# Short- and long-term effects of synchronized metronome training in children with hemiplegic cerebral palsy: A two case study

ANNA-MARIA JOHANSSON<sup>1</sup>, ERIK DOMELLÖF<sup>1,2</sup>, & LOUISE RÖNNQVIST<sup>1</sup>

<sup>1</sup>Department of Psychology, Umeå University, Umeå, Sweden, and <sup>2</sup>Kolbäcken Child Rehabilitation Centre, Umeå, Sweden

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#### Abstract

*Background*: Children with cerebral palsy (CP) require individualized long-term management to maintain and improve motor functions. The objective of this study was to explore potential effects of synchronized metronome training (SMT) on movement kinematics in two children diagnosed with spastic hemiplegic CP (HCP).

*Method*: Both children underwent 4-weeks/12 sessions of SMT by means of the Interactive Metronome (IM). Optoelectronic registrations of goal-directed uni- and bimanual upper-limb movements were made at three occasions; pre-training, post completed training and at 6-months post completed training.

*Results*: Significant changes in kinematic outcomes following IM training were found for both cases. Findings included smoother and shorter movement trajectories in the bimanual condition, especially for the affected side. In the unimanual condition, Case I also showed increased smoothness of the non-affected side.

*Conclusions*: The observed short- and long-term effects on the spatio-temporal organization of upper-limb movements need to be corroborated and extended by further case-control studies.

**Keywords:** Hemiplegic cerebral palsy, intervention, synchronized metronome training, kinematic, motor control, motor co-ordination

# Introduction

Activity limitations in hemiplegic cerebral palsy (HCP) stem from an interaction of primary and acquired impairments involving muscular and somatosensory systems [1] as well as motor planning ability [1,2]. Thus, deficits in multiple modalities are often present including sensory, perceptual and motor functions [3]. Methods targeted at improving motor planning ability coupled with strengthening and synchronization of sensory-motor connections appears promising when even small improvements would be beneficial for the individual child's daily activities.

The Interactive Metronome<sup>®</sup> (IM; Sunrise, Florida) is a synchronized metronome training (SMT) method that has received great clinical interest during the last decade, especially in North America. Effects of IM have been documented in terms of improvement in rhythmic motor performance (as measured by the IM equipment) [4–7]. Further, transfer effects to an unrelated motor task in adult golfers [4], improvements in visoumotor

control, balance, upper-limb coordination and upper-limb speed in children with ADHD-related difficulties [5,8] improved arm and hand function and perceived quality-of-life in two adults suffering from chronic stroke [9] and reading efficacy and fluency in 7-10-year-old children [6] have been reported. Additionally, reports from rehabilitation centres suggest positive effects of IM training on behaviours in various clinical populations. However, scientific evaluations of IM are still scarce and, to the authors' knowledge, none have involved children with CP, nor investigated possible effects on kinematic properties. Thus, there is a great need for more detailed empirical investigations concerning the effects of IM on movement performance and possible transfer effects to other behaviours, especially in clinical populations.

IM was originally developed to improve motor control and rhythmicity in musicians through rhythmic activation and timing of movements. To facilitate better timing, feedback cues related to performance are presented immediately after the

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Correspondence: Anna-Maria Johansson, Department of Psychology, Umeå University, SE- 901 87 Umeå, Sweden. Tel: +46 (0)90 786 78 27. Fax: +46 (0)90 786 66 95. E-mail: anna-maria.johansson@psy.umu.se

movement has been registered. The method includes elements of rhythmic auditory stimulation (RAS) which have been shown to improve timing of leg movements in gait, including velocity, stride length and symmetry, in a sample of children with HCP [10]. RAS has been shown to entrain central motor processors to couple with the imposed rhythmic timekeeper of the auditory system and to stabilize motor control, independent of neuropathology [11]. Accordingly, perceptual rhythmic training has been shown to affect performance on an untrained rhythmic motor task, confirming the close sensory-motor connections [12]. Accuracy feedback has been shown to be crucial for improved movement performance in HCP and in typically-developing children [13].

All successfully performed goal-directed movements are relying on the precise temporal organization and co-ordinated control over different groups of muscles and the synchronization between sensory cues with proprioceptive information, cognition and time representation [14,15]. As such, motor planning entails co-ordination of the involved bodily segments and muscular forces in relation to the goal that, when successful, results is an accurate, energy efficient, smooth and fast movement [16]. It is thus reasonable that IM, aimed at training motor planning and timing to advance the temporal synchronization of movements, could prove beneficial for improvement of upper-limb movement quality and organization in children with HCP.

