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The development of anticipatory action planning in children with unilateral cerebral palsy



Hilde Krajenbrink^{a,b}, Ali Crichton^{a,e}, Bert Steenbergen^{b,c}, Brian Hoare^{a,d,e,*}

^a Department of Paediatrics, Monash University, Clayton, Victoria, 3168, Australia

^b Behavioural Science Institute, Nijmegen, the Netherlands

^c Australian Catholic University, School of Psychology, Melbourne, Australia

^d School of Occupational Therapy, La Trobe University, Bundoora, Victoria, 3168, Australia

^e Victorian Paediatric Rehabilitation Service, Monash Children's Hospital, 246 Clayton Rd, Clayton, Victoria, 3168, Australia

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ABSTRACT

Background Previous studies suggest that compromised bimanual performance experienced by children with unilateral cerebral palsy (CP) is not only due to difficulties in action execution but may also be a result of impaired anticipatory action planning.

Aims The effect of age and side of hemiplegia were examined and the relationship between anticipatory action planning, unimanual capacity and bimanual performance was explored.

Methods and procedures Using a multi-centre, prospective, cross-sectional observational design, anticipatory action planning was analyzed in 104 children with unilateral cerebral palsy, aged 6–12 years, using the sword task.

Outcomes and results Anticipatory action planning did not improve with age in children with unilateral CP, aged between 6–12 years. No differences were found between children with left or right hemiplegia. Finally, anticipatory action planning was not related to unimanual capacity or bimanual performance.

Conclusion and implications This study demonstrates anticipatory action planning, measured using the sword task, does not improve with age in children with unilateral CP and is not related to bimanual performance or laterality. Future studies of anticipatory action planning in children with unilateral CP should consider using measures that require effective anticipatory action planning for successful task completion rather than end state comfort.

What this paper adds

The results from this study demonstrate that children with unilateral cerebral palsy (CP), aged 6 to 12 years old, do not naturally develop anticipatory action planning skills. This provides support for the need for therapy focused on improving anticipatory action planning, not just motor execution. Further evaluation of these interventions is necessary to identify whether anticipatory action planning is amenable to improvement from targeted training. In addition, we found that anticipatory action planning is not lateralized in children with unilateral CP and anticipatory action planning was not related to unimanual capacity or bimanual performance. Future studies of anticipatory action planning in children with unilateral CP are advised to use measures such as the hexagonal knob task that require effective anticipatory action planning for successful task completion rather than end state comfort.

* Corresponding author at: Department of Paediatrics, Monash University, Clayton, Victoria, 3168, Australia.

E-mail address: brianhoare@cpteaching.com (B. Hoare).

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1. Introduction

The most common subtype of cerebral palsy (CP) is unilateral CP, which is characterized by deficits in the posture and motor function of one side of the body, as a consequence of brain damage that primarily affects one hemisphere (Stanley, Blair, & Alberman, 2000). Previous studies suggest that compromised task performance experienced by children with unilateral CP is not only due to problems in action execution, but may also be a result of problems with anticipatory action planning (Mutsaerts, Steenbergen, & Bekkering, 2006; Mutsaerts, Steenbergen, & Bekkering, 2005; Steenbergen & Gordon, 2006; Steenbergen, Verrel, & Gordon, 2007; Steenbergen, Jongbloed-Pereboom, Spruijt, & Gordon, 2013).

Anticipatory action planning can be defined as the ability to anticipate the end of an upcoming action prior to commencing movement (Johnson-Frey, McCarty, & Keen, 2004). Planning movements in advance is especially advantageous in sequential tasks that involve the use of tools (Steenbergen, Meulenbroek, & Rosenbaum, 2004). Consequently, a commonly used method to study anticipatory action planning involves grasping an object with the intention of placing it in a specific goal position and/or in a specific orientation at the end of the task (Steenbergen et al., 2013). It has been repeatedly found that individuals tend to sacrifice comfort of the start posture so that the task is finished with the hand and arm in a biomechanically efficient and comfortable end-posture. This effect is known as the End-State Comfort (ESC) effect (Cohen & Rosenbaum, 2004; Rosenbaum, Barnes, Vaughan, & Jorgensen, 1992). By avoiding extreme joint angles, ending in a comfortable posture allows more precision to be exerted at the end of the task (Short & Cauraugh, 1999) and results in a more coordinated, graceful performance.

The ESC effect is a well-established phenomenon in healthy adult populations (Rosenbaum, Chapman, Weigelt, Weiss, & van der We, 2012). In typically developing children, however, results have been inconclusive with regard to the onset of the ESC effect during development (Wunsch, Henning, Aschersleben, & Weigelt, 2013). Some studies suggest the ESC effect is already evident in 3 year-old children (Jovanovic & Schwarzer, 2011; Knudsen, Henning, Wunsch, Weigelt, & Aschersleben, 2012) with increasing percentages of comfortable end postures with increasing age (Jongbloed-Pereboom, Nijhuis-van der Sanden, Saraber-Schiphorst, Craje, & Steenbergen, 2013; Stöckel, Hughes, & Schack, 2012; Thibaut & Toussaint, 2010; Wunsch, Pfister, Henning, Aschersleben, & Weigelt, 2016). Other studies have found a low percentage of comfortable end postures in children aged 2–7 years with little improvements with increasing age (Adalbjornsson, Fischman, & Rudisill, 2008; Manoel & Moreira, 2005; van Swieten et al., 2010). These conflicting results are likely due to differences in the type and complexity of the tasks that have been used to assess the ESC effect, which include the cup manipulation task (e.g., Adalbjornsson et al., 2008), the bar transport task (e.g., Stöckel et al., 2012), the handle rotation task/hexagonal knob task (e.g., van Swieten et al., 2010), the grasp height task (e.g., Wunsch et al., 2016), and the sword task (e.g., Jongbloed-Pereboom et al., 2013). These tasks substantially differ in the extent to which precision is needed at the end of the task which is known to affect outcomes (e.g., Jongbloed-Pereboom, Spruijt, Nijhuis-van der Sanden, & Steenbergen, 2016).

