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Modified Constraint-Induced Movement Therapy combined with Bimanual Training (mCIMT–BiT) in children with unilateral spastic cerebral palsy: How are improvements in arm-hand use established?

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ABSTRACT

A recent randomized controlled trial indicated that modified Constraint-Induced Movement Therapy followed by Bimanual Training (mCIMT–BiT) is an effective intervention to improve spontaneous use of the affected upper limb in children with unilateral spastic cerebral palsy (CP). The present study aimed to investigate how the above-mentioned improvements as a result of 8 weeks mCIMT–BiT were established. 52 children with unilateral spastic CP with Manual Ability Classification System (MACS) scores I, II or III and aged 2.5–8 years were randomly allocated to either mCIMT–BiT ($n = 28$) or Usual Care (UC) ($n = 24$). Developmental disregard ('learned non-use') and upper limb capacity and performance scores were derived from the Video Observations Aarts and Aarts, module Determine Developmental Disregard. Active and passive range of motion at the affected wrist and elbow were assessed using goniometry during isolated movements. Upper limb capacity and performance demonstrated significantly greater improvements after mCIMT–BiT compared to UC, which lasted up to 8 weeks follow-up, whereas developmental disregard and passive and active range of motion did not show differential effects. The results support the notion that improvement of capacity and performance of the upper limb through mCIMT–BiT in children with unilateral spastic CP is based on a better utilization of existing motor functions of the affected arm and hand. However, enhancement of the overall amount of use (or the reduction of learned non-use) may still be suboptimal leaving room for improvement of this treatment.

1. Introduction

In children with unilateral spastic CP, an increasing number of studies has indicated positive effects of (modified) Constraint-Induced Movement Therapy ((m)CIMT) on the potential of the affected arm to assist the unaffected arm during bimanual activities (Eliasson, Krumlinde-sundholm, Shaw, & Wang, 2005; Wallen, Ziviani, Herbert, Evans, & Novak, 2008) as well as on the quality, speed, and dexterity of upper limb function (Bonnier, Eliasson, & Krumlinde-

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sundholm, 2006; Charles, Wolf, Schneider, & Gordon, 2006; Crocker, MacKay-Lyons, & McDonnell, 1997; Deluca, Echols, Law, & Ramey, 2006; Naylor & Bower, 2005; Sung et al., 2005; Taub, Ramey, DeLuca, & Echols, 2004; Wallen et al., 2008; Willis, Morello, Davie, Rice, & Bennett, 2002), the spontaneous use of the affected arm (Charles et al., 2006; Crocker et al., 1997; Taub et al., 2004), and the level of independence in self care (Brandao, Mancini, Vaz, de Melo, & Fonseca, 2010; Charles et al., 2006; Deluca et al., 2006; Sung et al., 2005; Taub et al., 2004; Wallen et al., 2008; Willis et al., 2002). Nonetheless, a recent Cochrane review (Hoare, Imms, Carey, & Wasiak, 2007a) concluded that, although these results are encouraging, they are still inconclusive due to methodological limitations related to small sample sizes, group allocation bias, and the influence of non specific (mainly intensity) effects. It was recommended that the effectiveness of (m)CIMT should be revealed in future, sufficiently powered trials using uniform, objective and valid outcome measures.

Recently, we conducted a randomized controlled trial (RCT) (Aarts, Jongerius, Geerdink, van Limbeek, & Geurts, 2010) in 52 children with unilateral spastic CP showing that 6 weeks mCIMT followed by 2 weeks of task-specific bimanual training (mCIMT–BiT) improves the spontaneous use of the affected limb during play and self-care activities as assessed with the Assisting Hand Assessment (AHA) (Krumlinde-sundholm, Holmefur, & Eliasson, 2007) and the ABILHAND-Kids (Arnould, Penta, Renders, & Thonnard, 2004), respectively. In addition, significant improvements were obtained in terms of experienced daily life problems and individually tailored functional goals as assessed with the Canadian Occupational Performance Measure (Law et al., 2005) and Goal Attainment Scaling (Steenbeek, Ketelaar, Galama, & Gorter, 2007). Apparently, children receiving 8 weeks mCIMT–BiT improved the spontaneous use of their affected hand in most areas of daily functioning. From a neurophysiological perspective, this result imposes the question how these improvements were established? For instance, did these children improve the underlying active range of motion (aROM) or passive range of motion (pROM) at critical joints as measures at the 'bodily functions' level of the International Classification of Functioning Disability and Health (ICF) (World Health Organisation, 2010)? Or did they improve their upper limb capacity leading to better spontaneous use at the 'activity level' of the ICF, without true restoration of underlying motor functions? There is also the possibility that the children's upper limb capacity essentially remained the same, but that those receiving mCIMT–BiT improved the amount of use of the upper limb due to a reduction of so-called 'learned non-use' or 'developmental disregard'.

