# EFFICACY OF HOME-BASED INTENSIVE BIMANUAL TRAINING FOR CHILDREN WITH UNILATERAL SPASTIC CEREBRAL PALSY

**Claudio Luis Ferre**

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ABSTRACT

EFFICACY OF HOME-BASED INTENSIVE BIMANUAL TRAINING FOR CHILDREN WITH UNILATERAL SPASTIC CEREBRAL PALSY

Claudio Luis Ferre

Neuroplasticity research suggests intervention at early developmental stages is optimal for maximizing recovery of function in children with unilateral spastic cerebral palsy (USCP). Intensive bimanual training is an effective method for improving upper-extremity function in children with USCP when provided in massed-practice day camps. Given the challenges young children face sustaining attention and their susceptibility to fatigue, adapted models using distributed practice are required. The aim of this study was to perform a randomized trial comparing home-based hand-arm bimanual intensive training (H-HABIT) with a control group receiving an intervention of equal duration, intensity, and social interaction. Twenty-four children with USCP (age range 2 yr., 6 mos. - 10 yr. 1 mos.) were randomized to participate in either 90 hours of H-HABIT (*n*=12) or an equivalent dose of functional lower-limb training (FLL-control; *n*=12). Caregivers were trained by experienced interventionists to administer either H-HABIT or FLL-control. Caregivers then performed activities with children in their own home 2 hrs./day, 5 days/week for 9 weeks (90 hrs. total). Caregivers were supervised remotely once a week for one hour using telerehabilitation. Dexterity was assessed using the Box and Blocks test. Bimanual hand function was measured using the Assisting Hand Assessment (AHA). Canadian Occupational Performance Measure (COPM) was used to assess caregiver perception (performance

and satisfaction) of functional goals. All measures were assessed immediately prior to (pretest) and immediately after (posttest) the intervention and analyzed with a 2(group) x 2(session) repeated measures ANOVA. There were no statistical differences between the two groups at baseline. Children in H-HABIT showed greater improvement (pretest=9.0±5.8; posttest=14.5±7.8) than children in FLL-control (pretest=10.6±7.2; posttest=11.9±6.1) on the Box and Blocks test (for interaction, $F(1,20)=18.53$, $p<.001$). Neither group demonstrated change on the AHA ($F(1,22)=0.89$, $p>.05$) (H-HABIT=60.5 AHA units±10.1; FLL-Control=52.8 AHA units±17.1). COPM data revealed a significant test session by group interaction ($F(1,22)=10.82$, $p<.01$) with caregivers of children in H-HABIT rating higher goal performance (pretest=2.9±1.0; posttest=6.8±1.3) relative to FLL-control (pretest=2.7±1.0; posttest=4.5±1.7). Caregivers in both groups showed equal improvement between the two sessions ($F(1,22)=115.63$, $p<.001$) in ratings of satisfaction of goal performance (pretest=3.8±1.8; posttest=7.3±1.1 for H-HABIT and pretest=2.3±1.0; posttest=4.7±1.8 for FLL-control). Children in H-HABIT made greater improvements in dexterity and parent-rated goal performance. This is the first randomized trial to examine the efficacy of intensive bimanual training with caregivers as interventionists—a model which permits intervention at younger ages when there may be greater potential for improving hand function. Using caregivers as a way to implement intensive interventions provides a cost-effective alternative to expensive clinic-based interventions. Home-based models provide a valuable intervention approach to add to the repertoire of options clinicians have to chose from when developing individualized treatment programs for children and their families.

