



## Action planning in typically and atypically developing children (unilateral cerebral palsy)

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### ABSTRACT

In the present study, we investigated the development of action planning in children with unilateral Cerebral Palsy (CP, aged 3–6 years,  $n = 24$ ) and an age matched control group. To investigate action planning, participants performed a sequential movement task. They had to grasp an object (a wooden play sword) and place the sword in a hole in a wooden block. Our main dependent variable was the grip type that participants used, i.e., did they adapt their initial grip choice such that they would reach a comfortable posture at the end of the action? This end-state comfort effect has been abundantly shown in research on action planning, and is taken as evidence for anticipatory planning. The first aim of the study was to investigate the development of action planning in the unilateral CP group and the control group. Our hypothesis was that action planning improves with age in the control group, but not in the unilateral CP group. The results showed that planning was impaired in the unilateral CP group compared with the control group. Consistent with our hypothesis, we found an age effect in the control group, but not in the unilateral CP group. In the control group 5 and 6 years olds showed more anticipatory planning compared with the 3 and 4 years olds. The second aim of this study was to examine whether an intervention for children with unilateral CP (i.e., constrained induced movement therapy combined with bimanual training) affected action planning. The children with unilateral CP were therefore measured on the experimental task before and after an 8-week intervention period. The results showed that planning improved after the intervention. This finding suggests that action planning ability in young children with unilateral CP may be sensitive to improvement. These findings are discussed within the context of typical and atypical development of action planning and further guidelines for intervention in children with unilateral CP are given.

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### 1. Introduction

The age span between 3 and 10 years is critical for the development of motor control in children, as evinced by both behavioural studies (Ferrel, Bard, & Fleury, 2001; Hay, 1979; Hay, Bard, Ferrel, Olivier, & Fleury, 2005; Smyth & Mason, 1997; Thibaut & Thoussaint, 2010) as neuroimaging studies (Casey, Galvan, & Hare, 2005). In this age period, motor and sensory

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areas develop first, followed by higher order areas, such as the prefrontal cortex, which develop later (Casey, Tottenham, Liston, & Durston, 2005). An important aspect of motor control is motor planning. Motor planning can be defined as the ability to take the upcoming task demands into account when first taking hold of an object (Johnson-Frey, McCarty, & Keen, 2004; Mutsaerts, Steenbergen, & Bekkering, 2005, 2006; Steenbergen, Meulenbroek, & Rosenbaum, 2004). For example, a cup that is placed upside down and that needs to be turned over is initially grasped with an uncomfortable posture (thumb down, supination of forearm), such that the arm is in a comfortable posture (thumb up, pronation of the forearm) when the cup is turned over, i.e., at the end of the task. This phenomenon implies that participants planned the end of the action. Several studies showed that (adult) participants prefer to end an action with a 'comfortable end posture' and sacrifice comfort of the initial posture in order to attain this goal (e.g., Rosenbaum, Vaughan, Barnes, & Jorgensen, 1992).

Until now, the development of action planning in sequential tasks in children has only received limited attention, and results have been inconclusive. For example, Adalbjornsson, Fischman, and Rudisill (2008) studied two cohorts of children (2–3 and 5–6 years) that had to rotate a cup in order to pour water in it. They found that only a minority (11 of 40) of the children adapted their start posture in order to end the movement in a comfortable posture. No differences between these age groups were found. These findings suggest that action planning does not develop until age 6 (for consistent findings, see also Manoel & Moreira, 2005). In contrast, Smyth and Mason (1997) found that end posture planning developed in children between 3 and 8 years of age. Children had to rotate a bar, placed in different start orientations, into a target orientation. It was observed whether the children showed anticipatory planning, i.e., if they adapted the initial hand posture in order to reach a comfortable end posture. Planning improved with age, suggesting action planning develops between 3 and 8 years of age, although it has not yet reached adult levels at age 8. Consistently, Thibaut and Thoussaint (2010) showed that action planning increased from age 4 and till age 10. At age 10, a pattern of results similar to adults was observed.