The notion of a reduction of learned non-use has probably received the greatest attention in the literature on mCIMT in children with CP, but it has never been established with good empirical evidence (Brady & Garcia, 2009; Hoare et al., 2007a; Hoare, Wasiak, Imms, & Carey, 2007b; Huang, Fetters, Hale, & McBride, 2009). Children with unilateral spastic CP often display a form of learned non-use, as in daily life they experience too little incentive to use their affected upper limb during functional tasks, which often becomes apparent during bilateral activities (Gordon, Charles, & Wolf, 2005). The basic notion behind learned non-use following unilateral brain damage is that certain residual motor capacities of the affected extremity remain hidden due to a learning process favouring the easier movements with the non affected extremity (Taub, Uswatte, & Pidikiti, 1999). As a result, motor performance is often better during forced activities than during spontaneous activities of the affected upper limb (Taub, Uswatte, Mark, & Morris, 2006). In the paediatric literature, this phenomenon has been referred to as 'developmental disregard', because the learned non-use does not so much relate to a relatively short period of adaptation to an acute lesion (such as in stroke), but rather reflects a developmental process or strategy through which children with unilateral CP fail to integrate the potentials of their affected upper limb in daily life routines (Deluca et al., 2006; Hoare et al., 2007b; Sutcliffe, Logan, & Fehlings, 2009; Taub et al., 2004). In children with unilateral spastic CP, there may be a critical lack of movement stimulation during developmental periods when movement repertoires are rapidly being acquired in typically developing children. This creates a situation in which, in theory, new neural substrates for entire classes of behaviour are not well established, refined, and coordinated (Deluca et al., 2006). In addition to this lack of movement stimulation, children with unilateral CP often suffer from upper limb spasticity and loss of motor selectivity, leading to stereotypical movement patterns such as internal rotation of the shoulder, elbow flexion with pronation of the forearm, ulnar deviation and flexion of the wrist and thumb-in-palm and/or finger-swan neck deformities (Burtner et al., 2008). These children often tend to maintain the wrist in flexion and show difficulties in extending this joint during manual activities, even when they are able to actively extend the wrist and fingers at least 30 degrees from the resting position (Vaz et al., 2008). As a result, the wrist flexors and extensors may show tissue remodelling to generate more grip strength with the wrist in flexion (Vaz, Cotta, Fonseca, Vieira, & de Melo Pertence, 2006), after which a normal movement pattern of the hand is unlikely to return and children may become prone to develop developmental disregard.

The goal of this study was to investigate *how* the above-mentioned improvements in spontaneous use of the affected limb during play and self-care activities were established as a result of 8 weeks mCIMT–BiT. Developmental disregard was assessed with the Video Observations Aarts and Aarts module Determine Developmental Disregard (VOAA-DDD) (Aarts, Jongerius, Geerdink, & Geurts, 2009) as were upper limb capacity and performance as measures at the activity level of the ICF. Indeed, recent research has shown that both unimanual capacity and bimanual performance are important aspects of bimanual activities in children with CP (Sakzewski, Ziviani, & Boyd, 2010). In addition, active and passive range of (extension) motion of the affected wrist and elbow were assessed as measures at the ICF level of bodily functions. The results of a previous study (Sutcliffe et al., 2009) led us to the hypothesis that developmental disregard would be reduced or even resolved after mCIMT–BiT. In addition, it was hypothesized that changes in active or passive range of joint motion would not underlie the improvements found at the activity level.

2. Methods

2.1. Participants

The children for this study were recruited from eight rehabilitation centres in the Netherlands. Inclusion criteria were: (1) cerebral palsy with a unilateral or severely asymmetric, bilateral spastic movement impairment, (2) age 2.5–8 years, 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 programme, and (3) in ability to walk independently without a walking aid.

