants were asked to look at a target figure and choose the corresponding figure from among four rotated options presented alongside. The stimuli consisted of 2D figures of animals, and the task included 21 items. Participants had 5 min to complete the task. One point was awarded for each correct response. The accuracy was calculated as a proportion, i.e., the number of correct responses out of the total number of items. The higher the score the better the performance.

Procedure

Participants were tested in a quiet room during two individual sessions lasting ˜40 min each. The tasks were administered in a counterbalanced order. Instructions were given for each task, and participants practiced with each task before starting the experiment. The computerized tasks were administered using a laptop computer with a 15-inch LCD screen.

Data Analyses

Data were analyzed using R (R Core Team, 2015). First, the scores obtained from the sOPT task were modeled using a mixed-effects approach, and run using the "lme4" package (Bates et al., 2015). Both fixed and random effects were considered by means of a series of likelihood ratio tests for nested models based on the chi-square distribution (Pinheiro and Bates, 2000). For each model, the Akaike Information Criterion (AIC; Akaike, 1974) was reported and a lower AIC indicated a better model. The analyses were conducted considering every single trial for each participant and participants were included as random effects to consider their variability in the mixed-effect model. The following fixed effects and their interactions were tested: Group (2 levels: ASD, TD) and Angular disparity (3 levels: level 1 = 0–60°, level 2 = 60–120°, level 3 = 120–180°).

Then two different linear regression analyses were run to investigate the association between the dependent variable (sOPT) and the motor or visuospatial abilities considered, and to identify the most predictive combinations. First the measures of fine and gross motor abilities were included as predictors (i.e., Manual dexterity 1, MD 1; Manual dexterity 3, MD 3; Dynamic balance 1, BAL 1; Dynamic balance 2, BAL 2). Then the tasks measuring visuospatial abilities were considered (i.e., ROCFT; SSM; SSQM; Arrows; Animal Rotation). For both models, the main and interactive effect of Group (i.e., ASD, TD) was included as well (see **Table 2** for the descriptive statistics of each measure by group).

Additional analyses (differences between groups for each motor and visuospatial measure, correlations and skewness and kurtosis for the residuals of each regression model) were reported in section "**Supplementary Material**."

We adopted a model selection strategy for all the variables examined (as in Fox, 2008, for example), following the same procedure to detect the best-fitting model. First, starting from the full model (M0 – which included the main effects of motor or visuospatial tasks, and their interaction with the effect of Group), we built the various models by subtracting one effect at a time, so that all the possible models were fitted. Then the models were compared using the Akaike Information Criterion (AIC, Akaike, 1974) as a fit index following the procedure suggested by Burnham et al. (2011), where the best model coincided with the smallest AIC. The best model(s) were selected from the set of models tested by applying information-theoretic (I-T) approaches, considering the AIC and the relative likelihood (*l*) of each model (Burnham et al., 2011). The values of AIC_s, Δ^0 AIC_s [Δ^0 AIC = $AIC_{full} - AIC_i$], ΔAIC_s [$\Delta AIC = AIC_{bestmodel} - AIC_i$], and l_s [$l = \exp(\Delta AIC/2)$] were computed for each model: Δ^0 AIC greater than 0 meant that a particular model *i* fitted the data better than the full model; ΔAIC described the distance between the best model and the other models computed; *l* values greater than 1 indicated that the model considered was more plausible. Details of the selected models and the indexes guiding model selection are given in **Table 3**.

Graphical effects were obtained using the "effects" package (Fox, 2003).

Group Differences in the Short Object Perspective-Taking (sOPT) Task

No main effect of Group emerged for the sOPT task [$\chi^2(1) = 1.09$, $p = 0.30$ (full model: *AIC* = 13,849; model without Group: AIC = 13,848)], but the main effect of the angular disparity was significant [$\chi^2(2) = 614.23$, $p < 0.001$ (model without Angular disparity: AIC = 14,459)]. The model coefficients showed that participants' performance was more accurate for level 1 than for levels 2 and 3 (*p*s < 0.001), and it was more accurate for level 2 than for level 3 ($p < 0.001$). The analysis also revealed a significant interaction between Group and Angular disparity [$\chi^2(2) = 71.469$, $p < 0.001$ (model with interaction: AIC = 13,781)] (see **Figure 2**). The model coefficients showed that the ASD group's performance was less accurate than the TD group's on level 1 ($p = 0.04$), while the groups did not differ on levels 2 and 3 ($p = 0.77$ and 0.11, respectively). The group with ASD showed significant differences between the various levels of angular disparity: their performance was more accurate for level 1 than for levels 2 and 3, and it was more accurate for level 2 than for level 3 (*p*s < 0.001). The group with TD also showed significant differences in performance between level 1 and levels 2 and 3 (*p*s < 0.001), making fewer mistakes on the first level than on the other two, on which their performance did not differ ($p = 0.39$).

Short Object Perspective-Taking (sOPT) Task and Motor Abilities

Following the above-described model selection strategy, as shown in **Table 3**, our model fitting procedure showed that the best-fitting model was M3 sOPT ∼ MD1 + Group*MD3 + Group*BAL1 (**Figure 3**). The main effects of MD 1 emerged ($\beta = 17.87$, $t = 2.18$, $p = 0.03$): shorter times to complete the MD 1 task predicted larger errors in the sOPT task. The interaction between Group and MD 3 was also significant ($\beta = 22.03$, $t = 1.96$, $p = 0.05$), showing that lower scores in the MD 3 task predicted larger errors in the sOPT task for the group with ASD, but not for the TD group. A significant effect of the interaction between Group and BAL 1 emerged as well ($\beta = -47.89$, $t = -2.07$, $p = 0.04$), showing that lower scores in the BAL 1 task predicted larger errors in the sOPT task for the TD group, but not for the ASD group.

Short Object Perspective Taking (sOPT) Task and Visuospatial Abilities

Concerning the association between the sOPT and visuospatial tasks, the model-fit analysis shown in **Table 3** indicated that the best-fitting model was M6 sOPT ∼ SSM + SSQM + Group*Arrow (**Figure 4**). The main effects of the SSM ($\beta = -390.19$, $t = -2.39$, $p = 0.02$) and of the SSQM ($\beta = -488.07$, $t = -2.25$, $p = 0.03$) tasks came to light. In both groups, lower scores obtained in these tasks predicted larger errors in the sOPT task. The interaction between Group and Arrow was also significant ($\beta = -25.21$, $t = -2.64$, $p = 0.01$): lower scores in the Arrow task only predicted larger errors in the sOPT task for the TD group, not for the ASD group.

DISCUSSION

In previous studies on typical populations, motor and visuospatial abilities revealed a crucial influence on spatial perspective-taking performance (Johnston et al., 2004; Kaiser et al., 2008; Meneghetti et al., 2012). This involvement of motor and visuospatial skills has never been studied in participants with ASD, however, and the results of studies on their spatial perspective-taking abilities have been inconsistent (Hobson, 1984; Reed and Peterson, 1990; Tan and Harris, 1991; Yirmiya et al., 1994; Warreyn et al., 2005; David et al., 2010). Since the studies were heterogeneous, the findings generated to date underscore the need to further investigate the spatial perspective-taking abilities of participants with ASD, also considering the role of any underlying processes. The present study thus aimed to examine spatial perspective-taking abilities in children and adolescents with ASD but no ID, comparing them with a group of TD peers. The influence of motor and visuospatial abilities on perspective-taking performance was also considered to shed more light on this complex visuospatial domain.

TABLE 2 | Means (*M*) and standard deviations (*SD*) by group: children with autism spectrum disorders but no intellectual disability (ASD); and typically developing (TD) peers.

Tasks		ASD (*n* = 36)		TD (*n* = 39)		*Cohen's d*
		***M* (*SD*)**	**[Min–Max]**	***M* (*SD*)**	**[Min–Max]**	
sOPT degrees of error	Level 1	83.9 (52.83)	[5–180]	66.33 (53.55)	[0–171]	0.33
	Level 2	94.04 (55.26)	[1–176]	91.33 (57.79)	[0–177]	0.05
	Level 3	110.08 (65.44)	[0–180]	92.65 (67.52)	[2–180]	0.26
MD 1		4.72 (3.19)	[1.00–12.00]	5.95 (3.58)	[1.00–13.00]	0.36
MD 3		4.77 (4.11)	[1.00–12.00]	7.77 (4.03)	[1.00–13.00]	0.73
BAL 1		8.25 (3.93)	[1.00–12.00]	10.87 (1.87)	[4.00–12.00]	0.85
BAL 2		8.66 (3.96)	[1.00–12.00]	10.28 (2.42)	[4.00–12.00]	0.49
ROCFT		18.94 (8.53)	[4.50–32.00]	25.37 (6.26)	[11.50–35.00]	0.86
SSM		0.19 (0.22)	[0.01–0.99]	0.22 (0.16)	[0.03–0.70]	0.16
SSQM		0.15 (0.15)	[0.01–0.81]	0.18 (0.13)	[0.03–0.48]	0.21
Arrows		26.47 (8.24)	[4.00–38.00]	29.21 (3.78)	[16.00–38.00]	0.43
AR		0.75 (0.29)	[0.10–1.00]	0.76 (0.26)	[0.24–1.00]	0.04

sOPT, Short Object Perspective-Taking task; MD1, Manual dexterity 1; MD3, Manual dexterity 3; BAL1, Dynamic balance 1; BAL2, Dynamic balance 2; ROCFT, Rey-Osterrieth Complex Figure Test; SSM, Spatial-Simultaneous Matrices; SSQM, Spatial-Sequential Matrices; AR, Animal Rotation.

TABLE 3 | Model comparison investigating the association between the sOPT (dependent variable) and motor or visuospatial tasks (predictors).

Models		AIC	Δ°*AIC*	ΔAIC	*l*	Adjusted R^2
Motor skills						
M0	sOPT ∼ Group (MD1 + MD3 + BAL1 + BAL2)	826.65	0	-4.38	0.11	0.18
M1	sOPT ∼ MD1 + Group*MD3 + Group*BAL1 + Group*BAL2	824.68	1.97	-2.41	0.30	0.19
M2	sOPT ∼ MD1 + BAL2 + Group*MD3 + Group*BAL1	823.08	3.57	-0.81	0.67	0.20
M3	**sOPT ∼ MD1 + Group*MD3 + Group*BAL1**	**822.27**	**4.38**	**0**	**1**	**0.20**
Visuospatial abilities						
M0	sOPT∼ Group (Arrow + ROCFT + AR + SSM + SSQM)	806.88	0	-6.57	0.04	0.38
M1	sOPT∼ SSM + Group*Arrow + Group*ROCFT + Group*AR + Group*SSQM	805.08	1.8	-4.77	0.09	0.39
M2	sOPT∼ SSM + SSQM + Group*Arrow + Group*ROCFT + Group*AR	803.76	3.12	-3.45	0.18	0.40
M3	sOPT∼ ROCFT + SSM + SSQM + Group*Arrow + Group*AR	803.24	3.64	-2.93	0.23	0.39
M4	sOPT∼ ROCFT + AR + SSM + SSQM + Group*Arrow	802.36	4.52	-2.05	0.36	0.39
M5	sOPT∼ ROCFT + SSM + SSQM + Group*Arrow	800.81	6.07	-0.5	0.78	0.40
M6	**sOPT∼ SSM + SSQM + Group*Arrow**	**800.31**	**6.57**	**0**	**1**	**0.40**

AIC, Akaike Information Criterion; Δ°AIC, difference in AIC with respect to the full model (M0); ΔAIC, difference in AIC; l, relative likelihood with respect to best target model [i.e., exp(ΔAIC/2)]. The higher the ΔAIC and the adjusted R^2, the better the model. sOPT, Short Object Perspective-Taking task; MD1, Manual dexterity 1; MD3, Manual dexterity 3; BAL1, Dynamic balance 1; BAL2, Dynamic balance 2; ROCFT, Rey-Osterrieth Complex Figure Test; AR, Animal Rotation; SSM, Spatial-Simultaneous Matrices; SSQM, Spatial-Sequential Matrices.

We first checked for differences in the spatial perspective-taking performance of our two groups (children with ASD vs. TD controls), taking the angular disparity of the stimuli into account. Then we looked into the role of fine and gross motor skills, and several visuospatial abilities (i.e., visuo-constructive abilities, visuospatial working memory, visual imagery, and mental rotation) in predicting spatial perspective-taking performance.

The sOPT task was used to assess our participants' spatial perspective-taking abilities. Based on generalized mixed-effects models, both groups showed a significant effect of the angular disparity of the stimuli, showing that errors were larger greater the angular disparity. This result is consistent with previous findings (Kessler and Thomson, 2010) of individuals' performance in spatial perspective-taking tasks worsening as the angular disparity between the egocentric and target viewpoints increased (Huttenlocher and Presson, 1973; Levine et al., 1982; Kozhevnikov and Hegarty, 2001; Zacks and Michelon, 2005, for a review). Our results also revealed differences between the two groups' perspective-taking performance, with larger errors for the ASD group than for the TD group, but only for stimuli with an angular disparity in the range of 0–60°. There were no such differences between the groups when the task involved greater degrees of angular disparity (60–120°, 120–180°). These results partially overlap with previous reports of spatial perspective-taking abilities being intact (Hobson, 1984; Reed and Peterson, 1990; Tan and Harris, 1991; David et al., 2010) or impaired (Yirmiya et al., 1994; Warreyn et al., 2005) in participants with ASD, highlighting the influence of angular disparity. Looking at the performance of the two groups reported in **Figure 2**, we can see that children with ASD showed a constant worsening

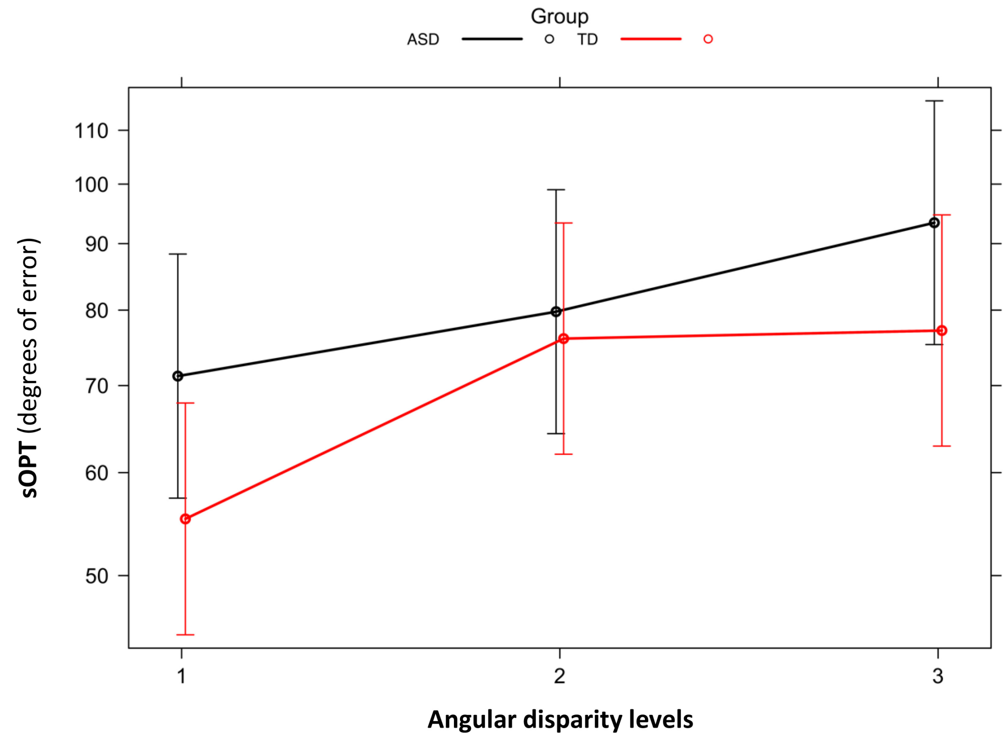

FIGURE 2 | Short Object Perspective Taking task (sOPT). Degrees of error by group (ASD, TD) and level of angular disparity. Error bars represent 95% confidence intervals. ASD, autism spectrum disorder; TD, typically developing; sOPT, Short Object Perspective-Taking task; level 1 = 0–60°; level 2 = 60–120°; level 3 = 120–180°.

performance as a function of the increase of the angular disparity. Differently, the TD group performance started to deteriorate when the angular disparity increased beyond 60°, showing a preserved performance when the angular disparity was lower. Our results for the TD group are consistent with previously published findings, which indicated that the performance of TD individuals in the sOPT remained fairly constant at lower angles, then – beyond an angular disparity of around 60–90° – their performance deteriorated (e.g., Kozhevnikov and Hegarty, 2001; Keehner et al., 2006; Kessler and Thomson, 2010). No previous studies, to our knowledge, explored the effect of the angular disparity in a perspective-taking task, considering children with ASD. Although, some similarities could be drawn from the study conducted by Brunyé et al. (2012), which explored the effect of autistic traits on the perspective-taking performance of adults. They found a pattern of deterioration in performance as a function of angular deviation, particularly for adults with high ASD traits. This pattern of performance showed by Brunyé et al. (2012) was similar to the pattern showed by our children with ASD, confirming a constant deterioration of the perspective-taking performance as a function of the increase of the angular disparity.