Cerebral Palsy (CP) is a developmental disorder of movement and posture (Bax, Goldstein, Rosenbaum, Leviton, & Paneth, 2005). With a prevalence of 2.0–2.5 per 1000 living births, CP is the most common cause of severe disability in childhood (Blair & Watson, 2006). One of the most frequently occurring forms of CP is unilateral CP, where one vertical body side is affected, as a consequence of brain damage that primarily affects one hemisphere. Recently it has been proposed that the compromised action performance of children with unilateral CP is not only due to problems in action execution, but is also related to problems with action planning (Steenbergen & Gordon, 2006; Steenbergen, Verrel, & Gordon, 2007). Participants with unilateral CP were shown to be compromised in their capacity to be engaged in anticipatory action planning when using their unaffected arm (Mutsaerts et al., 2006; Steenbergen, Hulstijn, & Dortmans, 2000; Steenbergen et al., 2004). Instead of planning the end of the action they were shown to use a step-by-step planning strategy. That is, they first plan the movement towards the target object, and only after having grasped the object the next movement is subsequently planned (Mutsaerts et al., 2005; Steenbergen & Van der Kamp, 2004). This is in contrast with control participants that plan the entire action sequence prior to the start of the first movement. Rehabilitation efforts in children with unilateral CP are predominantly aimed at facilitation of the motor execution problems of the affected side. The beneficial effects of rehabilitation programs are often established by measures of movement *execution*, for example, the assessment of wrist flexion and extension, motor proficiency and speed, or ratings of movement quality (Charles & Gordon, 2007; Eliasson & Gordon, 2000; Gordon, Charles, & Wolf, 2006; Taub, Ramey, DeLuca, & Echols, 2004). However, the potential beneficial effects of therapeutic programs on motor *planning* have never been scrutinized.

The first aim of the present study was to investigate action planning in young children (aged 3–6) with and without unilateral CP as this age range is critical for the development of planning in typically developing children. Based on previous literature we expected to find an increase in end posture planning with age in the typically developing children. In contrast, as ample evidence suggests that action planning is impaired in adolescents with unilateral CP (Crajé, Van der Kamp, & Steenbergen, 2009; Mutsaerts et al., 2005, 2006), we expect no developmental improvement in action planning in the children with unilateral CP.

The second aim of our study was to examine whether action planning in children with unilateral CP is prone to change after intervention. Until now, it has not been investigated whether action planning capacities can be improved by therapeutic programs. This is surprising given the constraining effects of compromised planning on action performance (Steenbergen & Gordon, 2006). Therefore, our second aim of the present study was to explore the potential beneficial effect of an 8-week period of intensive hand function training on motor planning in children with unilateral CP (Aarts, Jongerius, Geerdink, Van Limbeek, & Geurts, *in press*). Despite the fact that the training was mainly focused on the affected side, we hypothesize that it may alleviate motor planning of the less-affected side based on two lines of evidence. First, anticipatory planning is based on previous manipulatory experience with an object (Salimi, Hollender, Frazier, & Gordon, 2000) and variability of practice, a facet that is central in CIMT, may further promote anticipatory planning (Schmidt & Wrisberg, 2000). Second, anticipatory planning can be transferred between both body sides in both healthy children and adults (Gordon, Forssbergh, & Iwasaki, 1994; Westling & Johansson, 1984). Specifically, weight and friction information of an object gained during previous lift with one hand can be used to scale the fingertip forces during subsequent manipulations with the contralateral hand. More importantly, in a recent study, Gordon, Charles, and Steenbergen (2006) studying children with unilateral CP, showed that performance related to anticipatory fingertip force control can be improved in the less-affected side if movements are first performed with the affected hand. Based on these two lines of evidence we hypothesize that intensive and variable upper limb training may be beneficial for motor planning of the less-affected side.