2.2. Study design

Within 48 h after inclusion, each participant was randomized to the mCIMT–BiT or UC group by throwing a dice with equal probabilities. All children underwent a comprehensive upper limb evaluation before the start of the intervention period (week 0), at the end of the intervention period (week 9), and at the end of the study protocol (week 17). After the study protocol (week 17), the children who had been allocated to the UC group were as yet offered the opportunity to participate in a mCIMT–BiT group. The study was approved by the regional Medical Ethical Committee for Research Involving Human Subjects. Oral and written informed consent was obtained from all parents or caregivers.

2.3. Interventions

In the *mCIMT–BiT group*, also named the 'Pirate group', training to improve the functional performance of the affected arm and hand was given at the primary rehabilitation centre (St. Maartenskliniek) during 3-h afternoon sessions, three days per week, for eight weeks. Approximately 50% of the training was individual occupational therapy (OT) or physical therapy (PT), whereas the remaining 50% of the programme was given in small groups. During the first six weeks, restraint of the unaffected arm and hand was applied, while the affected arm had to be used for all activities. In all therapy sessions the principles of shaping and repetitive task practice (Gordon et al., 2005; Taub et al., 2004) were implemented. During the last 2 weeks, the emphasis was on goal-directed task-specific bimanual training without any restraint. In addition to these therapy sessions, parents were asked to stimulate their child to use the affected arm and hand at home as much as possible and to register the duration of specific periods of stimulation on the child's daily record form.

The children in the *UC group* received a regular rehabilitation programme in one of the participating rehabilitation centres. Bimanual hand use of similar intensity and duration was stimulated at home and in the school environment during eight weeks. The programme included individual OT and/or PT given twice a week in 0.5–1 h sessions (total therapy time: 1.5 h/week), while another 7.5 h/week stimulation of bimanual hand use was given at home by the parents or in (pre)school groups according to predetermined instructions. Parents and teachers were asked to register the duration of specific periods of stimulation on the child's daily record form.

Details of the mCIMT–BiT and UC treatments have been published in a previous article (Aarts et al., 2010). Due to the nature of both interventions, it was impossible to blind either participants or therapists with regard to treatment allocation. All assessments were performed by one blinded OT (YG).

2.4. Outcome measures

2.4.1. ICF level of activities

The VOAA-DDD allowed to assess developmental disregard as well as capacity and performance scores of the affected arm and hand using one test. In this test, both the duration and the frequency of spontaneous use of the affected arm and hand were systematically observed during two selected activities: 'stringing beads' (demanding the use of both hands) and 'decorating a muffin' (stimulating but not demanding the use of both hands) (Aarts et al., 2009). It was presumed that the more demanding beads task reflects the child's optimal capacity of the affected arm and hand (Fig. 1a). In contrast, the less demanding muffin task was regarded as an indicator of the child's natural performance with the affected arm and hand

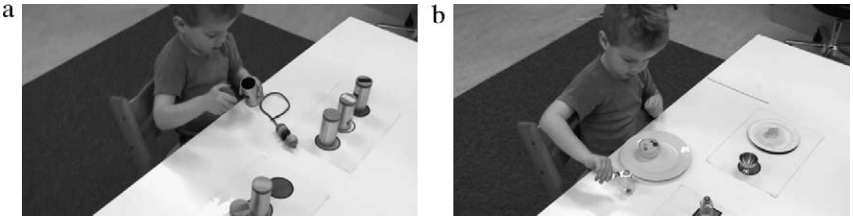

Fig. 1. (a) A boy (left side affected) removing the lid from a can bimanually during the beads task. (b) The same boy during the muffin task (same assessment).

(Fig. 1b). The difference in the duration of hand use between the beads and the muffin task was calculated as a measure of developmental disregard because, in the literature, learned non-use has typically been defined in terms of reduced amount (duration) of use (Taub, 2004). In addition, the frequency of the behaviours "grasps", "holds" and "releases" was assessed for both tasks and calculated as a percentage of the maximum number of possible observations (see for a detailed description Aarts et al. (2009)). As a measure of upper limb capacity, only movements executed with selective active wrist extension (not maintaining the wrist in a fixed flexion pattern) of the affected arm and hand were counted during the beads task. As a measure of performance, all movements irrespective of movement pattern were counted during the muffin task. How the various outcome measures were derived from the VOAA-DDD is illustrated in Fig. 2. Both the beads and muffin tasks were considered attractive and age appropriate and showed a maximum performance time of 5 min. The construct validity as well as the inter-rater, intra-rater and test–retest reliability of the VOAA-DDD have been reported to be good (Aarts et al., 2009).