The main objective of the present case study, including two children with HCP, was to explore if 4 weeks of IM training may affect the quality of goaldirected upper-limb movements as expressed in movement kinematics during conditions unrelated to the IM training. An additional aim was to investigate potential long-term effects after the training period concluded in terms of effect stability and/or consolidation effects.

# Methods

#### **Participants**

Two children with spastic HCP participated in the study. Case I was a 17 year old girl with paresis affecting primarily the right side of the body (Manual Ability Classification Score [17], MACS, II; Gross Motor Function Classification Score [18], GMFCS, II). In concordance, MR findings showed a substance defect in the lateral ventricle and partial cortical dysplasia in the temporal lobe of the left hemisphere. Case II was a 13 year old boy with paresis affecting primarily the left side of the body (MACS, II; GMFCS, I). Further, Case II had left-sided homonymous hemianopsia and was born at

full term as twin 1. No brain imaging had been performed on this child. Both Cases I and II had a score of 2 for elbow and wrist, respectively, on the Modified Ashworth Scale (as determined by their occupational therapist). They had no diagnosed learning disabilities and attended regular schooling. Case II received upper-limb botulinum toxin treatment between Post1 and Post2, although not in close adjunct to the Post2 occasion. Recruitment was based on willingness to participate in the study and ability to follow verbal instructions. Informed consent was obtained verbally from the children and in writing from the parents. The study was approved by the local medical ethical committee and conducted in accordance with the Declaration of Helsinki.

#### Study design

Figure 1 illustrates the overall study design. Both cases partook in 4 weeks comparable but individually adjusted IM training (12 sessions, ~30 minutes/session). Circa 16000 repetitions were induced in the training regime for both cases. All sessions were supervised by a trained IM instructor (A-MJ). The training involved bilateral and unilateral rhythmic movements of upper- and lower-body extremities, with instant feedback of timing synchronization. A metronome beat (54 bpm) was setting a rhythm presented through headphones where the volume was individually pre-adjusted. A set of pressureactivated sensors, attached to either limb, then allowed the child to clap, press or toe-tap in synchrony with the rhythmic tone. While responding to the beat, the participant was guided by both tonally and spatially changing feedback (corresponding sound cues in headphones and visual cues on a laptop computer screen). The feedback occurred immediately after the sensors were pressed which allowed for corrections of movement timing in response to the next beat. At start of each session, two 2-minute baseline assessments (clapping to a pre-set beat of 54 bpm), with and without guide sounds, were performed. Over sessions, this allowed a measure of individual changes in timing and rhythmic performance by changes in error magnitude to the auditory signal and in the variability of motor responses, respectively.

Outcome assessments were made before the training period began (Pre) and at two occasions after the 4-week training period (Post1 and Post2; see Figure 1). At these occasions goal-directed upper-limb movements were recorded by a six-camera 3D optoelectronic registration system (ProReflex, Qualisys Inc., Gothenburg, Sweden) with additional synchronized video recordings. The set-up and application of this system are described



Figure 1 Schematic illustration of the study design.



Figure 2 The extension-flexion/adduction-abduction task and marker placement.

elsewhere [19]. A total of nine markers were affixed with skin-friendly adhesive tape to the left and right shoulders ( $29 \text{ mm}^{3*}$ ), elbows ( $19 \text{ mm}^{3*}$ ), wrists ( $12 \text{ mm}^{3*}$ ) and knuckles of index finger ( $7 \text{ mm}^{3}$ ) (Figure 2). One marker ( $12 \text{ mm}^{3}$ ) was also affixed to the participant's forehead for measurement of head movements (\* markers included in statistical analyses). The sampling frequency was 120 Hz and the pre-set recording time was individually adjusted to suit the movement duration of the child.