**Table 1**  
Participant information.

Participant	Affected hand	Age	Sex	Melbourne score
1	R	3	M	51
2	R	3	M	43
3	L	3	F	50
4	L	3	F	61
5	L	3	F	76
6	L	3	M	46
7	R	4	M	71
8	R	4	M	73
9	R	4	M	38
10	L	4	M	47
11	L	4	M	62
12	L	4	M	65
13	R	5	F	74
14	R	5	F	56
15	R	5	F	48
16	R	5	F	68
17	R	5	F	65
18	L	5	M	62
19	R	6	F	61
20	R	6	F	28
21	R	6	M	67
22	L	6	M	61
23	L	6	F	69
24	L	6	M	73

The Melbourne measures upper limb capacities of the affected hand, with a minimum score of 0 and a maximum score of 100.

## 2. Methods

### 2.1. Participants

The unilateral Cerebral Palsy (CP) group consisted of 24 children between 3 and 6 years of age ( $n = 6$  for each age group, see Table 1 for participant information). Eleven children had their left arm affected (left unilateral CP), and 13 children had their right arm affected (right unilateral CP). All children were recruited from an upper limb training program for young children (3–6 years of age) with unilateral CP. Upper limb function of the affected arm was assessed with the Melbourne Assessment of unilateral upper limb function (Randall, 1999). The age matched control group consisted of 24 children (5 left handers). Parents gave permission for their children to participate. The study was approved by the local ethics committee.

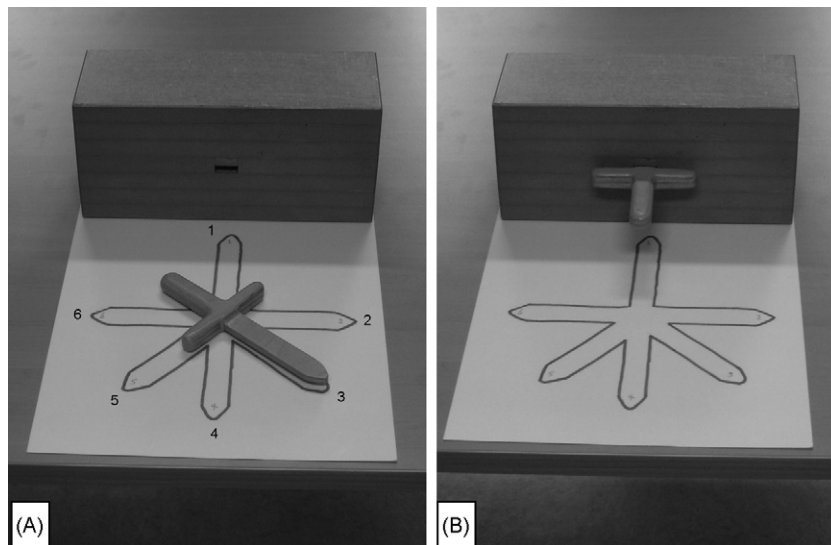
### 2.2. Upper limb training

All children with unilateral CP were enrolled in the so-called ‘Pirate group’ at the ‘Sint Maartenskliniek’ in Nijmegen, The Netherlands. This child centered intervention consisted of a combination of 6 weeks constrained induced movement therapy (CIMT) followed by 2 weeks of bimanual training (BiT), for 9 h a week (Aarts et al., in press). These 2 weeks provide the opportunity to apply the use of the affected hand in bimanual activities. Individual therapy was given in groups of 6 children by 4 occupational therapists, 1 physical therapist and 1 therapy assistant using shaping and repetitive task practice. In the training program the children were told to be pirates that are wounded on the less-affected arm. The training consisted of (among others) actions that were related to the pirate setting, like using a sword, beat the drums, sweep the deck, and cook for the other pirates. Inclusion criteria for the Pirate group were: (1) cerebral palsy with a unilateral or severely asymmetric, bilateral spastic movement impairment, (2) age 2–8 years<sup>1</sup> and (3) Manual Ability Classification System (MACS, Eliasson et al., 2006) scores I, II or III. Exclusion criteria were: (1) intellectual disability such that simple tasks could not be understood or executed (i.e., developmental age below 2 years), (2) inability to combine the study protocol with the regular school program, and (3) ability to walk independently without a walking aid.