## TABLE OF CONTENTS

CHAPTER

List of Tables ..... iii

List of Figures ..... iv

I. INTRODUCTION ..... 1

II. METHODS ..... 10

- i. Participants ..... 10
- ii. Randomization ..... 11
- iii. Study Design ..... 11
- iv. Intervention Protocols ..... 12
- v. Caregiver Training ..... 14
- vi. Participant Classification ..... 18
- vii. Primary and Secondary Outcome Measures ..... 19
- viii. Sample Size Calculation ..... 22
- ix. Statistical Analysis ..... 23

III. RESULTS ..... 23

- i. Patient Flow ..... 23
- ii. Treatment Characteristics ..... 28
- iii. Consistency Between Caregiver-and Supervisor-directed Assessments ..... 33
- iv. Dexterity ..... 33
- v. Quality of Spontaneously Using Affected Hand as an Assisting Hand ..... 34
- vi. Caregiver-rated Functional ..... 37
- vii. Predictors of improvement ..... 41

IV. DISCUSSION ..... 43

- i Home-based Interventions as a tool for Intensive Rehabilitation ..... 43
- ii Home-based Programs are Feasible, but for whom? ..... 45
- iii Feasibility of Lower-limb Training as a Control Group ..... 48
- iv Children in H-HABIT Make Unimanual but not Bimanual Improvements ..... 49

v Does Participation in an Intervention Increase Parental Satisfaction? .....52
vi Limitations and Future Considerations .....53
REFERENCES .....58
Appendix A .....66
Appendix B .....90
Appendix C .....96
Appendix D .....111
Appendix E .....121
Appendix F .....132
Appendix G .....139
Appendix H .....143
Appendix I .....146
Appendix J .....154

LIST OF TABLES

I. Participant Characteristics .....27
II. Responses to Daily Log Feasibility .....31
III. Categorization of Activities Performed During H-HABIT .....32
IV. Results of Hand Function Assessments by Testing Session.....35
V. Functional Goals Identified by Caregivers .....38

LIST OF FIGURES

I. Patient Flow .....26
II. Box and Blocks by Testing Session.....36
III. COPM-Performance by Testing Session .....39
IV. COPM-Satisfaction by Testing Session.....40
V. Relationship Between Change in AHA and .....42

# DEDICATION

Dedicado a mis viejos. Gracias Papá por enseñarme que “hay que meterle duro” y gracias Mamá por enseñarme que “no se puede esperar que se abran las puertas, hay que patearlas abiertas”.

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# I. INTRODUCTION

Brain injury during early development can affect descending motor pathways that control upper-limb movements. Individuals with unilateral spastic cerebral palsy (USCP) display motor impairments predominantly lateralized to one side of the body as a result of early brain injury (Himmelmann, Hagberg, & Uvebrant, 2010; Stanley, Blair, & Alberman, 2000). Given that most functional activities require the finely timed and coordinated use of both hands, individuals with USCP have difficulty performing activities of daily living independently (Skold, Josephsson, & Eliasson, 2004). Children with USCP in particular, require increased assistance with functional tasks (e.g., eating, dressing) and are delayed with respect to their peers in educational, social, and play activities. In addition to unilateral impairments, an underlying factor that contributes to their functional limitations is a deficit in movement planning and bimanual coordination (Gordon & Steenbergen, 2008; Hung, Charles, & Gordon, 2004; Utley, Steenbergen, & Sugden, 2004).

A recent review of the rehabilitation literature for children with cerebral palsy indicates that the most effective intervention approaches are motor learning-based paradigms delivered at high intensity with a focus on function (Novak et al., 2013). Two intensive approaches that have been successfully used to treat children with USCP are constraint-induced movement therapy (CIMT) and intensive bimanual training. CIMT involves concurrent physical restraint of the less-affected hand and structured, unilateral training of the hemiplegic hand (Taub & Wolf, 1997). CIMT, when provided at high levels of intensity, leads to improvements in amount and/or quality of unilateral upper extremity movement (Chen et al., 2014; Hoare, Wasiak, Imms, & Carey, 2007; Huang,

Fetters, Hale, & McBride, 2009; Novak et al., 2013). Hand-arm bimanual intensive therapy (HABIT) is a form of intensive bimanual training focused on improving the amount and quality of involved hand-use within the context of bimanual tasks (Charles & Gordon, 2006). Studies of HABIT in children with USCP have shown the therapy to improve the effectiveness with which children make use of their affected (assisting) hand during bimanual activities (Gordon, Schneider, Chinnan, & Charles, 2007), and their ability to coordinate the timing of the movements of each extremity (Hung, Casertano, Hillman, & Gordon, 2011; Hung et al., 2004). Studies directly comparing equivalent doses of intensive bimanual training and CIMT have demonstrated comparable efficacy between the two approaches (Facchin et al., 2011; Gordon et al., 2011; Sakzewski et al., 2011). These findings suggest that treatment intensity might be the driving factor in successful intervention paradigms.