The participants were seated in a chair in front of a custom-made test platform with 10 integrated easy to press light-switches (Figure 2). The participants

performed goal-directed movements (switch on light) with either the affected or non-affected armhand (unimanual condition) or both arm-hands simultaneously (bimanual condition). The task consisted of, beginning from a specified starting point, pressing three light-switch buttons in a sequential order. The light-switches were to be pressed either vertically from the bottom to the top (extension), vertically from top to bottom (flexion), horizontally from the midline and out (adduction) or horizontally from the button furthest away and in (abduction). As differentiated finger movements were strenuous for both participants, instructions were to perform the task with a clenched fist. The order of the sequences was determined by a randomized contra-balanced block design. Each block consisted of three trials for each of the four directions, resulting in a total of 36 trials for each testing occasion. If mistakes were made, the participants were asked to repeat the trial at the end of the block to maximize the number of useable trials. Both participants received two practice trials for each condition before the experimental measurement session began.

The onset and offset of each successful goaldirected sequential movement were identified from the 3D movement trajectory and the tangential velocity profile of the wrist marker. The onset of the movement was defined as the frame when the wrist marker crossed a velocity limit of 20 mm/s and increased over the next five frames and its offset at the frame when the wrist marker had a velocity of 100 mm/s that was increasing after the last successful light-switch press. The duration of the movement was extracted and converted to seconds. All 3D movement co-ordinates were analysed by custom written software in Matlab (The Mathworks Inc., Boston, MA) where data also were smoothed, prior to analysis, using a second-order 10 Hz dual pass Butterworth filter. The following kinematics was extracted from the upper-limbs (shoulder, elbow and wrist): the 3D distance and the movement segmentation by means of number of movement units (MUs; see Figures 3(a-c)). A MU is defined as an acceleration phase followed by a deceleration phase with an accumulated increase or decrease in velocity of at least 20 mm s<sup>-1</sup> and an acceleration or deceleration exceeding 5 mm/s [20]. Further, to investigate general effects on arm/hand kinematics, overall values ([shoulder + elbow + wrist]/3) were calculated in relation to 3D distance and MUs. To make values comparative for the statistical analyses, all duration and kinematic values were divided by the number of successful light-switch presses for each trial.

#### Statistical analyses

The outcome data was analysed for each case by Wilcoxon matched pairs tests with an alpha value of 0.025 where effects between Pre and Post1 and Pre and Post2 were analysed. Effect sizes were calculated on the statistically significant results by Pearson's correlation coefficient. All analyses were based on trial level data. The uni- and bimanual conditions were analysed separately by side (affected/nonaffected). A total of 17 trials were excluded from analyses due to premature movement onset and/or measurement errors. The final analyses are based on a total of 105 trials for Case I and 104 trials for Case I on the Post1 occasion was not analysed due to measurement error. Significant effects, means (M) and standard deviations (SD) are reported by side (affected/non-affected) and shown in Tables I for Case I and II for Case II.

# Results

# Case I

During the IM training period, Case I showed task learning in both self-phased rhythmicity when error feedback cues were present (Figure 4(a)), as reflected by a reduction in millisecond (ms) deviation from the metronome. The level of learning remained at Post2 for both conditions.

Unimanual condition. А significantly shorter duration at Post2 in comparison to Pre for the non-affected side (T=4, p < 0.025, r=0.76) was apparent. Further, a reduction in the number of MUs between Pre and Post2 for the elbow (T=1.5, p < 0.01, r = 0.45) and wrist (T = 0, p < 0.01,r = 0.89) of the non-affected side was also shown. Accordingly, an overall effect (derived from collapsed data from the shoulder, elbow and wrist markers) for MUs was apparent between Pre and Post2 (T=3, p < 0.01, r=0.80). The 3D distance of the non-affected side remained unchanged, whereas an increase was noted for the shoulder between Pre and Post1 (T=3, p < 0.01, r=0.90). A statistical trend was noted for a reduction of the movement duration of the non-affected side between Pre and Post2 (p = 0.04).

Bimanual condition. Case I showed significant reduction in movement duration at Post2 compared with Pre (T = 6, p < 0.025, r = 0.72) for the affected side. Further, there was a reduction in the number of MUs of the shoulder of the affected side at both post-test occasions, significant between Pre and Post2 (T=0, p < 0.01, r=0.89). Accordingly, the MUs of the elbow decreased significantly between Pre and Post2 (T = 1.5, p < 0.01, r = 0.45) and for the wrist between Pre and Post1 (T=7.5, p < 0.025, r = 0.69) and Pre and Post2 (T = 2, p < 0.01, r=0.84; see Figures 3(a-c)). In agreement, an overall effect was apparent between Pre and Post2 for the affected side (T=1, p < 0.01, r=0.86). Only the effects of the elbow between Pre and Post2 could be tested for the affected side due to measurement errors of the elbow marker. Similar to the affected side, although non-significant, a reduction in movement duration was shown between Pre and Post2 (p=0.04). Further, the number of MUs was reduced for the non-affected side between Pre and