2.4.2. ICF level of bodily functions

The active and passive range of extension motion at the affected wrist and elbow were measured simultaneously by two therapists (one assessing OT and one assisting PT) using a standard goniometer (Stuberg, Fuchs, & Miedaner, 1988) with the child in a seated position. All measurements were done according to a standard protocol during which the aROM was measured first, followed by the pROM. Wrist extension measurements were started with the elbow 90° flexed, the forearm fully pronated, and the upper arm alongside the trunk. Measurement of elbow extension started with the shoulder in 90° anteflexion, the elbow in full flexion with the fingertips on or near the ipsilateral shoulder and the elbow supported by the assisting PT. The active movements were demonstrated by the assessing OT, after which the child performed the required elbow or wrist extension. The assisting PT maintained the maximally reached joint position, while the assessing OT recorded the aROM joint angle in steps of 5°. Then, the assisting PT moved the respective joint towards the maximum passive position and the assessing OT recorded the pROM joint angle again in steps of 5°. Based on Klingels et al. (2010) results, test–retest reliability of measuring joint ROM was considered very good (ICCs 0.81–0.94). Based on the degree of active wrist extension (AWE) at baseline, children were categorized into three subgroups: AWE 1: 135–180° (good wrist control), AWE 2: 95–130° (moderate wrist control), and AWE 3: 0–90° (poor wrist control).

2.5. Statistical analysis

For all outcome measures, the two groups were compared with regard to functional changes between pre (week 0) and post treatment (week 9) using ANCOVA in which differences at baseline, even when 'insignificant', were used as covariates. Cohen's *d* values (Cohen, 1988) were calculated to obtain a pre-post intervention effect size. To determine whether possible effects remained constant 8 weeks post intervention, student *t*-tests (paired samples) were used to compare the results between week 9 (post treatment) and week 17 (at 8 weeks follow up). To compare pre-post intervention changes for different subgroups based on the MACS and AWE scores, one way ANOVA was used. All data handling and analyses were carried out by an independent statistician who was blinded for group allocation. Power calculation indicated that 52 children needed to be included (Aarts et al., 2010). SPSS version 17.0 was used for computerized analysis (SPSS, Inc., Chicago, IL).

3. Results

3.1. Participants

A total of 52 children with unilateral spastic CP were included (Aarts et al., 2010), of which 28 children were allocated to the mCIMT–BiT group and 24 to the UC group. Immediately after randomization, two children withdrew from the UC group

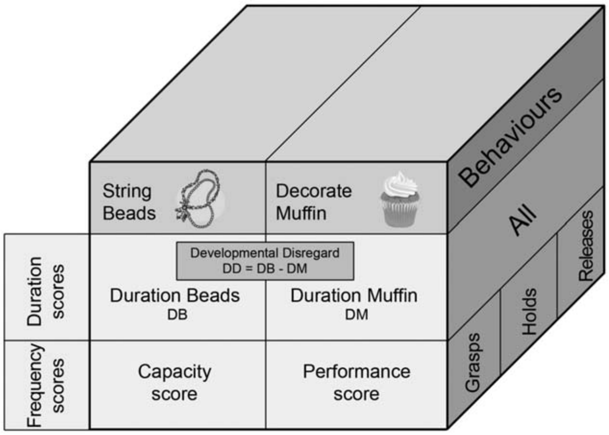

Fig. 2. VOAA (Video Observations Aarts and Aarts) with three dimensions (Tasks, Focus of scoring, Behaviours) and three different outcomes.

due to family circumstances. Thereafter, no participants were lost to follow-up or changed group allocation. Hence, the data of 22 participants in the UC group are presented. The mCIMT–BiT group ($n = 28$) and the UC group ($n = 22$) had similar baseline characteristics (age, gender, affected side, MACS, AWE, Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997)). In addition, no significant differences between groups were found for the baseline values of the outcome measures (Table 1). According to the children's daily record forms, the mCIMT–BiT group received on average 9 h per week therapy and 3.3 h additional stimulation at home (total stimulation time = 12.3 ± 1.9 h). The UC group received on average 1.5 h per week therapy and 11.2 h additional stimulation at home or at (pre)school (total stimulation time = 12.7 ± 2.1 h).