Studies on anticipatory action planning in children with unilateral CP suggest that anticipatory action planning does not improve with age (Craje, Aarts, Nijhuis-van der Sanden, & Steenbergen, 2010; Janssen & Steenbergen, 2011; Lust, Spruijt, Wilson, & Steenbergen, 2017). Consistent with findings that found dominant networks for planning processes in healthy adults that involve temporal, parietal, and frontal areas within the left hemisphere (e.g., Johnson-Frey, 2004; Schluter, Krams, Rushworth, & Passingham, 2001), adolescents with right unilateral CP have been found to have greater impairment than children with left unilateral CP, indirectly supporting left hemisphere specialisation for anticipatory action planning (Craje, van der Kamp, & Steenbergen, 2009; Steenbergen et al., 2004). However, these results need to be interpreted with caution given the relatively small sample sizes of both studies ($n = 22$ and $n = 11$, respectively). In addition, outcomes reported by Craje et al. (2009) were also based on whether participants used a consistent planning strategy based on their initial grip type distribution during the bar transport task. Both planning according to ESC (i.e., always ending with a comfortable end posture and switching between pronated and supinated start postures) and forearm pronation at start (i.e., always using a pronated start posture and switching between comfortable and uncomfortable end postures) were examined. There was no significant difference between children with right or left hemisphere damage when evaluating ESC effect alone. In a larger ($n = 76$) study, Kirkpatrick, Pearse, Eyre, and Basu (2013) also did not find differences in anticipatory action planning between children with left or right unilateral CP using the hexagonal knob task, suggesting a more distributed system for anticipatory action planning across both hemispheres.

Compromised anticipatory action planning is considered to be an underlying cause of impaired action execution in children with unilateral CP (Steenbergen et al., 2013), however, little is known about the relationship between them. In a group of typically developing children, Jongbloed-Pereboom et al. (2013) did not find a relationship between anticipatory action planning and unimanual capacity as measured with the Box and Blocks Test (BBT), $r = .28$. Similarly, in a group of children with CP, Mutsaerts et al. (2006) also did not find a relationship between the number of failures on an anticipatory action planning task and BBT scores. When the motor requirements for the BBT are considered, it is perhaps not surprising that a repetitive unimanual grasp and release task does not require high level cognitive skills as opposed to more complex bimanual tasks. However, Kirkpatrick et al. (2013) also did not find a relationship between anticipatory action planning and the ABILHAND-Kids questionnaire for manual ability. As the authors suggest, outcomes from a parental questionnaire that focuses on whether the child can perform a task, rather than the manner in which this task is performed, may also not reflect a requirement for anticipatory action planning. In our study, we will use the Assisting Hand Assessment (AHA), an observational-based measure that includes activities which impose a higher cognitive demand compared with unimanual grasp/release, to examine the association between anticipatory action planning on bimanual performance.

1.1. Aim

The broad aim of this study is to examine the effect of age and side of hemiplegia on the development of anticipatory action

planning in a large group of children with unilateral CP. We will also explore the relationship between anticipatory action planning and the Assisting Hand Assessment (AHA), an observational measure of bimanual performance.

1.1.1. Primary aim

Using the sword task, examine the developmental trajectory of anticipatory action planning in a large group of 6–12-year-old children with unilateral CP.

1.1.2. Secondary aims

Investigate the effect of side of hemiplegia on anticipatory action planning in children with unilateral CP.

1.1.3. Tertiary aims

Investigate the relationship between anticipatory action planning with two validated measures of unimanual capacity and bimanual performance: the BBT for manual dexterity and the AHA for bimanual performance in children with unilateral CP.

1.2. Hypothesis

1.2.1. Primary hypothesis

In children with unilateral CP, there will be no significant effect of age on development of anticipatory action planning.

1.2.2. Secondary hypothesis

In children with unilateral CP, there will be no significant effect of the side of hemiplegia on development of anticipatory action planning.

1.2.3. Tertiary hypothesis

In children with unilateral CP, there will be a relationship between anticipatory action planning and the AHA but not for the BBT.

2. Material and methods

2.1. Trial registration

This study has been registered (*information has been removed to prevent author identification*).

2.2. Design

Prospective, cross-sectional observational design.

2.3. Participants

The current study is part of a larger research project examining cognition and bimanual performance in children with unilateral CP. Children were recruited from five pediatric treatment centers (*information has been removed to prevent author identification*). Children were eligible to participate if they had a diagnosis of congenital unilateral CP; age 6 to 12 years at time of assessment; and had sufficient cooperation to complete the assessment. Children who otherwise met the inclusion criteria were excluded if they had upper limb surgery within 12 months of assessment or received Botulinum toxin-A injection within three months of assessment.

2.4. Ethics

Ethical approval was received from all sites. Further information has been removed to prevent author identification. Parents or guardians of all participants provided informed written consent for their child to take part in the study.

2.5. Data collection

Clinical assessments were undertaken by trained occupational therapists at each participating site. For descriptive purposes, children were classified using the Manual Ability Classification System (MACS; Eliasson et al., 2006), the Gross Motor Function Classification System (GMFCS; Palisano, Rosenbaum, Walter, & Russell, 1997), and the Wechsler Intelligence Scale for Children (4th Ed) (WISC-IV; Wechsler, 2003). The sword task was scored from video footage by two independent raters who were blinded to other scores. Consistent with Jongbloed-Pereboom et al. (2013), the inter-rater reliability in our sample was good with an average measure ICC of .85, 95% [.84, .86], $F(2260) = 6.70$, $p < .001$, calculated based on a mean-rating ($k = 2$), absolute-agreement, two-way mixed effects model.