Figure 3 (a) Example of a 3D spatial plot of the affected (right) distal arm trajectories (wrist marker) during the same light-switching task made at the pre- (solid line) and post1-test (dotted line) occasion by Case I (superimposed in the same plot). (b) The velocity profile of the pre-test performance (same as in a) with the onset and offset of each movement unit (MU) identified by round dots (30 MUs in total) and (c) the post-test performance (same as in a) with 15 MUs in total. Note; the total duration of the task performance was 5 seconds at the pre-test and 3.6 seconds at the post1-test.

	Case I																		
	Non-affected side									Affected side									
	Pre		Post1			Post2			Pre		Post1			Post2					
	М	SD	D	М	SD	D	M	SD	М	SD	D	М	SD	D	М	SD			
Unimanual																			
Duration (s)	1.12	0.11		1.00	0.14		0.97	0.14	1.66	0.19		1.78	0.16		1.49	0.18			
MUs (n): shoulder	3.6	0.8	_	3.7	1.1	_	3.1	0.8	5.9	0.7	+	6.9	1.8	+	5.9	1.4			
MUs (n): elbow	3.7	0.74	_	3.0	1.4	_	2.4	1.0	5.5	0.8		na	na	_	4.9	1.5			
MUs (n): wrist	4.0	0.5	_	3.5	0.94	_	2.8	0.5	4.6	0.5	+	5.1	0.6	_	4.2	1.1			
MUs (n): overall	3.8	0.6	_	3.4	1.0	_	2.7	0.7	5.4	0.4		na	na	_	5.0	1.1			
3D distance: shoulder	68	19	_	61	15	_	63	15	78	12	+	95	15	+	85	15			
3D distance: elbow	163	40	_	162	30	+	162	40	177	30		na	na	+	192	31			
3D distance: wrist	244	49	_	234	35	_	162	40	251	40	+	274	31	+	195	31			
3D distance: overall	158	34	—	152	24	—	147	30	168	25		na	na	+	179	23			
Bimanual																			
Duration (s)	1.93	0.25		1.86	0.22		1.70	0.18	1.98	0.24		1.84	0.19		1.67	0.19			
MUs (n): shoulder	9.0	2.2	+	10.5	2.7	_	7.2	1.0	10.0	2.2	_	8.5	1.6	_	6.8	1.4			
MUs (n): elbow	7.8	2.5	_	5.7	1.6	_	5.1	1.1	9.6	2.9		na	na	_	6.2	1.3			
MUs (n): wrist	7.6	1.8	_	5.6	1.7	_	4.6	0.7	8.9	2.0	_	6.3	1.6	_	5.7	1.0			
MUs (n): overall	8.1	2.0	_	7.2	1.8	_	5.7	0.7	9.5	2.3		na	na	_	6.1	1.0			
3D distance: shoulder	126	24	_	113	14	_	93	21	117	19	_	96	16	_	84	18			
3D distance: elbow	223	39	_	204	24	_	195	30	226	28		na	na	_	195	26			
3D distance: wrist	294	49	_	287	37	_	195	30	307	44	_	282	28	_	195	26			
3D distance: overall	214	34	_	204	21	_	188	26	217	28		na	na	_	189	24			

Table I. Durations, MUs and 3D distances for Case I in the uni-bimanual condition presented by side and occasion.

s, seconds; MU, mean number of movement units; M, mean; SD, standard deviation; D, direction of change relative pretest (+ -); na, missing values. Significant effects (p < 0.025) relative pre-test occasion are indicated in bold.

Table II. Durations, MUs and 3D distances for Case II in the uni-bimanual condition presented by side and occasion.

