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Motor learning curve and long-term effectiveness of modified constraint-induced movement therapy in children with unilateral cerebral palsy: A randomized controlled trial

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ABSTRACT

The goal of this study was to determine the progression of manual dexterity during 6 weeks (54 h) (modified) constraint-induced movement therapy ((m)CIMT) followed by 2 weeks (18 h) bimanual training (BiT) in children with unilateral spastic cerebral palsy (CP), to establish whether and when a maximal training effect was reached and which factors might influence the motor learning curve. In addition, long-term retention of effects was determined. In a randomized controlled trial of 52 children with CP, aged 2.5–8 years, comparing mCIMT-BiT to conventional therapy, 28 children were allocated to the mCIMT-BiT group. This group was assessed weekly with the Box and Block test. Long-term effectiveness was determined by collecting follow-up data of the primary (Assisting Hand Assessment, ABILHAND-Kids) and secondary (Melbourne, COPM) outcomes at six months and one year after intervention. Fifteen children (53.6%) reached a maximum training effect within the mCIMT period. This group differed from others with respect to age, but not gender, affected side or manual ability. Children younger than five years had a greater chance to reach a maximum score within 6 weeks mCIMT (OR = 6.67, 95%CI = 1.24–35.71) that stabilized already after four weeks; older children showed a longer progression and tended to decline afterwards. In both age groups, beneficial effects were retained in the long term. The findings suggest that children of 5 years and older might profit from a longer period of mCIMT than 54 h to reach their maximum unimanual capacity and to retain this capacity during subsequent bimanual training.

1. Introduction

Evidence is cumulating that constraint-induced movement therapy (CIMT) has beneficial effects on the capacity as well as the use of the affected upper limb in children with unilateral spastic cerebral palsy (CP) (Aarts, Jongerius, Geerdink, van Limbeek, & Geurts, 2010; Case-Smith, DeLuca, Stevenson, & Landesman Ramey, 2012; Charles, Wolf, Schneider, & Gordon, 2006; Deluca, Echols, Law, & Ramey, 2006; Eliasson, Shaw, Berg, & Krumlinde-Sundholm, 2011; Gordon et al., 2011; Sakzewski et al., 2011b; Taub, Ramey, DeLuca, & Echols, 2004). In addition, several studies have

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shown that the beneficial effects of (modified) CIMT/(m)CIMT are retained at six months (Case-Smith et al., 2012; Charles et al., 2006; Gordon et al., 2011; Taub et al., 2004; Wallen et al., 2011) and even one year (Sakzewski et al., 2011a) post intervention. However, relatively little is known about the learning curve during the period of constraint, for instance, at what point in time (maximal) effects are reached. Systematic reviews on (m)CIMT and forced use therapy have concluded that studies vary widely in terms of intervention characteristics, in particular with regard to the type and overall duration of constraint and the total dosage of unilateral upper-limb training (Hoare, Imms, Carey, & Wasiak, 2007; Huang, Fetters, Hale, & McBride, 2009; Sakzewski, Ziviani, & Boyd, 2009). Because mCIMT programmes are very intensive and, thus, demanding in terms of time and effort invested by the child and the family, adequate dosing of an mCIMT programme is essential to prevent unnecessary burden to the participants and to assure optimal cost-effectiveness. In addition, a well targeted dosage of mCIMT can minimize the potential risk of damage to the immature brain as a consequence of restraining the use of a healthy limb in a young child (Martin, Choy, Pullman, & Meng, 2004).