### 2.3. Procedure

The experimental task to measure anticipatory action planning was developed to attract the attention of the ‘pirates’. It consisted of stinging a wooden sword into a tight hole in a wooden block. The experimental task was not specifically exercised during the training. During the experiment, each child sat on his/her own height adjusted ‘tripp-trapp chair’ such that the feet were supported and the child could rest the underarms on the table. A wooden sword (length 18.0 cm, width

<sup>1</sup> Notably, not all children were included in the present study, as the group sizes of the 2, 7 and 8 years olds were too small.

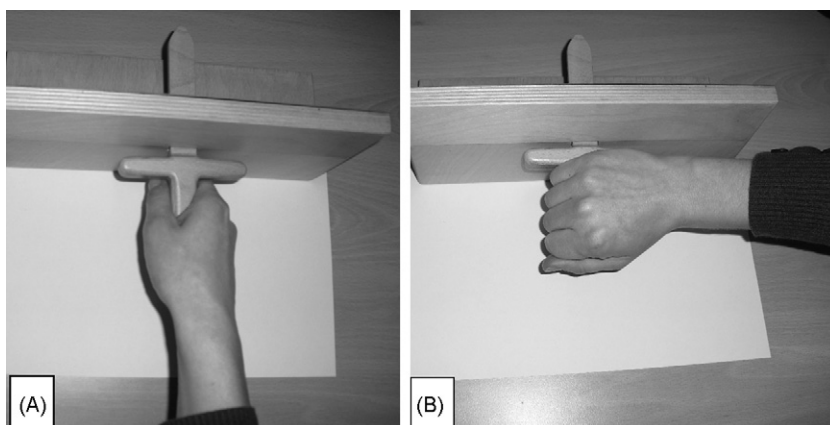


**Fig. 1.** The experimental setup from the participants' perspective, with the sword at start position 3 in this particular trial (A). The required end position is shown in (B). Start positions were numbered 1–6 in a clockwise direction. The sword position with the blade towards the target location was designated as position 1.

2.0 cm, height 1.2 cm, length handle 9.5 cm) was placed on the table and had to be stung into a tight fitting hole in a wooden block (27.0 cm × 13.0 cm × 13.0 cm, hole: 2.0 cm × 0.8 cm). The sword was always presented on a sheet of paper (30 cm long and 28 cm width) with a mold on it of 6 possible sword rotations (see Fig. 1). Only the long side of the sword (the blade), which was more flat than the sword handle, could be inserted in the hole. The child was told that we wanted to learn from a real pirate how a sword had to be placed in a wooden block, and the child was asked to show this. The experiment always started with a simple trial that did not require any sword rotation (position 1). After successful performance, we asked to show us the trick again, but now when the sword was placed in a different start rotation. Every rotation was repeated three times in random order, resulting in a total of 18 trials per child. The children performed the task with the less-affected hand. Control children used the dominant hand. No specific instructions were given about the way in which the task should be performed. The experimental session, that never exceeded 10 min, was registered with a digital video camera for off line data analysis. In the unilateral CP group two sessions were performed, one prior to, and one immediately following the training (8 weeks later). Control children performed one session.

#### 2.4. Data analysis

We were interested in the grip choice as a function of the rotation angle of the sword. Therefore, it was scored whether the posture of the hand at the end of the action was comfortable, i.e., with the thumb towards the end goal (see Fig. 2A), or uncomfortable, i.e., with the thumb opposite to the end goal (see Fig. 2B). For analyses we distinguished between critical trials and control trials. Critical trials were defined as trials where an uncomfortable start posture was needed to allow a



**Fig. 2.** Comfortable end posture (A) and uncomfortable end posture (B).

comfortable end posture. Control trials were trials where a comfortable start posture resulted in a comfortable end posture. Hence, planning was especially required in the critical trials. For data analyses the proportion of comfortable end postures in the critical conditions and the control conditions was used. For the critical conditions sword orientations 2 and 3 were used for the right handers, and sword orientations 5 and 6 for the left handers. The remaining orientations were regarded as control conditions (i.e., for the left handers orientation 1, 2, 3 and 4, and for the right handers orientations 1, 4, 5 and 6). Thus, for every participant there were two scores, an average for the critical conditions and an average for the control conditions.