The second aim of the present study was to see whether similar or different underlying motor or visuospatial processes might account for the two groups' performance in the spatial perspective-taking task. To do so, we looked first at how fine and gross motor skills predicted spatial perspective-taking performance, then at the involvement of various visuospatial abilities (i.e., visuo-constructive abilities, visuospatial working memory, visual imagery, and mental rotation) in the same task.

Consistently with previous studies, our results showed that motor skills significantly affected both our groups' spatial perspective-taking performance (Johnston et al., 2004; Kaiser et al., 2008), but not precisely in the same way. Shorter times taken to complete a manual dexterity task (MD 1) predicted larger errors in the sOPT task for both groups. Lower scores (i.e., more errors) in a manual dexterity task assessing visuomotor abilities (MD 3) also predicted larger errors in the sOPT task, but only for the group with ASD. Thus, results from the MD 3 task suggests that better fine abilities predicted better spatial perspective-taking abilities for children with ASD. Another possible explanation to consider for this result could be that both tasks require the same type of response, that is to draw. Differently, results from the MD 1 task seemed to be inconsistent with this finding. However, it is worth noting that, differently from the MD 3 task, in the MD 1 task no difference between groups emerged (see **Supplementary Table S1**). In this case, the role of motivational variables could be considered. Probably children did not consider the task as a challenge, perceiving it as easy and distracting. In line with what is claimed by Elosúa et al. (2017), the lack of motivation in performing the MD 1 task would have made it possible for them to get more distracted in the task. Consequently, this has led to unexpected results for this task. On the other hand, lower scores in a gross motor task assessing

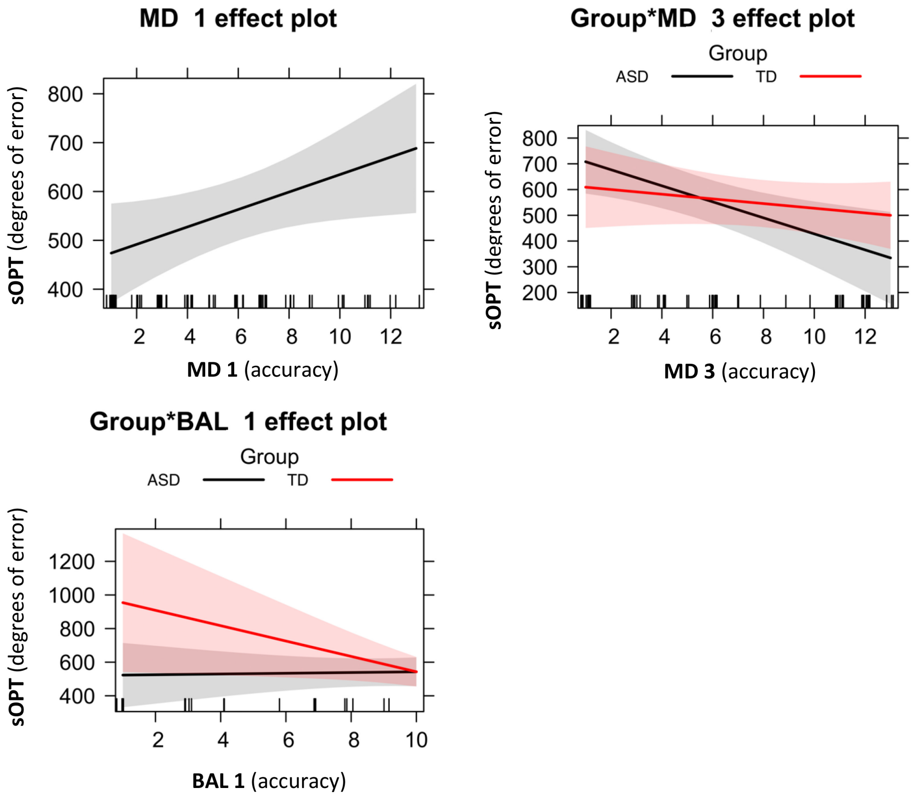

FIGURE 3 | Significant effects of the best-fitting model for degrees of error in the sOPT: M3 = sOPT ∼ MD1 + Group*MD3 + Group*BAL1. Error bands represent 95% confidence intervals. ASD, autism spectrum disorder; TD, typically developing; sOPT, Short Object Perspective-Taking task; MD1, Manual dexterity 1; MD3, Manual dexterity 3; BAL1, Dynamic balance 1.

balance (BAL 1), based on the ability to walk forward heel-to-toe or backward toe-to-heel, predicted larger errors in the sOPT task, but only for the TD group. To our knowledge, no previous studies investigated the role of motor skills in predicting children's spatial perspective-taking performance, but some interesting similarities with our results emerged from a study conducted by Lehmann et al. (2014) to correlate children's motor skills, working memory and mental rotation abilities. Their results showed a positive association between balance and mental rotation abilities in TD children. Mental rotation and spatial perspective-taking abilities are known to be related (Hegarty and Waller, 2004). Judging from our results, the same is true of TD children's balance (in terms of the ability to walk heel-to-toe or toe-to-heel) and perspective-taking abilities (Lehmann et al., 2014).

Concerning the role of visuospatial tasks in predicting spatial perspective-taking performance, a significant effect of visuospatial simultaneous and sequential working memory emerged for both our groups, showing that weaker abilities in these domains predicted greater difficulties in the spatial perspective-taking task. These results are consistent with previous reports supporting a relationship between perspective-taking ability and VSWM in the TD population (i.e., Johnston et al., 2004; Kaiser et al., 2008; Meneghetti et al., 2016; Eilam and Alon, 2019), and extend these findings to children with ASD. On the other hand, it was only in our TD children that we found a predictive effect of visuospatial processing on their perspective-taking performance, with lower scores in the Arrow task coinciding with larger errors in the sOPT task. The Arrow task assesses the ability to create and manipulate a mental representation of an object. In order to perform correctly the task, children have to imagine the path the arrow must take to get to the center of the target, considering the spatial relationships among the elements in the figure. Thus, spatial imagery abilities [i.e., the ability to represent the spatial relationships between the parts of an object and the location of objects in space or their movement (Van Garderen, 2006)] are involved in performing this task. Our result is in line with a previous report of a TD population's perspective- taking performance being predicted by spatial visualization ability and a preference for a spatial imagery strategy (Meneghetti et al., 2012). It is worth noting that no effect of spatial imagery on perspective-taking performance emerged for our participants with ASD, suggesting that our two groups shared some visuospatial processes underlying their spatial perspective-taking performance (i.e., visuospatial working memory), but probably used different strategies. Previous research on perspective-taking suggested that different strategies might be used by children with ASD comparing them with TD children. Pearson et al. (2016) found that perspective-taking (albeit visual perspective-taking as opposed to spatial perspective-taking) was driven by differential mechanisms in

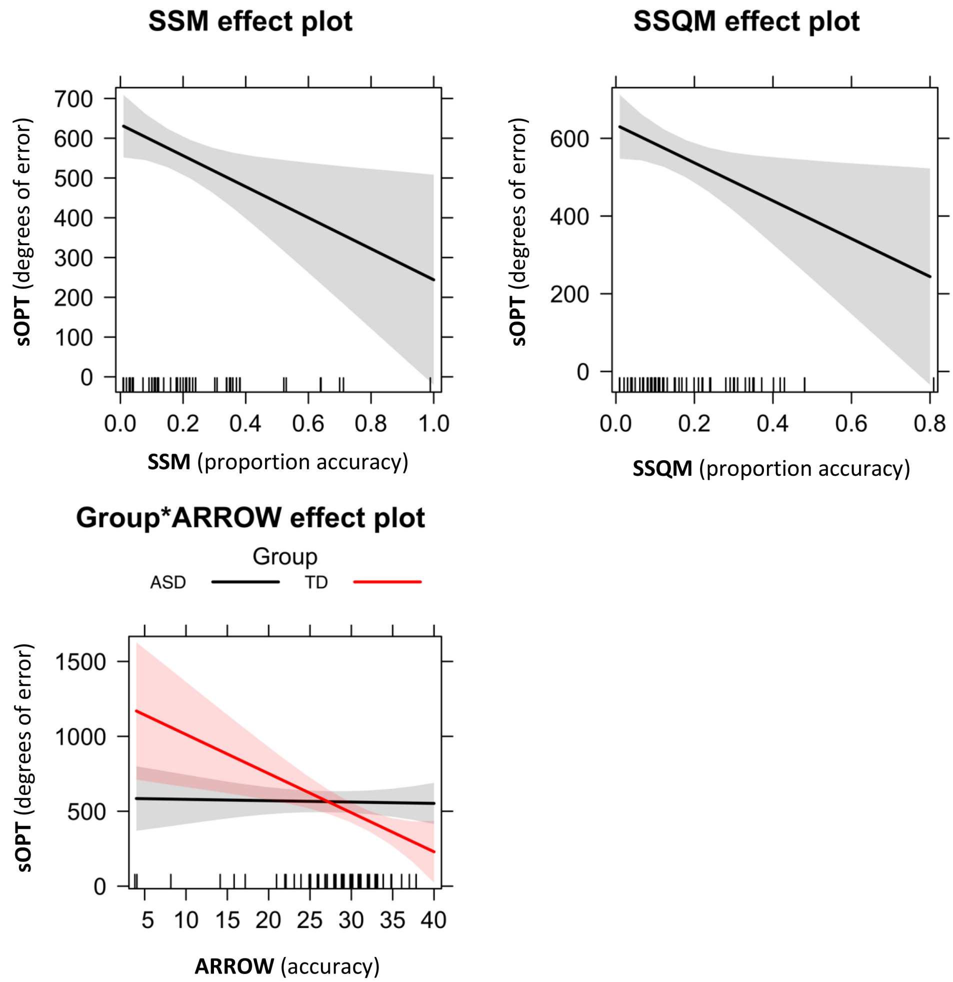

FIGURE 4 | Significant effects of the best-fitting model for degrees of error in the sOPT: M6 = sOPT∼ SSM + SSQM + Group*Arrow. Error bands represent 95% confidence intervals. ASD, autism spectrum disorder; TD, typically developing; sOPT, Short Object Perspective-Taking task; SSM, Spatial-Simultaneous Matrices; SSQM, Spatial-Sequential Matrices.

these two groups. Children with TD used an embodied egocentric transformation strategy to perform a perspective-taking task. They imagined to move their own position in the space and to see the world through a different perspective. This strategy involves the ability to mentally manipulate body representations. On the contrary, children with ASD were supposed to use a mental rotation strategy, drawing on their good spatial skills. They imagined scene rotating, using a cognitive demanding spatially grounded strategy as opposed to the embodied strategy used by the children with TD. Our results are in line with the study by Pearson et al. (2016), showing that the spatial perspective-taking abilities of TD children were sustained by different processes (i.e., spatial imagery abilities) as compared with the children with ASD. We did not find the effect of mental rotation abilities on the perspective-taking performance of our group with ASD, as Pearson et al. (2016) have showed. A possible explanation for this inconsistency between the studies may relate to the different tasks used to assess mental rotation. The mental rotation task proposed by Pearson et al. (2016) used the same material of the perspective-taking task. On the contrary our mental rotation task was quite different from the sOPT. Nevertheless, both the studies suggest the importance of considering different strategies in understanding spatial perspective-taking abilities of children with ASD and children with TD, providing interesting ideas for future research.

Taken together, our findings intriguingly suggest that different abilities might be involved in explaining the spatial perspective-taking performance of children with ASD and their TD peers. Further studies will be needed to confirm and extend our results, and to overcome certain limitations of the present study, one of which concerns the small size of our samples. Given that some papers on ASD made the distinction between ASD with and without speech onset delay to account for the heterogeneity of the spectrum regarding visuospatial abilities (e.g., Nader et al., 2015; Chiodo et al., 2017), further research should take into account the effects of the speech onset delay on the perspective-taking performances of children with ASD. In addition, previous findings provided evidence for executive dysfunctions in ASD (e.g., Berenguer et al., 2018), thus it might be interesting to consider also the effect of executive functions on the perspective-taking performance of children with ASD. Finally, in order to better explain the high variability of the clinical sample, a further reflection should be made on the possibility of comparing studies that use different statistical approaches (i.e., cluster analysis or individual analysis).

We nonetheless believe that our findings shed more light on the spatial perspective-taking abilities of children with ASD as compared with their TD peers, and may help us to clarify the former's performances in this domain. Our findings may also have some clinical and educational implications. Given the strong impact of spatial perspective-taking abilities on people's everyday functioning – in environment learning (Allen et al., 1996; Pazzaglia and De Beni, 2006), navigation and wayfinding (Kozhevnikov et al., 2006), for instance – elucidating the strengths and weaknesses of children with ASD could lead to training activities tailored to their specific needs.

To sum up, the present findings contribute to our knowledge of the spatial perspective-taking abilities of children with ASD, how they cope with angular disparity, and in what ways they differ from their TD peers. In particular, our ASD group was relatively inaccurate at all angles, instead of reflecting the TD group's decline in performance beyond angles of around 60°. We also confirmed the importance of examining the influence of various motor and visuospatial processes in predicting spatial perspective-taking performance as it differed in our two groups in some respects. Fine motor skills and visuospatial working memory were significant predictors for both groups, while gross motor skills and complex visuospatial abilities seemed to sustain spatial perspective-taking performance only in the TD group, not in the children with ASD. This would suggest that the two groups shared some processes but also differed in other predictors of perspective-taking performance. Hence, ASD could be considered as a form of human neurodiversity which manifests in a set of strengths and difficulties in performing a spatial perspective-taking task that may differ to the typical population.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

Ethical approval was obtained from the Research Ethics Committee at the University of Padua, Italy (protocol number: 2811). All parents had given prior written consent to their children's participation by signing an informed consent form. Following parental consent, the participants were tested individually either at specialized centers or at their school.

AUTHOR CONTRIBUTIONS

RC and IM have made substantial, direct and intellectual contribution to the work. CE contributed to data analysis. All authors listed approved the work for publication.

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We declare that the theoretical framework concerning visuospatial abilities is based on RC's Ph.D. dissertation.

SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fnhum.2020.00208/full#supplementary-material

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The Acquisition of Survey Knowledge by Individuals With Down Syndrome

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People with Down syndrome often exhibit deficiencies in wayfinding activities, particularly route learning (e.g., Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015). Evidence concerning more sophisticated survey learning has been sparse. In the research reported here, two experiments are reported that evaluated survey learning of youth with DS and typically developing children (TD) matched on mental age. In Experiment 1, participants learned two overlapping routes consisting of three turns each through a virtual environment depicting 9 square city blocks. Following acquisition, they were tested on multiple measures of survey knowledge: finding a shortcut, identifying the direction of landmarks not currently visible from their location in the environment, and recognizing a bird's-eye representation of the overall environment. Under these conditions, which should provide relatively optimal opportunities for survey learning, the participants with DS performed comparably to TD participants matched on non-verbal ability on all of our measures of survey learning. Hence, we concluded that people with DS can acquire some survey knowledge when tasked with learning a small environment and given the opportunity to do so. In Experiment 2, the experimenter navigated participants through a large, relatively complex, virtual environment along a circuitous path, beginning and ending at a target landmark. Then, the participants were placed at a pre-specified location in the environment that they had viewed previously and instructed to navigate to the same target (a door) using the shortest possible path from their current location. They completed the task three times: once after being shown the environment one time, once after three exposures, and once after five exposures. Results indicated that the participants with DS exhibited significantly less skill at identifying the shortcut than did the TD participants, with differences emerging as the number of exposures increased. Participants with DS were also less able to recall landmarks at the end of the experiment. Overall, however, the performance of both groups was relatively poor in both experiments – with the performance of participants with DS being worse as conditions became less optimal. These results were discussed in terms of underlying mechanisms that may account for variations in survey learning as environmental complexity increases.

