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Intensive upper limb intervention with self-management training is feasible and promising for older children and adolescents with unilateral cerebral palsy



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ABSTRACT

This study explored the feasibility and preliminary effectiveness of a short (one week) intensive intervention combining Constraint Induced Movement Therapy (CIMT) and bimanual training (BiT) to improve upper limb capacity and bimanual performance guided by individual goal setting in children and adolescents with unilateral cerebral palsy aged 8–18 years. Self-management training was added to the intervention to maximize the effect of training and to empower the participants in self-monitoring the effective use of their affected hand. Functional goals (Canadian Occupational Performance Measure), unimanual capacity (Box and Block Test), bimanual performance (ABILHAND-Kids, Children's Hand-use Experience Questionnaire (CHEQ)) and amount of use (Video Observation Aarts and Aarts – determine developmental disregard (VOAA-DDD-R)) were measured at baseline, one week and four months post intervention. Twenty children (mean age 9.5 years) participated. Repeated measures ANOVA was used to measure effects over time. Compared to baseline, there were significant improvements on all outcome measures. The largest effect sizes were found for the COPM-performance and COPM-satisfaction (Cohen's $d = 2.09$ and $d = 2.42$, respectively). The effect size was large for the ABILHAND-Kids ($d = 0.86$), moderate for the CHEQ ($d = 0.70$) and Box and Block Test ($d = 0.56$), and small for the VOAA-DDD-R ($d = 0.33$). All effects were retained at the four months post intervention assessment. The results of this study indicate that one-week (36 h) intensive CIMT-BiT combined with self-management training is a feasible and promising intervention for improving the capacity of the upper limb and its use in bimanual activities in older children and adolescents with unilateral CP.

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1. Introduction

Unilateral cerebral palsy (CP) is the most common type of CP in children (Cans, 2000). It can lead to a wide variety of problems in upper limb (UL) capacities and subsequent limitations in the performance of unimanual and bimanual activities of daily life. During the last decade, intensive activity-based interventions such as Constraint Induced Movement Therapy (CIMT) and bimanual training (BiT) have become available as UL rehabilitation approaches. In short, CIMT involves the use of restraining the unaffected UL emphasizing unimanual practice with the affected hand, whereas BiT involves UL training through bimanual tasks. Both interventions have in common that they offer high-intensity training to improve UL capacities and achieve individual goals. There is a wide variation in the intensity and duration of these UL interventions (Sakzewski, Ziviani, & Boyd, 2014). The effects of both types of intervention have been studied in several RCTs and were shown to be equally effective compared to usual care (Gordon et al., 2011; Sakzewski et al., 2011a; Sakzewski et al., 2011b; Sakzewski et al., 2014b). Yet, there are indications that CIMT is somewhat superior to BiT with regard to unimanual capacity (Sakzewski et al., 2011b) and BiT superior to CIMT with regard to both individual goal attainment (de Brito Brandao, Gordon, & Mancini, 2012) and bimanual coordination (Hung, Casertano, Hillman, & Gordon, 2011). Recently, it has been suggested that a combination of CIMT followed by BiT might offer additive value to optimize the performance level of bimanual activities (Boyd et al., 2013; Dong, Tung, Siu, & Fong, 2013; Gordon et al., 2011; Sakzewski et al., 2011b). This sequential approach was effectuated in the 'hybrid' CIMT-BiT intervention "the Pirate group" (Aarts, Jongerius, Geerdink, van Limbeek, & Geurts, 2010) presuming an optimal motor learning process by starting with unimanual training (CIMT) followed by intensive and goal-directed bimanual training (BiT). This hybrid approach has shown to improve UL capacity, bimanual performance and individual goals in young children (2.5–8 years) with unilateral CP (Aarts et al., 2010), but the effects of this CIMT-BiT combination have not yet been studied in older children.

School children and adolescents (8–18 years) with unilateral CP need more time to acquire the necessary motor skills of daily life than their typically developing peers (Vos et al., 2013). At this age, new challenges arise when the limited capacity of the affected UL hinders the acquisition of age-appropriate bimanual skills, such as working with a ruler, precise cutting with scissors, buttoning tight trousers, using cutlery for meat, using make-up, and cooking and baking. At the same time, parents of older children with unilateral CP often express their concern about the diminishing amount of use of the affected UL as their children gradually acquire greater skills to unimanually perform many daily activities with their unaffected hand (Fedrizzi, Pagliano, Andreucci, & Oleari, 2003). This discrepancy between the need to learn new age-appropriate skills and the tendency of older children to disregard the capacity of their affected UL calls for effective UL (booster) interventions to maintain and improve both the unimanual capacity and bimanual performance of age-appropriate activities.

In contrast to younger children, the school program prevails for children eight to eighteen year of age. As a consequence, high intensity interventions must be restricted to short time periods during school holidays. To accommodate the needs of this group of children with unilateral CP, we developed a day-camp intervention that combines high intensity CIMT and BiT condensed in one week (40 h) during school holidays. Until now, day-camps with only one type of intervention (CIMT or BiT) have been described for younger and older children with unilateral CP providing 30 to 90 h of training during one to three weeks. Reports were based on controlled trials studying the effects of CIMT compared to BiT (Gordon et al., 2011; Sakzewski et al., 2011b; Sakzewski, Provan, Ziviani, & Boyd, 2014) and on either controlled (Gordon, Schneider, Chinnan, & Charles, 2007) or uncontrolled trials (Green et al., 2013) studying the effects of BiT compared to no training. All day-camps were found to be effective with regard to improving UL capacity and performance, except a 30-h training program in 5 days (Sakzewski et al., 2014a). None of these day-camps offered a combination of CIMT and BiT. Because the exact critical dose of CIMT or BiT to achieve meaningful changes in either unimanual capacity or bimanual performance is still a matter of debate (Sakzewski et al., 2014b), we decided to test whether a short combined CIMT-BiT approach would be feasible and effective to improve both unilateral UL capacity and spontaneous use of the affected UL during bimanual activities in older (8–18 years) children with unilateral CP, guided by individual goal setting. Given the limited duration of the training (one week) it was deemed essential to maximize the effect of the intervention (Taub, Uswatte, Mark, & Morris, 2006; Taub et al., 2013), in terms of amount of UL use, by incorporating self-management training. Indeed, for older children and adolescents, it would be in the line of their maturation to learn to self-monitor the (supportive) use of their affected UL during daily activities and become less dependent on being prompted by their parents and/or teachers. Thus, this study aimed to determine the feasibility and preliminary effectiveness of a one-week CIMT-BiT group intervention combined with self-management training to improve UL capacity and bimanual performance in children with unilateral CP aged 8–18 years.

2. Methods

2.1. Participants and design

All children who previously participated in studies on UL training interventions in our pediatric rehabilitation center, or who had requested information about such interventions, were eligible. To be included, children had to be diagnosed with unilateral CP, aged between 8 and 18 years, and able to walk independently. In addition, they had to be able to formulate individual goals related to relevant and age-appropriate activities involving the UL, which was determined by means of a parent and child combined Canadian Occupational Performance Measure (COPM) interview. All participants attended regular education or special education for children with physical disabilities. No formal data on intellectual ability was

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