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Topographical Working Memory in Children with Cerebral Palsy

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ABSTRACT. Forty children with cerebral palsy (CP) and 120 typical developing children (TD) performed a topographic working memory (WalCT) test requiring to move their body in a walked vista-space and a visuo-spatial test (CBT) requiring just reaching movements. WalCT score was significantly higher in GMFCS II/III than in TD. CBT score was significantly lower in GMFCS I than in III/IV but lower than TD in all CP groups. Similar results in WalCT between GMFCS I and TD and GMFCS II and III/IV respectively indicate that mobility is associated with topographic working memory. Differently in CBT, the absence of bodily movement allows using different cognitive strategies. Children should be provided with opportunities and active participation to enhancing spatial awareness and navigational skills.

Keywords: motor disorder, spatial orientation, navigation, environmental representation

Delays in object interaction, sitting, and locomotion are often present and related to future broader delays in infants and children with different neurological diseases (Andersen et al., 2014). Consequently, it is mandatory to focus on the child's perceptual-motor behavior to enhance the future development facilitating to entering into relationship with people, and during events in social situations (Lobo et al., 2013). Recently it has been confirmed that assistive technology-based programs were effective for promoting participation and independence of children with cerebral palsy functioning at different levels (Stasolla et al., 2019).

The diagnosis of cerebral palsy (CP) includes a primary impairment of movement and posture, with activity limitations ranging from walking to participation in society (Rosenbaum et al., 2007). However, accompanying impairments in sensation, perception, cognition, may at times produce even greater activity limitation than the motor impairments that are the hallmark of CP (Rosenbaum et al., 2007). Some studies focus on the sensory part of the movement disorder in children with CP. For example, freezing of posture during a postural demanding task in sitting was found to be associated with perceptual disorders (Ferrari et al., 2010). This sign has been defined as characteristic for perceptual disorders in children with spastic CP (Ferrari et al., 2014). Moreover, proprioception deficits in the lower limbs in children with CP have been associated with motor disturbances during standing and walking (Bartonek et al., 2018; Damiano et al., 2013; Lidbeck et al., 2016;

Wingert et al., 2009) where in the latter study during turning gait, difficulties in spatial orientation was believed to negatively influence locomotion.

Undoubtedly, locomotion is more than organizing a pattern of movements to make forward progression; it is in effect a close understanding of the spatial layout and the mutual spatial relationship between the individual and the environment (Andersen et al., 2014). What infants remember about how a space is mapped out is inseparably linked to their movements through that space. Maintaining an orientation to the external layout, memorizing landmarks and recalling the objects location, are features of spatial cognition, making functional locomotion related to navigation and memory (Clearfield, 2004). Spatial cognition includes the mental representation of near, peri-personal and reaching space and that of far, extra-personal and navigational space, processed by nearly partially segregated neurocognitive systems (Berti & Frassinetti, 2000; Nemmi et al., 2013; Rizzolatti et al., 2000) which seem to develop differently during growth (Piccardi, Palermo, et al., 2014; Piccardi, Leonzi, et al., 2014). Spatial cognition includes the ability to code spatial information (spatial coding), to remember positions and locations of objects and of environmental features (spatial memory), to use the memorized information for acting or moving in space (spatial planning), as well as spatial awareness. Within spatial cognition, topographic working memory enables encoding and maintaining online sequences of environmental cues that are central during navigation (Piccardi, Palermo, et al., 2014; Piccardi et al., 2015). Owing to topographic memory, the individual can reach various places in the environment, find the shortest way between two positions, recognize spatial layouts and orient in familiar environments (Palmiero & Piccardi, 2017).

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Taking into account the importance of locomotion for navigation, navigation skills have been investigated in individuals with CP. Stanton et al. (Stanton et al., 2002) examined the effects of early exploratory experience on spatial learning in teenagers with different histories of physical mobility of which 16 participants with CP. They found that disabled participants whose mobility was more limited early in development were poorer at the task than those whose mobility had deteriorated with age. For children with CP specifically they argued that neurological damage can include spatial brain structures however, poor spatial learning in these children may be secondary to their neurological conditions. Pavlova et al. (Pavlova et al., 2007) studied premature-born adolescents with CP and periventricular leukomalacia finding that the severity of right frontal periventricular damage and leg-dominated motor disorders can serve as independent predictors of the visual navigation disability. Based on their findings the authors proposed early training through active spatial exploration and wayfinding in familiar and unfamiliar surroundings in patients with motor disabilities (Pavlova et al., 2007).