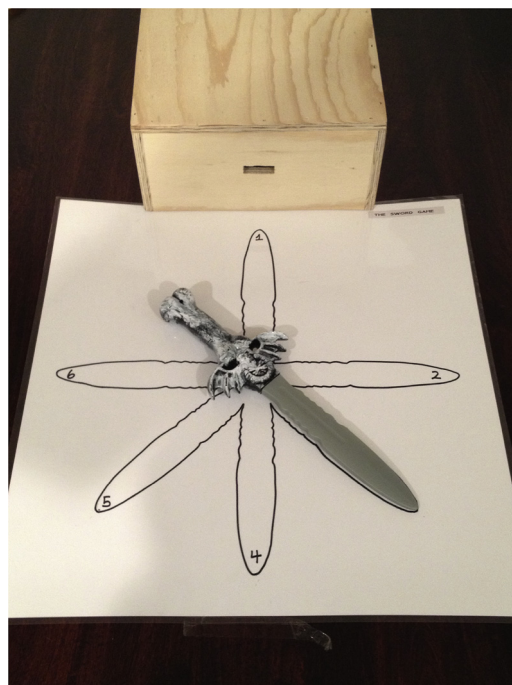


Fig. 1. Setup of the sword task, sword in orientation 3.

2.6. Tasks

2.6.1. Anticipatory action planning

Anticipatory action planning was assessed using the sword task (Craje et al., 2010). The sword task has previously been used in a study of children with unilateral CP (Craje et al., 2010) and found to be valid and reliable in typically developing children (Jongbloed-Pereboom et al., 2013). Administration was designed to be interesting and motivating for children (Craje et al., 2010). During the design phase of this study in 2011, the sword task was deemed to be the most appropriate measure of anticipatory action planning for the study population, especially in a clinical setting. Sitting at a table, children were asked to use their dominant hand to grasp the handle of a plastic sword that had been placed randomly in one of six positions on a template board (see Fig. 1). Children were required to place the sword into a hole in a wooden block. Two orientations served as experimental or critical orientations, in which children were required to sacrifice comfort of the start position in order to be able to end the task in a comfortable posture (orientation 2 and 3 for right-handed participants and orientation 5 and 6 for left-handed participants). The other four orientations served as control orientations, in which comfort at the start resulted in comfort at the end. To avoid the confounding influence of musculo-skeletal impairment children were instructed to use their dominant hand to pick up the sword using a power grip which was demonstrated by the experimenter. Three blocks of six trials (one for each orientation) resulted in a total of 18 trials. Trial orientations were randomized within each block. Trials were recorded with a video camera for offline scoring by two independent raters. The percentage of comfortable end postures in both, the critical orientations and the control orientations, were used in the analysis. Outcomes were only included when the sword task was administered correctly. Outcomes were excluded when at least one of the raters considered two or more of the three trials for a certain orientation were performed incorrectly.

2.6.2. Unimanual capacity

The Box and Blocks Test (BBT) was used to measure unimanual capacity of both upper limbs (Mathiowetz, Volland, Kashman, & Weber, 1985). In 60 seconds, children were required to move as many 2.5 cm blocks as possible from one compartment, over a low partition, to another. The number of blocks transported with each hand were used in the analysis. Validity of the BBT and test-retest reliability of the right hand and left hand were good, $r = .98$ and $r = .94$, respectively (Mathiowetz et al., 1985).

2.6.3. Bimanual performance

Bimanual performance was assessed using the Assisting Hand Assessment (AHA School Kids Version 5.0; Holmefur & Krumlinde-Sundholm, 2016) The School Kids AHA is a standardized, criterion-referenced test for use with children aged 6 to 12 years. In the context of one of two board games, the AHA aims to measure how effectively a child uses their more-affected hand in bimanual activities using a 4-point rating scale across 20 items. The session was videotaped and later scored from video observation by a certified AHA rater (occupational therapist). Raw scores are converted to internal level data using Rasch analysis. Rescaled logit-based AHA units ranging from 0 to 100 were obtained and used in the analysis. Validity of the AHA and test-retest reliability were

excellent, ICC = .99 (Holmefur & Krumlinde-Sundholm, 2016; Holmefur, Aarts, Hoare, & Krumlinde-Sundholm, 2009; Krumlinde-Sundholm, Holmefur, Kottorp, & Eliasson, 2007).

2.7. Data analysis

Shapiro-Wilk tests indicated that percentages of comfortable end-postures in both the critical orientations, skewness of 0.29 ($SE = 0.24$) and kurtosis of -1.37 ($SE = 0.47$), and the non-critical orientations, skewness of -1.72 ($SE = 0.24$) and kurtosis of 1.97 ($SE = 0.47$), were not normally distributed. Therefore, non-parametric tests were used. Alpha level was set at .05. All analyses were performed using SPSS version 22 (IBM Corp Released, 2013).

To analyze the effect of age on development of anticipatory action planning, a Kruskal-Wallis test was performed for both the percentage of comfortable end postures in the critical orientations and in the non-critical orientations. Differences between children with left or right hemiplegia and those with the presence or absence of impaired ESC were analyzed with Mann-Whitney U test. Finally, the association between anticipatory action planning and unimanual and bimanual performance were tested with Spearman's correlation coefficients. Correlation coefficients greater than 0.70 were considered as high, 0.50 to 0.70 as moderate, 0.30 to 0.50 as low and less than 0.30 as negligible (Mukaka, 2012).