3.2. ICF level of activities

Table 2 presents the outcome measures derived from the VOAA-DDD. After the intervention period, the capacity score had increased by 58% in the mCIMT–BiT group versus 3.6% in the UC group. The mean group difference in change scores (corrected for differences at baseline) was 13.6 (95% CI 3.77–23.51). Cohen's *d* indicated a moderate effect size (0.55). Subgroup analysis (Table 3) indicated that the capacity scores increased most strongly (17.0) in children with classification AWE 2, although this subgroup effect did not reach significance. Fig. 3 illustrates a good response in a typical child. At follow-up, the capacity score had decreased by 11% in the mCIMT–BiT group versus 12% in the UC group, which group difference was not significant.

As for the performance score, the mCIMT–BiT group improved by 15% after the intervention period, whereas the UC group showed 1% deterioration. The mean group difference in change scores (corrected for differences at baseline) was 12.8 (95% CI 4.82–20.83), indicating a moderate effect size (Cohen's *d* 0.65). Subgroup analysis (Table 4) indicated that the performance scores increased most strongly in children with classifications MACS II (12.2) and III (18.0) (subgroup effect $p = 0.044$). At follow-up, the observed effects further improved by 3% in the mCIMT–BiT group, whereas the UC group showed a 9% improvement. This group difference was not significant.

Developmental disregard showed a 31% decrease in the mCIMT–BiT group after the intervention period versus 2% decrease in the UC group. The mean group difference in change scores (corrected for differences at baseline) was −6.4 (95% CI −13.61 to 0.72) (Cohen's *d* 0.33), which effect did not reach significance. Subgroup analysis (Table 4) indicated that developmental disregard decreased specifically in children with classification MACS III (−21.0) (subgroup effect $p = 0.007$). At follow-up, developmental disregard had again increased by 33% in the mCIMT–BiT group, whereas the UC group showed a decrease of 17%. This group difference was not significant.

3.3. ICF level of bodily functions

The results for the ROM outcomes are presented in Table 5. Although the mCIMT–BiT group showed a positive trend towards improvement of active (5%) and passive (2%) wrist extension after the intervention period, these differences were not significant compared to the UC group. At follow up, active and passive wrist extension almost returned to baseline values in the mCIMT–BiT group. Any differential effects of mCIMT–BiT on active or passive elbow extension were negligible and non significant.

Table 1
Baseline characteristics of both groups.

Characteristics	mCIMT–BiT ($n = 28$)	UC ($n = 22$)
Gender		
Male	14 (50)	14 (64)
Female	14 (50)	8 (36)
Age (mean ± SD in years)	4.8 ± 1.3	5.1 ± 1.7
Affected side		
Left	14 (50)	14 (64)
Right	14 (50)	8 (36)
GMFSC		
I	27	21
II	1	1
MACS		
I	9	7
II	12	10
III	7	5
AWE		
1	11	7
2	15	9
3	2	6

Values are numbers (percentages), unless otherwise indicated. mCIMT, modified Constraint-Induced Movement Therapy; UC, Usual Care; GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System; AWE, Active Wrist Extension.

Table 2
VOAA-DDD outcome measures at all assessments, change scores, and group differences of change scores.

Outcome measures	mCIMT–BiT	ΔmCIMT–BiT	UC	ΔUC	Mean group difference (95% CI)[a]	Effect Size[b]	mCIMT–BiT p-value	UC p-value
Capacity score (range 0–100%)								
Baseline; mean ± SD	25.7 ± 23.1		27.6 ± 30.2	1.0 ± 13.6				
Week 9	40.5 ± 29.2	14.9 ± 19.7	28.6 ± 28.8		13.6 (3.77–23.51)	0.55		
Week 17	36.1 ± 24.9	10.4 ± 21.0	25.3 ± 26.4	−2.3 ± 12.3			0.290	0.084
Performance score (range 0–100%)								
Baseline; mean ± SD	60.5 ± 29.4		51.1 ± 31.0					
Week 9	69.6 ± 21.4	9.1 ± 17.0	50.4 ± 28.5	−0.7 ± 16.0	12.8 (4.82–20.83)	0.65		
Week 17	71.4 ± 19.8	10.9 ± 14.2	55.1 ± 31.5	4.0 ± 13.7			0.596	0.184
Development disregard (DD) (range 0–100%)								
Baseline; mean ± SD	21.4 ± 14.1		21.7 ± 14.8					
Week 9	14.7 ± 11.6	−6.6 ± 14.8	21.3 ± 14.8	−0.5 ± 17.4	−6.4 (−13.61–0.72)	0.36		
Week 17	19.6 ± 15.3	−1.7 ± 13.2	17.7 ± 13.0	−4.0 ± 18.3			0.072	0.253