The need for intensity creates difficulties in the delivery of therapy because of logistical and economical challenges associated with the schedules and personnel needed to run an intervention. Intensive therapies are typically delivered in settings that require one-to-one supervision by an interventionist for several hours a day (i.e., massed practice). In some cases, in order to achieve the desired intensity characteristic of functional improvements, children must be engaged in structured activities for a significant portion of the day (~6 hrs. per day for ~3 weeks). Such massed practice models might not be appropriate for younger children, are not available in many communities, and present a burdensome financial challenge. Thus, the structure of intensive interventions creates barriers between rehabilitation services and families. One way to circumvent barriers is to adapt intensive clinic-based rehabilitation to a context

that might be more feasible for a broader range of the population. This includes delivering therapy in more naturalistic settings or within the child's typical environment.

The majority of studies that have embedded therapy into a more naturalistic setting have been adaptations to CIMT delivered during the child's routine at home or at school (Al-Oraibi & Eliasson, 2011; Chen et al., 2013; Eliasson, Shaw, Berg, & Krumlinde-Sundholm, 2011; Gelkop et al., 2015; Klingels et al., 2013; Rostami & Malamiri, 2012a; Taub et al., 2011; Wallen et al., 2011). In addition to the setting in which the activities are performed, researchers have modified the dosing schedule of massed-practice models by reducing the amount of daily hours (down to ~2 hours) and distributing the intervention over a longer duration (~2 months). They have also incorporated caregivers (i.e., school teachers or parents) in the delivery of therapy activities. Children receiving CIMT in home and/or preschool settings consistently demonstrate gains in upper-limb function. Intensive bimanual approaches and functional goal training have also shown promise when provided at home or school (Ferre, Brandao, Hung, Carmel, & Gordon, 2015; Gelkop et al., 2015; Novak, Cusick, & Lannin, 2009; Novak, Cusick, & Lowe, 2007; Wallen et al., 2011).

Although having caregivers administer and intervention might seem demanding, these studies suggest that having them play an active role in the intervention and performing the activities in a home-based setting is an effective approach for eliciting improvements in hand function. There is also evidence to indicate that caregivers can participate in home-based interventions without disrupting the psychosocial dynamic that exists between them and their children. One study examining the feasibility of home-based bimanual training found stable levels of parental stress throughout the course of the

intensive intervention (Ferre et al., 2015). As opposed to creating a burden, working with their own children and identifying improvements in hand function might even serve as a motivating tool for caregivers to comply with the requirements of home-based activities (Novak et al., 2009). Caregivers also show a high rate of compliance with home-based therapy schedules when they perceive health professionals to be genuinely interested in their child's situation and when they are given information which helps them carry out home programs (Gajdosik & Campbell, 1991; Law & King, 1993; Moxley-Haegert & Serbin, 1983). Thus, training caregivers to perform therapies at home might serve as a feasible alternative to massed practice models that involve one-on-one supervision by a clinician for several hours a day. Delivering therapy in the context of the home is also consistent with family-focused interventions—interventions sensitive to the needs of families and that place the caregiver in the role of "expert" with the child (Wallen, Ziviani, Herbert, Evans, & Novak, 2008).

Despite the growing trend of performing interventions in the home setting, few studies have examined intensive bimanual training when provided exclusively at home using caregivers as the primary interventionist (Ferre et al., 2015). In order to examine the feasibility of a caregiver-directed intervention, we developed and piloted a home-based hand-arm bimanual intensive training (H-HABIT) (Ferre et al., 2015). Using a schedule in which local families were asked to perform 90 hours of bimanual activities at home over a period of 9 weeks, we examined whether caregivers would adhere to the requirements of the intervention and do so without increasing their parental stress levels. In order to supervise the activities and provide ongoing training/support to caregivers, weekly home visits were scheduled with an experienced supervisor. Caregivers

demonstrated high levels of compliance, parental stress remained stable, and children showed improvement in the quality of bimanual hand-use and functional goal performance.