Keywords: down syndrome, survey-knowledge, landmark memory, spatial abilities, MA comparison

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INTRODUCTION

Down syndrome (DS) results from the presence of a full or partial copy of extra chromosomal material associated with chromosome 21. It is the most common genetic syndrome associated with Intellectual Disability (ID) (Dykens et al., 2000), with a prevalence reported at 1 in 700 births (Parker et al., 2010). The expression of DS includes physical, cognitive, and neuroanatomical abnormalities. Characteristic physical features may include poor overall muscle tone, flattened facial features, upward slanting eyes, wide short hands and fingers, small head and ears, and a protruding tongue (Bull and The Committee On Genetics, 2011). Cognitive impairments in DS include well documented deficits in speech and language (e.g., Fowler, 1990; Martin et al., 2009) and problems with verbal short-term and long-term memory (e.g., Wang and Bellugi, 1994; Jarrold et al., 1999; Godfrey and Lee, 2018). More recently, evidence has been presented suggesting that some aspects of visuo-spatial processing may also be impaired in individuals with DS (see Yang et al., 2014). In particular, researchers have identified poor performance with mental rotation (Meneghetti et al., 2018), the acquisition of visuo-spatial knowledge (Meneghetti et al., 2017), and the use of navigation and wayfinding skills (e.g., Courbois et al., 2013; Davis et al., 2014; Farran et al., 2015). Neuroanatomically, DS is characterized by smaller brain volumes, particularly associated with the cerebellum, frontal lobes, and temporal lobes (e.g., Pinter et al., 2001b; White et al., 2003; Dierssen, 2012). In addition, studies have reported smaller volumes for the hippocampus and corpus callosum (Aylward et al., 1999; Pinter et al., 2001a), as well as the entorhinal cortex (Dierssen, 2012; Guidi et al., 2018). Longitudinal differences in hippocampal volume have been associated with decreased cognitive functioning (Pujol et al., 2018).

There is considerable overlap between brain regions that are impacted by DS and those regions that support wayfinding/navigation activities. Wayfinding is generally thought to involve a fairly distributed network of brain regions (Boccia et al., 2014). The temporal lobes appear to play an important role in memory-guided navigation (Pine et al., 2002). The cerebellum has recently been identified as contributing to both motor and cognitive aspects of navigation (e.g., Iglói et al., 2015). Further, because navigation is a goal-oriented activity, evidence indicates a necessary role for frontal lobes in navigation (Ciaramelli, 2008) which may be related to the ability to keep the goal in mind during navigation activities and making navigation plans (Spiers, 2008).

There is ample neuroscientific and behavioral evidence that wayfinding depends heavily on two distinct but related mental representations, often termed route and survey knowledge. Route representations involve ordered connections of landmarks, whereas survey knowledge is a more sophisticated and flexible form of environmental representation that involves acquiring knowledge of the directions and relative distances between objects and locations within an environment that is independent of any specific route (Siegel and White, 1975), and is related to Tolman's (1948) conception of a cognitive map. Spatial relational processing, which is important for developing survey knowledge, seems to rely on a distributed network in the hippocampal region. The hippocampus proper is known to play a prominent role in the learning and memory of novel and recently learned environments (e.g., Claessen et al., 2019), responsible for considerable spatial relational processing (Kumaran and Maguire, 2005), and engaged in the learning of survey representations, which provide a mental representation of the physical environment (Schinazi et al., 2013). Specifically, the left hippocampus seems to be important for encoding relations between landmarks (Wolbers and Büchel, 2005), whereas the right hippocampus is associated with retrieval of relational information (Mellet et al., 2000). At the cellular level, place cells in the hippocampus provide a mechanism for encoding relative spatial location (O'Keefe and Nadel, 1978), which can be updated through movement and head direction (see Burgess, 2008). A full path integration model can be developed at the cellular level when also including the medial entorhinal cortex (McNaughton et al., 2006). Given neuroanatomical abnormalities and associated cognitive weaknesses that people with DS exhibit related to the hippocampus, it would not be at all surprising to find that people with DS would exhibit difficulties with wayfinding.

A small number of studies have supported the view that people with DS exhibit relatively poor wayfinding skills. Several of these studies focused on the acquisition of route knowledge, which involves the acquisition and memory of a fixed sequence of landmarks and turns to get from a starting location to a designated target location (Siegel and White, 1975). For example, Purser et al. (2015) examined route learning in adolescents and young adults with DS relative to typically developing (TD) children and participants with Williams Syndrome (WS). They found that their participants with DS could learn a six-turn route. However, the performance of the participants with DS depended on their non-verbal ability level. Participants with DS who were relatively low in non-verbal ability performed below that of the TD participants, whereas those who exhibited higher levels of non-verbal ability performed at levels similar to TD participants. Davis et al. (2014) reported two studies that indicated adolescents and young adults with DS exhibited more errors during route learning and took longer to learn routes than did a comparison group of participants with mixed etiology ID and a group of typically developing children with whom they were matched on non-verbal mental age. Farran et al. (2015) assessed route learning of adolescents and young adults with DS relative to TD children and adolescents and young adults with WS. They found that the participants with DS committed more errors during acquisition of two overlapping routes than did the TD children, although they performed similarly to the participants with WS.

Courbois et al. (2013) and Farran et al. (2015) evaluated the ability of people with DS to form shortcuts following the learning of two short routes. Courbois et al. (2013) reported that only two of seven participants with DS were able to identify the shortest path along two overlapping routes to find a target from the start of one learned route to the end of the second learned route. However, five of nine TD children matched on mental age (MA) were able to do so. Similarly, Farran et al. (2015) reported that only 10% of participants with DS were able to identify the shortest route using a novel path along across two previously learned routes, whereas 59% of their TD children found the shortest route. Hence, it appears that the acquisition of survey knowledge may also be a problem for people with DS. However, this conclusion is based on two studies that have focused on one aspect of survey knowledge, the ability to identify a shortcut that was not explicitly taught. One additional study has been reported that involved presenting survey knowledge in the form of sketch maps to assist environmental learning (Meneghetti et al., 2017). These researchers found that although their participants with DS benefited from the presence of the maps they did so to a lesser degree than did TD children. In our study, we expand upon the available research by looking at the learning of survey knowledge following different levels of exposure to the environment and evaluating multiple aspects of survey knowledge.

One goal of the current investigation was to replicate and extend our understanding of survey learning performance of people with DS. Hence, we examined performance using multiple measures of survey knowledge under optimal conditions of environmental learning, where participants were required to learn two routes through a virtual environment prior to assessing survey knowledge. For Experiment 1 we selected a relatively small, predictable environment (i.e., 9 square city blocks). We then provided sufficient exposure that allowed all participants to learn two overlapping routes that traversed the full environment. Finally, we evaluated several different measures of survey knowledge after the two routes had been sufficiently learned. More specifically, whereas previous research conducted on people with DS have used finding shortcuts as the primary measure of survey knowledge, there are several additional ways that knowledge of the environment can be assessed. For example, survey knowledge is demonstrated when making direction or distance estimates and identifying maps of the overall environment, in addition to finding the most efficient route to a target (Blades, 1997; Montello, 1998).

The use of additional measures of survey knowledge may provide greater insight into the kind of survey knowledge encoded by people with DS. More specifically, finding a shortcut requires that individuals recognize similarities across routes that they directly encountered. Direction estimation requires a more abstract representation that requires an understanding of relations between objects in the environment that were not directly perceived. Choosing a bird's eye view map representation of the overall environment requires fully integrating the segments into a coherent whole over multiple experiences. Therefore, to assess survey knowledge we asked participants to find a shortcut, identify the direction of landmarks not currently visible from their location in the environment, and identify a bird's eye representation of the overall environment. Given the conditions of Experiment 1, the results would be expected to reveal whether or not participants with DS have the basic capacity to acquire survey knowledge and use that knowledge to navigate a small, predictable environment. If participants are better at using some aspects of survey knowledge relative to others, it may be possible to identify specific mechanisms and strategies that operate differently during survey learning for people with DS.

A second goal of the current investigation was to identify differences in the acquisition of survey knowledge across multiple exposures to the environment in TD children and people with DS. We were specifically interested in whether differences emerged under less optimal conditions of learning. Therefore, we created conditions that were less optimal for environmental learning in Experiment 2. The environment was both more complex and less predictable in overall layout in the second study. Our evaluation of ongoing learning was prompted by recent studies indicating that learning of survey knowledge can begin with initial exposures to the environment. Although early perspectives of wayfinding suggested that survey knowledge develops from route knowledge (e.g., Siegel and White, 1975), Montello (1998) has argued that survey representations develop more gradually and become more accurate over repeated exposures to novel environments rather than in distinct stages. Ishikawa and Montello (2006) demonstrated that individuals can develop at least some survey knowledge at their first exposure to a new environment. In that study, the researchers drove participants through two novel large-scale environments once a week for 10 weeks. The routes were connected but participants were not made aware of that connection until week 3. Participants learned the location of buildings on each route and were asked to complete several measures of survey knowledge, including pointing to the location of unseen buildings, making distance estimates, and drawing sketch maps of the environment. Ishikawa and Montello (2006) reported that individuals performed better than chance levels when sketching a map of the environment after their first exposure. This was interpreted to mean that some survey knowledge was being developed in conjunction with route learning, contrary to the strictly hierarchical view suggested by Siegel and White (1975). However, as indicated by the researchers, the use of heuristics may lead to better than chance performance, as many environments are structured similarly. Additionally, Buchner and Jansen-Osmann (2008) showed that distances between landmarks can support route learning (see also Latini-Corazzini et al., 2010). This provides some evidence that survey learning occurs concurrently with, rather than subsequent to, other kinds of environmental knowledge.

In Experiment 2, individuals with DS and a group of TD children approximately matched on non-verbal ability (MA) experienced an environment multiple times. Their ability to navigate the environment was examine after one, three, and five exposures. This allowed us to examine whether survey knowledge accrued at different rates for participants with DS relative to TD participants matched on non-verbal ability. Based on previous research (e.g., Davis et al., 2014) indicating a slower accrual of route knowledge by participants with DS, we expected that the participants with DS in Experiment 2 would exhibit slower acquisition of survey knowledge as well.

We also evaluated landmark learning in Experiment 2. Landmarks are integral to learning about the environment for navigation purposes (Presson and Montello, 1988). There has been a considerable amount of research demonstrating the benefit of landmarks to route learning and wayfinding (Walkowiak et al., 2015). However, the process of identifying landmarks is fairly complex. Research has shown that visibility, frequency among multiple contexts, and relation to decision points are all important factors influencing landmark selection

(Caduff and Timpf, 2008). Further, specific positions of landmarks at intersections can influence the utility of the landmark for reproducing a path versus navigating an alternative route such as a return path (Karimpur et al., 2016; Balaban et al., 2017).

Although our focus was on shortcut finding relative to finding a return path, it is reasonable to expect that difficulties with perspective taking and selection of landmarks at locations more relevant to the original path participants are shown compared to the shortcut test may impede identifying a shortcut. Further, persons with DS who exhibit difficulties with executive function processes that are responsible for visual perspective taking (Rowe et al., 2006; Lanfranchi et al., 2010) may be expected to have greater difficulties choosing paths from alternative directions than TD children. This difficulty would be compounded by difficulties in remembering landmarks in general, as would be predicted based on Davis et al. (2014).

EXPERIMENT 1

Experiment 1 was designed to investigate the type of survey knowledge acquired during route learning under relatively optimal conditions by people with DS compared to typically developing (TD) children with whom they were matched on level of intellectual functioning. Participants learned two routes through a simple, predictable virtual environment consisting of depictions of city streets. Once the routes were learned, they engaged in three new tasks that were presumed to assess different aspects of survey knowledge. First, they were asked to find a target in the environment using a shortcut route that they had not learned and was available only if they had integrated the two individual routes into a combined, connected representation of the environment. Second, they were asked to point to the location of an unseen object in the environment using straight line direction judgments. This measures the extent to which participants represented landmarks allocentrically – in relation to each other rather than in relation to the self – in their survey representation of the environment. Third, they were shown several depictions of the environment and asked to identify which depiction most accurately represented the environment they had just learned to navigate. This measures the extent to which their spatial knowledge includes an allocentric representation of the entire environment.

Method

Participants

The participant groups consisted of 12 adolescents/young adults with DS and 12 TD children. Participants were paid $5.00 for completing the tasks. The participants with DS were recruited from the University of Alabama Intellectual Disabilities Participant Registry and from local service providers. Parents or guardians confirmed a diagnosis of DS. The TD participants were recruited from local preschool programs. The groups were approximately matched on KBIT-2 Matrices Raw scores. The participants with DS performed slightly better on the KBIT Matrices (21.0: SD = 9.2 vs. 19.8: SD = 3.9, for DS and TD, respectively), although this difference was not significant (see section "Results"). However, the participants with DS were clearly more variable in performance on the KBIT-2 than were the TD children. The mean age of the participants with DS was 19 years and 2 months (SD = 26 months) and for the TD children was 5 years and 0 months (SD = 4 months).

Measures

Kaufman brief intelligence test-2

Raw scores on the KBIT-2 Matrices subtest were used to match groups on non-verbal ability. The Matrices subtest consists of a 2 × 2 or 3 × 3 grid of pictures with one element missing. Participants are asked to choose which one of five pictures best completes the grid. The KBIT-2 was selected because it has good reliability [between 0.87 and 0.91 based on split-half and 0.76 and 0.89 based on test–retest reliability for TD participants in the age range tested and participants with Intellectual Disability (Kaufman and Kaufman, 2004)]. Further, it correlates well with the Leiter-R (r = 0.62) in children with special needs (e.g., Scattone et al., 2012).

Virtual environment task

The virtual wayfinding task was created using the Valve Hammer editor version 4.1 and presented using Portal 2. The environment consisted of twelve square blocks along city streets. The environment was selected to be consistent with previous studies (e.g., Courbois et al., 2013), to allow for overlapping routes, and to ensure a reasonable likelihood that participants could learn the simple routes in a few exposures (see **Figure 1**). There were 13 landmarks spaced throughout the environment that participants would encounter along the routes to which they were exposed (see **Figure 2**). All of the landmarks were visible from the trained routes and were identified by the experimenter as they traveled the routes the first time. Within the environment we created two overlapping routes that included 4 turns each. These were the routes that participants were taught during the training phase. These are also shown in **Figure 1**. A third route, the Shortcut, was constructed to include one segment from Route 1 only, one segment from Route 2 only, and one segment used on both routes. One specific landmark, a green trash can, was located along Route 2 and was the target for the Shortcut route. This landmark was identified as an important landmark to remember as participants learned Route 2. All computer tasks in Experiment 1 were completed on a Dell Inspiron 7548 laptop with a 15.6″ monitor and a screen resolution of 1,920 × 1,080 pixels. All participants sat approximately 60–75 cm from the display.

Route learning errors

Participants received up to six trials to learn each route. Wayfinding errors were recorded for all trials. An error consisted of taking one step down an incorrect segment, whether by making a wrong turn or going straight when a turn was required. If participants made an error, they were verbally redirected back to the correct path by the experimenter to finish the trial. If participants completed the route successfully prior to making all six trials, they moved on to the next part of the procedure. However, we ultimately used only the number of errors across the first three trials for each route as our measures

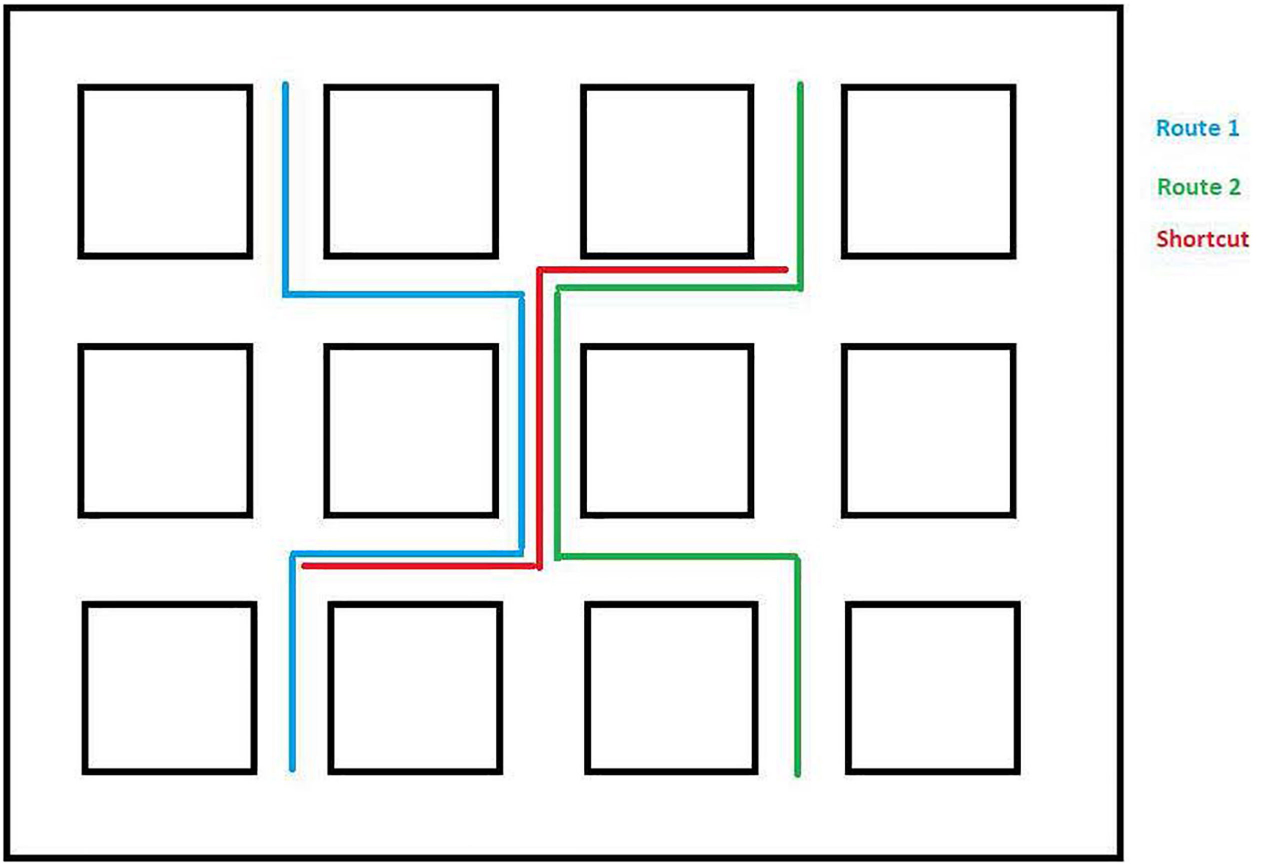

FIGURE 1 | Overview of Experiment 1 environment with learned routes and shortcut route depicted.

of wayfinding errors because more than half of the participants did not need the last three trials when learning Route 1 (7 participants in each group).