In children with CP, deficits in spatial cognition and in spatial memory have been proposed to be related to motor deficits. Belmonti, Fiori, et al., (2015) studied cognitive strategies for locomotor navigation in children with typical development and children with spastic CP with independent walking without aids. They found that visuo-spatial memory was significantly lower in children with CP than in the matched control group, whereas navigation skills did not differ between groups. Yet, they concluded that further exploration of navigational space could prove useful in enhancing spatial representation and reference-frame manipulation in CP. Belmonti, Fiori, et al. (2015) also studied the development of anticipatory orienting strategies and trajectory formation in goal-oriented locomotion in children and adults (Belmonti et al., 2013). From these results, they proposed that navigational skills, such as path planning and shifting from ego- to allocentric spatial reference frames are proposed as necessary requisites for mature locomotor control. To better understand perceptual, cognitive and emotional factors that influence walking in children with CP, the authors included walking conditions of independent as well as accompanied locomotion with the examiner walking behind the subject without touching the child. Their results showed only minor effects between groups of independent versus accompanied locomotion and were mainly considered as useful for future investigation on children with motor and perceptual impairment. When distinguishing locomotor trajectory formation from head anticipation in children with CP, navigation performance seemed reduced in children with right-brain impairment and spatial integration was suggested to be considered more during rehabilitation (Belmonti et al., 2016).

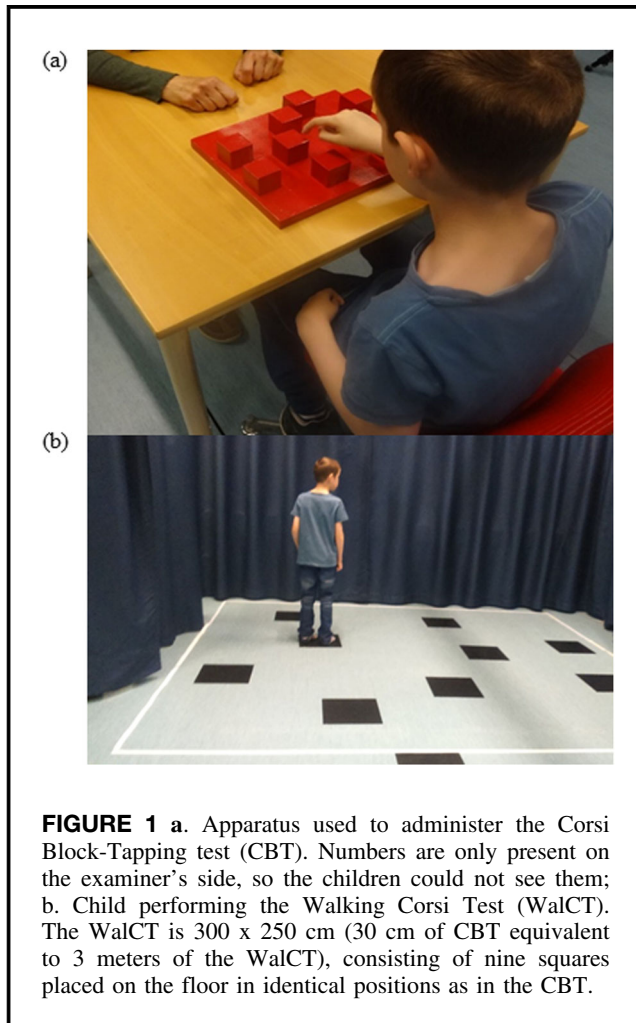
To support an utmost and adequate autonomy in terms of locomotion, we were interested to know how children with CP with various motor performance manage navigation. We started with studying a larger sample of children with various motor disabilities including peripheral and orthopedic disabilities as well as with central nervous system involvement (submitted data). The main aim of the present study was to focus on children and adolescents with CP, exploring topographic working memory in children with various degrees of motor performance levels. Children with typical development are able to track their own movements to locate formerly visited places in the environment after reaching a new location already at the end of the first year (Schmuckler & Tsang Tong, 2000). Considering that, we hypothesized that children with CP would have a poorer topographic working memory than typical developing children (TD) due to the lack of motor experience. Based on findings during studying turning gait (Bartonek et al., 2018) we also hypothesized that children with CP functioning at a low mobility level would show increasingly more difficulties in navigation tasks than children with independent locomotion in the community. Visual perception disorder, has been considered one of the cognitive functions often impaired in children with CP (Ego et al., 2015). Furthermore, remembering a sequence of block-tapping on a baseboard versus a sequence of steps in a route rely on different processes (Nemmi et al., 2013; Piccardi et al., 2008) and require differential cognitive strategies (Belmonti, Cioni, et al., 2015). For that reason, a further purpose was to explore visuo-spatial working memory with respect to spatial memory in navigational space in children with CP.

