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Topographical working memory in children and adolescents with motor disabilities

Åsa Bartonek[1]*, Cecilia Guariglia[2,3] and Laura Piccardi[2,3]

*Corresponding author: Åsa Bartonek, Motoriklab Q2:07, Karolinska University Hospital, Karolinska Vägen 37, 17176 Stockholm, Sweden
E-mail: asa.bartonek@ki.se

Abstract: Aim: The aim of the present study was to investigate topographical working memory in individuals with motor disabilities. Methods: Topographical working memory was investigated using the Walking Corsi Test in 89 participants with motor disability, mean age 11.5 years, of which 40 with cerebral palsy, 31 with spina bifida, and 18 with orthopaedic or peripheral symptoms. The participants were grouped according to everyday mobility, i.e. walking outdoors, walking indoors, and using wheelchair. A control group constituted 120 typically developing participants, mean age 9.9 years. Results: Individuals with spina bifida, orthopaedic or peripheral symptoms as well as typically developing participants performed significantly larger walking spans than the cerebral palsy group. With respect to mobility, those walking outdoors had significantly larger span than those walking indoors and those using wheelchair for mobility. Conclusions: Participants with outdoor walking in the community, apart from type of motor disability, seem to have improved topographic memory compared to individuals who don't walk outside and individuals who are mobile through wheelchair. The results highlight the question of development of spatial cognition to enhance participation in social environments. Future research should focus on prematurity in the cerebral palsy group, and on hydrocephalus in the spina bifida group.
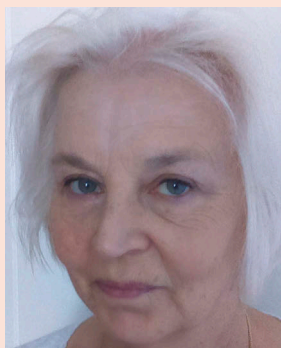
ABOUT THE AUTHOR

Åsa Bartonek received her PhD from Karolinska Institutet Sweden in 2001 and became associate professor in Physiotherapy in 2010. Her clinical experiences expands over several European countries in congenital and neurodevelopmental disabilities in children. Her research fields include neuro-orthopedic diagnoses where her findings regarding gait and orthoses have been implemented in clinic. In children with brain lesions, the perceptual-motor system perspective has come to determine her view of motor development, also as a book-author. Her research articles on the above mentioned subjects cover methods from laboratory gait analysis to prospective follow-up studies and parent-reported quality of live. During the years she has also been supervisor of doctoral students and is presently involved in several projects as well as in teaching of students on various levels. The topic of the current article on navigation, and that mobility should be utilized to increase participation in social environments, will lead to further projects.

Åsa Bartonek

PUBLIC INTEREST STATEMENT

Locomotion ability is important to interact in the environment. In children with motor disabilities, ambulation in society is often demanding, also in children with walking ability. Participation in social and environmental settings may therefore be less active than in able-bodied peers. Due to the children's compromised mobility, there are less possibilities of early locomotor experience and understanding of the spatial layout. Today, in middle and high income countries, many children are equipped for utmost locomotion. The aim of the present study was to investigate spatial cognition during moving the body in a navigational space. The study indicates that children who practice outdoor walking, apart from type of motor disability, seem to have developed improved navigational skills compared to children who don't walk outside and children who are mobile extensively through wheelchair. Children are suggested to be provided with daily opportunities to actively maximize their interactions with objects, people, and events.

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Subjects: Cognitive Neuroscience of Memory; Children and Youth; Movement Disorders; Physiotherapy

Keywords: Spatial orientation; navigational memory; motor disorder cerebral palsy; spina bifida; orthopaedic

1. Introduction

In the habilitating work in children with motor disabilities, much effort has traditionally been put on supporting the child to utmost locomotor ability. Since locomotion is more than organizing a pattern of movement to make forward progression, a close understanding of the spatial layout, and his or her relation to it is required by the individual (Andersen et al., 2014). The infant's locomotor experience, already during prone progression when crawling, affects the infant's autonomy, wilfulness and social cognition (Campos et al., 2000). What infants remember about how a space is mapped out is suggested to be inseparably related to their movements through the space. Maintaining an orientation to the external layout, remembering landmarks, and recalling the objects location are features of spatial cognition, making functional locomotion related to navigation and memory (Clearfield, 2004). In healthy pre-school children who had not yet achieved independent locomotion nor were able to point a direction while seated in a pushchair, poorer performance in spatial memory tests were found than in children who actively directed their own route from a pushchair, or were led on foot to positions selected by the experimenter (Foreman et al., 1990).