We were also interested in determining caregiver interest in home-based programs from a larger subset of the population. Thus, we developed and administered a survey asking caregivers over a large geographical area to rate the importance of various functional daily activities and their willingness to be involved in a home-based intervention (Appendix F). Interestingly enough, despite caregivers rating home-based interventions as particularly challenging, an overwhelming majority (75%) indicated they would participate if the intervention were provided in a distributed practice model (i.e., 2 hrs./day, 5 days/week, 9 weeks total). Conversely, about 60% of caregivers indicated they would be unable to participate if they performed the intervention in a massed practice model (i.e., 6 hrs./day, 5 days/ week, 3 weeks total). Thus, caregivers are more likely to participate in an intensive intervention if the practice is distributed as opposed to massed. The results of the survey and our pilot study suggest there is high caregiver interest in home-based upper-limb rehabilitation and that such models are feasible. While these results are promising, the data from the feasibility study involved a single-group pre-post design. In order to test the efficacy of home-based bimanual training in a more rigorous manner, a randomized control trial is needed.

One challenge in testing H-HABIT in a randomized control trial is identifying an appropriate control group that is meaningful for participants. In many studies of interventions for children with CP, researchers use a no-treatment control or a group that receives usual and customary care (UCC). Unfortunately, no-treatment control groups are

limited given that they effectively compare intensive intervention to a group that receives nothing, thereby making it difficult to conclude whether changes in the intervention group are a direct result of the intervention or simply the increased amount of interaction between the child and interventionist. Studies using a UCC group (Eliasson, Krumlinde-sundholm, Shaw, & Wang, 2005) suffer from the limitations that it is often unclear as to the type of therapy children in this group receive, there is a great deal of variation in the amount of UCC, and that the dosage of UCC is typically so low that it is very unlikely that children in the control group would receive an equal amount of training in the course of 9 weeks that children in the intervention group receive.

To avoid the limitations of using a UCC control group, some studies have used comparison groups with children who receive an equal duration and intensity of an alternative type of intervention (Facchin et al., 2011; Gordon et al., 2011; Sakzewski et al., 2011), crossover experimental designs (Eliasson et al., 2011), or a social control group that accounts for the increased social interaction that accompanies intensive intervention (Heathcock, Lobo, & Galloway, 2008). The latter study using the social control group was conducted with 2-month old at-risk infants and involved only 10 min/day of social interaction activities between the caregiver and infants. This approach might be less appropriate for interventions involving children with USCP given the much higher daily time commitment associated with H-HABIT. Cross-over designs suffer from the limitations of “carryover” and “order” effects. Although using a washout period of sufficient duration or statistical analyses that account for order effects can reduce these limitations, there is increased burden for the family associated with the added time involvement of washout periods. One way to decrease the burden on families

participating in studies is to use comparison groups that provide an equally intensive intervention that is evidence-based and used widely throughout clinical practice.

The present study attempted to determine the efficacy of home-based hand-arm bimanual intensive training (H-HABIT) in comparison to a control group receiving an alternative therapy of equal intensity and duration that controlled for the amount of interaction between caregivers and their child. Children in the control group received intensive home-based lower-limb training. The advantage of such a comparison group is that it is an evidence-based approach that incorporates motor learning principles, controls for the amount of caregiver interaction and also provides training for lower-limb activities/function that caregivers deem as equally important as upper-limb activities/function (as determined by a caregiver survey administered at our center, Appendix F).

An effective home-based bimanual intervention would be an economically and logistically feasible alternative to a day-camp intervention or massed-practice interventions that require constant one-to-one supervision. A home-based distributed-practice intervention provides the flexibility to suit the developmental constraints of children. Thus, an adapted version of HABIT would permit intervention at younger ages when the developing nervous system exhibits considerable plasticity. It would also increase access to a greater range of the population.