Shortcut navigation task

Following exposure to both routes, participants were placed at a location along Route 1 (see **Figure 1**), and asked to travel

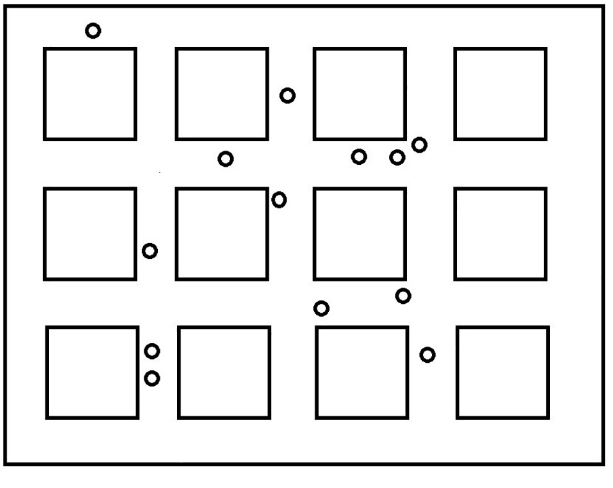

FIGURE 2 | Locations of landmarks used during the direction-of landmarks task.

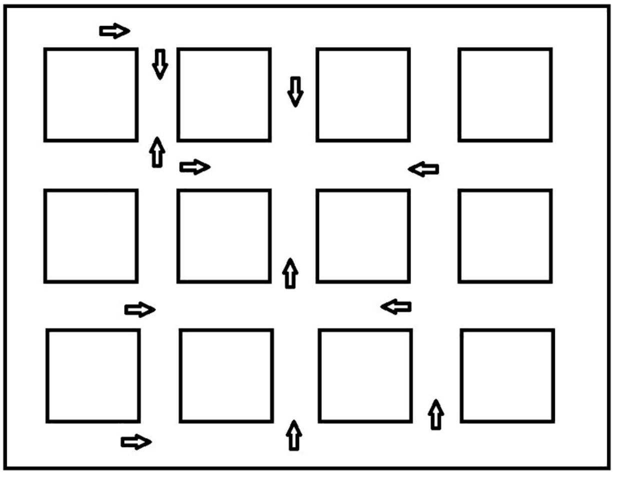

FIGURE 3 | Pointing locations and facing directions for the landmark directions task.

the shortest distance to get to the target (a green trash can) that was along Route 2. The target was specifically identified as the participants traveled Route 2 during training (see section "Procedure") to ensure they knew what the target was and where it was located. They were given one attempt at finding the shortest route. The number of segments traveled to reach the target was used as the dependent measure. The shortest possible route was 3 segments long. Note that there was an alternative to the shortcut (see **Figure 1**) that was also three segments long that did not include the overlapping segment. However, none of our participants took that route. Hence, this possibility was not considered in the analysis.

Direction of landmarks

Following the shortcut task, participants were placed at different locations along one of the routes and asked to locate various landmarks. **Figure 3** presents the different locations where the participant was placed in the environment. They were positioned to look straight ahead down a street and saw a scene that included 13 small black squares in a line across the screen (see **Figure 4** for the participant's view). The squares represented a range of approximately 45 degrees from the left to the right of the monitor. Squares were approximately 3.0 degrees of visual angle apart from each other from center to center. Participants were shown a picture of one of the landmarks and asked to point to the black square that was closest to where they thought the landmark would be from where they were standing, even if they could not see it. The first pointing trial was considered a practice trial and the landmark was visible from the participant's location. After each trial, the participant was invisibly moved to a new location (we told them they were transported), viewed a new scene with squares, and shown a new landmark to locate. We recorded which square the participant indicated. This was repeated for all 12 remaining landmarks. We wanted to be sure that participants understood the basic pointing instructions and were not simply responding randomly. We reasoned that they should be more accurate for visible landmarks if this was the case. Therefore, during testing, four landmarks were visible to the participant and 8 were not visible to the participant. We recorded the square that was identified by the participants as the one closest to the landmark. The dependent measure was divergence from the actual location of the landmark in terms of visual angle (3.0 degrees for each square from the target landmark). Then we averaged across trials to calculate an average degree of divergence for each participant. In a given trial, it was possible for maximum pointing errors to range from 0 degrees (if the target object was directly behind the selected square) to 36 degrees of visual angle (if the target object was at either extreme right or left location and the participant chose the opposite extreme). However, in most trials, the maximum pointing error was less than 36 degrees because the correct square was closer than the edge of the screen. After averaging the trials, an error of 27 degrees was the maximum score and 0 degrees was the minimum score. Completely random responding would result in an average error of 13.5 degrees.

Map recognition task

For the last measure, participants were shown five depictions of a bird's eye view of the environmental layout (see **Figure 5**). They were asked to select the picture that most closely resembled the

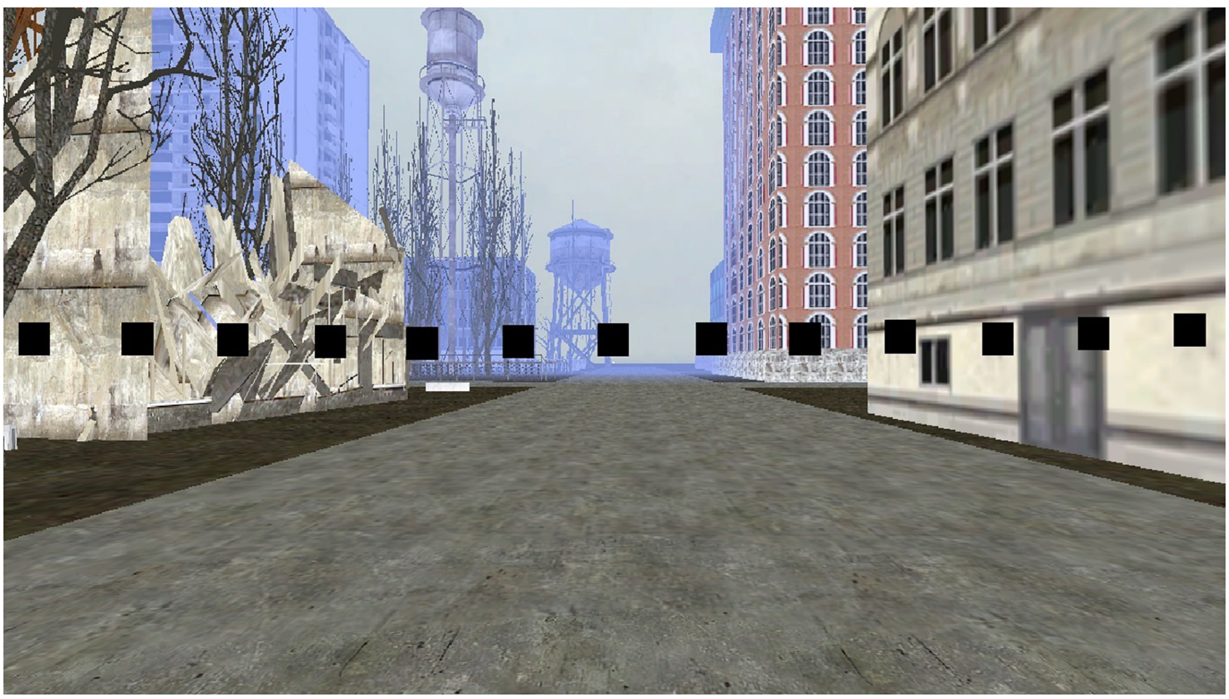

FIGURE 4 | Participant view in the direction of landmarks task.

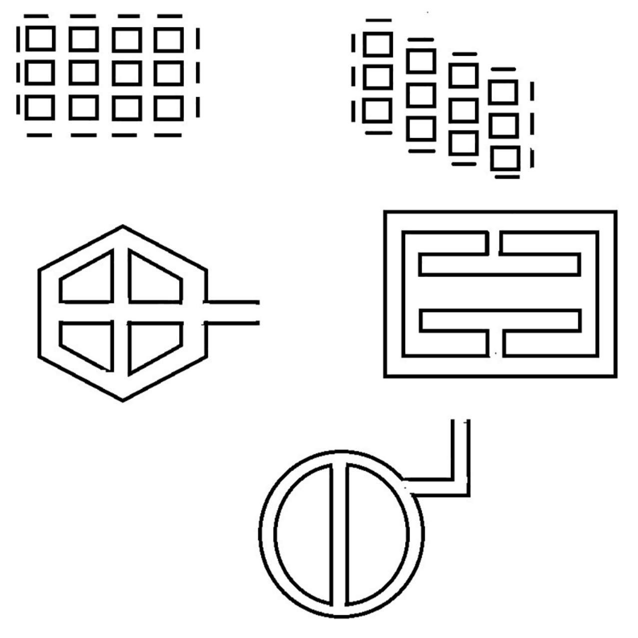

FIGURE 5 | Stimuli for the map recognition task. Correct figure in the upper left.

layout they have been navigating. We recorded whether or not the participant selected the correct picture.

Procedure

Parents or guardians gave consent and all participants gave assent. All participants were first administered the Matrices Subtest of the KBIT-2. Then they were presented the virtual environment. Following familiarization with the mouse controls, they were trained on the two overlapping routes. During the training phase, participants were first shown training Route 1 by the experimenter. This was experimenter controlled, rather than presented in video format, to allow the experimenter to refocus the participant's attention to the route if they appeared to look away during the initial viewing of the route. Then they were given three trials to retrace the path on their own. If they were unable to retrace the path after three attempts, they were shown Route 1 a second time and given three more trials to retrace the route. Following a maximum of six trials, they were switched to Route 2. However, if they were able to successfully retrace Route 1 without error, they were immediately switched to Route 2 without having to make all six attempts. Route 2 was presented in the same manner as Route 1. They were exposed to the route, given up to six trials to navigate the route successfully, then they were exposed to the route a second time if needed prior to achieving a successful attempt without error to a maximum of six trials. An error consisted of entering an incorrect segment. When an incorrect segment was entered during the route learning phase participants were told "This is not the way I went. Can you turn around and go the way I went?" this was repeated for all errors. The number of errors was recorded for each attempt. Landmarks used in the pointing task were identified as the participants were shown the routes. A green trash can was specifically identified as a landmark to remember when learning the second route. This was the target of the shortcut task.

Following a maximum of six trials to learn each of the two routes, the shortcut task was presented. Participants were placed at the beginning shortcut location on the first path (see **Figure 1**). They were told that "Someone dropped something important in the green trash can that you saw along the second path. Can you find the shortest way to the green trash can?" The number of segments traveled was recorded.

Following the shortcut task, participants completed the direction of landmarks task and the map recognition task in the order. The order of presentation was fixed to prevent participants from using information in the later tasks to perform the earlier tasks.

Results

KBIT-2

Primary data for Experiment 1 are presented in **Table 1**. A preliminary analysis of MA scores revealed that groups were not significantly different on measured KBIT-2 Matrices raw scores [$F(1,21) = 0.163$, $p = 0.690$, $\eta_p^2 = 0.008$]. However, because of the variability in KBIT-2 scores between groups, all analyses performed using standard Analysis of Variance procedures were followed by Analysis of Covariance using KBIT-2 scores as a covariate. No differences between any statistical comparisons were found using ANCOVA relative to ANOVA. Hence, only ANOVA results are reported.

Route Learning

Growth curve analysis was used to analyze the number of errors taken to find the target during the route learning task. This approach provides several benefits over a traditional repeated measures ANOVA for data that is clustered within an individual, including the ability to handle missing data and to account for individual variation in the statistical effects. For the purposes of this study, the main benefit of growth curve analysis is the ability to examine trends across the repeated trials. For a brief discussion of growth curve analysis, see Curran et al. (2010). A more detailed discussion can be found in Snijders and Bosker (2012).

We started with a basic model and then sequentially added the fixed effects and compared the new model to the previous model. The improvements on model fit were evaluated using -2 times the change in log-likelihood. Statistically significant improvements in model fit indicate that the model with the added variable better explains the observed results. If the change in model fit was not significant, we retained the original model. All model coefficients were estimated using maximum likelihood estimation using the lme4 (Bates et al., 2015) package in R.

The model we started with included a random intercept for each participant, a fixed effect of trial (coded 0–2), and a random effect for trial. The random intercept was necessary because of the repeated measures research design. A fixed effect of trial was included based on previous research (Ishikawa and Montello, 2006; Courbois et al., 2013) and theoretical evidence (Montello, 1998) suggesting that route learning improves across trials. Barr et al. (2013) argue that maximizing the random effects structure allows for better generalization of the results. Therefore,

TABLE 1 | Mean results from Experiment 1 for each Group and Task.

Group	Task[1]	Measure	Mean[2]
Down syndrome	Route learning (12)	Route 1 errors	
		Trial 1	2.4 (1.9)
		Trial 2	1.2 (1.4)
		Trial 3	2.6 (2.2)
		Route 2 errors	2.3 (4.0)
		Trial 1	1.5 (2.1)
		Trial 2	1.0 (1.2)
		Trial 3	2.0 (2.7)
	Shortcut (10)	Segments	4.5 (1.6)
	Direction of visible landmarks (10)	Degrees of divergence	7.5 (3.6)
	Direction of non-visible landmarks (10)	Degrees of divergence	8.4 (3.6)
	Map recognition (10)	# of participants choosing correctly	5
TD children			
	Route learning (12)	Route 1 errors	
		Trial 1	2.9 (2.2)
		Trial 2	1.9 (2.0)
		Trial 3	1.6 (1.4)
		Route 2 errors	
		Trial 1	1.4 (1.2)
		Trial 2	1.3 (1.6)
		Trial 3	1.7 (2.0)
	Shortcut (11)	Segments	6.8 (1.9)
	Direction of visible landmarks (12)	Degrees of divergence	7.5 (2.7)
	Direction of non-visible landmarks (12)	Degrees of divergence	11.7 (5.7)
	Map recognition (12)	# of participants choosing correct map	6

Map recognition reflects number of participants who selected the correct map. [1]Number of participants completing each Task in parentheses. [2]Standard deviation in parentheses.

we allowed the effect of trial to differ among participants as a random slope. By only allowing the effect of trial to vary among participants, we also ensured that all models compared have the same random effects structure. However, we also conducted the same model comparisons without using a random effect for trial. The results did not differ. A visual inspection of residual plots did not reveal any major deviations from the assumptions of our initial model.

We first tested the effect of route, which was dummy coded with the first route serving as the reference group. This effect was significant, indicating that participants made more errors when navigating route 1 (**Table 2**). We then tested the effect of group (DS and TD) on performance, which was dummy coded with DS as the reference group. This effect was not significant and was not included in the model. We also tested all two-way interactions and a three-way interaction, none of which were significant.

Although the groups showed no mean difference on the KBIT-2 Matrices subtest, the DS group was more variable than the TD group, so we also tested the fixed effect of non-verbal ability as a potential covariate. One participant with DS was given a mean replacement score because of missing data. That test was not significant. Further, it could be that changes across trials are not linear, so an orthogonal polynomial for trial was created to test the quadratic trend. The quadratic trend was not significant. The results of the likelihood ratio tests are presented in **Table 2**.