There is only sparse knowledge about the relation between movement and spatial relations in children with motor disorders. Compared with non-disabled classmates, a small group of motor impaired children in mainstream school were found deficient in drawing landmarks on a classroom map and pointing in the direction of landmarks on the school campus, demonstrating poor awareness of the environmental space (Foreman et al., 1989). Commonly seen patient groups in habilitation services worldwide are individuals with cerebral palsy (CP), spina bifida (SB), and various diagnoses comprising orthopaedic disabilities. CP is today the most common congenital neurodevelopmental disorder with a prevalence of 1.77 per 1,000 live births in Europe (Sellier et al., 2012). Previous definitions of CP have not given sufficient prominence to the non-motor neurodevelopmental disabilities of performance and behavior that commonly accompany CP. Therefore, other motor-associated factors have been included in the latest classification (Rosenbaum et al., 2007), of those perceptive impairment has been identified as fear during moving and difficulty to tolerate the surrounding space (Ferrari & Cioni, 2010). During walking this difficulty has been reported as spatial insecurity, and required visual control due to deficits in proprioception and sensation of the lower limbs (Bartonek et al., 2018). In premature-born adolescents with CP and periventricular leukomalacia, navigational skills have been found to be inversely related to the severity of leg-dominated bilateral spastic CP. Based on their findings the authors proposed early training through active spatial exploration and way finding in familiar and unfamiliar surroundings in patients with motor disabilities (Pavlova et al., 2007). Since deficits in spatial cognition may be related to motor deficits in children with CP, tests during navigation and locomotion involving the manipulation of multiple spatial reference frames have been proposed (Berthoz & Zaoui, 2015). A navigation task in children with bilateral CP while walking requiring real body motion, however, could not confirm differences to typically developing children (Belmonti et al., 2015).

SB is a congenital malformation involving the central nervous system with need for ventriculoperitoneal shunting due to hydrocephalus, as well as leg muscle paresis, orthopedic, and urologic abnormalities (Kahn et al., 2015). Prevalence of SB is reported as ranging from 3 to 5 cases/10,000 (Williams et al., 2005). In myelomeningocele (MMC), the most severe form of SB, consequences of muscle paresis level are most discriminating for ambulation. Consequences of secondary damages of the spinal cord, such as spasticity, as well as of hydrocephalus, have additionally impact on walking ability (A Bartonek et al., 2005; Bartonek & Saraste, 2001). In children with SB, route

knowledge in a virtual environment was acquired later than in non-disabled children, possibly due to their missing mobility experiences (Wiedenbauer & Jansen-Osmann, 2006). In adolescents with hydrocephalus and SB, mobility experiences were found to be important for route learning (Simms, 1987) as well as in patients with hydrocephalus without SB, in those, less accurate route-learning, poor memory for landmark objects and less accurate spatial updating was found (Lowry et al., 2010).

In orthopaedics, the rare disease of arthrogryposis multiplex congenita (AMC) occurs in between one in 3,000 to one in 5,000 live births (Lowry et al., 2010). AMC implies contractures in multiple body areas involving the limbs with an intact sensory system, and are usually non-progressive (Hall, 2014). Consequences of muscle weakness and joint contractures in the lower limbs influence walking ability. With orthoses most children achieve functional ambulation and various ambulatory groups with their orthotic needs have been identified (Eriksson et al., 2010). Stanton et al. (2002) studied wayfinding choice in a computer-simulated test in participants with varying histories of mobility impairment, including among other disabilities, participants with CP, SB, and AMC. Groups were analysed with respect to independent exploration during the period from birth to autonomous walking, compared with their present mobility level. Disabled participants whose mobility was more limited early in development were poorer at the task than those whose mobility had deteriorated with age.

Spatial cognition is a multi-faceted domain including at least two different types of space, namely, (a) near, peri-personal or reaching space and (b) far, extra-personal or navigational or environmental space, which are processed by at least partially segregated neurocognitive systems (Nemmi et al., 2013). Spatial cognition comprises a cluster of skills, including the ability to code spatial information (spatial coding), to memorize positions and locations of objects and of environmental features (spatial memory), to use the memorized information for acting or for moving in space (spatial planning), as well as the conscious knowledge of spatial features (spatial awareness). Spatial cognition for reaching and navigational space seem to develop with various rate during childhood (Piccardi, Leonzi et al., 2014). The development of spatial cognition for navigational space is also depending from the experience of individuals derived by moving in the environment. Some aspects of spatial cognition, such as spatial memory in navigational space (thereafter defined as "topographical memory"), may therefore differ from that of typical developing children. Topographical working memory takes in account encoding and maintaining online sequences of environmental cues that are crucial during navigation (Palmiero & Piccardi, 2017; L Piccardi, Palermo et al., 2015). It allows the individual to reach different places in the environment, to find the shortest way connecting two locations, to recognize familiar spatial layouts, and to orient in familiar environments. When testing topographical working memory on healthy adults interfering a dual task, the results suggested that motor and spatial-motor tasks did not interfere with performing the test, whereas a spatial-environmental task may disrupt the topographic working memory (Piccardi, Nori et al., 2015). However, children's knowledge of environment develops together with their ability to move in it, making us hypothesize that navigational memory may be affected by motor impairments in developmental age.