Increasing the range of families involved in an intervention however creates a challenge in terms of supervision, as the families are spread out over a large geographical area. Novak et al. (2007) found that one of the primary factors involved in a family's decision to participate in a home program was the increased guidance provided by an

experienced supervisor. Families found the supervisor's direction reassuring and led to increased confidence in their interactions with their children. In-home visits have been demonstrated to increase parental compliance with a home program (Mayo, 1981). In our study examining the feasibility of home-based bimanual training with local families, a supervisor made weekly home visits to provide feedback, lend support, and model activities for caregivers (Ferre et al., 2015). Compliance was high with caregivers completing on average 97% of the required hours of intervention. During their exit interviews, caregivers expressed that the weekly home visit was an important component of the intervention for the added motivation it provided to children and for the encouragement caregivers received. These finding might suggest that participation in and compliance with home-based interventions is limited to families near clinics that might provide such supervision.

One alternative for providing ongoing supervision when home visits are not possible is by monitoring caregivers remotely via telerehabilitation. Telerehabilitation involves the use of communication and information technologies as a means of decreasing the logistical gaps between vulnerable populations and clinicians (Kairy, Lehoux, Vincent, & Visintin, 2009). Telerehabilitation techniques could potentially offer a time- and cost-saving technique to logistical challenges (Cooper et al., 2001; Torsney, 2003; Wakeford, Wittman, White, & Schmeler, 2005). Although the majority of studies using telerehabilitation come from adult clinical populations, the technique has also been used successfully with children. Cason (2009) successfully piloted an early intervention program with a set of rural families over a 12-week period in which an occupational therapist remotely directed a caregiver and facilitated the caregiver's interaction with the

child. Caregivers expressed a high level of satisfaction with the program as a result of the increased intensity of services afforded by the technique. Sicotte, Lehoux, Fortier-Blanc, and Leblanc (2003) demonstrated the feasibility of videoconferencing for delivering speech therapy to children with stuttering problems along with improvements in fluency. Telerehabilitation has even been used to coach parents to provide intervention to their children diagnosed with autism spectrum disorder (Baharav & Reiser, 2010). Finally, internet-based modules are also being examined in which children with CP perform upper-limb tasks and games while feedback about performance is sent to a remote supervisor (Boyd et al., 2013). It should be noted however, that this approach and other remote gaming interventions can lack the direct contact with supervisors rated so highly by caregivers. Despite the paucity of large, well-designed studies using telerehabilitation, the delivery model appears to offer increased accessibility of services. Whether or not the technique can be used to provide direct, face-to-face supervision to caregivers administering an intensive home-based bimanual intervention to children with USCP is yet to be determined. By using a telerehabilitation service delivery model, this study will include children from a broad geographical range.

The specific aim of this study is to determine **if children receiving H-HABIT make greater improvements in upper extremity function in comparison to a control group receiving a lower-limb training of equal intensity and duration.** We hypothesized that children in the H-HABIT group will demonstrate improvements in the quality of bimanual hand-use, functional goal performance, and dexterity in comparison to the children in the control group. We also hypothesized that there will be a greater number of individuals who demonstrate clinically meaningful changes in the quality of

bimanual hand-use and functional goal performance in the H-HABIT group in comparison to the lower-limb training group.

## II. METHODS

### i. Participants

Participant recruitment occurred in two phases. The initial recruitment phase involved families attending a Children's Hemiplegia and Stroke Association (CHASA) retreat. Families planning to attend the retreat were provided with a study announcement that included the study requirements and inclusion/exclusion criteria. The remaining families were recruited from clinics in the New York City area, our research center's website (http://www.tc.edu/centers/cit/), and online support groups (i.e., CHASA). In order to ensure that children qualified for the study, potential participants were screened via e-mail and/or telephone. Children between the ages of 2.5 and 12.5 years with unilateral spastic cerebral palsy were invited to complete a screening. Inclusion criteria included: 1) ability to grasp and release with affected hand, 2) ability to follow two-step instructions and complete testing, and 3) ability of caregiver to provide one-to-one attention to the child during the daily activities. The primary investigator confirmed the child's impairments using a recorded video screening. If caregivers were local, they were given the option to come in for an in-person screening. Parents were asked to send a short video clip of the child 1) taking the cap off a marker, 2) grasping a small object (e.g., cereal), and 3) grasping a large object (e.g. stuffed animal) with the whole hand. Participants included children with hemiplegia between the ages of 2.5 – 12.5 years. Exclusion criteria included: 1) any associated cognitive delays that would prevent