3. Results

A total of 131 children with unilateral CP participated in this study. Outcomes from five children were excluded due to missing data for the sword task. A further 22 children were excluded as at least one rater considered two or more of the three trials for a certain orientation were performed incorrectly. In total, data from 104 children (63 males; 48 right hemiplegia; mean age = 9y,2 m; $SD = 1y,10 m$; range: 6y,1 m - 12y,8 m; MACS I: $N = 25$, II: $N = 74$, III: $N = 5$; GMFCS I: $N = 79$, II: $N = 25$) with unilateral CP were included in the analysis (see Table 1). Thirty three percent of the children were born preterm (< 37 weeks) and 31% had a history of seizures.

Table 2 shows an overview of the mean, median, and range of the percentage of comfortable end postures in the critical and the non-critical orientations. In addition, the mean percentage of comfortable end postures in the action planning task as a function of age are shown in Fig. 2. Scores varied between 0–100%, with most children either ending 0% (28 in total, 27% of the sample) or 100% (22 in total, 21% of the sample) of critical trials in a comfortable end posture. There were no differences between the different age groups in the percentages of comfortable end postures in both the critical orientations, $H(6) = 4.06$, $p = .668$, and the non-critical orientations, $H(6) = 1.72$, $p = .944$.

The percentage of comfortable end postures in the critical orientations did not differ for participants with left hemiplegia ($Mdn = 50.24$, $IQR = 83.33$) or right hemiplegia ($Mdn = 55.14$, $IQR = 66.67$), $U = 1217.50$, $p = .403$.

There was no association found between anticipatory action planning and unimanual capacity (BBT) using the more-affected hand ($\rho = -.136$, $p = .169$) or the less-affected hand ($\rho = -.004$, $p = .964$). There was also no significant association between anticipatory action planning and bimanual performance (AHA) ($\rho = -.146$, $p = .140$). Based on the sword task data, we identified two subgroups: children that ended 0% of the critical trials in a comfortable end posture ($n = 28$) or children that ended 100% of the critical trials in a comfortable end posture ($n = 22$) and examined whether these groups differed in unimanual capacity and bimanual performance using Mann-Whitney U tests. There were no differences in BBT scores or AHA scores between groups.

4. Discussion

The main aim of this study was to investigate the development of anticipatory action planning in a large group of children ($N = 104$) with unilateral CP. Consistent with previous studies that used much smaller sample sizes, the present study demonstrates that anticipatory action planning does not improve with age in children with unilateral CP (Craje et al., 2010; Janssen & Steenbergen, 2011; Lust et al., 2017). No differences were found in the percentage of comfortable end postures in the critical orientations for children between 6 to 12 years of age, suggesting a lack of development of anticipatory action planning skills. Using outcomes from 351 typically developing children aged 3 to 10 years as a reference (Jongbloed-Pereboom et al., 2013), in our study children with unilateral CP appear to perform at a similar level as 4 to 9 year-old typically developing children. However, Jongbloed-Pereboom et al. (2013) found that 10 year-old children performed significantly better than 9 year-old children suggesting that performance may further improve with age, because a ceiling effect was not yet observed for 10 year-old children. The results of our study, and those discussed above, suggest that unlike typically developing children, children aged 6 to 12 years with unilateral CP do not naturally develop anticipatory action planning skills. This supports the need for intervention focused on anticipatory action planning, an important determinant for performing activities of daily living (Steenbergen & Gordon, 2006). An increasing body of knowledge from behavioural and neurophysiological studies (Caeyenberghs, van Roon, Swinnen, & Smits-Engelsman, 2009; Steenbergen et al., 2013; Zielinski, Jongasma, Baas, Aarts, & Steenbergen, 2014) suggests that children with CP have a neurocognitive deficit affecting their motor skill learning in general, and motor planning in particular (Kurz, Becker, Heinrichs-Graham, & Wilson, 2014; Zielinski et al., 2014). Aside from established interventions such as bimanual therapy (Hoare & Greaves, 2017), promising interventions to improve motor planning include Cognitive Orientation to daily Occupational Performance (CO-OP; the learning of a global problem solving strategy - goal, plan, do check; Cameron et al., 2017), Motor Imagery (MI; internal rehearsal of future motor actions without overt motor output; Steenbergen, Craje, Nilsen, & Gordon, 2009), and Action Observation (AO; observation of the action performed by someone else; Gatti et al., 2013; Kirkpatrick, Pearse, James, & Basu, 2016; Sgandurra et al., 2017). The results from this study warrant

Table 1
Characteristics of participants separated by age group.

Age group	Side of hemiplegia (L/R)	Gender (M/F)	Classification		Bimanual Performance		Unimanual Capacity		IQ scores	
			MACS (I/II/III)	GMFCS (I/II)	AHA Units (SD)	BBT More-affected hand (SD)	BBT Less-affected hand (SD)	WISC FSIQ (SD)	WISC GAI (SD)	
6 (n = 16)	11/5	4/12	2/14/0	15/1	58.56 (14.21)	13.75 (6.88)	28.63 (5.90)	96.00 (15.10)	94.44 (23.91)	
7 (n = 22)	13/9	12/10	7/14/1	16/6	56.50 (17.46)	18.55 (10.25)	34.32 (8.14)	90.91 (14.61)	94.36 (12.60)	
8 (n = 12)	7/5	8/4	2/8/2	8/4	54.17 (18.23)	15.33 (10.02)	35.17 (12.03)	83.67 (22.47)	84.75 (22.38)	
9 (n = 13)	5/8	9/4	5/8/0	7/6	58.46 (17.36)	21.31 (11.97)	37.23 (6.13)	90.31 (17.21)	90.31 (19.31)	
10 (n = 22)	11/11	13/9	3/18/1	18/4	60.18 (17.10)	22.86 (11.56)	42.64 (8.85)	83.91 (16.05)	86.64 (18.16)	
11 (n = 11)	7/4	10/1	2/8/1	9/2	60.00 (21.04)	22.64 (17.06)	41.55 (9.37)	88.30 (19.44)	84.55 (24.90)	
12 (n = 8)	2/6	7/1	4/4/0	6/2	68.88 (24.78)	26.75 (15.38)	43.50 (7.23)	83.00 (20.17)	80.63 (27.34)	
Total (n = 104)	56/48	63/41	25/74/5	79/25	58.89 (17.85)	19.76 (11.94)	37.13 (9.57)	88.34 (17.36)	89.03 (20.31)	