mCIMT–BiT, modified Constraint-Induced Movement Therapy combined with Bimanual Training; UC, Usual Care; VOAA-DDD, Video Observations Aarts and Aarts, module Determine Development Disregard; SD, Standard Deviation.

[a] Mean group difference (95% confidence interval) (corrected for difference at baseline).

[b] Effect size (Cohen's d): "small, $d = .2$," "moderate, $d = .5$," and "large, $d = .8$".

Table 3
VOAA-DDD outcome measures: pre-post intervention change scores for subgroups AWE 1–3.

AWE	Mean change T2–T1(SD)					
	Capacity score		Performance score		Developmental disregard	
	mCIMT–BiT	UC	mCIMT–BiT	UC	mCIMT–BiT	UC
1	12.64 (15.91)	6.57 (13.35)	7.91 (21.05)	−6.00 (14.28)	−4.18 (14.67)	5.00 (19.69)
2	17.00 (22.68)	−5.11 (13.59)	10.47 (15.25)	−2.33 (17.88)	−8.53 (13.53)	3.22 (17.25)
3	11.00 (24.04)	3.67 (12.16)	6.00 (8.49)	7.83 (13.89)	−6.00 (32.53)	−12.33 (9.85)

mCIMT–BiT, modified Constraint-Induced Movement Therapy combined with Bimanual Training; UC, Usual Care; SD, Standard Deviation; AWE, Active Wrist Extension, AWE 1: 135–180° (good wrist control), AWE 2: 95–130° (moderate wrist control), and AWE 3: 0–90° (poor wrist control); T1, pre-intervention assessment (baseline); T2, post-intervention assessment (week 9).

4. Discussion

In a previous publication it was shown that, in young children with unilateral spastic CP, 6 weeks of modified Constraint-Induced Movement Therapy followed by 2 weeks of task-specific Bimanual Training (mCIMT–BiT) improved the spontaneous use of the upper limb during play and self-care activities in both qualitative and quantitative terms more than usual care of the same duration (Aarts et al., 2010). The goal of the present study was to investigate *how* the above-mentioned improvements were established. Using the VOAA-DDD as an integral test, the greatest improvements were found for the capacity score that specifically assessed the frequency of selective motor control at the level of the wrist during a beads task that demanded the use of both hands. Children with moderate wrist control at baseline (AWE 2)

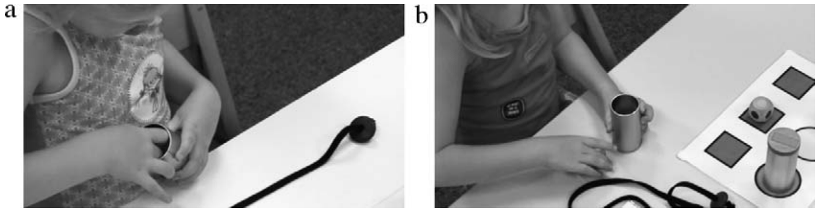

Fig. 3. (a) A girl using a flexed position of the affected wrist during the beads task (left side) before treatment. (b) The same girl using an extended wrist during the beads task after treatment.

Table 4
VOAA-DDD outcome measures: pre-post intervention change scores for subgroups MACS I–III.