METHODS

Participants

In total 89 participants with motor disabilities of various diagnosis, were consecutively included in a project between January 2014 and December 2016 to get a comprehensive picture of topographical working memory in children with motor disabilities. Inclusion criteria were ability of independent ambulation with or without technical aid, and ability to understand instructions required to perform the tests. To determine the sample size, a power calculation was performed (Faul et al., 2007), suggesting a sample size of 90 participants of which 89 were available to fit with the inclusion criteria.

In this study, we report on 40 participants with CP (24 males; 16 females) of which 33 participants were diagnosed with spastic bilateral CP, 4 with spastic unilateral CP, and 3 with dyskinetic CP. Of the 40 children with CP, gestational age data was available in 35 children. Of those 23 children (65.71%) were born pre-term (prior to 37 weeks gestational age). The participants were



classified according to gross motor function classification system (GMFCS) describing the gross motor function of children and youth with cerebral palsy on the basis of their self-initiated movement with particular emphasis on sitting, walking, and wheeled mobility (Palisano et al., 1997; Palisano et al., 2008). Seventeen participants were classified at GMFCS level I walking without limitations (mean age 10.4, S.D.=3.3), 12 at level II walking with limitations (mean age 11.8, S.D.=3.62), 11 at level III/IV using a hand-held mobility for walking or a wheelchair for self-mobility (mean age 13.2, S.D.=2.8). One hundred and twenty typically developing individuals (TD), 57 males, 63 females, mean age 9.89 (S.D.= 3.11) constituted a control group. All participants with CP were patients at Karolinska University Hospital, Stockholm. The participants in the control group were recruited among siblings of inpatients and through advertisements 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 the study.

Visuo-Spatial and Topographic Working memory

All participants performed two working memory tests, a visuo-spatial memory test (Corsi Block-tapping Test, CBT: Corsi, 1972) and a topographic working memory test (Walking Corsi Test, WalCT: Piccardi et al., 2008; Piccardi, Palermo, et al., 2014) in randomized order.

The CBT consists of nine wooden blocks (3×3 cm) fixed on a baseboard (30×25 cm) in a scattered array, and numbered on the examiner's side for ease of identification, not visible to the participant (Figure 1a). 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 (span) 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 allows to test the topographic memory in a vista-space, namely, the space the individual can see from a single location or with only little exploratory movements, such as single rooms or town squares (Wolbers & Wiener, 2014). It is a larger version of the CBT (namely the WalCT is 300×250 cm; that is to say 30 cm of CBT equivalent to 3 meters of the WalCT) consisting of nine squares placed on the floor in identical positions as in the CBT. The WalCT area was set up in a part of a room visualized by straps on the floor as well as encircled by textile curtains in front and at the sides of the WalCT area (Figure 1b). Both examiner and participant start from the same starting point. The examiner illustrates the sequence by walking on the squares and stopping on each of them for two seconds, avoiding to walk on squares not included in the sequence. 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 to continue the test with the following sequence. The WalCT score is equivalent to the longest sequence (span) repeated correctly by the participant.

Ten out of eleven participants in GMFCS level III and IV performed the WalCT in their habitually used manual wheelchair. They were instructed to stop on the square in a manner that the square was still observable by her/himself. This was possible by observing the square through the footplate of the wheelchair.