The final model included fixed effects of trial and route. The estimates for the fixed effects of the final model are presented in **Table 3**. The individuals' intercepts varied with an SD of 0.97 and the fixed effect of trial varied across individuals with a SD of 0.03. This indicates that little variability in route learning was explained by differences in the effect of trial. The SD of error not accounted for in the study was 1.52. These results suggest that learning errors decreased across trials and that this negative linear trend did not differ between the two routes or between the DS and TD groups. In addition, learning errors for Route 1 was higher than for Route 2; this difference was the same for all three trials and for both groups.

Survey Learning

Correlations among survey learning tasks and the KBIT-2 are presented in **Table 4**. The only correlation that reached statistical significance ($r = -0.592$, $p = 0.01$) was the association between KBIT-2 scores and performance on the pointing task. However, all other correlations were in the expected direction. Specifically, each measure of survey learning was associated with better performance on all other measures of survey learning.

Shortcut navigation

The analysis of shortcut learning was conducted using a One-Way between-subjects ANOVA with Group (DS and TD) as the independent variable and number of segments traveled to reach the target as the dependent variable. The analysis indicated a significant effect of Group, $F(1,19) = 8.894$, $p = 0.008$, $\eta_p^2 = 0.319$. The participants with DS walked fewer segments to get to the target than did the TD children. The significant main effect remained following an analysis with KBIT scores as a covariate. In addition, inspection of the data indicated that four participants with DS and none of the TD participants took the shortest route to the target, confirming that the DS participants as a group actually performed better than the TD participants on the shortcut task.

Direction of landmarks

The analysis of the Direction of Landmarks performance was conducted using a Group × Visibility (landmark visible vs. landmark not visible) mixed effects ANOVA, with Visibility treated as a within-subjects variable. Because we were not interested in responses to individual trials, we averaged the errors across landmarks by Visibility. In this case, multiple analytical approaches would be appropriate. We settled on a mixed effects ANOVA, as opposed to the modeling approach used above, because the two tests would produce similar results, but the ANOVA focuses on the "average" effect across participants in each group. The main effect of Visibility was significant, $F(1,20) = 5.35$, $p = 0.0314$, $\eta_p^2 = 0.211$. However, neither the main effect of Group, $F(1,20) = 1.218$, $p = 0.283$, $\eta_p^2 = 0.057$, nor

TABLE 2 | Likelihood ratio tests for growth curve model comparisons.

	Model fitting criteria	Likelihood ratio tests			
Fixed effects in model	**AIC**	**−2 log-likelihood**	**χ^2**	***df***	***p*-value**
Experiment 1					
Trial (linear)	413.44	401.44	–	–	–
Trial (linear) + route	408.28	394.28	7.16	1	0.007
Trial (linear) + route + group	410.07	394.08	0.20	1	0.651
Trial (linear) × route + group	410.65	392.64	1.63	2	0.443
Trial (linear) + route × group	411.94	393.94	0.34	2	0.844
Trial (linear) × route × group	414.96	390.96	3.31	5	0.652
Trial (linear) + route + KBIT	409.83	393.84	0.44	1	0.506
Trial (quadratic) + route	407.64	391.64	2.63	1	0.105
Experiment 2					
Trial (linear)	152.56	140.56	–	–	–
Trial (linear) + group	149.78	135.78	4.77	1	0.029
Trial (linear) × group	146.86	130.86	4.93	1	0.026
Trial (quadratic) × group	142.21	122.21	8.65	2	0.013
KBIT + trial (quadratic) × group	144.10	122.10	0.10	1	0.748

TABLE 3 | Fixed effects for the growth curve models.

Experiment 1				Experiment 2			
Term	***b (SE)***	***t***	**CI**	**Term**	***b (SE)***	***t***	**CI**
Intercept	2.40 (0.33)	7.31	1.75–3.05	Intercept	4.63 (0.07)	64.31	4.49–4.77
Trial (linear)	−0.39 (0.20)	−1.96	−0.79–0.01	Trial (linear)	−0.11 (0.54)	−0.21	−1.17–0.87
Route 2	−0.89 (0.32)	−2.83	−1.57–0.26	Trial (quadratic)	−1.26 (0.49)	−2.58	−2.15 – −0.29
				TD group	−0.25 (0.11)	−2.32	−0.46 – −0.03
				Trial (linear) × TD group	−1.87 (0.81)	−2.31	−3.43– −0.23
				Trial (quadratic) × TD group	2.16 (0.72)	3.02	0.74–3.55

CI = 95% bootstrap estimated confidence intervals.

the two-way interaction, $F(1,20) = 1.255$, $p = 0.276$, $\eta_p^2 = 0.059$, was significant. Both groups located the visible landmarks more easily than they did the not visible landmarks (7.5 degrees vs. 9.9 degrees divergence, respectively). However, the groups did not differ from each other. The performance of both groups was also significantly better than would be expected if they were responding in random fashion: $t(20) = 6.33$, $p < 0.001$, $d = 1.06$ for the visible and $t(20) = 4.84$, $p < 0.001$, $d = 1.42$ for the not visible landmarks.

Map recognition

The analysis of Map Recognition performance was conducted using two Chi Square analyses. First, we conducted a Test of Independence to determine whether the groups differed in their ability to select the most accurate map representation. As expected from visual inspection of the data, the groups did not differ, χ^2 (1) = 0.0063, $p = 0.94$. Second, we conducted a Goodness of Fit test to determine if the performance of our participants (both groups combined) differed from chance performance (i.e., choosing the correct response at a rate of greater than 20%). The results of this analysis yield a significant effect, $\chi^2(1) = 12.68$, $p < 0.0004$. Hence, overall the participants performed above chance on the map recognition test, but groups did not differ.

Discussion

The results of the Experiment 1 indicated that the participants with DS performed no worse on any of our measures of route or survey learning than did the TD children. This was a little surprising given that Davis et al. (2014) found the people with DS performed below expected levels on route learning relative to mixed ID participants and to TD children. However, in our current study the participants with DS committed numerically fewer errors on several individual trials across both routes and traveled significantly fewer segments to reach the target in the shortcut task.

TABLE 4 | Correlations among survey learning tasks in Experiment 1.

Measure	**1**	**2**	**3**
(1) KBIT			
(2) Shortcut	0.186		
(3) Map recognition	−0.022	−0.140	
(4) Not visible	−0.592*	−0.218	−0.147

*KBIT, KBIT-2 matrices subtest raw score; not visible, direction of landmarks task for landmarks not visible on screen; *p = 0.01.*

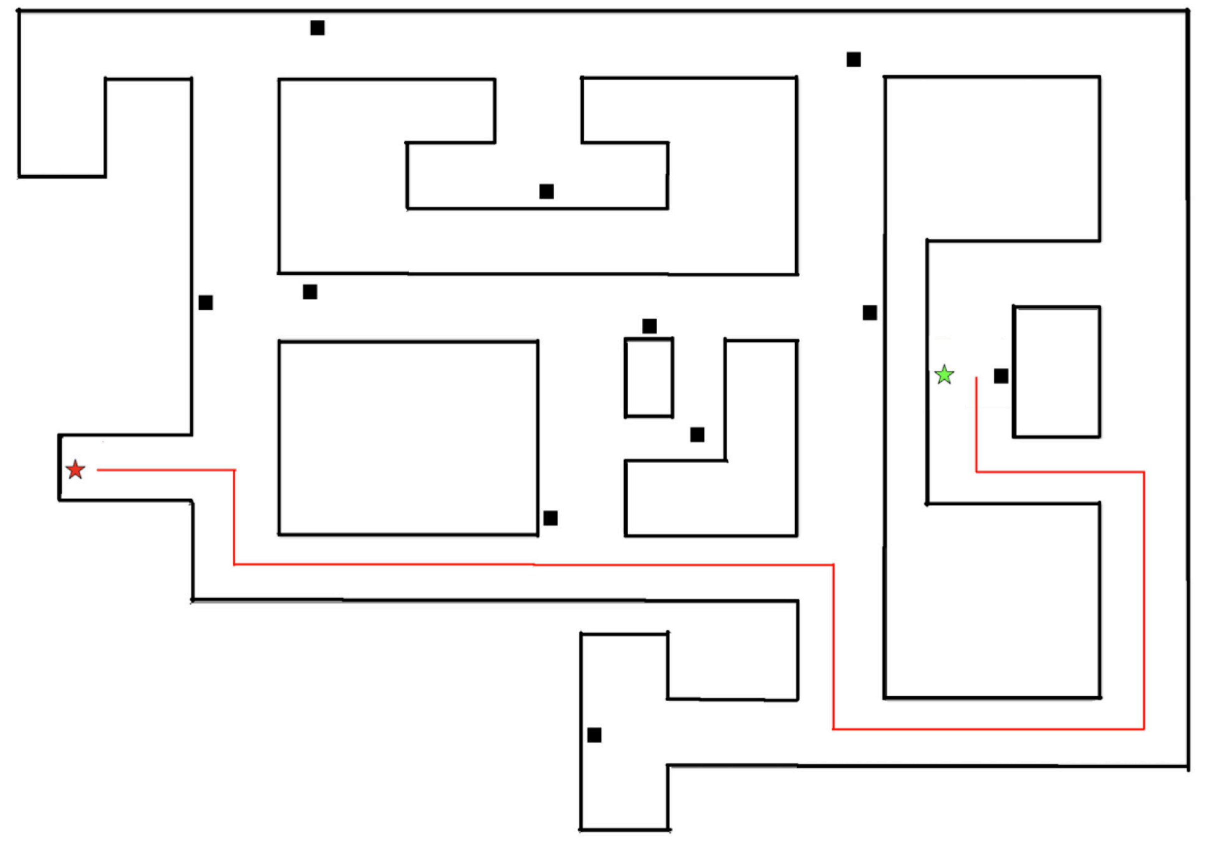

FIGURE 6 | Overview of Experiment 2 environment. The target is depicted by a red star and the starting point for the shortcut trial is depicted by the green star. The fastest possible path to the target is depicted by the red line.

One reason for this discrepancy is likely that we created environments that were easier for all participants to learn so we could focus on survey knowledge. This provided participants with an optimal opportunity to demonstrate survey knowledge following the learning of the routes. If they were unable to learn the routes, then they would not have been able to access very much survey knowledge. It would be interesting to identify the mechanisms of wayfinding responsible for magnifying group differences as environments get more complex. There are certainly a number of plausible personal factors (e.g., spatial working memory, sequence memory, route integration, etc.). Nevertheless, our data indicate that survey learning need not be a deficiency in DS relative to TD children under optimal learning conditions using simple and predictable routes. Purser et al. (2015) also found that non-verbal abilities could explain many of the differences between people with DS and TD children on numerous spatial tasks. It may be that our choice to match on non-verbal ability limited the likelihood of observing differences in performance on our wayfinding measures. The choice of matching criteria is always important. In this case, we wanted to know if wayfinding skills present a unique problem for people with DS relative to other non-verbal abilities. At least for the conditions of Experiment 1, where we used a small, predictable environment, the answer appears to be no. Further, this was true for multiple measures of survey knowledge.

One limitation of our results may be that the sample size was relatively small. This is especially problematic when assessing the correlations among survey learning tasks. All of the correlations were in the expected direction, but few reached statistical significance. We feel confident in concluding that the tasks did indeed measure the same underlying construct, survey knowledge, even if the correlations were largely inconclusive. It could also be argued that there were few significant differences between groups on the wayfinding tasks because of a lack of statistical power. However, given the current results, it is not clear that adding participants would increase the possibility of obtaining a significant result in favor of the TD children. In fact, the performance of the participants with DS was numerically better than the TD participants on several measures. That is, the difference was in the opposite direction as is typically found. Although the participants with DS performed better than the TD participants on the shortcut learning task, there was insufficient evidence to identify group differences on any of the other tasks. For this reason, we are most comfortable in concluding that our participants with DS performed at least as well on our measure of route learning and most measures of survey knowledge as the TD participants. However, it is interesting that the participants with

DS outperformed the TD participants when navigating using a shortcut, at least under optimal learning conditions. Still, it will be necessary to replicate these results using multiple samples and methods for verification, and to identify the variables that may have led to this particular result.

EXPERIMENT 2

Experiment 2 compared individuals with DS to TD children on measures of survey learning and landmark recognition over multiple exposures to the environment without first requiring them to learn individual routes. The groups of participants were approximately matched on non-verbal ability using the KBIT-2 Matrices subtest (Kaufman and Kaufman, 2004). An experimenter navigated a circuitous path through a virtual environment five times while participants watched. All presentations began and ended at a specific target (i.e., the only door in an office building). After the first, third, and fifth presentation of the environment, participants were tasked with finding the shortest path possible to a known target. Distance traveled at each trial served as a measure of survey learning. Consistent with past research (Farran et al., 2015; see also Courbois et al., 2013), we expected that participants with DS would travel a longer distance to find the target than would the TD participants. We also expected that participants with DS would recognize fewer landmarks.

Method

Participants

The participant groups consisted of 20 adolescents/young adults with DS and 17 TD children. Participants were paid $5.00 for completing the tasks. The participants with DS were recruited from the University of Alabama Intellectual Disabilities Participant Registry and from local service providers. Different service providers were recruited to limit targeting the same participants in each study. Nevertheless, three participants with DS were included in both studies. Because the time between studies was greater than 8 months for these participants and because the studies involved very different environments and procedures, we determined that practice effects across studies would be minimal. Further, analyses conducted with and without the three overlapping participants yielded results that were largely identical. Therefore, except as noted only the analyses including all of the participants are presented below. Parents or guardians confirmed a diagnosis of DS. The TD participants were recruited from local preschool programs. The mean age of the participants with DS was 19 years and 8 months (SD = 37 months) and for the TD children was 5 years and 5 months (SD = 11 months). The groups were poorly matched on gender (DS: 4 females; TD: 13 females). Two TD participants chose not to complete the landmark recall task, resulting in 15 TD participants for that test.

Measures

Kaufman brief intelligence test-2 – matrices

All participants completed the Matrices subtest of the KBIT-2.

Survey-learning task

Participants were tasked with finding the shortest path to a target in a virtual environment (**Figure 6**). The environment was constructed using the FPSCREATOR software. The environment was modeled after a typical office building and contained nine thematically appropriate landmarks. There were nine unique landmarks, including a door (the target of the short cut task), small blue cabinet, conference table, computer workstation, large black cabinet, painting on wall, water cooler, flip chart, desk, and chair. In addition to these nine unique landmarks, there were three identical couches that appeared. This was done to better mimic real-world environments, in which not every landmark is unique. The specific environment was chosen after pilot testing with typical adults and young adults with intellectual disability but not Down syndrome demonstrated that the environment was unlikely to result in ceiling or floor effects. All computer tasks in Experiment 2 were completed on an Acer Aspire 5253 laptop with a 15.6″ monitor and a screen resolution of 1,280 pixels × 1,024 pixels. All participants sat approximately 60–75 cm from the display.

All participants were able to familiarize themselves with the navigational controls prior to beginning the task. The experimenter presented the environment by navigating along a circuitous path that began and ended at the same landmark in the environment. The participant was placed in the environment at a location along the path taken by the experimenter (see **Figure 6**) and tasked with finding the shortest possible path to the target (Trial 1). Once the participant found the door, the experimenter presented the environment two additional times and participants were again placed at a starting point and traversed to the target (Trial 2). The experimenter again presented the environment two times, followed by the participants' task of finding the shortest path (Trial 3).

The environment was constructed by connecting a discrete number of equal sized blocks, which appeared to participants as a continuous environment. The total distance traveled was measured by counting the number of blocks traversed on each trial. The shortest possible path to the target was 47 blocks. If participants successfully navigated the shortest path to the target on the first or second trial, no additional trials were completed.

Landmark recognition task

Following the survey learning task, participants completed a measure of landmark recognition. Participants were shown two potential landmarks side-by-side. One was a landmark from the environment and the other was a similar object that did not appear in the environment. All pictures were shown from the same perspective that the object would have been viewed during the experimenter's presentations of the environment. Participants were asked to point to the object that they had seen in the virtual environment. There were nine unique trials presented in random order for each participant. The dependent variable was the total number of landmarks correctly recognized.

Procedure

Parents or guardians gave consent and all participants gave assent. Participants completed the KBIT-2 Matrices subtest,

TABLE 5 | Descriptive statistics for Experiment 2 (means with standard deviations in parentheses).

Group	KBIT	Trial 1	Trial 2	Trial 3	Landmarks
DS (*n* = 20)	14.1 (5.1)	108.9 (59.2)	137.4 (69.6)	105.2 (56.7)	6.1 (1.7)
TD (*n* = 17)	15.3 (4.5)	118.1 (54.3)	76.5 (36.2)	73.7 (31.5)	7.5 (0.9)

KBIT, KBIT-2 matrices subtest raw score; Trial, distance traveled in the survey learning task; Landmarks, landmarks successfully recognized.

then they were presented with the virtual environment. To navigate the environment, participants used a mouse to look around and the 'w' or 'up-arrow' key to move forward. To become better acquainted with the controls, a researcher demonstrated how to look in each direction and move forward, then the participant mimicked the behaviors of the researcher. The participants were then tasked with navigating a short 'L' shape, then turning around and going back without researcher assistance.