A few studies (Case-Smith et al., 2012; Gordon, 2011) have been undertaken to find evidence for the best composition of mCIMT, tailored to the needs and capacities of the children. A recent randomized controlled trial (RCT) (Case-Smith et al., 2012), comparing a six hours/day with a three hours/day protocol (both 21 intervention days) in *young* children ($n = 18$, age 3–6 years), found no differential effects and suggested that a three hours/day training during four weeks (a total of 63 h) would be sufficient for an optimal effect. On the other hand, a review (Gordon, 2011) comparing two mCIMT protocols (60 h/10 days (Charles et al., 2006) vs. 90 h/15 days (Gordon et al., 2011)) for *older* children ($n = 21$, age 4–13 years, and $n = 20$, age 3–10 years, respectively) showed a larger improvement of hand function as assessed with the Jebson–Taylor test in the group receiving 90 h of training. Hence, the conclusion is justified that the optimal intensity of mCIMT is still unknown for various age groups with unilateral spastic CP. In addition, it is unknown whether optimal (maximal) effects are being reached at all and which factors (e.g. age, manual ability at baseline, training intensity) might influence the motor learning curve as well as the retention of treatment effects in the long term.

Previously, we have reported on the efficacy of six weeks mCIMT followed by two weeks bilateral training (mCIMT-BiT) in 28 children with unilateral spastic CP, aged 2.5–8 years, providing results up to two months post intervention (Aarts et al., 2010). Our mCIMT-BiT programme was embedded in a playful 'Pirate group' programme which facilitated both constraint of the 'wounded' (i.e. non-affected) arm and unilateral training of the 'sound' arm handling the sword (i.e. the affected arm). This programme consisted of 54 h of unilateral upper-limb training followed by 18 h of bimanual training to ensure that training effects would be integrated into daily life bimanual activities (Aarts, Hartingsveldt, et al., 2011; Aarts, Jongerius, Geerdink, van Limbeek, & Geurts, 2011). Although this intervention turned out to be highly effective to improve the use of the affected upper limb as assessed with the Assisting Hand Assessment (AHA) (Krumlinde-Sundholm, 2003) and the ABILHAND-Kids (Arnould, Penta, Renders, & Thonnard, 2004), we could not draw any conclusions based on these measures with regard to the *learning curve* of the children during the mCIMT period or the BiT period. For this purpose, a different outcome measure was collected every week during the entire training period of eight weeks, i.e. the Box and Block test. This separate measure, focusing on the main functions of the hand (i.e. grasp, hold and release), was selected in order not to influence the assessments of the primary outcomes (AHA, ABILHAND-Kids) due to repeated testing. To the best of our knowledge, there are no previous studies that reported on the learning curve *during* an mCIMT intervention.

Hence, the primary goal of this study was to investigate the progression of manual dexterity as assessed with the Box and Block test during a six-weeks mCIMT programme followed by a two-weeks BiT programme that has proven its effectiveness in children with unilateral spastic CP, aged 2.5–8 years, in order to determine whether and when a maximum training effect was reached and what factors might influence the learning curve. The ultimate goal was to extract information based on which the optimal dosage of constraint and unilateral upper-limb training could be determined. In addition, we compared the follow-up data of the original primary and secondary outcome measures at six months and one year post intervention with the previously published results one week post intervention for all children that participated in the original study (Aarts et al., 2010).

2. Methods

2.1. Participants and design

Fifty children with unilateral spastic CP, aged 2.5–8 years, participated in this study. The selection process has been described previously (Aarts et al., 2010). Table 1 shows the demographic and clinical characteristics of the participants. The Manual Ability Classification System (MACS) was used to classify manual ability, focusing on actual performance of handling objects (Eliasson et al., 2006). Twenty-eight children took part in the mCIMT-BiT group and 22 in the usual care group (UC) after random allocation. Only the 28 subjects participating in the mCIMT-BiT group were tested weekly with the Box and Block test to assess changes in manual dexterity during the intervention. As for the original primary and secondary outcome measures, all children were assessed at baseline and at one week, two months and six months after the intervention. In addition, the mCIMT-BiT group underwent a follow-up assessment of the primary outcome measures one year after the intervention. In the present study, the follow-up assessments at six months and one year post intervention are compared to the data one week post intervention.