L, left; R, right; M, male; F, female; MACS, Manual Ability Classification System; GMFCS, Gross Motor Function Classification System; AHA, Assisting Hand Assessment; BBT, Box and Blocks Test; WISC, Wechsler Intelligence Scale for Children; FSIQ, Full Scale Intelligence Quotient; GAI, General Ability Index.

Table 2
Percentage comfortable end postures of both the critical and non-critical orientations separated by age group.

Age group	Critical orientations			Non-critical orientations		
	Mean	Median	Range	Mean	Median	Range
6	48.96	41.67	100.00	96.88	100.00	16.67
7	46.97	36.67	100.00	96.78	100.00	25.00
8	36.11	33.33	83.33	95.08	100.00	33.33
9	60.26	83.33	100.00	94.23	100.00	25.00
10	38.79	16.67	100.00	94.70	100.00	25.00
11	36.36	33.33	100.00	95.32	100.00	25.00
12	41.67	41.67	100.00	92.71	100.00	25.00

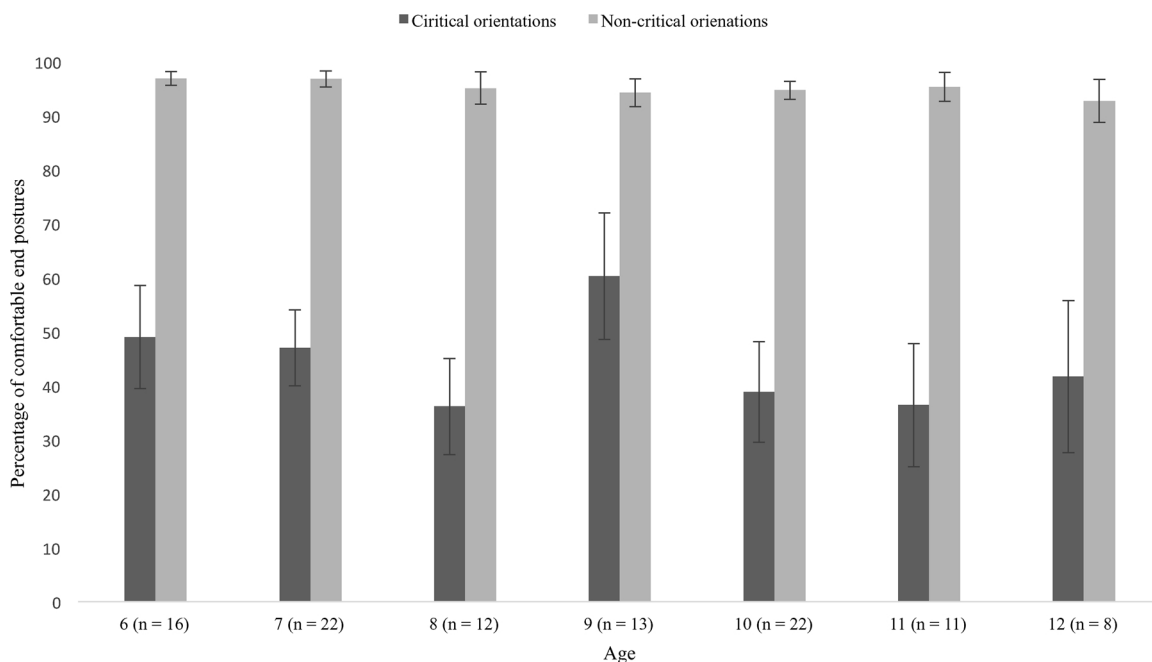


Fig. 2. Visualization of the mean percentage comfortable end postures of both the critical and non-critical orientations, for each age group separately. Error bars represent standard errors.

further testing of these interventions in children with unilateral CP. This will assist in identifying whether anticipatory action planning is amenable to improvement from targeted training. Future research should also include a broader age range to examine development of anticipatory action planning over a longer developmental period.

As a second aim, we examined the possible differential effect of side of hemiplegia on anticipatory action planning ability. Previous studies investigating this relationship showed equivocal results. Initially, smaller studies in children with unilateral CP by [Steenbergen et al. \(2004\)](#) and [Craje et al. \(2009\)](#) report a disproportionate action planning impairment among adolescents with lesions in the left hemisphere. However, a more recent study by [Kirkpatrick et al. \(2013\)](#) of 76 children with unilateral CP, demonstrated that anticipatory action planning was not lateralized. These outcomes were used to guide our hypotheses which was supported by our results. We found no difference in the percentage of comfortable end postures between children with left or right hemiplegia, supporting the idea of a distributed system of anticipatory action planning across hemispheres and/or cortical re-organization following brain damage in early life ([Kirkpatrick et al., 2013](#)).