	Case II																		
	Non-affected side									Affected side									
	Pre		Post1			Post2			Pre		Post1			Post2					
	М	SD	D	М	SD	D	М	SD	М	SD	D	М	SD	D	М	SD			
Unimanual																			
Duration (s)	2.25	0.39		1.29	0.27		1.27	0.19	2.37	0.41		2.33	0.30		2.32	0.40			
MUs (n): shoulder	10.3	5.8	_	5.3	1.0	_	6.0	2.27	11.0	2.7	+	12.8	2.8	_	10.8	2.4			
MUs (n): elbow	5.8	3.4	_	3.1	1.2	_	3.79	1.5	7.6	1.9	+	8.3	1.9	+	8.4	2.0			
MUs (n): wrist	5.4	2.7	_	2.8	0.7	_	3.73	1.0	8.4	1.7	+	8.8	1.6	_	8.1	2.3			
MUs (n): overall	7.7	4.0	_	3.6	0.9	_	4.2	1.5	9.0	1.8	+	9.9	1.9	_	8.8	2.0			
3D distance: shoulder	73	30	_	67	37	_	53	13	124	27	+	127	25	_	111	30			
3D distance: elbow	179	107	_	171	26	_	158	49	233	35	_	231	36	_	215	69			
3D distance: wrist	280	97	_	263	71	_	272	107	317	42	_	309	51	+	340	98			
3D distance: overall	172	85	_	138	25	—	128	30	225	31	_	222	35	-	218	59			
Bimanual																			
Duration (s)	2.19	0.39		2.3	0.37		2.01	0.13	2.46	0.43		2.47	0.39		2.46	0.22			
MUs (n): shoulder	11.6	3.5	_	10.4	3.2	_	9.7	1.5	10.9	2.0	+	13.7	3.8	_	10.0	1.2			
MUs (n): elbow	10.2	4.1	_	8.3	3.2	_	10.0	1.6	7.9	2.0	+	8.0	2.9	+	8.1	0.9			
MUs (n): wrist	9.3	2.6	_	8.2	2.1	_	8.5	2.0	8.9	2.2	_	8.4	1.9	_	7.3	1.0			
MUs (n): overall	10.9	3.8	_	8.9	2.4	_	9.2	1.4	10.8	3.2	_	10.3	2.3	_	8.7	0.9			
3D distance: shoulder	127	22	_	118	36	_	96	22	143	16	_	133	33	_	119	24			
3D distance: elbow	199	24	+	214	63	_	171	38	281	46	_	263	69	_	208	41			
3D distance: wrist	256	52	+	360	100	_	208	26	381	82	_	360	130	_	306	62			
3D distance: overall	208	40	-	192	53	-	161	31	300	72	-	243	62	_	207	37			

s, seconds; MU, mean number of movement units; M, mean; SD, standard deviation; D, direction of change relative pretest (+ -); na, missing values. Significant effects (p < 0.025) relative pre-test occasion are indicated in bold.



Figure 4 Changes in rhythmic performance (self-phased and with guide-sounds) at Pre, Post1 and Post2.

Post2 for the elbow (T=3, p < 0.01, r=0.80) and wrist (T=0, p < 0.01, r=0.89). An overall effect was also apparent between Pre and Post2 (T=4, p < 0.01, r=0.78). Concerning the 3D distance of the movement trajectory for the affected side, a reduction was apparent between Pre and Post2 for the shoulder (T=3, p < 0.01, r=0.81) and elbow (T=7, p < 0.025, r=0.70). The wrist also showed a reduction in 3D distance, although non-significant. However, an overall effect was apparent between Pre and Post2 (T=6, p < 0.01, r=0.72). Similar changes between Pre and Post2 were shown for the shoulder (T=2, p < 0.01, r=0.84), elbow (T=4, p < 0.01, r=0.84). p < 0.01, r=0.78), wrist (T=6, p < 0.025, r=0.73) and overall (T=4, p < 0.01, r=0.80) of the non-affected side.

# Case II

Case II showed some improvement in self-phased rhythmicity, but demonstrated a greater ability to learn how to use the guide-sounds effectively (Figure 4(b)). The achieved level of synchronization with the metronome, both self-phased and with guide sounds, remained at Post2.

Unimanual condition. A significant reduction in movement duration for the non-affected side between Pre and Post1 (T=1, p < 0.01, r=0.85) and Pre and Post2 (T=0, p < 0.01, r=0.89) was apparent. Further, a trend towards a reduction in the number of MUs was noted for the wrist between Pre and Post1 (p = 0.025). Accordingly, the number of MUs was reduced for the shoulder and elbow and, overall, a trend towards significance was noted (p=0.036). This trend remained to Post2 where the overall reduction reached statistical significance (T=2, p < 0.025, r=0.81). A reduction in 3D distance was apparent for the shoulder, elbow and wrist of the non-affected side (non-significant). The performance of the affected side remained unchanged.