Visuo-Spatial Reasoning

Additionally, all patients performed the Raven's Colored Progressive Matrices (CPM: Raven, 1938;

TABLE 1. Gender, age and spans of Walking Corsi test (WalCT) and Corsi Block Test (CBT) and Raven score (CPM) with respect to the GMFCS levels and TD group.

	TD N = 120	GMFCS I n = 17	GMFCS II n = 12	GMFCS III/IV n = 11	p-value	TD - I	TD - II	TD - III	I - II	I - III/IV	II - III/IV
Gender	m = 57, f = 63	m = 11, f = 6	m = 9, f = 3	m = 4, f = 7	0.135						
Age (mean, SD) (min-max) years	9.89 (3.11) (5-16)	10.41 (3.32) (6-17)	11.8 (3.62) (5-17)	13.18 (2.83) (7-16)	0.004	0.525	0.048	0.001	0.525	0.025	0.296
WalCT span (mean, SD)	4.19 (1.5)	4.18 (1.2)	3.33 (0.9)	3.27 (0.7)	<0.001	0.967	0.047	0.041	0.117	0.102	0.919
CBT span (mean, SD)	4.79 (0.9)	4.24 (0.8)	3.92 (0.9)	3.45 (1.5)	<0.001	0.031	0.004	<0.001	0.394	0.043	0.265
CPM score (mean, SD)	-	25.18 (7.6)	22.67 (8)	22.18 (6.5)	p = .518	-	-	-	ns	ns	ns

m: male; f: female; GMFCS: level of gross motor classification system; TD: typical developing child; WalCT: Walking Corsi test; CBT: Corsi Block-Tapping test; CPM: Raven total score; ns: non-statistical significance.

Pueyo et al., 2008) to assess their ability to reason on visuo-spatial material. Indeed, the CPM allows measuring clear-thinking ability for young children ages 5 to 11 years and elderly individuals, as well as 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 CPM produces a single raw score that can be converted to a percentile based on normative data collected from various groups.

Statistical Analysis

The children at GMFS levels III and IV were analyzed as one group, since all 11 participants performed the WalCT test with technical aids, 10 with manual wheelchair and one child at GMFCS III with a walking aid.

Basic statistical analyses (e.g., descriptive statistics, analysis of variance) were conducted using IBM SPSS software version 24. The participants' gender as well as presence of prematurity among the GMFCS levels were analyzed with a Chi-Square test. Influence of prematurity on WalCT and CBT was performed through an ANOVA with Group (present or not present) as independent variables and span on WalCT or CBT as dependent variables. To examine possible differences between GMFCS mobility levels in WalCT and CBT we performed separated Analysis of Variance (ANOVA). In these analyses we maintained age and CPM total score as covariates and

we excluded the TD group since we aimed at investigating the weight of mobility and cognitive functions in the children with CP. The alpha level chosen for considering a statistical test as significant was $p < .05$.

RESULTS

A Chi-square test revealed no difference in gender between TD and GMFCS levels I-III/IV ($p = .135$) (Table 1).

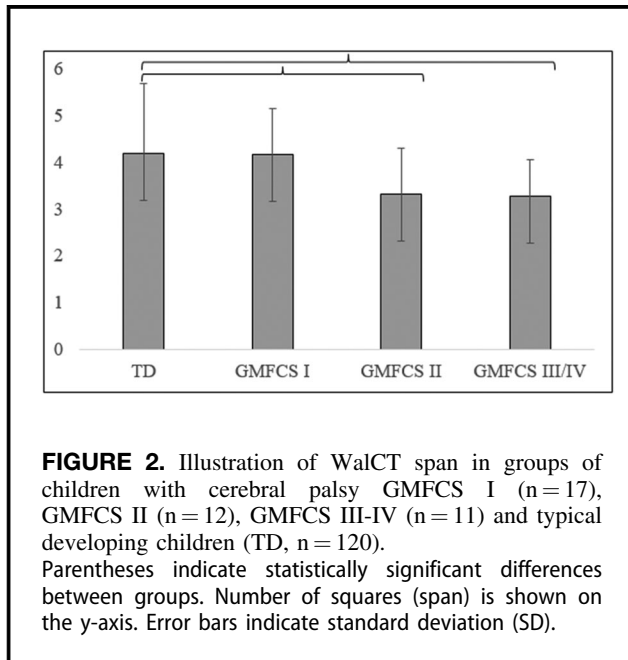
The ANOVA on age with post-hoc analyses test (Fisher's Least Significant Difference, LSD) showed that children at GMFCS level I were significantly younger than GMFCS level III/IV and that the TD group was significantly younger than GMFCS levels II and III/IV, $F(3,156) = 4.626$; $p = .004$; partial $g^2 = .082$ (Table 1).