The concept of grounded cognition emphasizes the idea that it is not sufficient to assess whether a child can perform a behavior in isolation. Important is how the child uses this behavior to explore objects, to enter into relationship with people, and during events in social situations. Focus of early intervention should thus be to facilitate global development through important early behaviors such as object interaction and locomotion. (Lobo et al., 2013). To support as good autonomy as possible, it thus seems reasonable to pay attention to the child's ability to recognize spatial layouts and orient in its environment. For all these reasons, we were interested to get an overview of spatial cognition among children with motor disabilities who had access to relevant up-to-date equipment for utmost locomotion. The main aim of the present study was therefore to investigate spatial cognition, especially topographical memory, during moving the body in a navigational space. Since topographical working memory allows maintaining online environmental information during navigation, we expected that children with motor disabilities would perform worse in topographical working memory

tasks than typical developing children. Also lower performance during visual-spatial memory tasks have been reported in children with motor disabilities (Berthoz & Zaoui, 2015; Norrlin et al., 2003; Pavlova et al., 2007). For that reason, a secondary purpose was to explore visual-spatial working memory with respect to their topographical working memory in the studied children.

2. Methods

2.1. Participants
In total 89 individuals (49 males, 40 females) with motor disabilities, mean age 11.53 (S.D. = 3.25) years were consecutively included in the study between January 2014 and December 2016. All participants with motor disabilities were recruited at Karolinska University Hospital, Stockholm. Inclusion criteria were ability to follow verbal instructions required for the tests. The participants were allowed to use walking aids or a wheelchair, but were requested to have the ability to steer their technical aids independently.

One hundred and twenty typically developing individuals (TD), 57 males, 63 females, mean age 9.89 (S.D. = 3.11) constituted a control group. The participants in the control group were recruited among siblings of inpatients and through advertisement in the hospital. The study was approved by the Regional Ethical Review Board in Stockholm. Parents gave informed written consent and children provided a verbal assent before taking part in to the study.

The individuals with motor disabilities were designated a motor disability group with respect to their diagnosis. Forty participants were diagnosed with CP, 31 with SB, and 18 participants had orthopaedic or peripheral symptoms (ORT/PERI). Of the participants with CP, 33 were diagnosed with bilateral CP, 4 with spastic unilateral CP, and 3 participants with dyskinetic CP. Twenty-two (55%) were born prior to 37 weeks gestational age, and gestational age data was missing in three participants. In the SB group, all 31 participants had MMC of which 19 (61%) had shunted hydrocephalus. Of the 18 participants in the ORT/PERI- group, 10 individuals had AMC, 1 child had severe orthopaedic joint deformities, 4 had peripheral motor disturbances in the lower limbs or feet following hereditary periphery paraplegia and myelitis of the spinal cord, and 3 individuals had closed neural tube defects.

All participants with motor disabilities were grouped according to their everyday mobility, i.e. WalkOUT group, commonly walking both in-and outdoors; WalkIN group, frequently walking indoors and using wheelchair outdoors; and the WalkNO group using wheelchair for all mobility and transfer. Table 1 shows age and gender as well as distribution of mobility with respect to motor disability groups. There were no significant differences in age between the disability groups.

2.2. Instruments and procedure
All participants performed two working memory tests, a visuo-spatial memory test (Corsi Block-tapping Test, CBT: Corsi, 1972 (Corsi, 1972) and a topographical working memory test (Walking Corsi Test, WalCT: (Piccardi et al., 2013, 2008; Piccardi, Palermo et al., 2014) in randomized order. The CBT consists of nine wooden blocks (4.5 × 4.5 cm) fixed on a baseboard (30 × 25 cm) in a scattered array, and numbered on the examiner's side for ease of identification (Figure 1(a)).

The examiner taps a number of blocks at a rate of one block per 2 seconds, lifting the hand straight up before moving it to the next block, after which the participant is expected to tap the block sequence in the same order. Starting from a two-block sequence, the examiner gradually increases the length of block numbers. Five trials of each block sequence are presented, of which three trials must be correctly performed. The CBT score is equivalent to the longest block sequence repeated correctly by the participant. The participants were tested individually in a quiet room, seated on a height-adjustable chair in front of the CBT baseboard facing the experimenter. The WalCT is a larger version of the CBT (3 × 2.5 m; scale 1:10 of the CBT) consisting of nine squares placed on the floor in identical positions as in the CBT (Figure 1(b)). Both examiner and participant

	CP *n* = 40	SB *n* = 31	ORT/PERI *n* = 18	Total *N* = 89
Age (mean, SD)	11.59 (3.41)	11.95 (3.29)	10.66 (2.79)	11.53 (3.25)
Male	24	16	9	49
Female	16	15	9	35
WalkOUT	16	10	5	31
WalkIN	17	8	13	38
WalkNO	7	13	0	20

Table 1. Age and gender with respect to motor disability group and distribution of mobility among the motor disability groups. CP = cerebral palsy; SB = spina bifida; ORT/PERI = non-neural orthopedic deformities and peripheral neurological disturbances. WalkOUT = commonly walking both in-and outdoors; WalkIN = walking indoors and using wheelchair outdoors; WalkNO = using wheelchair for all mobility and transfer

CP = cerebral palsy; SB = spina bifida; ORT/PERI = orthopedic deformities and peripheral neurological disturbances.