children from following one-step instructions 2) any health diagnoses which are not associated with cerebral palsy, 3) visual problems that would prevent them from carrying out the intervention or testing tasks, 4) the inability to grasp or make extensive use of the more affected upper extremity as a stabilizer (determined by screening videos), and 5) caregivers who were not willing or unable to commit to the entire duration of the intervention. The project supervisor verified the child met the criteria following observation of the screening videos and phone interviews with parents. Caregivers responsible for the attention or care of more than one child were required to find childcare for their other children in order to ensure one-to-one attention with the child with hemiplegia during the two daily hours of the intervention.

**ii. Randomization**

Children were matched into pairs based on age and randomized using concealed allocation into either the H-HABIT group or lower-limb intensive functional training group (LIFT-control). A computer generated (Microsoft Excel, Version 2007) set of random numbers was used to create an allocation sequence. An individual without any direct participation in the clinical aspects of the study performed the randomization, random number generation, and preparation of allocation materials off-site.

**iii. Study Design**

To examine changes in hand function resulting from the intervention, children were assessed at 2 different time points: once prior to the intervention (pretest) and immediately after the intervention (posttest). A separate assessment was also performed during caregiver training and before the intervention to examine reliability of caregiver-administered assessments (see Outcome Measures). Caregivers were trained to perform

H-HABIT prior to the first assessment. Upon receiving training, caregivers were required to engage their children in either H-HABIT or LIFT-control activities for 2 hrs./day any 5 days/week for a total of 9 weeks. The two daily hours did not need to be consecutive—although caregivers were asked to structure the activities for at least 30-minute sessions. An extra week was allotted at the end of the 9 weeks for children who became ill (e.g., caught cold) and had to miss time.

#### iv. Intervention Protocols

For the H-HABIT group, caregivers engaged participants in bimanual tasks. The aims of the bimanual tasks were to improve reaching, grasp, releasing, in-hand manipulation, and movements related to active wrist extension, forearm supination, gross movements of the shoulder and elbow, individual finger movements, and using the affected hand as an assisting hand. Children performed the activities in the context of games and child-friendly play. H-HABIT is based on 1) research delineating the mechanisms underlying hand impairments in children with cerebral palsy, 2) intensive and structured practice, and 3) principles of motor learning and neuroplasticity (Gordon & Friel, 2009).

During H-HABIT, children are engaged in bimanual activities for a period of 90 hours. Rather than encouraging compensatory use of the involved hand, children are encouraged to use the hand as a typically developing child uses their non-dominant hand (i.e., as an assist). The intervention involves specific bimanual activities and shaping of the environment to promote bimanual hand-use. An important feature of H-HABIT is grading task demands in order to ensure children can successfully perform the tasks. The child's caregiver provides positive reinforcement in order to motivate performance and

reinforce specific movements. In order to maintain the child's motivation, the focus of the child is to complete a game or play with a specific toy whereas the focus of the caregiver structuring the activity is to create an enjoyable situation involving sufficient difficulty to challenge and numerous opportunities for repetition (Eliasson et al., 2005).

For the FLL group, caregivers engaged children in functional lower-limb tasks. The aims of the lower-limb tasks were to improve balance, strength, and coordination with an emphasis on the involved leg. Activities were embedded in the context of games and child-friendly play or performed during functional tasks (e.g., walking up an inclined surface). For example, lower-limb activities included ball kicking, jumping through squares (hopscotch), or walking through fun obstacle courses with a series of lower-limb challenges. Similar to H-HABIT, LIFT-control is based on principles of motor learning, caregivers were required to complete 90 hours of activities, supervisors ensured that activities were functional, activities were graded to child's abilities, and caregivers were encouraged to focus on making the activities child-friendly and fun.