After explaining the instructions for the task, an experimenter presented the environment to participants by navigating a circuitous path along the environment, pausing to look at and naming each landmark in the environment. Further, the experimenter paused at each choice point and looked both directions while verbalizing the action to participants (e.g., "I am going to stop and look right, then look left. I am going to go this way."). The experimenter's path began and ended at the only door in the environment, which served as the target that participants were to find. The presentation of the environment was again experimenter controlled to allow the e experimenter to refocus the participant's attention as needed during the presentation. The shortcut task was then completed by participants. The experimenter traversed the environment two additional times, then the participants completed the second trial of the shortcut task. The experimenter traversed the environment twice more and the participant completed the third trial of the shortcut task. If a participant found the target in the fewest number of blocks, then no more trials were completed. Following the maximum number of three trials, the landmark recognition task was presented. The total procedure took approximately 45–60 min.

Results

KBIT-2

Descriptive statistics for the primary variables are presented in **Table 5**. A preliminary analysis of KBIT-2 Matrices Subtest scores revealed the groups were not significantly different on measured non-verbal ability [$F(1,35) = 0.62$, $p = 0.436$, $\eta_p^2 = 0.017$]. Qualitatively, data from the shortcut learning task revealed that one participant with DS reached the target in the shortest possible distance, and did so on the first trial. In addition, six TD participants found the target in the shortest possible path, with two participants doing so on the second trial and four on the third trial. Quantitative analyses were conducted on distance traveled to evaluate changes in ability locate the target location as a function of increased exposure to the environment.

Survey Learning

Growth curve analysis was used to analyze the distance (number of blocks) taken to find the target over the course of the three trials in the survey learning task. We used the same modeling approach used in Experiment 1. Specifically, we started with a basic model and then sequentially added fixed effects and compared the models. The model we started with included a random intercept for each participant, a fixed effect of trial (coded 0–2), and a random effect for trial. Our reasoning for starting with this model is the same as in Experiment 1. We conducted the model comparisons without a random effect of trial and the results remained consistent. A visual inspection of residual plots revealed deviations from the assumption of normality in our initial model. Therefore, a natural log transformation was used on the distance variable. A visual inspection of the new residual plots did not reveal major deviations from homoscedasticity or normality. The model statistics presented below are for the transformed data.

We first tested the effect of group (DS and TD) on performance, which was dummy coded with DS as the reference group. This effect was significant, meaning that there was a difference in performance between the two groups, and was included in the model (**Table 2**). We then tested a trial by group interaction, which was also significant. Specifically, the TD participants improved their performance across trials, but the participants with DS showed little change. However, we had no reason to assume that the change across trials should be linear. An orthogonal polynomial for trial was created to test the quadratic trend and a trial by group interaction was tested again. This model was significant, indicating that the quadratic trend was a better fit to the data. Finally, although the groups showed no mean difference on KBIT-2 Matrices subtest, we tested the fixed effect of non-verbal ability as a potential covariate. That test was not significant and not included in our final model. The results of the likelihood ratio tests are presented in **Table 2**.

The final model included an orthogonal quadratic effect of trial, group, and an interaction between the terms as fixed effects. The estimates for the fixed effects of the final model are presented in **Table 3** and a graph of the interaction is presented in **Figure 7**. The individuals' intercepts varied with an SD of 0.29 and the fixed effect of trial varied across individuals with a SD of 0.13. The SD of error not accounted for in the study was 0.36.

When we reanalyzed the data without the three overlapping participants from Experiment 1, the results were largely similar but contained one difference. Specifically, trial by group interaction was no longer statistically significant ($p = 0.104$), though the *AIC* did suggest an improved model fit when the interaction term was included. The quadratic trial by group interaction did provide a statistically significant ($p = 0.008$) improvement to model fit compared to a model without an interaction, though, so we retained the same model in both sets of data.

Landmark Recognition

The data for number of landmarks correctly recognized did not meet the normality or homogeneity of variance assumptions for a parametric test. Therefore, a non-parametric test was used. The

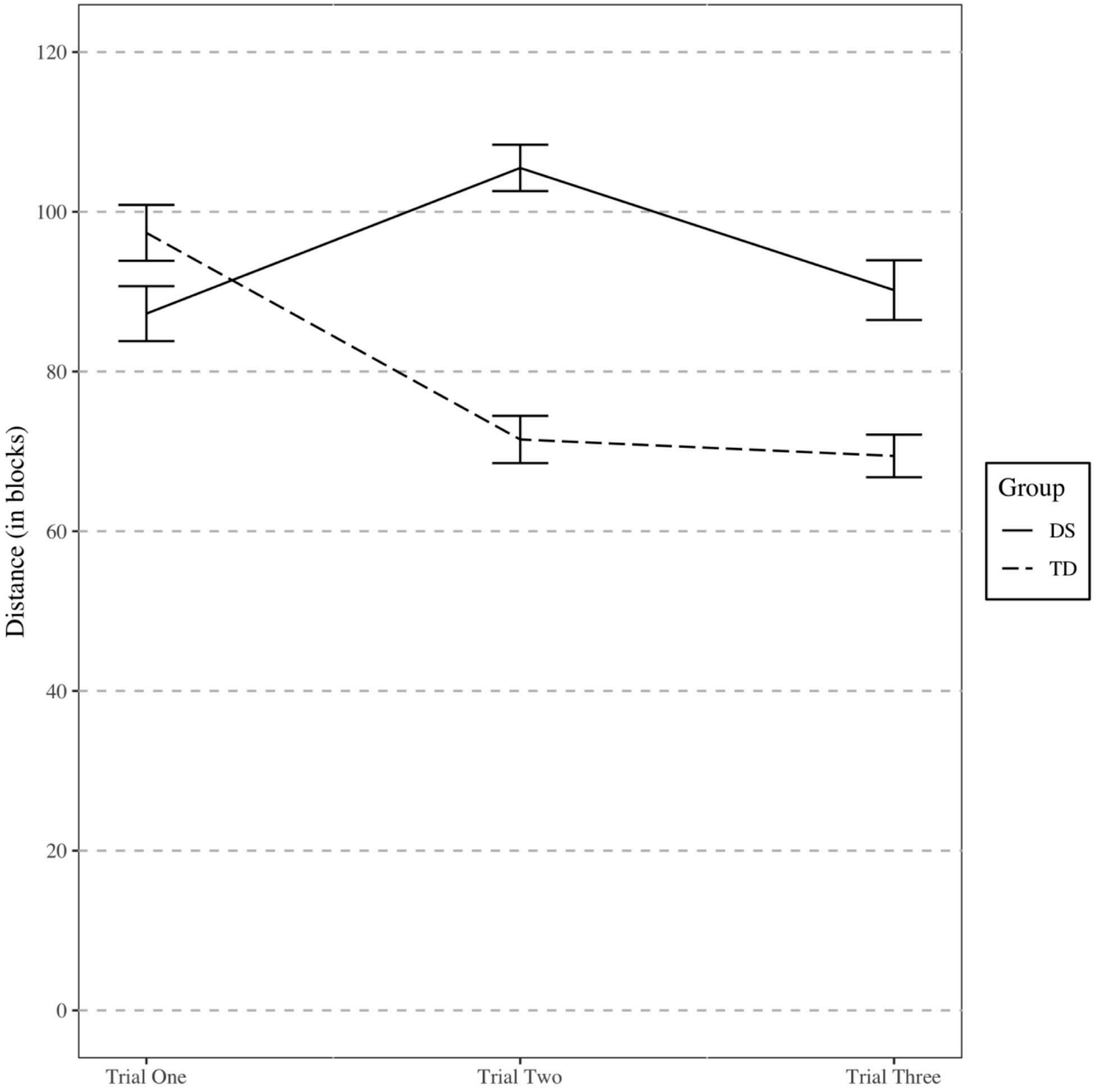

FIGURE 7 | Distance traveled during Experiment 2 shortcut task. Error bars represent one standard error.

median number of landmarks recalled in the DS (median = 6, $n = 20$) and TD (median = 8, $n = 15$) groups differed significantly (Mann–Whitney $U = 69$, $p = 0.006$, Cliff's $d = -0.588$).

Discussion

The results from Experiment 2 are consistent with the hypotheses and previous research. Specifically, participants with DS were similarly able as TD children to find the shortest path to a target after their first exposure to an environment. On subsequent trials, though, the participants with DS were less able to find the target as efficiently as the TD children. Further, participants with DS recognized fewer landmarks. Hence, our results are consistent with previous research in demonstrating a relative weakness in landmark memory (Courbois et al., 2013; Davis et al., 2014) and survey knowledge (Farran et al., 2015) in individuals with DS. Further, they demonstrate that survey knowledge develops more slowly over successive exposures to the environment for individuals with DS relative to TD children.

Interestingly, on the first trial, participants with DS did not travel a longer distance to find the target than TD. However, TD participants generally exhibited improved performance across repeated trials, whereas participants with DS generally did not. This resulted in better performance for TD participants in trials two and three. Thus, the pattern of results is suggestive of an inefficiency in accumulating survey knowledge through repeated exposures to an environment or in effectively using a survey representation to take a novel shortcut. This is consistent with the findings of Courbois et al. (2013), who found that participants with DS did not differ significantly from TD children in their first trial on a shortcut task. Unlike the current results, though, Courbois et al. (2013) also did not find a significant difference in the final trial whereas the current study did. This difference may be due to the relatively low number of participants that participated in that part of their study ($n = 16$).

A quadratic trend was found to match the data better than a linear trend. For the participants with DS, performance was better on the first trial than on the second trial. The descriptive statistics revealed much more variation in performance for participants with DS, beginning after the first trial. This could explain the pattern of getting worse on trial 2. The quadratic trend was also driven by the TD participants, who tended to much more learning from trial 1 to trial 2 than from trial 2 to trial 3. This pattern could indicate that these participants learned most of what they were able to learn within the first few exposures to a novel environment. Alternatively, it could be that the TD children did not understand the task in trial 1, but better understood what was asked of them by trial 2.

The current findings are consistent with an explanation that abnormal development of the hippocampal regions is associated with the observed weakness in survey learning. Specifically, the hippocampus (Maguire et al., 1998) and medial entorhinal cortex (McNaughton et al., 2006) have been associated with the formation of survey representations (see Burgess, 2008 for a review). The hippocampal region has been identified as being smaller relative to overall brain size in individuals with DS (Aylward et al., 1999; Pinter et al., 2001a) compared to TD individuals. Pennington et al. (2003) showed that individuals with DS perform less well on measures associated with hippocampal functioning than would be expected given their cognitive phenotype, suggesting that the neurodevelopmental abnormalities are accompanied by specific behavioral weaknesses.

We also found that participants with DS correctly recognized fewer landmarks than TD children. This finding is consistent with Courbois et al. (2013) and Davis et al. (2014). Individuals with DS seem to be less selective when attending to landmarks, resulting in them disproportionately focusing on fewer landmarks located at choice points (Davis et al., 2014). In the current study, there was no prescribed path that participants had to take. However, the individuals with DS may have not relied as much on information from landmarks, which may have made their performance less consistent across trials. Because we only measured landmark recognition after participants finished the shortcut task, we are not able to conclusively determine the relationship between landmark recognition and online survey learning. However, it is reasonable to believe that such a relationship exists and that being able to recognize landmarks from different perspectives plays an important role in identifying alternative routes to a target location (Karimpur et al., 2016).

GENERAL DISCUSSION

In two experiments, we observed similarities and differences in the acquisition of survey knowledge by people with DS and TD children with whom they were approximately matched on non-verbal MA.

In Experiment 1, we were able to identify a number of similarities in what types of survey information was acquired following explicit route learning in a simple environment. After learning two overlapping routes in the environment, the participants with DS exhibited learning that was at least as high as the TD participants on three different measures of survey knowledge: shortcut performance, identifying the direction of unseen landmarks from a designated location in the environment, and selecting a map of a bird's eye view of the environment. Hence, it appears that once the two routes for the environment were learned, the majority of the participants with DS were able to exhibit some general knowledge of the overall environmental layout. Further, the degree of knowledge exhibited by these participants was similar to what may be expected based on their level of general cognitive performance. Taken together, the two experiments suggest that even though the acquisition of survey learning may take more time when people with DS are simply exploring the environment, under conditions of explicit learning they can acquire at least some survey knowledge through the learning of overlapping routes. This is consistent with Courbois et al. (2013) who found that some participants with DS could identify a shortcut after learning routes in a similar simple environment.

Although several of the participants with DS could exhibit shortcut learning, their overall performance was likely much less than that of similar CA participants without intellectual disability (see Courbois et al., 2013). That result and our results still suggests important limitations on everyday wayfinding and navigation for people with DS. Only a third of our participants actually found the shortest route in the shortcut task, and only about half were able to identify a bird's eye view of the environment. Even performing at a level roughly the same as the MA matched TD children may not be particularly consequential. Indeed, the TD children evaluated in our study are likely just beginning to show the learning of survey knowledge themselves. For example, Cousins et al. (1983) found that tests of survey knowledge that involved estimating the positions of landmarks was late developing compared to route and landmark learning in a test of 7, 10, and 13-year-old children with only a portion of the oldest group effectively demonstrating this knowledge. However, it does appear that some survey knowledge can be learned by children as young as 4-years-old (e.g., Hazen et al., 1978; Huttenlocher et al., 2008). For example, Huttenlocher et al. (2008) found that children between 3 and 4 years of age were able to use a scale model to find an object in a larger scale environment. Hence, it is likely that both the participants with DS and TD children were exhibiting rudimentary survey knowledge at best. Further, while it may be expected that the TD children will gradually acquire additional abilities to represent survey knowledge with increasing CA and experience with environmental learning, the same outcome cannot be assumed for people with DS. They have already had much more general experience with the environment than have the TD children and are still exhibiting beginning level survey knowledge.

In Experiment 2, we found an important difference in that it took longer for our participants with DS to acquire survey knowledge relative to the TD children when they were exposed to the environment over a series of trials. We patterned this task to mimic being walked around the neighborhood and incidentally acquiring knowledge of the overall environment. The results clearly suggested that our participants with DS had more difficulty learning the environment under these conditions than did the TD children. It may be that people with DS are less likely to focus on information relevant to navigation unless explicitly told to do so when they experience an environment. This was true in spite of the fact that the participants were aware that a portion of the task was to navigate the environment after being led around. However, because we stopped the experiment after 5 exposures, it is not clear whether or not the participants with DS might eventually achieve a level of performance similar to that of the TD children. Further, we acknowledge the possibility that the complexity of the route may have adversely affected the participants with DS more than it did the TD children. We would need to replicate these results across different levels of complexity and different numbers of exposure to determine the generality of this conclusion.

With respect to survey learning in particular, we think the possible discrepancy between Experiment 1 and Experiment 2 may be more apparent than real. In Experiment 2, the focus was on how rapidly survey learning takes place for persons with DS. In Experiment 1, we asked about the kind of survey knowledge that could be accessed following learning under optimal conditions. Hence, a different pattern of results may be expected. Indeed, Experiment 1 suggests that survey knowledge of a learned environment may be similar for participants with DS and TD children. Experiment 2 suggests that, at least for longer and less predictable environments, it may take more time for the participants with DS to acquire that knowledge. One real possibility is that the larger and more unpredictable environment involved more visual processing resources to be completed than did the small environment of Experiment 1. Research has clearly demonstrated that visual working memory is important to constructing survey representations of the environment (see for example, Wen et al., 2013; Piccardi et al., 2019). Recent research has indicated that people with DS may have some weaknesses in some aspects of visual processing involving spatial memory and visuoconstructive tasks (Fidler, 2005). Hence, we might expect greater differences in tasks that require a greater use of these processes as in Experiment 2.

A big question that remains is whether it is possible to build on existing skills of survey knowledge acquisition as identified in Experiment 1 to promote better survey learning in people with DS. As noted in the introduction, there is considerable overlap between brain abnormalities reported in DS and those regions of the brain known to support wayfinding activities (e.g., Maguire et al., 1998; Aylward et al., 1999; Pinter et al., 2001a). To what degree do these differences constrain environmental learning in DS? One interesting feature of spatial ability performance is that spatial abilities appear to be relatively malleable across a range of ages and ability levels in persons without DS. In a meta-analysis of over 200 studies, Uttal et al. (2013) found that training and experience can produce positive and lasting effects in adults and children. For example, video game activities (e.g., Green and Bavelier, 2003; Spence and Feng, 2010), puzzle completion (Levine et al., 2012), and building activities (Coxon, 2012) promote the development of spatial abilities such as mental rotation, perspective taking, and spatial visualization in TD children and adults. To the extent that spatial abilities that are involved in acquiring survey knowledge of the environment are malleable in people with DS, then it may be possible to promote the acquisition of survey knowledge during environmental learning by people with DS. It is important for future research to explore this possibility.