Figure 1. (a) Child performing the Walking Corsi Test. The scale was 1:10 of the Corsi Block-tapping test (CBT) and the apparatus measured 3 × 2.5 m; black squares were 30 × 30 cm. (b) Apparatus used to administer the Corsi Block-tapping test (CBT). Numbers are present on the examiner's side only, not visible to the children.
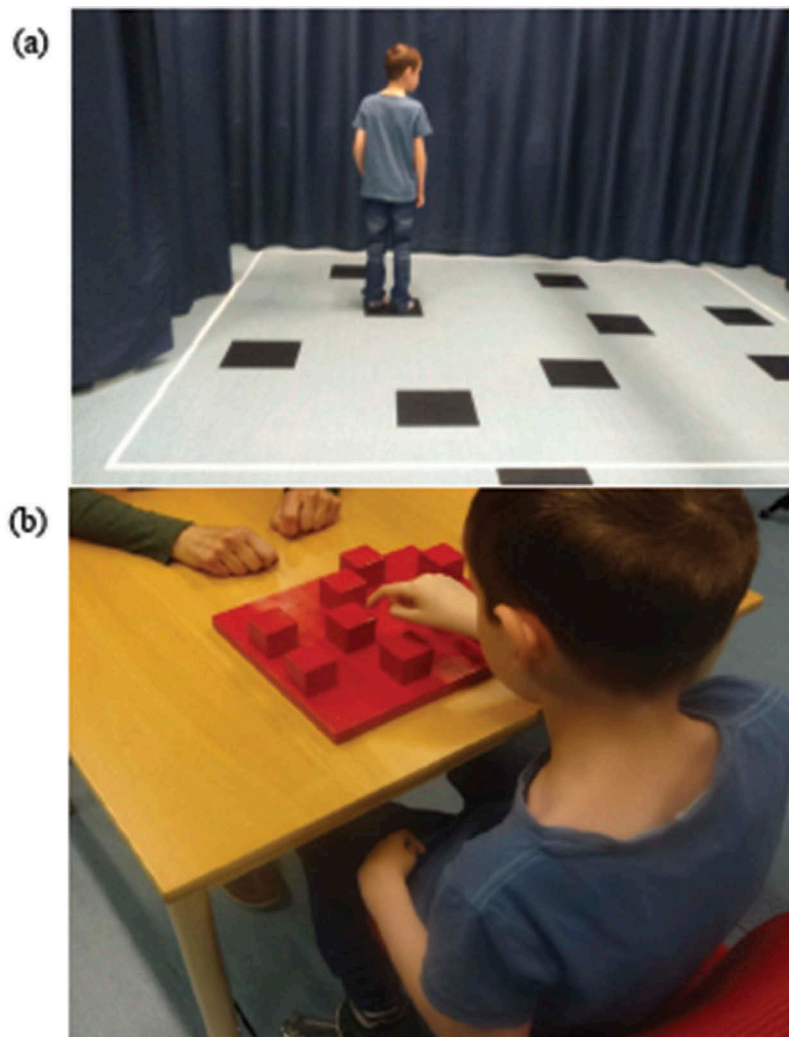
start from the same point. The examiner illustrates the sequence by walking on the squares and stopping on each of them for two seconds. Starting from a two-square sequence, the examiner gradually increases the length of square numbers. Five trials of each square sequence are presented, of which three trials must be correctly performed. The WalCT score is equivalent to the longest sequence repeated correctly by the participant. The WalCT area was set up in one part of a motion laboratory room visualized by straps on the floor as well as encircled by textile curtains in front and at the sides of the WalCT area. WalCT and CBT were performed in randomized order. All children in the WalkNo group performed the WalCT in their habitually used manual wheelchair. The participant was instructed to stop on the tile in a manner that the tile was still observable by her/himself. This was possible through the footplate of the wheelchair, which was constructed with a metal frame. It could be assumed that participants who are tested in a wheelchair would require more time to perform the test than those who are able to perform the sequence by walking. We therefore tested the WalkNo group with manual use of a laser pointer from a static position. Since there were no differences in WalCT scores between wheeling and pointing, we assumed wheelchair mobility to be relevant to use in this study. This decision was confirmed by the results of De Nigris et al. (2013) who found no difference between pointing a WalCT sequence and performing the route by walking.

Additionally, a visuo-spatial reasoning test (Raven's Coloured Progressive Matrices, CPM (Pueyo et al., 2008; Raven, 1938) was performed in all participants with motor disabilities. The CPM assesses non-verbal abilities at three levels by measuring clear-thinking ability for young children ages 5:0–11:0 years and elderly individuals, and mentally and physically impaired individuals. The test consists of 36 items in three sets with an administration time of 15–30 minutes per individual. The CPM items are arranged to assess cognitive development up to the stage when a person is sufficiently able to reason by analogy and adopt this way of thinking as a consistent method of inference. The Raven's CPM produces a single raw score that can be converted to a percentile based on normative data collected from various groups. CPM data was missing in one participant with SB, who however attended the regular school, like most children with disabilities did.