In addition, caregivers in each group were asked to perform activities that *only incorporated the use of either the upper- or lower-limbs*. That is, caregiver in the H-HABIT group only performed activities that involved the use of the hands/arms and caregivers in the LIFT-control group only performed activities that involved the use of the legs. In order to ensure these criteria were met, supervisors asked caregivers in the H-HABIT group to perform tasks while at a table or while sitting, whereas caregivers in the LIFT-control group were asked to avoid doing tasks that involved the use of the hands. Caregivers in both groups were instructed to avoid performing activities that involved the

whole body (i.e., riding a bike) during treatment time. Supervisors monitored daily logs in order to ensure there was no treatment overlap.

Caregivers were required to perform either H-HABIT or LIFT-control for two hours a day and 5 times a week (total of 10 hours). Although the two hours did not need to be performed continuously, the activities were performed in blocks of no less than 30 minutes. Based on our previous camp-based models, we suspect that some of the success of an intervention is based on the one-to-one ratio of child to interventionist. Therefore, the caregiver performing the activities with the child was asked to not be responsible for attending to another child during the two daily hours. If the child receiving the training had siblings, parents were instructed to arrange for another caregiver to attend to the siblings during the daily two hours. Prior to the onset of the intervention and during caregiver training, a list of potential items/games to be used during the intervention was discussed with the caregiver. Efforts were made to incorporate toys, games, or activities already owned by the caregivers. In addition, as part of the goal of the intervention is to improve functional independence, parents were encouraged to work on functional activities. For example, this included putting on shoes, putting on pants with both hands, or using both hands during mealtime.

**v. Caregiver Training**

Caregivers were trained over three sessions (see below for details). For families recruited from the CHASA retreat, caregivers were trained in a hotel meeting space. A primary supervisor with experience administering intensive bimanual interventions (both camp- and home-based) and a senior occupational therapist (OT) with experience administering HABIT trained caregivers in the H-HABIT group. A physical therapist

(PT) with experience administering intensive lower-limb interventions and a separate, senior PT that designed the lower-limb program trained caregivers in the LIFT-control group. A third PT assisted with training caregivers and modeling lower-limb activities. For the families recruited from the CHASA retreat, caregivers were trained in large groups (*n*=9 in each group) for the first training session. Caregivers in both the H-HABIT and LIFT-control groups were trained in groups of 2-3 caregiver-child dyads for the second session.

The primary supervisor for H-HABIT and the primary PT for LIFT-control trained subsequent cohorts. These families were trained at our center at Teachers College, Columbia University. The senior OT assisting with the H-HABIT group and the senior PT assisting with the LIFT-control group continued to advise the primary supervisor in their respective groups. Caregivers from these cohorts were trained individually (*n*=3 in H-HABIT; *n*=3 in LIFT-control), in pairs (*n*=4 in H-HABIT group; *n*=4 LIFT-control) or groups of three (*n*=0 in H-HABIT; *n*=6 in Lift-Control) for the first session. Participants who were randomized but did not attend training accounted for asymmetric training group sizes across the two study groups (see Patient Flow).

Caregivers received 3 training sessions before the treatment began (5 hours total). The first training occurred without the child present and lasted 2 hours. Caregivers were provided with a reference manual of potential activities, strategies, and games that could be used for the intervention activities. An experienced interventionist discussed the requirements and demands of the intervention. We were also concerned about potentially disrupting the psychosocial dynamic between caregivers and children. In order to avoid disruptions, caregivers were encouraged to use a subtle cue to the child (e.g., H-HABIT

hats or scarves) to indicate that the daily 2 hours of training were separate from the child's typical routine. Caregivers were also encouraged to use a separate space in the home and activities/toys that were only used during the daily 2 hours. This created a context for therapy in the home environment that was discrete from the child and caregiver's typical routine. Caregivers were also encouraged to avoid using constant prodding and requests to perform certain behaviors of interest outside of the daily therapy schedule. Safety procedures and child-friendly strategies were also outlined. For example, caregivers were told to keep the area in which they provided the treatment clutter-free (i.e., not too many objects or toys provided at once).