CONCLUSION

Our conclusions must be considered in the context of some experimental limitations. We used TD children matched on non-verbal ability as a comparison group to evaluate the acquisition of environmental knowledge in people with DS. However, there are many differences between young adults with DS and TD children beyond just those directly associated with the DS genotype. For example, our participants with DS may have a wider range of experiences navigating unfamiliar environments, which may lead to the developing more efficient heuristics. These heuristics may work relatively well in the simple environment presented in Experiment 1, but not in the irregular and complex environment presented in Experiment 2. There are other differences between these young adults with DS and TD children that may also impact our results. It is important for future studies to use alternative comparison groups, such as other adults with intellectual disability, to help rule out certain confounds. Another limitation is that the study was conducted in a virtual reality environment. Navigation in the real world provides additional aids that are not available in virtual environments, such as proprioception (Waller et al., 2004), peripheral visual cues (Alfano and Michel, 1990), and other sensory cues (Chrastil and Warren, 2013). Many studies have shown an approximate equivalence between real world and virtual environmental learning (e.g., Coutrot et al., 2019), including for atypical populations (Claessen et al., 2016). However, it is still an open question whether individuals with DS have specific difficulties navigating in virtual environments.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by The University of Alabama IRB. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

EM, ZH, FC, BR, and YY contributed to design of research. EM, ZH, FC, and TR contributed to stimulus preparation and final execution of procedure. EM, ZH, FC, BR, YY, and TR contributed to data analysis and interpretation. EM, ZH, FC, BR, YY, and AR contributed to the final production of the manuscript. All authors contributed to the article and approved the submitted version.

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Peripersonal Visuospatial Abilities in Williams Syndrome Analyzed by a Table Radial Arm Maze Task

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Williams syndrome (WS) is a genetic deletion syndrome characterized by severe visuospatial deficits affecting spatial exploration and navigation abilities in extra-personal space.To date, little is known about spatial elaboration and reaching abilities in the peripersonal space in individuals with WS. The present study is aimed at evaluating the visuospatial abilities in individuals with WS and comparing their performances with those of mental age-matched typically developing (TD) children by using a highly sensitive ecological version of the Radial Arm Maze (table RAM). We evaluated 15 individuals with WS and 15 TD children in two different table RAM paradigms: the free-choice paradigm, mainly to analyze the aspects linked to procedural and memory components, and the forced-choice paradigm, to disentangle the components linked to spatial working memory from the procedural ones. Data show that individuals with WS made significantly more working memory errors as compared with TD children, thus evidencing a marked deficit in resolving the task when the mnesic load increased. Our findings provide new insights on the cognitive profile of WS.

Keywords: spatial exploration, spatial memory, ecological behavioral task, children, navigation abilities

INTRODUCTION

Williams syndrome (WS) is a rare genetic disorder, with a prevalence of 1 in 7,500–1 in 20,000 (Stromme et al., 2002), without gender preference, caused by a microdeletion on chromosome 7q11.23 (Ewart et al., 1993; Koehler et al., 2014).

WS has drawn the attention of cognitive neuroscientists as a result of an uneven cognitive profile with selective weak points in visuospatial abilities, and relative strength points in verbal abilities and face recognition (Atkinson et al., 2001; Bellugi and St George, 2001; Vicari et al., 2001; Searcy et al., 2004). In relation to their visuospatial deficit, WS individuals fail selectively on tasks requiring to decipher, judge, recall, and reconstruct the relationship between forms and objects (e.g., draw a house, replicate a block design, recall where an object was previously seen on a page, determine the orientation of a line; Bellugi et al., 1999; Mervis et al., 2000; Vicari et al., 2005; Martens et al., 2008; Farran et al., 2013; Broadbent et al., 2014a; Farran and Dodd, 2015).

In the last decade, behavioral studies based on large-scale spatial tasks elucidated how the visuospatial deficits of WS individuals influence exploration of large environments (Mandolesi et al., 2009a; Smith et al., 2009; Farran et al., 2010, 2012; Farran and Dodd, 2015; Farran et al., 2019; Foti et al., 2011; Broadbent et al., 2014b, 2015).

Importantly, the exploration and orientation in new and known environment represent spatial abilities needed in everyday life and, therefore, a prerequisite to autonomy and social integration. In the same way, it is important to be able to explore, know, and understand how peripersonal space is organized. These abilities allow to interact with objects by correctly interpreting what they are for. Hence, the study of spatial exploration proves relevant for the implementation of intervention programs in cognitive disabilities in general, and in WS in particular.

To explore an environment, the subject has to gain knowledge of the position of the environmental cues and of his/her own position with respect to these (O'Keefe and Nadel, 1978; Stupien et al., 2003). This type of spatial knowledge is referred as "declarative" (Jarrard, 1993). In wayfinding and navigation task, the declarative knowledge is mainly related to egocentric and allocentric encoding. Egocentric coordinates refer to the positions of environmental cues with respect to the subject, while allocentric coordinates represent the position of a cue in relation to another cue and then irrespective of the position of the subject (Arleo and Rondi-Reig, 2007).

On the other hand, the subject needs to understand how to move in the environment to reach (or avoid) specific cues (Mandolesi et al., 2009b). This type of knowledge is referred to as "procedural" (Foti et al., 2011) and in wayfinding and navigational tasks the procedural knowledge is highly correlated to exploration strategies.

It is well accepted that declarative and procedural spatial abilities are equally necessary for an efficient exploration and for the construction of a spatial map (O'Keefe and Nadel, 1978; Mandolesi et al., 2009b). Furthermore, these processes are strongly correlated with spatial working memory (Sorrentino et al., 2019). In conclusion, given its complex and multi-faceted nature, the study of spatial exploration requires taking into account all the spatial abilities involved.

Many researchers investigating spatial exploration in WS have mainly focused on the different facets of declarative knowledge, analyzing egocentric and allocentric encoding by means of wayfinding and navigational tasks (Bernardino et al., 2013). From these studies, it emerges that individuals with WS have difficulty to estimate the relation between landmarks and specific items within an environment (Farran et al., 2010; Broadbent et al., 2014a), and to employ a sequential egocentric strategy to guide the learning and retracing of a route (Broadbent et al., 2015). This evidence suggests a deficit in allocentric and egocentric encoding that could be explained by anatomical and functional alterations in the hippocampus (Meyer-Lindenberg et al., 2005), and by the well-documented deficit in the dorsal stream (Atkinson et al., 2001; Meyer-Lindenberg et al., 2005)—the neural pattern related to the link between perception and action.

Along with behavioral studies on WS, our research group mainly focused on the study of explorative abilities of WS individuals by means of two highly ecological walking spatial tasks: the Radial Arm Maze (RAM) and the Open Field with multiple rewards (OFmr). Both spatial tasks revealed the presence of procedural and memory deficits in WS (Mandolesi et al., 2009a; Foti et al., 2011).

While exploration abilities in WS have been extensively studied with the large-scale tasks, little is known about the exploration of peripersonal space in this syndrome. This space has a key functional role, as it is where all physical interactions with objects in the environment occur (Serino, 2019). The binding of visual information arising outside the body with tactile information arising on the body allows the representation of the space lying in between. This peripersonal space is often the theater of interactions with objects, supports self-location, contributes to bodily self-consciousness, and mediates higher-level cognitive functions (Serino, 2019).

The objective of the present study is to study whether the deficit in visuospatial information processing in the extra-personal space evidenced in WS individuals was also present during the exploration of the peripersonal space. Hence, in the present research, we evaluated the peripersonal visuospatial abilities in individuals with WS by using a table version of the Radial Arm Maze task (table RAM) and compared their performances with those of mental age-matched typically developing (TD) children.

MATERIALS AND METHODS

Participants

Fifteen individuals with WS (nine males and six females) and 15 mental age- and gender-matched TD children were recruited to participate in the study. All WS individuals (mean chronological age, 18.1 years ± 5.2) and TD children (mean chronological age, 6.5 years ± 0.5) were right-handed and native Italian speakers. Participants with WS were recruited at the Children's Hospital Bambino Gesù in Rome and at WS Association Marche in Fano (PU, Italy). Clinical diagnosis of WS was confirmed by genetic investigation (fluorescence *in situ* hybridization) demonstrating the deletion on the chromosome band 7q11.23. All participants live home with their families in Italy. The participants' cognitive level was measured using the short version of the Leiter-R intelligence scale (Roid and Miller, 2002). Mean mental age in the WS group was 6.2 years ± 0.8 and in the TD group was 6.5 years ± 0.6, whereas mean intelligence quotient (IQ) was 55.7 ± 7.3 and 103.5 ± 6.0, respectively. Overall, the groups differed in chronological age ($F_{(1,28)} = 73.9$, $p < 0.00001$, $\eta_p^2 = 0.73$) and IQ ($F_{(1,28)} = 386.4$, $p < 0.00001$, $\eta_p^2 = 0.93$), but not mental age ($F_{(1,28)} = 1.6$, $p = 0.22$, $\eta_p^2 = 0.054$).

Written informed consent was obtained from all the parents of the participants. The study had been approved by the Ethics Committee of Bambino Gesù Children's Hospital, Rome, Italy (protocol number 486 LB) and was carried out in accordance with the declaration of Helsinki.

Table Radial Arm Maze (table RAM)

The table RAM is made of a round central platform (5 cm in diameter). Eight green arms (3 cm wide × 25 cm long) depart the central platform like the spokes of a wheel (**Figure 1**). At the end of each arm, a small black round cap (1 cm in diameter × 2 cm height) covered the reward (a little colored wooden ladybug).

The table RAM was placed on a desk while the extra-maze cues (windows, paintings, posters, doors, and experimenter) were held in constant spatial relations throughout the experiment. The arms were virtually numbered clockwise, arm 1 being in front of the subject. Participants had visual access to the table RAM only during the experiment.

Although the participant is seated in front of the RAM, this task can be considered as a peripersonal visuospatial task because it forces the child to explore a portion of space accessible with the limbs.

Experimental Procedure

The table RAM task was presented as "Ladybug game." The child had to move the older sister ladybug ("Ladybug"), placed on the central platform, to find its sisters hidden inside the caps at the end of each arm.

To increase motivation, at the end of each trial the child received a reward (a coin) in exchange for the ladybugs discovered.

The children were evaluated in two different table RAM paradigms: the *free-choice paradigm*, to analyze the peripersonal procedural and memory components, and the *forced-choice paradigm*, to disentangle the components linked to spatial memory from the procedural ones. The choices made by the participants in each trial of both paradigms were videotaped and registered manually.

Free-Choice Paradigm

Each child could explore the eight arms freely to find the ladybugs hidden inside the caps at the ends of each arm. A trial was counted as successful when all eight ladybugs were collected. Afterwards, the child could keep the Ladybug for a short time to verify whether he/she was aware of having finished the trial. In other words, the experimenter observed whether the child revisited a previously explored arm to find a further ladybug in the upturned caps. After a supplementary incorrect visit, the child was informed that the game was over.

A trial ended when all eight ladybugs had been collected, when 20 choices had been made, or after 5 min from the start of the task. Since each cap contained one ladybug, the optimal performance entailed eight visits to the arms, that is, visiting each upturned cap only once.

If the subject visited the same arm twice during the same trial, it was considered as an error. All participants performed three trials in 1 day (one session). The inter-trial interval was at least 1 h long.

At the beginning of the first trial, the experimenter used the same simple verbal instructions to explain the task to each participant ("Now we will play a little game in which the Ladybug has to find its sisters hidden in these black upturned caps. On my mark, make the Ladybug move. Remember not to let it get out of the corridors, and always return it to the center. Then, put its sisters at the center so they won't hide anymore. Go and enjoy yourself!"). Immediately after the instructions were given, the participants started the task and no further instructions and verbal encouragement were provided. All participants displayed no hesitation when starting the task. In fact, all participants did not spend more than 1–2 s before moving the Ladybug on the central platform. The trial was void if the child made the Ladybug exit the maze. All participants ended the trials in the required time. To assess the subjects' performance in the free-choice paradigm, we evaluated the following parameters: *search efficiency*, defined as the percentage of correctly visited arms divided by the total number of visits; *the longest sequence of correctly visited arms* (the sequence ranged from 1 to 8); *error-free trials*, calculated as the number of trials without errors; *adjacent visits*, calculated as the percentage of visits in adjacent arms (i.e., visiting arm 3 and then arm 4) divided by the number of visits; *declarative mastery*, a binary index classifier of the participant's behavior at the end of the trial, defined as 0 if they wrongly continued the search, and as 1 otherwise.

Forced-Choice Paradigm

The day after the session of the free-choice RAM paradigm, the participants were tested in the forced-choice paradigm of table RAM. In the first phase, although all arms contained the ladybugs on them, only four arms (for example, arms 1, 3, 4, 7) were accessible because the remaining four arms were closed by a Lego cube (2 cm × 2 cm × 2 cm) at the proximal end of each arm. Different angles separated the opened arms to avoid the subjects reaching the solution through the employment of a procedural strategy, such as for example performing only adjacent angles. The table RAM task started with the Ladybug placed on the central area of the maze and the participant was allowed to explore the four open arms by moving the Ladybug to collect the four accessible ladybugs. Afterwards, the participant was invited to interrupt the game and she/he was kept in a separate place without seeing the game and chatting with the experimenter for 120 s before the second phase of the task started. In the second phase, the participant was allowed moving the Ladybug in all arms, but only the four previously closed arms were rewarded (since the other four ladybugs had been collected in the previous phase). The successes in visiting only the rewarded arms essentially depended on remembering which arms had already been visited, stressing the memory component and neglecting the search patterns.

Each participant performed three trials a day for two consecutive days (two sessions), with an inter-trial interval of at least 1 h. In each of the six trials, a different configuration of closed arms was used. The arms opened in the first phase were never contiguous nor separated by regular patterns of angles (e.g., the opened arms never were 1, 2, 3, 4, or 1, 3, 5, 7).

At the beginning of the first trial, the experimenter explained the task to each participant using the same simple verbal instructions ("Do you remember the Ladybug that had to find its sisters under the black upturned caps? Well, the mischievous sisters have hidden themselves again. However, they do not know

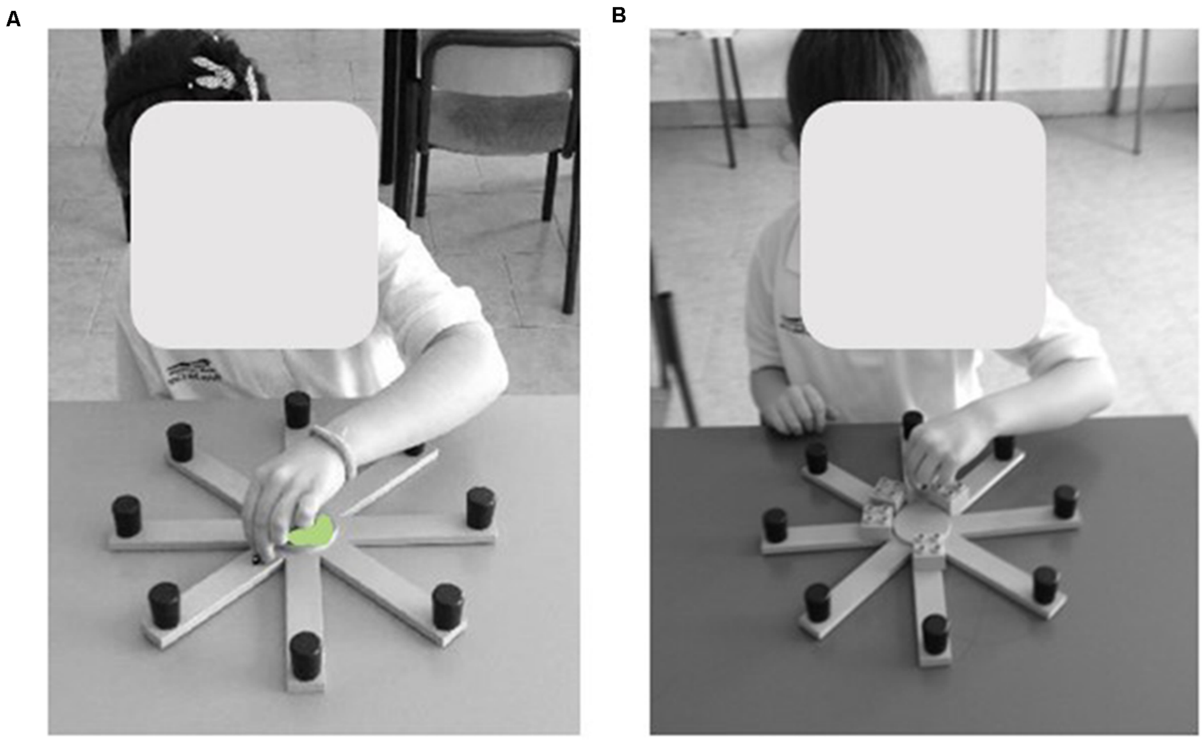

FIGURE 1 | Views of the table Radial Arm Maze (RAM). The figure shows two typically developing (TD) children performing the "Ladybug game" by means of table RAM in the free-choice **(A)** and forced-choice **(B)** paradigms of the task. Written informed consent was obtained from the parents of children for the publication of this image.

you're there to help the Ladybug find them. Now, some corridors are blocked. You have to let the Ladybug enter only in the arms that are open. Go!"). The verbal instructions after the 120-s interval were: "Uh! Something now has changed, there are no Lego bricks anymore. Then, the Ladybird can freely go and look for the other sisters! Good job!").