2.3. Data analysis and statistics
Basic statistical analyses (e.g., descriptive statistics, analysis of variance (ANOVA)) were conducted using Statistica software (version 2.5 for parallel desktops Windows for MACOS X version 10.10.5). To examine possible differences between groups in topographical and visuo-spatial working memory, we performed separated ANOVA. In order to investigate the effect of children's motor disability group, we performed two ANOVAs with Group (TD, CP, SB, and ORT/PERI) as independent variable and span on WalCT or on CBT as dependent variables followed by post hoc analysis (LSD test) when required. In both analyses we also used age as covariate in order to reduce the effect of age on performance. To investigate the effect of the children's mobility, we performed two ANOVAs with Group (WalkOut; WalkIn; WalkNo) as independent variables and span on WalCT or CBT as dependent variables. In this analysis, we maintained age as covariate and we excluded the TD group since we aimed at investigating the weight of mobility in the children with disabilities.

To ensure that there were no gender influence, we also investigated the effect of gender on WalCT and CBT through an ANOVA with Group (males and females) as independent variables and span on WalCT or CBT as dependent variables. The ANOVA on gender did not show any significant differences on CBT ($F_{1,206}$ =.38; p = .54) nor on WalCT ($F_{1,206}$ = .15; p = .70).

To analyse the presence of differences in CPM in children with disabilities, we performed ANOVA with Group (CP, SB, ORT/PERI) as independent variables and Raven's CPM total score as dependent variable.

The alpha level chosen for considering a statistical test as significant was p < .05.

3. Results

3.1. Motor disability groups
Spans of WalCT and CBT according to motor disability groups are shown in Table 2 and are illustrated in Figure 2.

With respect to the motor disability groups, the ANOVA on WalCT span evidenced a significant difference between groups ($F_{3,204}$ = 8.92; p < .001; η^2 = .116; observed power = .995). A post-hoc analyses test (Fisher's Least Significant Difference, LSD) test showed that the TD group, and SB and ORT/PERI disability groups performed longer spans than the CP group.

The ANOVA on CBT evidenced a significant difference between groups ($F_{3,204}$ = 16.42; p < .001; η^2 = .194; observed power = 1). The LSD test showed that the TD group, and the SB and ORT/PERI disability groups performed significantly longer visuo-spatial spans than the CP group. Neither WalCT nor CBT spans differed significantly among TD, SB, and ORT/PERI groups.

The ANOVA on total Raven CPM Mean (SD) score 23.6 (7.4), 25.9 (7.8), and 27.4 (5.9) in CP, MMC, and ORT/PERI, respectively, did not evidence any significant differences between motor disability groups ($F_{(2,85)}$ = 1.98; p = .14).

3.2. Mobility groups
Spans of WalCT and CBT according to mobility groups are shown in Table 3 and are illustrated in Figure 3.

The ANOVA performed on WalCT span revealed a significant difference between groups ($F_{2,85}$ = 4.34; p = .016; η^2 = .092; observed power = .738). The LSD test evidenced that WalkOUT group had longer span than WalkIN and WalkNO groups. The ANOVA on CBT showed no significant differences between the WalkOUT, WalkIN, and WalkNo mobility groups ($F_{2,85}$ = 1.34; p = .27).

4. Discussion
This study is a first attempt to get an overview of the ability to recognize spatial layouts and orient in the environment in children and adolescents with various motor disabilities. The question is relevant when considering new proposed physiotherapy approaches putting focus on the child in its environment, facilitating it to interact with people during social occasions (Lobo et al., 2013). Based on the knowledge that an infant's locomotor experience influences the infant's autonomy (Campos et al., 2000), this development may likely be delayed in children with motor disability whose early motor development in many cases is altered.

To study locomotion related to navigation and memory, we choose the method of topographical working memory (WalCT) (Palmiero & Piccardi, 2017; Piccardi, Palermo et al., 2015). Other than in visual-spatial memory tests where the subject typically is located facing the experimenter when performing the test, during the WalCT, the subject has to move its body in space thus acting from a continuously changing direction. Contrary to our hypothesis, not all of the participants with disabilities performed worse in topographical working memory tasks than typical developing participants. In the SB group, we found somewhat lower mean span in WalCT than the TD

Table 2. WalCT (Walking Corst test) and CBT (Corsi block-tapping Test) spans (mean, SD) according to motor disability groups				
	TD **N = 109**	**CP** **N = 40**	**SB** **N = 31**	**ORT/PERI** **N = 18**
WalCT	4.19 (1.5)	3.68 (1.14)	4 (1.15)	4.17 (1.2)
CBT	4.79 (0.94)	3.93 (1.14)	4.84 (1.09)	4.61 (0.85)