We conducted the following analyses. First, we compared the unilateral CP group with the age matched typically developing children (3–6 years) using a repeated measures ANOVA with 1 within subjects factor (Condition: critical versus control) and 2 between subjects factors (Age [3, 4, 5, and 6 years] and Group [unilateral CP and control]). Second, the effect of training was evaluated within in the unilateral CP group using a 2 (Condition: critical versus control)  $\times$  2 (Measurement: pre versus post) repeated measures ANOVA with Age (3, 4, 5, and 6 years) as between subjects factor.

### 3. Results

Our main dependent variable was the planning strategy that the children used to solve the task. Trials in which children were not paying attention or were playing, were not used in the analysis (82 trials, 7%). The data were normally distributed.

#### 3.1. Development of action planning in the unilateral CP group and the control group

The proportion of comfortable end postures in the control and critical conditions, separated for age and group, is depicted in Fig. 3. First, there were significant between subjects effect of Group ( $F(1, 32) = 7.88, p < .01$ ) and Age ( $F(3, 32) = 3.207, p < .01$ ). The effect of Group indicates that the proportion of comfortable end postures was higher in the control group compared with the unilateral CP group, whereas the effect of Age indicates that the proportion of comfortable end postures increased with age. Second, there were significant within subject effects of Condition and an interaction effect of Condition  $\times$  Group  $\times$  Age. These effects indicate that the proportion of comfortable end postures was higher in the control conditions, compared with the critical conditions (main effect of Condition ( $F(1, 40) = 284.79, p < .001$ )). This effect was the same in the control group and in the unilateral CP group, as there was no interaction effect of Condition  $\times$  Group ( $F < 1$ ). However, the 3-way interaction effect of Condition  $\times$  Group  $\times$  Age ( $F(3, 40) = 4.70, p < .01$ ) indicates that this difference was not similar for all age groups within the two groups. Post hoc comparisons with Bonferroni correction showed that the proportion of comfortable end postures was different for control and critical conditions for all age groups in the unilateral CP group, but only for the 3 and 4 years old in the control group. Thus, for the 5 and 6 years olds in the control group there was no significant difference between control and critical conditions. This finding suggests that the proportion of comfortable end postures increases with age in the control group, but not in the unilateral CP group.

#### 3.2. Effect of training

We found a significant main effect of Measurement ( $F(1, 20) = 13.77, p < .01$ ) which indicates that the proportion of anticipatory planned trials was higher in the post measurement compared with the pre measurement (see Fig. 4). A significant main effect of Condition ( $F(1, 20) = 145.80, p < .01$ ) indicated that proportion anticipatory planned trials was higher in the critical trials compared with the control trials. This is as expected, as planning is especially required in the critical trials. Planning improved in both the control and the critical conditions, as there was no interaction effect of Measurement  $\times$  Condition. Finally, there were no (interaction) effects of Age, indicating the improvement was similar in the age groups.

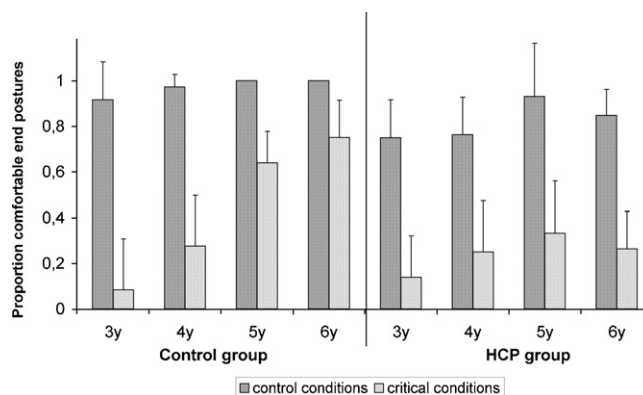


Fig. 3. The proportion of comfortable end postures for the Control group (left) and the unilateral CP group (right), separated for age. Dark grey bars represent control conditions, whereas light grey bars represent critical conditions. Error bars represent 2SE.