MACS	Mean change T2–T1(SD)					
	Capacity score		Performance score		Developmental disregard	
	mCIMT–BiT	UC	mCIMT–BiT	UC	mCIMT–BiT	UC
I	13.33 (18.97)	−3.86 (20.45)	−1.78 (7.95)	−11.14 (18.59)	−0.67 (10.86)	2.43 (19.88)
II	21.83 (22.74)	1.00 (6.46)	12.17 (16.91)	6.70 (14.52)	−2.75 (16.27)	−5.30 (15.88)
III	4.86 (9.96)	7.80 (12.26)	18.00 (20.16)	−1.00 (5.48)	−21.00 (3.92)	5.20 (17.85)

mCIMT–BiT, modified Constraint-Induced Movement Therapy combined with Bimanual Training; UC, Usual Care; SD, Standard Deviation; MACS, Manual Ability Classification System; T1, pre-intervention assessment (baseline); T2, post-intervention assessment (week 9).

Table 5
ROM outcome measures at all assessments, change scores, and group differences of change scores.

Outcome measures	mCIMT–BiT	ΔmCIMT–BiT	UC	ΔUC	Mean group difference (95% CI)[a]	Effect size[b]	mCIMT–BiT *p*-value	UC *p*-value
ROM active wrist extension								
Baseline mean ± SD	127.9 ± 21.2		117.5 ± 36.7					
Week 9	133.8 ± 21.0	5.9 ± 13.5	118.9 ± 39.4	1.4 ± 17.3	5.4 (−3.41–14.29)	0.25		
Week 17	128.2 ± 22.0	0.4 ± 17.5	114.8 ± 38.7	−2.7 ± 29.1			0.062	0.393
ROM passive wrist extension								
Baseline mean ± SD	177.7 ± 7.0		178.2 ± 6.6					
Week 9	180.4 ± 7.6	2.7 ± 8.7	177.3 ± 10.7	−0.9 ± 5.9	3.5 (−0.82–7.76)	0.33		
Week 17	179.8 ± 7.9	2.1 ± 6.7	176.4 ± 13.2	−1.8 ± 8.9			0.725	0.623
ROM active elbow extension								
Baseline mean ± SD	170.2 ± 15.4		172.1 ± 14.9					
Week 9	172.1 ± 10.3	2.0 ± 12.6	171.1 ± 14.1	−0.9 ± 7.5	2.1 (−2.85–6.99)	0.17		
Week 17	173.6 ± 10.4	3.4 ± 12.1	170.2 ± 17.6	−1.8 ± 8.5			0.434	0.611
ROM passive elbow extension								
Baseline mean ± SD	179.8 ± 7.9		180.9 ± 10.2					
Week 9	179.8 ± 7.5	0.0 ± 6.2	179.6 ± 11.4	−1.4 ± 5.2	1.2 (−2.07–4.46)	0.15		
Week 17	180.9 ± 6.4	1.1 ± 4.8	178.4 ± 12.5	−2.5 ± 5.3			0.297	0.397

mCIMT–BiT, modified Constraint-Induced Movement Therapy combined with Bimanual Training; UC, Usual Care; ROM, Range Of Motion in degrees; SD, Standard Deviation.

[a] Mean group difference (95% confidence interval) (corrected for difference at baseline).

[b] Effect Size (Cohen's *d*): "small, *d* = .2," "moderate, *d* = .5," and "large, *d* = .8".

appeared to be the best responders, although other children improved as well. To a lesser extent, improvements were observed for the performance score that assessed the frequency of all motor behaviours during a muffin task that merely stimulated bimanual activity. Here, only children with moderate to poor manual ability at baseline (MACS II and III) appeared to be responders. There was no significant reduction of developmental disregard defined as the difference in the amount (duration) of hand use between both tasks. There was, however, a significant effect of subgroup. Children with poor manual ability at baseline (MACS III) showed a large reduction of developmental disregard, whereas children with better manual ability hardly responded on this measure. No significant changes were observed in the underlying active or passive ranges of joint motion.