The third aim was to examine the relationship between anticipatory action planning, unimanual capacity and bimanual performance. In line with a recent study in typically developing children ([Jongbloed-Pereboom et al., 2013](#)), results from our study demonstrated anticipatory action planning was not related to the scores on the BBT. This is unsurprising given our previous comments on the nature of the BBT where a child is simply required to move blocks from one side to another. The requirement for planning is minimal. Bimanual task performance however, requires greater perceptual and cognitive demands, especially planning. Our study investigated the relationship between anticipatory action planning and an observational-based test of bimanual performance, the Assisting Hand Assessment (AHA). Contrary to our hypotheses, anticipatory action planning was not related to performance on the AHA. The most logical explanation for this may be that the AHA total score (summing 20 items) is too broad to sensitively reflect the specific components of bimanual performance that require anticipatory action planning. As a result, it seems

that the step by step planning process many children with unilateral CP use to plan action execution may be sufficient to complete bimanual tasks of the AHA.

Overall, the results from our study are not entirely consistent with previous studies on anticipatory action planning in children with unilateral CP. One likely explanation is that the sword task may not be the most valid measure of anticipatory action planning. The reason for this is that the mean percentage of comfortable end postures in the critical orientations has been found to be 80% and 60% for healthy adolescents and adults, respectively (Jongbloed-Pereboom et al., 2016). This suggests that environmental and cognitive factors, other than planning for ESC, may also affect initial grasp orientation of the sword. For example, the combination of a relatively high level of initial discomfort and relatively low accuracy requirements at the end of the sword task possibly allows children to make a decision not to sacrifice comfort of the start posture and still be able to perform the task reasonably well. Furthermore, the temporal aspects during task administration may also significantly influence a child's response. The sword task does not specify the speed in which children perform the task (three blocks of six trials). Review of videotaped sessions in our study indicates wide variation in speed of administration. As a result, it may be speculated that two children with equal anticipatory action planning capabilities score differently on the sword task due to a different speed-accuracy trade-off. In addition, although the goal of the task is straightforward, some children experienced difficulty following the rules for administration. Some tended to grasp the sword at the bottom instead of the handle and/or change their grip from ulnar to radial during the movement sequence, which led to administration errors. Exclusion of these children from our analysis may have introduced bias since these errors may be a reflection of correct or incorrect anticipatory action planning. Grasping the sword at the bottom possibly reflects anticipatory action planning since it makes the transport movement much easier. On the other hand, changing grip during the transport phase suggests a step-by-step anticipatory action planning strategy, where children did not plan the entire action before starting the execution. To overcome these issues, we suggest that future studies of anticipatory action planning in children with unilateral CP use the handle rotation task/hexagonal knob task (2006, Kirkpatrick et al., 2013; Mutsaerts et al., 2005). Here, instead of using relative comfort ratings, effective anticipatory action planning is necessary for successful task completion. Consistent methods to measure the ESC effect are essential for comparison across studies. At the same time, task aspects such as precision demands at the end of the movement, the amount of start positions, and sequence length, should be varied in a structured manner to be able to identify what aspects determine task complexity and thereby performance on anticipatory action planning tasks.

5. Conclusion

This study demonstrates that anticipatory action planning, measured using the sword task does not improve with age in children with unilateral CP, aged 6–12 years. Furthermore, anticipatory action planning is not lateralized in children with unilateral CP and there is no association between anticipatory action planning and the Assisting Hand Assessment. Future studies of anticipatory action planning in children with unilateral CP are advised to use measures such as the hexagonal knob task that require effective anticipatory action planning for successful task completion rather than ESC.

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References

- Adalbjornsson, C. F., Fischman, M. G., & Rudisill, M. E. (2008). The end-state comfort effect in young children. *Research Quarterly for Exercise and Sport*, 79(1), 36–41.
- Caeyenberghs, K., van Roon, D., Swinnen, S. P., & Smits-Engelsman, B. C. M. (2009). Deficits in executed and imagined aiming performance in brain-injured children. *Brain and Cognition*, 69(1), 154–161. <https://doi.org/10.1016/j.bandc.2008.07.001>.
- Cameron, D., Craig, T., Edwards, B., Missiuna, C., Schweltnus, H., & Polatajko, H. J. (2017). Cognitive orientation to daily occupational performance (CO-OP): A new approach for children with cerebral palsy. *Physical & Occupational Therapy in Pediatrics*, 37(2), 183–198. <https://doi.org/10.1080/01942638.2016.1185500>.
- Cohen, R. G., & Rosenbaum, D. A. (2004). Where grasps are made reveals how grasps are planned: Generation and recall of motor plans. *Experimental Brain Research*, 157(4), 486–495. <https://doi.org/10.1007/s00221-004-1862-9>.
- Craje, C., Aarts, P., Nijhuis-van der Sanden, M., & Steenbergen, B. (2010). Action planning in typically and atypically developing children (unilateral cerebral palsy). *Research in Developmental Disabilities*, 31(5), 1039–1046. <https://doi.org/10.1016/j.ridd.2010.04.007>.
- Craje, C., van der Kamp, J., & Steenbergen, B. (2009). Visual information for action planning in left and right congenital hemiparesis. *Brain Research*, 1261, 54–64. <https://doi.org/10.1016/j.brainres.2008.12.074>.
- Eliasson, A. C., Krumlind-Sundholm, L., Rosblad, B., Beckung, E., Arner, M., Ohrvall, A. M., ... Rosenbaum, P. (2006). The manual ability classification system (MACS) for children with cerebral palsy: Scale development and evidence of validity and reliability. *Developmental Medicine and Child Neurology*, 48(7), 549–554. <https://doi.org/10.1017/S0012162206001162>.
- Gatti, R., Tettamanti, A., Gough, P. M., Riboldi, E., Marinoni, L., & Buccino, G. (2013). Action observation versus motor imagery in learning a complex motor task: A short review of literature and a kinematics study. *Neuroscience Letters*, 540, 37–42. <https://doi.org/10.1016/j.neulet.2012.11.039>.
- Hoare, B., & Greaves, S. (2017). Unimanual versus bimanual therapy in children with unilateral cerebral palsy: Same, same, but different. *Journal of Pediatric Rehabilitation Medicine*, 10(1), 47–59. <https://doi.org/10.3233/PRM-170410>.
- Holmfur, M., & Krumlind-Sundholm, L. (2016). Psychometric properties of a revised version of the assisting hand assessment (Kids-AHA 5.0). *Developmental Medicine and Child Neurology*, 58(6), 618–624. <https://doi.org/10.1111/dmcn.12939>.
- Holmfur, M., Aarts, P., Hoare, B., & Krumlind-Sundholm, L. (2009). Test-retest and alternate forms reliability of the assisting hand assessment. *Journal of Rehabilitation Medicine*, 41(11), 886–891. <https://doi.org/10.2340/16501977-0448>.
- IBM Corp Released (2013). *IBM SPSS statistics for windows, version 22.0* (Released 2013) Armonk, NY: IBM Corp.
- Janssen, L., & Steenbergen, B. (2011). Typical and atypical (cerebral palsy) development of unimanual and bimanual grasp planning. *Research in Developmental Disabilities*, 32(3), 963–971. <https://doi.org/10.1016/j.ridd.2011.02.002>.