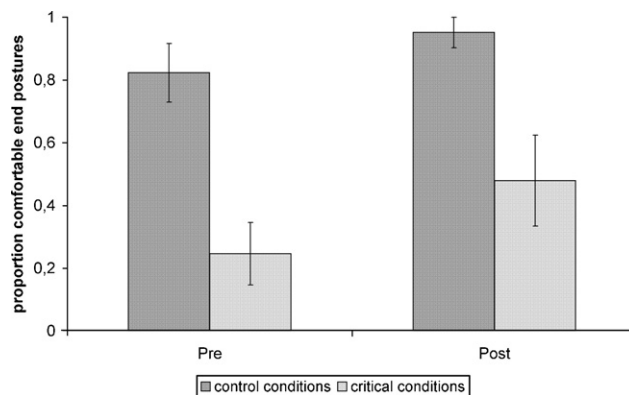


Fig. 4. The proportion of comfortable end postures in the unilateral CP group before and after intervention. Dark grey bars represent control conditions, whereas light grey bars represent critical conditions. Error bars represent 2SE.

### 3.3. Correlation with Melbourne scores

To investigate if the severity of hand function affected the improvement of the training, we calculated the Pearson's correlations between the Melbourne scores and the individual difference in proportion anticipatory planned trials before and after training, viz. planning improvement. A significant correlation was found between Melbourne and difference score for the Control trials ( $\rho = -.432, p < .05$ ), whereas the correlation between Melbourne and difference score for the Critical trials was not significant ( $\rho = -.354, p = .09$ ). This finding suggests that participants with lower Melbourne scores have more improvement on the relatively easy conditions.

## 4. Discussion

In the present study we investigated the development of action planning in typically developing children and young children with unilateral CP (four age cohorts, 3, 4, 5, and 6 years). In line with our hypothesis, we found that planning improved with increasing age in the typically developing children. The younger children (aged 3 and 4) had a low proportion of comfortable end posture in the critical conditions. In the majority of trials they used a comfortable start posture to grasp the sword, which resulted in an uncomfortable end posture. This finding suggests they did not plan the movement ahead. For the older children (aged 5 and 6), the proportion of comfortable end postures was, higher, and similar for the critical and control conditions, suggesting an increased level of anticipatory planning in comparison with the 3 and 4 years olds. However, overall, the older children did not reach a level that was similar to adults, as they did not show end posture planning in all (i.e., control and critical) trials, suggesting that planning is not adult-like at age 6. This finding replicates previous findings in the literature, which showed improvement in planning until age 10 (Manoel & Moreira, 2005; Thibaut & Thoussaint, 2010).

As anticipated, the proportion of comfortable end postures was lower in the unilateral CP group, compared with the control group. This finding is consistent with other studies in adolescents with unilateral CP that have repeatedly shown impaired action planning (e.g., Steenbergen & Gordon, 2006). We did not find an age effect on action planning in the unilateral CP group, suggesting the planning capacities did not change between 3 and 6 years of age. This was in line with our hypothesis, because compromised action planning has been shown in older children with unilateral CP (Mutsaerts et al., 2005, 2006). Interestingly however, our results showed that action planning in the unilateral CP group improved after an intervention period. This finding suggests that planning *can* be trained in children with unilateral CP. As far as we know this is the first study to show planning capacities can be improved in young children with unilateral CP. This is an important finding, as planning problems do not only affect the affected hand, but also the unaffected hand, which has a major impact on activities of daily life (Mutsaerts et al., 2006; Steenbergen et al., 2000, 2004). Further studies are warranted to investigate (1) the best form of intervention to train planning capacities; and (2) individual differences between children with unilateral CP that may benefit successful training. Below we will elaborate on this.