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Figure 2. WalCT (Walking Corsi test) and CBT (Corsi block-tapping Test) spans (mean, SD) according to motor disability groups and typical developing children (TD, *n* = 109), cerebral palsy (CP, *n* = 40), spina bifida (SB, *n* = 31), and orthopaedic/ peripheral lesions (ORT/PERI, *n* = 18).
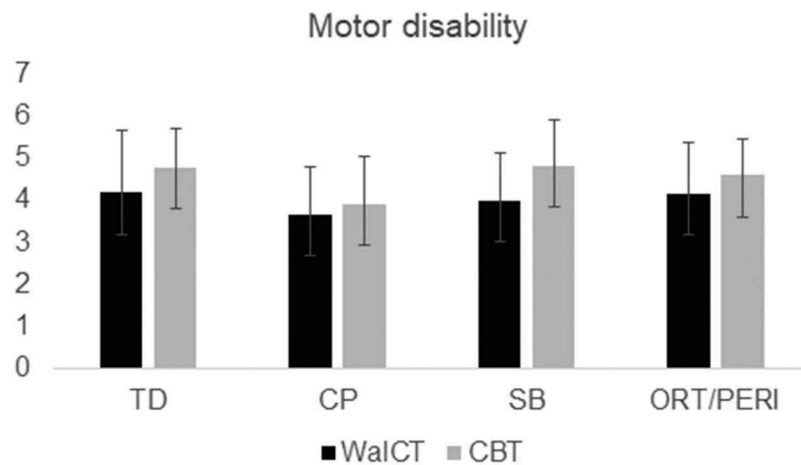

Motor disability

Table 3. WalCT (Walking Corst test) and CBT (Corsi block-tapping Test) spans (mean, SD) according to mobility groups. WalkOUT = commonly walking both in-and outdoors; WalkIN = walking indoors and using wheelchair outdoors; WalkNO = using wheelchair for all mobility and transfer

	WalkOUT *N* = 31	WalkIN *N* = 38	WalkNO *N* = 20
WalCT	4.35 (1.19)	3.57 (1.11)	3.74 (1.01)
CBT	4.58 (0.92)	4.14 (1.35)	4.48 (1.03)

Figure 3. WalCT (Walking Corsi test) and CBT (Corsi block-tapping Test) spans (mean, SD) according to mobility groups in children with disabilities. WalkOUT = commonly walking both in-and outdoors (*n* = 31); WalkIN = walking indoors and using wheelchair outdoors (*n* = 38); WalkNO = using wheelchair for all mobility and transfer (*n* = 20). The WalkOUT group had larger span than WalkIN and WalkNO groups (*p* <.05).

Mobility
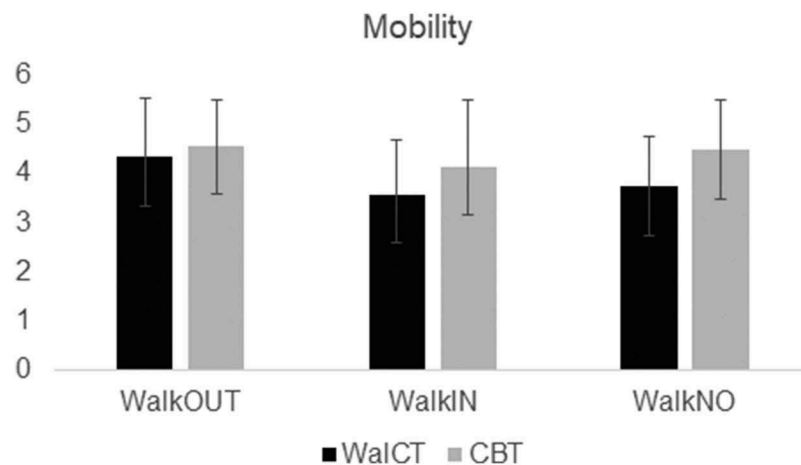
group, although not significantly. Wiedenbauer and Jansen-Osmann (Simms, 1987) examined spatial knowledge in children with SB in a computer-simulated environment. They found impaired route knowledge but not an impaired landmark knowledge, interpreting their results as that spatial impairment in children with SB being more accentuated towards their reduced mobility. Tests by Simms (1987) in adolescents with SB included route learning by both being driven and directing the route as well as drawing freehand sketch maps of the routes. They concluded that environmental experiences and independence training for mobility during childhood for those with SB and hydrocephalus is of importance. The ORT/PERI group performed similar good in WalCT as the TD group. The majority of participants in the ORT/PERI group were diagnosed with AMC. Stanton et al.

(2002) compared way-finding choices of able-bodied teenagers with physically disabled teenagers, including participants with CP, SB and arthrogryposis, and half of the participants presenting with conditions not commonly referred to central nervous system damage. The authors concluded that early independent exploration is important in the development of spatial knowledge, suggesting that the effects of limited early exploratory experience may persist in later years. Some of the children in the ORT/PERI group had other disabilities than orthopaedic deformities as defined in the group name, such as peripheral spasticity or muscle weakness in the feet muscles. The participants in ORT/PERI performed the WalCT with somewhat higher span than TD participants, indicating that the peripheral symptoms could not having disturbed navigation ability substantially.

In line with our hypothesis that individuals with motor disability would perform less during WalCT than the TD group, the CP group achieved significantly less span, not only than TD but also than SB and ORT/PERI groups. Belmonti et al. (2015) reported that children with spastic CP performed tasks in the navigational space without differences from typical developing children. Since among our patients also those with less functional level were included than those in the study of Belmonti et al. (2015), a close comparison with our patients cannot be made. Furthermore, the Magic Carpet used by Belmonti et al. (2015) even if derived by WalCT, cannot be fully compared to the WalCT itself as used in our study. In the Magic Carpet, stimuli are automatically delivered by LED switching (tiles are lit up) while the participant stands on starting point. Differently in the WalCT, the examiner demonstrates the sequence by moving its body through the space, also providing directional information concerning the following square in the sequence.