Prematurity

A Chi-square test revealed presence of prematurity in GMFCS level I, 9 versus 7 children, in GMFCS level II, 11 versus 0 children, and in GMFCS level III, 3 versus 5 children ($p = .010$). The ANOVA on prematurity among the children with CP showed significant differences on WalCT, $F(2,31) = 3.246$; $p = .052$; partial $g^2 = .173$ but not on CBT $F(2, 31) = .621$; $p = .544$; partial $g^2 = .039$.

Topographic Working Memory with Respect to GMFCS Levels

The ANOVA with age as covariate showed significant differences on WalCT, $F(3,155) = 12.621$; $p < 0.001$; partial $g^2 = 0.196$. A post-hoc LSD analyses test



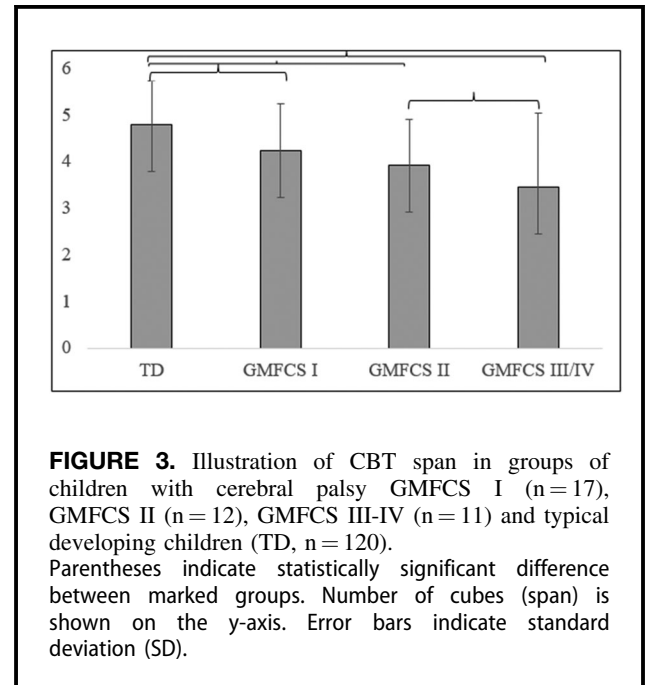
evidenced that GMFCS levels II and III had significantly shorter WalCT span the TD group had. Spans of WalCT in children with CP according to GMFCS levels I-IV and in TD children are shown in Table 1 and are illustrated in Figure 2.

Visuo-Spatial Working Memory with Respect to GMFCS Levels

The ANOVA with age as covariate showed significant differences on CBT, $F(3,155) = 20.402$; $p < 0.001$; partial $g^2 = 0.283$. A post-hoc LSD analyses test evidenced that the GMFCS levels I, II and III/IV had significantly shorter CBT span than TD group, and GMFCS level I had longer than GMFCS level III/IV. Spans of CBT in children with CP according to GMFCS levels I-III/IV and in TD children are shown in Table 1 and are illustrated in Figure 3.

Visuo-Spatial Reasoning Test

The ANOVA on total Raven CPM Mean (SD) score in GMFCS levels I-III/IV (25.18 (7.6), 22.67 (8), 22.18 (6.5) respectively) did not evidence any significant difference in visuo-spatial reasoning between GMFCS level groups, $F(2,37) = 0.669$; $p = .518$. The ANOVA with CPM score as covariate showed no significant differences on WalCT, $F(2,36) = 2.428$; $p = .103$; partial $g^2 = 0.103$ and CBT $F(2,36) = 1.085$; $p = 0.049$; partial $g^2 = 0.057$. CPM total score with respect to GMFCS levels is shown in Table 1.