- Johnson-Frey, S. H. (2004). The neural bases of complex tool use in humans. *Trends in Cognitive Sciences*, 8(2), 71–78. <https://doi.org/10.1016/j.tics.2003.12.002>.
- Johnson-Frey, S. H., McCarty, M. E., & Keen, R. (2004). Reaching beyond spatial perception: Effects of intended future actions on visually guided prehension. *Visual Cognition*, 11(2-3), 371–399. <https://doi.org/10.1080/13506280344000329>.
- Jongbloed-Pereboom, M., Nijhuis-van der Sanden, M. W. G., Saraber-Schiphorst, N., Craje, C., & Steenbergen, B. (2013). Anticipatory action planning increases from 3 to 10 years of age in typically developing children. *Journal of Experimental Child Psychology*, 114(2), 295–305. <https://doi.org/10.1016/j.jecp.2012.08.008>.
- Jongbloed-Pereboom, M., Spruijt, S., Nijhuis-van der Sanden, M. W. G., & Steenbergen, B. (2016). Measurement of action planning in children, adolescents, and adults: A comparison between 3 tasks. *Pediatric Physical Therapy*, 28(1), 33–39. <https://doi.org/10.1097/Pep.0000000000000211>.
- Jovanovic, B., & Schwarzer, G. (2011). Learning to grasp efficiently: The development of motor planning and the role of observational learning. *Vision Research*, 51(8), 945–954. <https://doi.org/10.1016/j.visres.2010.12.003>.
- Kirkpatrick, E., Pearse, J., Eyre, J., & Basu, A. (2013). Motor planning ability is not related to lesion side or functional manual ability in children with hemiplegic cerebral palsy. *Experimental Brain Research*, 231(2), 239–247. <https://doi.org/10.1007/s00221-013-3687-x>.
- Kirkpatrick, E., Pearse, J., James, P., & Basu, A. (2016). Effect of parent-delivered action observation therapy on upper limb function in unilateral cerebral palsy: A randomized controlled trial. *Developmental Medicine and Child Neurology*, 58(10), 1049–1056. <https://doi.org/10.1111/dmcn.13109>.
- Knudsen, B., Henning, A., Wunsch, K., Weigelt, M., & Aschersleben, G. (2012). The end-state comfort effect in 3- to 8-year-old children in two object manipulation tasks. *Frontiers in Psychology*, 3, 445. <https://doi.org/10.3389/fpsyg.2012.00445>.
- Krumlinde-Sundholm, L., Holmefur, M., Kottorp, A., & Eliasson, A. C. (2007). The assisting hand assessment: Current evidence of validity, reliability, and responsiveness to change. *Developmental Medicine and Child Neurology*, 49(4), 259–264. <https://doi.org/10.1111/j.1469-8749.2007.00259.x>.
- Kurz, M. J., Becker, K. M., Heinrichs-Graham, E., & Wilson, T. W. (2014). Neurophysiological abnormalities in the sensorimotor cortices during the motor planning and movement execution stages of children with cerebral palsy. *Developmental Medicine and Child Neurology*, 56(11), 1072–1077. <https://doi.org/10.1111/dmcn.12513>.
- Lust, J. M., Spruijt, S., Wilson, P. H., & Steenbergen, B. (2017). Motor planning in children with cerebral palsy: A longitudinal perspective. *Journal of Clinical and Experimental Neuropsychology*, 1–8. <https://doi.org/10.1080/13803395.2017.1387645>.
- Manoel, E. J., & Moreira, C. R. P. (2005). Planning manipulative hand movements: Do young children show the end-state comfort effect? *Journal of Human Movement Studies*, 49(2), 93–114.
- Mathiowetz, V., Volland, G., Kashman, N., & Weber, K. (1985). Adult norms for the box and block test of manual dexterity. *American Journal of Occupational Therapy*, 39(6), 386–391. <https://doi.org/10.5014/ajot.39.6.386>.
- Mukaka, M. M. (2012). Statistics corner: A guide to appropriate use of correlation coefficient in medical research. *Malawi Medical Journal*, 24(3), 69–71.
- Mutsaerts, M., Steenbergen, B., & Bekkering, H. (2005). Anticipatory planning of movement sequences in hemiparetic cerebral palsy. *Motor Control*, 9(4), 439–458. <https://doi.org/10.1123/mcj.9.4.439>.
- Mutsaerts, M., Steenbergen, B., & Bekkering, H. (2006). Anticipatory planning deficits and task context effects in hemiparetic cerebral palsy. *Experimental Brain Research*, 172(2), 151–162. <https://doi.org/10.1007/s00221-005-0327-0>.
- Palisano, R., Rosenbaum, P., Walter, S., & Russell, D. J. (1997). Gross motor function classification system for cerebral palsy. *Developmental Medicine and Child Neurology*, 39(4), 214–223.
- Rosenbaum, D. A., Barnes, H. J., Vaughan, J., & Jorgensen, M. J. (1992). Time course of movement planning - selection of handgrips for object manipulation. *Journal of Experimental Psychology-Learning Memory and Cognition*, 18(5), 1058–1073. <https://doi.org/10.1037/0278-7393.18.5.1058>.