A first question to ask is: What is the best way to train planning capacities? Our results have at least two suggestions regarding this issue. First, in our study children with unilateral CP were not explicitly trained on action planning, but still planning improved. This is surprising as the training that the children received was aimed at practicing/repeating a variety of tasks with the affected arm followed by goal-directed task-specific bimanual training, from gross motor skills to fine motor skills (Aarts et al., *in press*). The improvement in planning of the less-affected side, indicates that variable practice of motor tasks may be sufficient to improve motor planning (Schmidt & Wrisberg, 2000), even without specific motor planning training. It may be speculated that the variety of tasks with different complexities that were practiced during intervention provides the necessary ballpark of 'hands on experience' to improve planning. Stated differently, our study cannot disentangle the necessary prerequisites for planning improvement, but variety of task practice is a likely factor. Second, we

assessed motor planning in the relatively unaffected side, whereas the intervention was predominantly focused on the affected side. This finding may suggest an intermanual transfer (Gordon, Charles, et al., 2006) and points to the fact that action planning is a higher order cognitive function.

Because motor planning is a cognitive aspect of motor control, the use of motor imagery, may be a promising technique to train motor planning in children with unilateral CP (Steenbergen, Crajé, Nilsson, & Gordon, 2009). Motor imagery can be defined as the ability to mentally perform movements, without over motor output. Mental practice effects are thought to be a result of the rehearsing of the cognitive components of the motor task (Mulder, 2007). Johnson-Frey (2004) argued that the observed effects in motor imagery can be attributed to experience-dependent changes in higher-level brain regions involved in the planning, rather than the execution, of movements. It was recently suggested that motor imagery may be used as a 'backdoor' access to the motor system, or neural representation of movement (Sharma, Pomeroy, & Baron, 2006). Indeed, converging evidence supports the notion that motor imagery training may promote general rehabilitation of upper limb function in individuals with subacute and chronic stroke (Braun, Beurskens, Brom, Schack, & Wade, 2006; Crajé, De Graaf, Lem, Geurts, & Steenbergen, in press; Sharma et al., 2006), and in children with Developmental Coordination Disorder (Wilson, Thomas, & Maruff, 2002). However, a recent review showed that there is still a void in studies on the use of motor imagery for improving aspects of upper limb control in children with unilateral CP (Steenbergen et al., 2009).

The second issue that warrants further investigation, are individual differences related to age, side of lesion or severity of unilateral CP among the participants that may affect benefit of planning training. First, in the present study, we did not find age-related effects of the training, suggesting that planning improved similarly in all age groups. However, we did not measure children older than 6 years. As planning is a cognitive process, one might argue that older children may be more susceptible to learn planning strategies, and 'the-earlier-the-better' rule may not apply in this specific situation. This may be supported by the finding that in control children development of planning is not finished until the age of 10 (Thibaut & Thoussaint, 2010). Second, the side of lesion may play a role. Previous studies have shown that planning problems are more severe when the right body side is affected, i.e., left hemisphere lesions (Crajé et al., 2009; Steenbergen et al., 2004). One may suggest that children who are affected on the right body side have more capabilities for improving planning. Finally, the severity of unilateral CP may have an impact on the trainability of planning. For example, a recent study of Williams, Thomas, Maruff, and Wilson (2008), showed that children with mild DCD are better able to use MI than children with severe DCD (for similar ERP results with adults with unilateral CP from our own lab, see Van Elk et al., Invited revision submitted).

A final note of caution should be mentioned. As the present study is the first to study the potential effects of an existing intervention on action planning in unilateral CP it was set up as an experimental trial and not designed as a randomized controlled trial. Therefore, we cannot be conclusive about the underlying factor(s) for improvement in motor planning. For example, it is possible that the effect can be ascribed to the attention that the children received or to the specific tasks trained during the training. Also, the group size per age group was relatively small, and therefore the interpretation of the results regarding age must be taken with due caution. Collectively, these promising results warrant further study. In particular, they beg the question as to what extent more specific training may facilitate motor planning in these children.

### Conflict of interest statement

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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