DISCUSSION

In accordance with our hypothesis, we found that children with CP have more difficulties with topographic working memory than TD children have. Grouping the participants with CP according to GMFCS levels allowed us to discriminate between the participants' every day mobility between walking without or with limitations outdoors, to requiring a hand-held mobility for walking or a wheelchair for self-mobility. With regard to the functional mobility classified as GMFCS levels, we found that GMFCS level I showed similar ability to perform the WalCT span as the TD Group whereas both GMFCS levels II and III/IV reached significantly lower WalCT spans.

Children at various GMGCS levels also behaved differently in gait studies. When investigating gait during straight walking focusing an external target at the end of the walkway, GMFCS I group behaved similar as TD group, whereas GMFCS II behaved with less trunk rotation and more trunk flexion than TD, and GMFCS III group reduced movements and walking velocity compared to the other groups (Bartonek et al., 2016). During a demanding turning task around a target, GMFCS I group walked as fast as the TD group with similar head and trunk movements, GMFCS II rotated less between head and trunk than TD and GMFCS I, and in GMFCS III, head and trunk flexion during turning were larger than the other groups (Bartonek et al., 2018). From these studies was concluded that motor behavior of the children in GMFCS III could be seen as a consequence of lack of sensory information from lower limbs reflecting deficits in proprioception and sensation requiring visual

control of the lower limbs. Furthermore, in GMFCS II group, less rotation was reflected as spatial insecurity similar to freezing of posture, which has been described as a sign of perceptual disorder in children with spastic CP (Ferrari et al., 2014). In GMFCS III, during the turning task, the walker was considered to serve as an external reference frame, enabling functional ambulation despite difficulties with spatial perception (Bartonek et al., 2018).

According to the above mentioned studies, the children's behavior during the current study of topographical working memory assessed with the WalCT test must be taken into consideration. Stanton et al. (2002) suggested that even if children with CP may suffer neurological damage including spatial brain structures, poor spatial learning may be secondary to their neurological conditions and that any neurological impairment associated with the disability, may have implications for the development of spatial awareness. According to Belmonti, Fiori, et al. (2015) deficits in spatial cognition and in spatial memory have been proposed to be related to motor deficits. In our study also children with unilateral CP and dyskinesia as well as those with less functional level were included, thus, a close comparison with our patients cannot be made with that of Belmonti et al. Furthermore, the Magic Carpet used by Belmonti et al. 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, which was used in our study, the examiner demonstrates the sequence by moving its body through the space, also providing directional information concerning the following square in the sequence. The results of Belmonti et al. did not find that navigation skills differed significantly between children with CP and typically developing children, yet, they concluded that further exploration of navigational space could prove useful in enhancing spatial representation and reference-frame manipulation in CP. To better understand how perceptual, cognitive and emotional factors influence walking in children with CP, Belmonti et al. (2013) included independent as well as accompanied locomotion. In CP, accompanying impairments in sensation, perception, cognition, may contribute to activity limitation beyond the influence of motor impairments (Rosenbaum et al., 2007). Since as well path planning and shifting from ego- to allocentric spatial reference frames are necessary requisites for mature locomotor control (Belmonti et al., 2013), difficulties with topographical memory test are probable in this patient group. It is therefore reasonable to believe that perceptual disorder may have played a role when performing the WalCT test, in particular in the GMFCS III group.

Time duration to perform a test with locomotion involved, may tell us about a child's motor behavior. For instance, in children with CP with less lower limb proprioception, walking velocity was found slower than in those with less symptoms of sensory disturbances (Damiano et al., 2013; Bartonek et al., 2016, 2018). To which extent a child's ability to change body position in space may influence time required to perform the topographical memory test, and thereby the child's working memory, cannot be answered from the results in this study. Yet, the results observed in the various subgroups may enhance understanding of a child's situation in the environment. Five of six children with indoor walking ability with a walking aid preferred performing the topographical memory test using 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. During the WalCT, the participant must turn its body on the squares in various directions. The use a wheelchair could possibly have been a strategy to avoid spatial insecurity also from deficits in proprioception of the lower limbs as well as not being obliged to visually control the feet during steps allowing awareness of surroundings. Preferring wheelchair during the test may also have been associated with visual perception disorder, often present in children with CP (Ego et al., 2015). However, the finding in one child at GMFCS III who performed the WalCT with a walking aid as properly as TD children of similar age despite requiring more time to move her body through the WalCT test, indicates the necessity to be aware of variations in the CP group.