Bimanual condition. The number of MUs of the non-affected side (shoulder, elbow, wrist) was reduced, although non-significantly, between Pre and Post1. The reduction remained at Post2 for the wrist and shoulder. No changes in the number of MUs were apparent for the affected side, although statistical trends were noted between Pre and Post2 for the shoulder (p=0.041), elbow (p=0.028) and overall (p = 0.047). The relative 3D distance of the non-affected side was reduced for the shoulder from Pre to Post2, significant between Pre and Post2 (T=10, p < 0.025, r=0.77). The 3D distance of the elbow was unchanged between Pre and Post2 for the non-affected side, whereas a trend was apparent for the wrist (p = 0.038) and overall (p = 0.028). The 3D distance of the affected side was reduced for the shoulder, elbow, and wrist between all occasions, significant between Pre and Post2 (shoulder: T=2, p < 0.01, r = 0.82; elbow: T = 0, p < 0.01, r = 0.89; wrist: T=0, p < 0.01, r=0.82; overall T=0, p < 0.01, r = 0.89).

# Discussion

Beginning with the learning of the training task, the ability to learn motor skills through feedback has been shown to be governed by similar principles in children with HCP as in the population without such impairments [13]. In accordance, the two children with HCP in the present study showed motor learning by improving their synchronization with the metronome sound during 4 weeks individually adjusted IM training. The improvement was noticeable in both conditions (with and without error feedback) for Case I and specifically with error feedback for Case II (Figures 4(a) and (b)). The effects appeared to be consolidated as the achieved

synchronization level remained at 6 months after concluded training for both cases.

IM training incorporates many of the factors that have been described to be important in the induction of plastic changes in the brain [21] and, thus, appears to be a promising intervention method for persons with sensori-motor deviations. However, as most of the research on cortical plasticity has been focused on the study of how neurologically mature animal models recover in response to induced strokes or asphyxias, it remains unclear to what extent these mechanisms are applicable on lesions occurring early on in life as in CP. The results from the present case study are in agreement with previous findings from studies using the IM paradigm [4,6-8]. Thus, suggesting a behavioural manifestation of neuronal plasticity processes which may be governed by the plasticity inducing principles suggested by Nudo [21]. However, such improvements in synchronization abilities may also be related to increased focus of attention as an effect of IM training.

Turning to the kinematic findings, smoothness of movement trajectory has been shown to be a reliable measure with high test-re-test reliability in children with varying severity of CP [22]. Several kinematic studies have implicated measures of smoothness in investigations of motor control and planning in CP [19,23-25]. Although impairment of motor planning abilities in CP has been shown in terms of end state control [2,16,26] it is unclear to what extent smoothness of movement trajectory can be interpreted as a marker for constraints in motor planning ability and/or in biomechanical functions. Thus, the general reduction of the number of MUs and 3D distance between Pre and Post2 (Case I; in relation to the non-affected side in the unimanual condition and both sides in the bimanual condition; Case II; in relation to MUs of the non-affected side in the unimanual condition and 3D distance of the affected side in the bimanual condition) indicate increased movement control and/or decreased biomechanical constraint as a possible consolidation effect of IM training. In relation to quality of movements, most of the kinematic effects occurred with a simultaneous reduction in duration of movement. Taken together, this suggests a more dynamical and energy efficient movement performance. These findings are in adherence to the clinically significant effects of IM training on upper-limb function reported in patients with stroke-induced hemiparesis [9]. Case II showed fewer significant effects on the kinematic properties than Case I, possibly associated to the diminutive level of learning of self-phased rhythmicity as assessed by the IM apparatus. Differences in the children's clinical background and/or ability to focus their attention could possibly also account for these learning differences.

# Conclusions

Although the effects of IM training on kinematics varied among the two children, some improvements in spatio-temporal organization were observed. The differences in kinematic outcomes between the cases could probably be related to differing clinical background and/or the ability to learn and/or to consolidate the training task. Future studies, including larger samples of children with different severity level of HCP and also other types of CP are needed to evaluate the generalizability and stability of the present findings. Further, it is highly important to investigate underlying neural mechanisms of motor planning and control and how changes in brain plasticity activity and morphology through mechanisms mediate behavioural changes in this heterogeneous group of children. Such knowledge is important in furthering the development of intervention/training methods that are well adjusted and effective.

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