In the forced-choice paradigm, the parameter taken into account was the *short-term memory errors*, defined as the re-visits into already visited arms. This parameter was broken down further into two error subtypes: *across-phase errors*, defined as visits into an arm that had been visited during the first phase of the same trial; *within-phase errors*, defined as re-visits into an arm already visited in the same phase. We considered also *the longest sequence of correctly visited arms*. In this case, the sequence ranged from 1 to 4.

Statistical Analyses

All data were presented as the mean ± SD and were first tested for normality (Shapiro–Wilk's test) and homoscedasticity (Levene's test). When normally distributed, data were analyzed by using one-way or two-way analyses of variance (ANOVAs). When data were not normally distributed, non-parametric analyses (Mann–Whitney *U*, Wilcoxon's test) were used. The error-free trials and the declarative mastery were evaluated using χ^2 metric. Analyses were performed by using Statistica 8.0; the significance level was defined as $p < 0.05$. Giving the numerous analyses, controlling for the alpha inflation was needed. We controlled the proportion of type I errors among all rejected null hypotheses by setting the false discovery rate (FDR) to 0.05. The FDR was estimated through the procedure described in Benjamini and Hochberg (1995). In our results, the 0.05 level of significance reported was pre- and post-FDR correction.

RESULTS

Free-Choice Paradigm

Search Efficiency

The percentage of correct visits represents a general parameter that indicates the efficiency of processing of peripersonal space. Therefore, it can be considered as a parameter that expresses all the others in their entirety. Individuals with WS obtained significantly lower values of search efficiency than TD children (WS: median = 88.89, q_1 = 76.4, q_3 = 100; TD: 100, q_1 = 96, q_3 = 100; Mann–Whitney U = 68.5, $Z = -1.99$, p = 0.04, p_{FDR} = 0.049; **Figure 2A**).

Error-Free Trials

This parameter more precisely reflects the search efficiency, that is, the degree of correctness of the task. When the error-free trials were considered, the performance observed in WS and TD groups was significantly different. Indeed, the WS participants

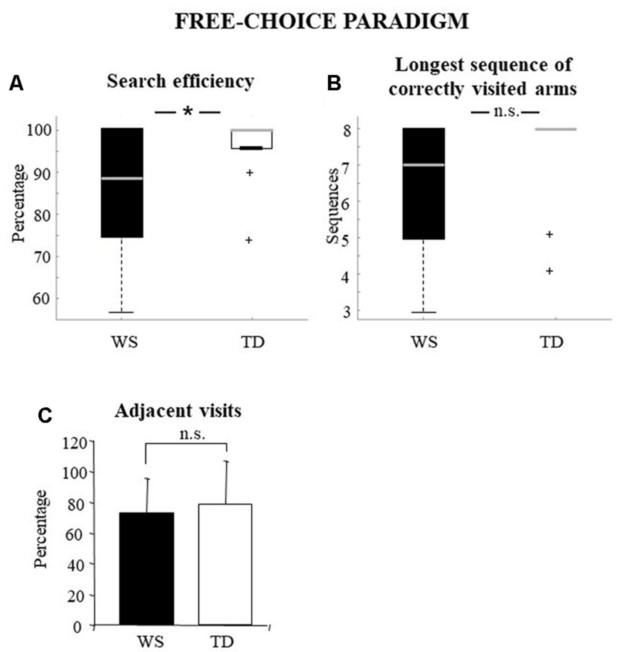

FIGURE 2 | Performances of the Williams syndrome (WS) participants and TD children in free-choice paradigm. Data are expressed as median and quartiles **(A,B)** and mean ± SD **(C)**. The asterisk indicates the significance level of Mann–Whitney *U* (*$p < 0.05$), +indicates outliers. ns = no significant difference.

performed a significantly lower number of error-free trials than TD participants (WS vs. TD: 6 vs. 12; $\chi^2 = 5$; $df = 1$; $p = 0.02$, $p_{FDR} = 0.027$).

The Longest Sequence of Correctly Visited Arms

This parameter indicates the longest sequence of correctly visited arms. High values could be obtained by exploiting the working memory and mapping abilities or by efficient explorative strategies. The longest sequence of correctly visited arms of WS and TD groups was not significantly different (WS: median = 7; $q_1 = 5$; $q_3 = 8$; TD: 8; $q_1 = 7.2$; $q_3 = 8$; Mann–Whitney $U = 78.5$, $Z = -1.53$, $p = 0.12$, $p_{FDR} = 0.132$; **Figure 2B**).

Adjacent Visits

The exploration of adjacent arms indicates how well the procedural strategy of research is put into action. High percentages indicate that the task is solved without (or with little) mnesic load and is correlated to the parameter reporting the longest sequence of correctly visited arms. The percentages of adjacent visits of WS and TD groups showed no significant difference (WS: $\bar{x} = 72.7 \pm 22.54$; TD: $\bar{x}$: 79.1 ± 28.09; one-way ANOVA: $F_{(1, 28)} = 0.42$, $p = 0.52$, $p_{FDR} = 0.520$, $\eta^2_p = 0.01$; **Figure 2C**).

Declarative Mastery

The awareness of having concluded the task represents a parameter correlated to the mapping abilities and/or to rules learned. A significantly higher proportion of WS individuals was not aware of having completed the task as compared to TD group ($\chi^2_{(df = 1)} = 2.16$; $p = 0.014$, $p_{FDR} = 0.022$).

Forced-Choice Paradigm

Short-Term Memory Errors

This parameter as a whole indicates a possible deficit in short-term memory processes without specifying the kind of the memory deficit. A one-way ANOVA was run on short-term memory errors performed in the second phase of the test when all the arms were opened and the participants could move the Ladybug without restrictions. Statistical analysis revealed that WS individuals made a significantly higher number of short-term memory errors than TD children (WS: = 4 ± 1.08; TD: $\bar{x} = 2.1 \pm 0.91$; $F_{(1, 28)} = 27.02$, $p = 0.00002$, $p_{FDR} = 0.00011$, $\eta^2_p = 0.49$; **Figure 3A**).

To better understand the kind of the memory deficit, we have analyzed the two subtypes of short-term memory errors: *across-phase errors* (visits into an arm that had been entered during the first phase) and *within-phase errors* (re-visits into an arm previously visited in the same phase). While across-phase errors reflect a short-term deficit, within-phase errors express a working memory deficit. Non-parametric analyses (Mann–Whitney) revealed that WS individuals made a significantly higher number of across-phase and within-phase errors compared with TD children (across-phase errors: $U = 31$, $Z = 3.38$, $p = 0.0007$, $p_{FDR} = 0.0001$; within-phase errors: $U = 38$, $Z = 3.09$, $p = 0.002$, $p_{FDR} = 0.004$). Moreover, non-parametric analyses (Wilcoxon's test) revealed that both groups made more across-phase errors than within-phase (WS: $Z = 3.29$, $p = 0.0001$, $p_{FDR} = 0.0004$; TD: $Z = 3.41$ $p = 0.0006$, $p_{FDR} = 0.002$; **Figure 3A**).

The Longest Sequence of Correctly Visited Arms

In this paradigm, this parameter ranged from 1 to 4. High scores in this parameter express a correct functioning of working memory processes. In fact, to prevent solving the task by using procedural strategies, in the second phase the correct arms were never contiguous nor separated by regular patterns of angles. WS individuals obtained significantly lower scores of the longest sequence of correctly visited arms than TD children (WS: $\bar{x} = 2.1 \pm 0.42$; TD: $\bar{x}$: 2.8 ± 0.24; one-way ANOVA: $F_{(1,28)} = 28.79$, $p = 0.00001$, $p_{FDR} = 0.0001$, $\eta^2_p = 0.51$; **Figure 3B**).

DISCUSSION

The present study focused on the analysis of the processing of the peripersonal space in individuals with WS by using a small-scale behavioral task that allows distinguishing memory components from procedural ones. In particular, we used a table version of the RAM that reproduces in small scale the classical walking RAM task (Overman et al., 1996; Mandolesi et al., 2009a). We tested WS individuals according to two RAM paradigms: the free-choice and forced-choice paradigms that allow to evaluate different facets (procedural and memory abilities) of the spatial function. In fact, while the free-choice paradigm allows to evaluate the possible presence of a global spatial deficit (without distinguishing procedural from memory abilities) and of impaired declarative abilities (by means of the declarative mastery parameter), the forced-choice paradigm allows to realize more precisely the nature (procedural or mnesic)

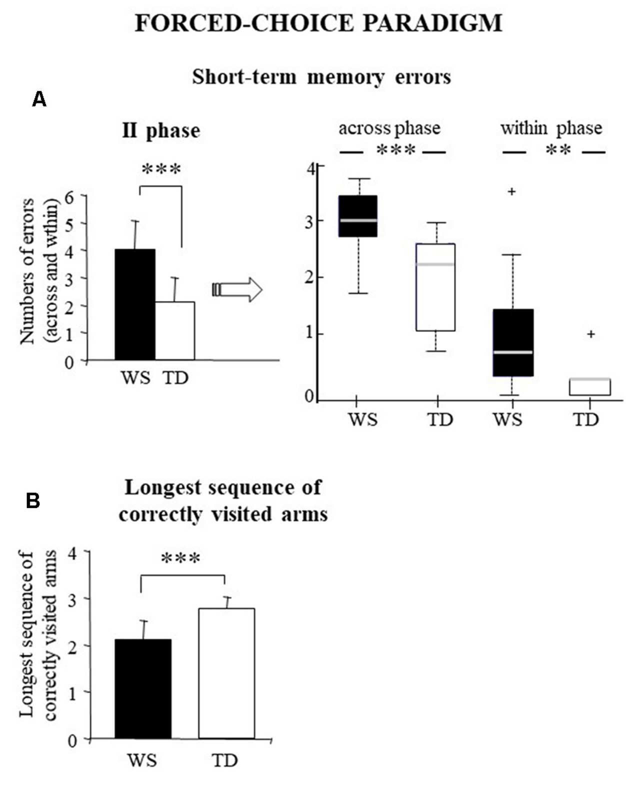

FIGURE 3 | Performances of the WS and TD participants in forced-choice paradigm. Data are expressed as mean ± SD: (left side of part **A** and **B**) and median and quartiles (right side of part **A**). The asterisks indicate the significance level of ANOVAs and Wilcoxon's test (**$p < 0.005$, ***$p < 0.00005$). +indicates outliers.

of the deficit, providing also the distinction between short-term and working-memory errors.

The main result of the present research is the deficit of WS individuals in the peripersonal space based mainly on impaired memory abilities. Such a memory deficit emerged clearly analyzing the performances of the WS individuals in the forced-choice paradigm that singles out short-term from working memory abilities. In fact, WS individuals made significantly more within-phase errors and obtained lower values of the longest sequence of correctly visited arms (**Figures 3A,B**) in comparison with TD children.

A memory deficit was evident, although more mildly, even in the free-choice condition, when WS participants obtained a success rate lower than TD children (**Figures 2A,B**). Again, when procedural abilities were considered, the WS participants performed similarly to the TD children. In fact, the two groups of participants made a comparable number of visits in adjacent arms (**Figure 2C**), thus putting in place an efficient search strategy. The procedural strategy of visiting adjacent arms is captured by the longest sequence of correctly visited arms, a parameter that in this paradigm was similar in both groups (**Figure 2B**). It is important to stress that, despite the WS individuals used exploration strategies similar to the TD children, unlike them, the WS participants did not realize that the task was finished (as indicated by the declarative mastery parameter), once again confirming the presence of a deficit in visuospatial information processing.

The data described in both our studies with walking RAM task (Mandolesi et al., 2009a) and table RAM task highlight that in WS the deficit in the elaboration of the allocentric space is different from that in the peripersonal space. While in the elaboration of the peripersonal space the WS individuals exhibited memory but not procedural deficits, in the elaboration of the allocentric space they exhibited, beside the memory deficit, also remarkable procedural deficits (Mandolesi et al., 2009a; Farran et al., 2012, 2016; Foti et al., 2013; Broadbent et al., 2014b). Such diversity of processing of near and far space may be explained by taking into account the sensorimotor and cognitive processes recruited as well as the neuronal circuitry involved in the two conditions. In fact, walking tasks involving the exploration of extra-personal space (as walking RAM task) require to process the proprioceptive, vestibular, visual information derived from signals related to locomotory movements, as well as path integration processes in which the self-motion signals are processed in conjunction with external location-based references.

Conversely, tasks involving the processing of body–objects interaction in peripersonal space (as table RAM task) imply not only low-level sensorimotor representations of the space around the different body parts (in the present case, mainly upper limbs), but also the coding of multisensory signals in reference frames, to which visual and proprioceptive signals on the body parts location in space strongly contribute. Notably, it has been reported that tactile and visual stimuli inside the peripersonal space elicit a stronger processing and induce a more powerful multisensory activation than stimuli outside the peripersonal space (Serino, 2019).

The spatial memory deficit exhibited by WS participants in the table RAM task finds correspondence in the deficits of WS individuals when performing the Corsi Block task or block construction tasks (Vicari et al., 1996; Jarrold et al., 1999; Farran et al., 2001; Hoffman et al., 2003; Farran and Jarrold, 2005; Sampaio et al., 2008).

In addition to the different processing of the information linked to self-motion signals or to body–object interaction, it has to be taken into account that, while in the walking RAM task, the participants are inside the maze and see it from inside, in the table RAM task, the participants see the whole maze from above. The different view of the maze (from inside or from above) triggers different mental processes related to the construction of the spatial map. In the first case (vision from inside/RAM walking task), the participant is compelled to build a spatial cognitive map of RAM to orient and move himself/herself in it. In this way, the declarative competence of a space is built through the procedural competence, as we previously showed (Mandolesi et al., 2003). In the second case (vision from above/table RAM task), the participant is facilitated in the construction of the spatial cognitive map because he/she sees the maze in its completeness; therefore, his/her declarative knowledge is promptly formed. This interpretation would explain why WS individuals do not show procedural deficits in the free-choice paradigm of the table

RAM task. In this line, it is intriguing to interpret the vision from above of the table RAM like an observation type that permits the observer to develop a sort of "perceptual blueprint" of the task to be learned (Bandura, 1977).

As last note, it is important to recall the functional role of dorsal stream in the spatial cognition (Ungerleider et al., 1998). fMRI studies showed that the dorsal stream projections to prefrontal, premotor, and medial temporal cortices through the posterior cingulate and retrosplenial cortices support spatial working memory, visually guided action, and navigation (Kravitz et al., 2011; Burles et al., 2018). Interestingly, in WS impairment of dorsal stream functionality, hypoplasia of the dorsal areas of the parietal cortex and weakening of fronto-parietal circuitry have been described (Bernardino et al., 2014). In the light of such an evidence, it is possible to hypothesize that in the resolution of the table RAM task a specific portion of the posterior cingulate cortex might be involved, linking thus the deficits of WS individuals to the vulnerability of the medial pattern of dorsal stream.

We are aware that a possible limitation of the present work concerns the relatively small sample size; however, it has to be considered that WS is a rare genetic disorder. In an attempt to obtain major consistency among performances, we carefully compared the WS individuals' performances with those of mental age-matched TD children.

Presenting the table RAM task as a game, we allowed the study of the searching behavior in peripersonal space in an ecological manner. Furthermore, the table RAM is an easy task, particularly suitable for clinical populations with evident deficits. However, despite its simplicity, the table RAM task allows the evaluation of different facets of the spatial abilities.

In conclusion, the present study highlights that the difficulties in processing visuospatial information typically displayed by individuals with WS have to be extended also to the processing of visuospatial information of the peripersonal space. Further research on spatial impairment in WS will be carried by using RAM paradigms in virtual reality in a rehabilitative perspective.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of Bambino Gesù Children's Hospital, Rome, Italy [protocol number: 486LB] and was carried out in accordance with the Declaration of Helsinki. Written informed consent was obtained from the minors' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

All authors designed the research. SM and MP tested the participants. All authors analyzed the data and discussed the data. FF, PS, LP, and LM wrote the article.

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