Within the aim of this study, a further purpose was to explore visuo-spatial working memory with respect to spatial memory in navigational space, in children with CP. To investigate these two aspects of memory in children with CP is noteworthy because they demonstrate the contribution of motor action in spatial representation during the development. During the WalCT the subject has to move its body in space acting from a continuously changing direction. Other when performing the CBT test during which the subject is located in a static body position facing the experimenter, thus the environmental perspective with control of the surrounding space is largely eliminated. In line with our hypothesis, the CP group achieved significantly shorter CBT span than the TD group, as well as continuously decreasing with GMFCS level, however higher span than in navigational space requiring body movements. This finding is in accordance with Belmonti et al. who found similar results in the reaching space using the same method of CBT (Belmonti, Fiori, et al., 2015). Performing a sequence of block-tapping on a baseboard versus a sequence of steps in a route, thus, rely on different processes and strategies (Piccardi et al., 2008) and requires

differential cognitive strategies (Belmonti, Cioni, et al., 2015). Even in children with TD a greater effort is required when reproducing a sequence of steps in a large-scale space than when reproducing a sequence of reaching movements in a small-scale context (Piccardi et al., 2017).

Visuo-spatial difficulties are reported to be involved in the motor deficits of children with CP (Belmonti, Fiori, et al., 2015). We therefore investigated this aspect by using Raven's Colored Progressive Matrices test (CPM) that intends to provide a measure of cognitive functions such as visuo-perceptual ability and visuo-spatial reasoning (Pueyo et al., 2008). CPM was investigated solely in CP participants since the intention was to rule out possible differences between children with CP about the capability to reason on visuo-spatial material. In this study, CPM score as covariate showed no differences between the GMFCS levels neither on the results of CBT nor on WalCT.

Concerning age differences between the groups, the TD children were younger than all CP groups and GMFCS level I participants were younger than of levels II and III/IV. We therefore did not assume any positive influence of age on topographical working memory in this study group, however, to control for age factor we used age as a covariate in our statistics. We also found increased presence of prematurity in approximately 65% of the children. The difference was most accentuated in GMFCS level II where all eleven children were born pre-term. This result may be in agreement with that of Pavlova et al. (2007) who suggested prematurity and leukomalacia to be predictors of visual navigation disability in children with CP (Pavlova et al., 2007). A weakness of the present study however, is that there is not sufficient data to make any conclusion about the role of gestational age on topographical working due to the small participant number.



Authors in the field promote early exploration of navigational space in children with CP (Stanton et al., 2002; Pavlova et al., 2007; Belmonti, Fiori, et al., 2015). In the current study group, the participants in GMFCS II and III/IV performed similar in WalCT, yet the GMFCS III/IV group is considered to have only sparse navigation experiences compared to GMFCS II, who walk outdoors but with limitations. Therefore, the question arises whether outdoor locomotion to a limited extent supports exploration of navigational space sufficiently. In contrast, the GMFCS I group, walking without limitations, performed similar with the TD group indicating possibilities of earlier environmental exploration than GMFCS II and III/IV groups. Lobo et al. (Lobo et al., 2013) recommended providing children with frequent daily opportunities to actively maximize their interactions with objects, people, and events. This approach is thus of significant importance since interactions in the environment

contributes to development of navigational skills. Implications for the development of spatial awareness were suggested even in children with CP who may suffer neurological damage including spatial brain structures, and poor spatial learning was considered as secondary to their neurological conditions (Stanton et al., 2002). However, attention should be paid to each child's perceptual-motor conditions since it is likely that sensory disturbances and spatial orientation difficulties are associated with performance of topographical memory tasks.

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