- Rosenbaum, D. A., Chapman, K. M., Weigelt, M., Weiss, D. J., & van der We, R. (2012). Cognition, action, and object manipulation. *Psychological Bulletin*, 138(5), 924–946. <https://doi.org/10.1037/a0027839>.
- Schluter, N. D., Krams, M., Rushworth, M. F. S., & Passingham, R. E. (2001). Cerebral dominance for action in the human brain: The selection of actions. *Neuropsychologia*, 39(2), 105–113. [https://doi.org/10.1016/S0028-3932\(00\)00105-6](https://doi.org/10.1016/S0028-3932(00)00105-6).
- Sgandurra, G., Lorentzen, J., Inguaggiato, E., Bartalena, L., Beani, E., Cecchi, F., & Consortium, C. (2017). A randomized clinical trial in preterm infants on the effects of a home-based early intervention with the 'CareToy System'. *PLoS One*, 12(3), <https://doi.org/10.1371/journal.pone.0173521>.
- Short, M. W., & Cauraugh, J. H. (1999). Precision hypothesis and the end-state comfort effect. *Acta Psychologica*, 100(3), 243–252. [https://doi.org/10.1016/S0001-6918\(98\)00020-1](https://doi.org/10.1016/S0001-6918(98)00020-1).
- Stanley, F. J., Blair, E., & Alberman, E. D. (2000). *Cerebral palsies: Epidemiology and causal pathways*. London, UK: Cambridge University Press.
- Steenbergen, B., & Gordon, A. M. (2006). Activity limitation in hemiplegic cerebral palsy: Evidence for disorders in motor planning. *Developmental Medicine and Child Neurology*, 48(9), 780–783. <https://doi.org/10.1017/S0012162206001666>.
- Steenbergen, B., Craje, C., Nilsen, D. M., & Gordon, A. M. (2009). Motor imagery training in hemiplegic cerebral palsy: A potentially useful therapeutic tool for rehabilitation. *Developmental Medicine and Child Neurology*, 51(9), 690–696. <https://doi.org/10.1111/j.1469-8749.2009.03371.x>.
- Steenbergen, B., Jongbloed-Pereboom, M., Spruijt, S., & Gordon, A. M. (2013). Impaired motor planning and motor imagery in children with unilateral spastic cerebral palsy: Challenges for the future of pediatric rehabilitation. *Developmental Medicine and Child Neurology*, 55, 43–46. <https://doi.org/10.1111/dmcn.12306>.
- Steenbergen, B., Meulenbroek, R. G. J., & Rosenbaum, D. A. (2004). Constraints on grip selection in hemiparetic cerebral palsy: Effects of lesional side, end-point accuracy, and context. *Cognitive Brain Research*, 19(2), 145–159. <https://doi.org/10.1016/j.cogbrainres.2003.11.008>.
- Steenbergen, B., Verrel, J., & Gordon, A. M. (2007). Motor planning in congenital hemiplegia. *Disability and Rehabilitation*, 29(1), 13–23. <https://doi.org/10.1080/09638280600947591>.
- Stöckel, T., Hughes, C. M. L., & Schack, T. (2012). Representation of grasp postures and anticipatory motor planning in children. *Psychological Research*, 76(6), 768–776. <https://doi.org/10.1007/s00426-011-0387-7>.
- Thibaut, J. P., & Toussaint, L. (2010). Developing motor planning over ages. *Journal of Experimental Child Psychology*, 105(1-2), 116–129. <https://doi.org/10.1016/j.jecp.2009.10.003>.
- van Swieten, L. M., van Bergen, E., Williams, J. H. G., Wilson, A. D., Plumb, M. S., Kent, S. W., ... Mon-Williams, M. A. (2010). A test of motor (not executive) planning in developmental coordination disorder and autism. *Journal of Experimental Psychology Human Perception and Performance*, 36(2), 493–499. <https://doi.org/10.1037/a0017177>.
- Wechsler, D. (2003). *WISC - IV Australian administration and scoring manual*.
- Wunsch, K., Henning, A., Aschersleben, G., & Weigelt, M. (2013). A systematic review of the end-state comfort effect in normally developing children and in children with developmental disorders. *Journal of Motor Learning and Development*, 1(3), 59–76. <https://doi.org/10.1123/jmld.1.3.59>.
- Wunsch, K., Pfister, R., Henning, A., Aschersleben, G., & Weigelt, M. (2016). No interrelation of motor planning and executive functions across young ages. *Frontiers in Psychology*, 7, 1031. <https://doi.org/10.3389/fpsyg.2016.01031>.
- Zielinski, I. M., Jongsmá, M. L. A., Baas, C. M., Aarts, P. B. M., & Steenbergen, B. (2014). Unravelling developmental disregard in children with unilateral cerebral palsy by measuring event-related potentials during a simple and complex task. *BMC Neurology*, 14. <https://doi.org/10.1186/1471-2377-14-6>.