Considering the various natural history of the disability groups, the division into motor disabilities groups allowed us to distinguish differences between the groups, although at the same time being aware about the variation in severity among a disability group itself. Stanton et al. (2002) suggested that children with CP may suffer neurological damage that can include spatial brain structures but that poor spatial learning in these children may be secondary to their neurological conditions. In our study, we observed a five year-old girl with dyskinetic form of CP who was completely dependent on a walking aid. She performed as good as TD children of similar age, despite requiring more time to move her body through the WalCT test. The finding in this girl may thus highlight the necessity to be aware of variations in the CP group. Some of the children with spastic CP who were able to use a walker choose to perform the WalCT test in their manual wheelchair. In these children, we may suspect that perceptive impairment leading to difficulties to tolerate the surrounding space made them do this choice (Ferrari & Cioni, 2010). When performing the WalCT, the subject has to turn its body on the tiles in various directions. During a task of turning in children with CP, a walker was found to compensate for spatial insecurity and deficits in proprioception of the lower limbs (Bartonek et al., 2018). This finding could thus be in line with the chosen strategy of some children to perform the WalCT in the wheelchair not being obliged to visually control the feet during steps allowing awareness of surroundings.

The main finding of the study can be pointed out as that participants in the WalkOUT group, with independent walking ability in the environment, performed significantly higher on the WalCT test than both children walking only indoors (WalkIN) and those using wheelchair for all mobility and transfer (WalkNO). It could however be assumed that participants who are tested in a wheelchair would require more time to perform the test than those who are able to perform the sequence by walking. It is thus reasonable to question how various pace during walking influences the child's topographical working memory. We therefore tested the WalkNo group manually using a laser pointer from a static position. Since there were no differences in WalCT scores between wheeling and pointing, we assumed wheelchair mobility to be relevant to use in this study. Moreover, in the SB group, children often have muscle paresis in large muscle groups requiring precisely body segment alignment with orthosis use with longer time to perform walked distances than able-bodied children. This was the case among same participants in our study in the WalkIN group, yet the WalCT mean results did not differ significantly from TD. Neither the time used to perform the

test in the previous mentioned girl with dyskinetic CP in the WalkIN group, however, did not seem to play a crucial role. However, in participants suspected to have motor-perceptual difficulties it is probable that time to move the body or steering the wheelchair was increased compared to autonomously moving participants which may influence working memory. These variations among subjects, on the other hand, tell us about the children's behaviour in the environment and will hopefully contribute to increased understanding of their situations in social situations. In this study, we did not measure manual ability objectively that would have been relevant both for the participants requiring wheelchair, as well as having impact on performance on CBT (discussed later), although this question did not appear as relevant in any of the participants.

The CP group performed significantly less on the WalCT compared to SB and ORT/PERI groups, yet the majority in the CP group were in the WalkOut group. It can therefore not be argued that a specific motor disorder overstated the results in this study, neither that the functional level merely determined the results. On the other hand, in the ORT/PERI group who performed WalCT as good as TD, no participant was in WalkNo group, most probably for the reason that children with only peripheral central nervous system involvement were included.

Previous authors investigating spatial knowledge in children with disabilities have emphasized the importance of mobility experiences (Smith & Buckley, 2012; Stanton et al., 2002; Wiedenbauer & Jansen-Osmann, 2006). In the concept of Karolinska hospital, each child is supposed to be supported with training and technical aids to achieve utmost walking ability or ambulation with respect to its resources. It is likely that mode of ambulation used in this study was also practiced in everyday life and that early independent exploration had been possible with respect to each child's possibility. Moreover, early exploration with the developmental benefits of independent locomotion through child driven powered-mobility equipment has been recommended (Lobo et al., 2013). Powered wheelchairs has been used in the Swedish habilitation during the last decades to enable the child to take part in social events being as independent as possible, given sufficient maturation of the child to handle it.

A secondary purpose of this study was to explore visual-spatial working memory with respect to topographical performance in the studied children by using the CBT test.

Our findings showed that the SB and ORT/PERI groups performed similar good as TD in CBT whereas the CP group had significantly lower CBT than the TD and the other disability groups. This finding concerning the CP group is in accordance with Belmonti et al. (2015) who found similar results in the reaching space using the same method of CBT (Corsi, 1972). Belmonti et al. (2015) concluded that in children with spastic CP, the distribution of impairment of spatial memory in reaching space is different from that in navigational space. Even if visuospatial deficits have been reported in children with MMC possibly leading to difficulty to get a comprehensive picture of the surroundings (Norrlin et al., 2003), this could not be confirmed by the CBT test in our study. As analyzed in this study, CBT spans were higher than WalCT spans both with respect to groups of motor disability and groups of mobility. This may be following the different test situations with CBT being testing from a fixed body position, whereas WalCT was performed during moving the body in space. This was experienced by a TD participant during testing where WalCT was randomized to be tested after CBT. The participant expressed spontaneously that she had to switch and think in another way when changing direction on the WalCT than during the CBT test during sitting. This indicates that remembering a sequence of block-tapping in a baseboard (CBT) and remembering a sequence of steps in a route (WalCT) do rely on different processes and strategies (Piccardi et al., 2008).