The hypothesis that developmental disregard would be reduced or even resolved after mCIMT–BiT was, thus, not corroborated. After the intervention, there were still several children who underused their affected arm during the muffin task, while these children did show improved capacity scores during the beads task. Apparently, their improvement of hand capacity did not reach a level of sufficient automaticity in order to enhance the overall amount of spontaneous use. As a result, a tendency to rely on the non affected hand still existed. This phenomenon of 'persistent non-use' was observed in all MACS subgroups. Nevertheless, children with poor manual ability (MACS III) appeared to be the best responders in terms of reduction of developmental disregard, although their improvement did not last until the follow up. This result is consistent with the findings of Eliasson et al. (2005) who found that children with poor manual ability improved considerably more on the AHA after CIMT than children with better hand ability. Interestingly, a previous study by Sutcliffe et al. (2009) concluded that developmental disregard at baseline predicted improvement of upper limb capacity and performance in bimanual tasks as well as improved grip strength after mCIMT in children with unilateral spastic CP. This conclusion, however, was based on the comparison of the Quality of Upper Extremity Skills Test (DeMatteo, 1992) and the Paediatric Motor Activity Log (an adaptation of the adult Wolf Arm Motor Ability test (Wolf, Lecraw, Barton, & Jann, 1989)) in merely five children without providing MACS classifications. As a result, a comparison with the results of this study is hard to make.

The second hypothesis that changes in active or passive ranges of joint motion would not underlie any improvements found in upper limb capacity and performance was substantiated by the results. Apart from a non significant trend towards small (2–5%) increases in passive and active wrist extension, the mCIMT–BiT group did not show different outcomes compared to the UC group. This pattern of results supports the notion that improvement of upper limb capacity and performance after mCIMT–BiT in children with unilateral spastic CP is based on a better utilization of existing motor functions of the affected arm and hand, rather than on true restoration of muscle strength or motor selectivity. Apparently, (m)CIMT promotes the use of latent but existing motor functions which is reflected in an improved quality and, to a lesser extent, enhanced frequency of upper limb movement. This ‘unmasking’ of latent motor functions may, however, not be strong enough to result in a significant increase in the overall amount (duration) of use of the affected hand, e.g. due to lack of automaticity. Thus, the results of this study provide a first indication that, in contrast with current definitions of developmental disregard that focus on the amount of use, the quality of upper limb control may be an equally important aspect of learned non-use that is more responsive to mCIMT–BiT than the quantitative aspects.

Certain limitations of this study warrant consideration. First, only active and passive range of (extension) motion at the affected elbow and wrist were measured as important underlying motor functions. As a result, possible changes in shoulder and trunk motions might have remained undetected. When performing a forward reach task, children with unilateral spastic CP typically display increased abduction and internal rotation at the shoulder, while the trunk shows increased flexion as a compensation for loss of elbow extension and shoulder anteflexion (Reid, Elliott, Alderson, Lloyd, & Elliott, 2010). It may be that, after mCIMT–BiT, the children in the present study improved the selective control in their affected shoulder and diminished compensatory trunk flexion accordingly, which might underlie their improved upper limb capacity and performance. In future studies, three-dimensional kinematic assessment of the upper limb and trunk will help to assess detailed changes in active joint motions such as proposed by Butler et al. (Butler et al., 2010) using the ‘Reach and Grasp Cycle’. Second, we did not specifically assess motor planning as a cognitive component of upper limb control. Indeed, Steenbergen, Verrel, and Gordon (2007) found evidence for the fact that activity limitations in children with CP are not solely caused by disorders in movement execution of the affected upper limb, but also by motor planning deficits. Future (m)CIMT studies in children with CP should try to take this neglected motor planning aspect into account as well. Third, the lack of a significant mCIMT–BiT effect on developmental disregard in this study may have been due to a type II error. Whereas the children with a MACS score I or II at baseline showed a negligible effect, the subgroup of children with an initial MACS score III showed a very clear reduction of developmental disregard, while the group as a whole almost showed a significant improvement. Hence, a sufficiently large and well selected group of children with poor initial manual ability might have yielded significant effects on developmental disregard or ‘learned non-use’ after treatment.

In conclusion, the results of this study indicate that the observed improvements of upper limb capacity and performance after 8 weeks mCIMT–BiT in children with unilateral spastic CP are based on a better utilization of existing motor functions of the affected extremity, rather than on true restoration of muscle strength or motor selectivity. Effects on the amount of use (reduction of ‘learned non-use’) are less clear and seem to be restricted to the children with poor manual ability at baseline (MACS III). Thus, there appears to be room for further improvement of the effectiveness of (m)CIMT-BiT in children with unilateral spastic CP. Perhaps the implementation of repeated training periods in the successive stages of the child’s development will lead to even better effects in terms of upper limb capacity and performance, but particularly to significant and lasting effects on the overall amount and automaticity of use. In addition, short booster sessions in between training periods might help to improve the automatization process. Future studies should address these important issues.

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