Raven's CPM intends to provide a measure of cognitive functions such as visuo-perceptual ability and visuo-spatial reasoning (Pueyo et al., 2008) that allows to rule out any difference between children with disability about the capability to reason on visuo-spatial material. We investigated this aspect since visuo-spatial difficulties are reported to be involved in the motor deficits of

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children with CP (Belmonti et al., 2015) as well as in children with SB (Norrlin et al., 2003). We did however not find any statistical difference between the motor disability groups, even if the lowest total CPM score was found in the CP group. CPM data was missing in one girl with SB from the WalkOUT group who however attended regular school. Based on the findings in our study, the children were believed to have sufficient visuo-spatial reasoning ability to perform the tests used in this study. The cognitive performance of the children with disabilities are therefore not considered to having influenced the study results.

A limitation of the study is that no analysis with respect to the difference in gestational age could be done in the CP group due to the small participant number. Neither any conclusions regarding the influence of hydrocephalus could be drawn in the SB group. This is a limitation since hydrocephalus in children has been found to be associated with navigational impairments (Smith & Buckley, 2012). A further limitation may be our choice to permit the participants to use wheelchair during the WalCT even if they managed indoor walking ability in everyday life. On the other hand, this decision allowed the participants to perform the topographical memory test without additional cognitive load of checking their leg movements when reproducing the route.

In children, the motor system contributes to build up their mental environmental representation allowing a continuous practice of the topographic memory system through a direct experience with the environment (Piccardi, Palermo et al., 2015). Consequently, much effort should be put on supporting children with motor disabilities to recognize spatial layouts and orient in its environment, besides improving motor function. Today, rehabilitation programs for individuals with CP increasingly use virtual reality environments to enhance motor practice. In a study of Nobre de Paula et al. (2018), children with CP were able to learn a virtual reality game by moving a mobile phone and to transfer the performance acquired in an opposite maze path. The test was performed during sitting and participants walked without limitation thus it is not comparable with the present study. Since children with CP are suggested to have deficits in spatial cognition related to motor deficits, tests should involve "egocentric" or "route" strategy requiring the use a body-centred reference frame (Belmonti et al., 2015). Belmonti et al. (2015) suggested promotion of early exploration of navigational space to increase multisensory representation of spatial relationships and shapes, which could be generalized across settings and spaces.

An indication from this study is that children who practice outdoor walking in the community, apart from type of motor disability, seem to have developed improved topographic memory compared to children who don't walk outside and children who are mobile extensively through wheelchair. Health-related quality of life (HRQL) was compared between ambulators and wheelchair users in a Swedish population with MMC in a social environment with high acceptance of early wheelchair use (Bartonek et al., 2012). The results of the parents' questionnaire revealed similarly perceived HRQL in ambulators and those with wheelchair dependence. These findings point to the importance of highlighting participation in social environments to contribute to development of spatial cognition even in young wheelchair users. This would be in line with Lobo et al. (2013) who recommended providing children with frequent daily opportunities to actively maximize their interactions with objects, people, and events. The interactions with environment could as well increase the children's engagement with the surroundings and thereby improve their relationship with the world around them. Furthermore, in future studies attention should also be paid to get a yet closer understanding of the topographical memory with respect to variations and characteristics in the various subgroups, such as prematurity, perceptual-motor ability, and hydrocephalus.

5. Conclusion

The CP group performed significantly shorter WalCT span than the TD, SB, and ORT/PERI groups. The children and adolescents walking outdoors (WalkOUT) had larger span than those walking only indoors (WalkIN) and those using wheelchair for all mobility (WalkNO). It may be indicated from this study that individuals who practice outdoor walking in the community, apart from type of

motor disability, have developed improved topographic memory compared to individuals who don't walk outside and those who are mobile only through wheelchair. The results highlight the relevance of practicing topographical memory to enhance spatial cognition and thus increase participation in social environments independent of using wheelchair or walking. In future research attention should also be given on variations in the various subgroups, such as prematurity, perceptual-motor ability, and hydrocephalus.

Author details
Åsa Bartonek[1]
E-mail: asa.bartonek@ki.se
Cecilia Guariglia[2,3]
ORCID ID: http://orcid.org/0000-0001-7474-6030
Laura Piccardi[2,3]
E-mail: laura.piccardi@cc.univaq.it
ORCID ID: http://orcid.org/0000-0002-1322-5538
[1] Neuropediatric Unit, Department of Women's and Children's Health, Karolinska Institutet, Stockholm, Sweden.
[2] Department of Psychology, Sapienza Universityof Rome, Rome, Italy.
[3] Cognitive and Motor Rehabilitation and Neuropsychology Unit, IRCCS Fondazione Santa Lucia, Rome, Italy.

Declaration of interest
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