Then, the experienced interventionist demonstrated and provided instruction about specific activities that could be incorporated as part of the intervention. For example, for H-HABIT, the experienced interventionist demonstrated how building with blocks could be performed as a bimanual activity by using the affected hand to pick up the blocks, transferring it to the unaffected hand, then stacking the blocks one by one. We also provided specific examples of how to grade task difficulty to match the ability of the child. For example, if a child demonstrated difficulty grasping the blocks because they slid around on the surface of a table when a grasp is attempted, caregivers were encouraged to place the blocks on a textured material that increased the stability of the object and facilitated grasping. Caregivers were instructed to increase the difficulty of the task (i.e., removing the cloth) as the child's abilities improved. They were also instructed to keep the task within the limits of the child's abilities in order to decrease the possibility of frustration or negative effects associated with failing to complete a task. In order to provide examples, caregivers were shown video of other caregivers performing H-

HABIT. Then, caregivers were guided through on how to keep track of the child's activities in an online daily log (Appendix G). Finally, caregivers were encouraged to ask questions or voice any concerns they had about performing the intervention.

After completing the first training session, the experienced interventionist administered a second training session that lasted another 2 hours. Caregivers were asked to perform the activities with their child while the experienced interventionist supervised. Caregivers were asked to engage their children in a range of bimanual activities discussed at the previous training session. Immediate feedback about performance was provided, and the experienced interventionist also modeled how to direct H-HABIT activities with the child. Each child received supervision from the same individual.

The third training session was overlapped with the first hour of the 90-hr intervention. Participants were monitored via webcam-based software (i.e., Adobe Connect) while they performed the activities in their own home for an hour. The purpose of the remote supervision was to ensure that caregivers administered the intervention correctly and to provide feedback to caregivers about their performance. Hourly supervision continued on a weekly basis for the entire duration of the intervention (i.e., 1 hr./week for 9 weeks). Caregiver activities were also monitored daily by the supervisor who checked activity logs submitted online (Appendix F). The logs provided the supervisor with information about whether the families completed the required 2 hr./day and whether or not caregivers had any questions about the activities. Each time caregivers submitted a log online, the project supervisor was notified via email with detailed information about the daily activities. Prior to commencement of the intervention, caregivers developed a schedule in cooperation with the supervisor specifying the days

they would be performing the activities. If caregivers failed to complete the log for more than 2 consecutive days or on the days that were specified on the schedule, the supervisor contacted the caregiver to ensure that the activities were being completed. For participants who did not have access to a computer for the weekly supervised sessions or daily logs, alternative arrangements were made to videotape portions of the intervention and to make daily reports of the activities by phone.

Separate social media groups were created for parents in the H-HABIT and LIFT-control group. The groups were private and only caregivers who agreed to be a part of the group were added. The purpose of the online groups was to provide a forum for caregivers to exchange ideas, provide encouragement to one another, and to share photos/videos of creative activities and accomplishments as motivation. Supervisors were also members of these groups, and would sometimes provide ideas or words of encouragement or praise.

#### vi. Participant Classification

The Manual Abilities Classification System (MACS) classifies the child's ability to handle objects in daily activities on one of 5 levels (Eliasson et al., 2006). The MACS has strong construct validity and high inter-rater reliability, intraclass correlation coefficient=0.97 (Eliasson et al., 2006). The Gross Motor Function Classification System (GMFCS) classifies the child's ability to carry out self-initiated movements related to sitting and walking across 5 levels (Palisano et al., 1997). The GMFCS has established construct validity (Palisano et al., 2000) and high inter-observer reliability (Morris, Galuppi, & Rosenbaum, 2004).

### vii. Primary and Secondary Outcome Measures

Two standardized clinical tests of unimanual and bimanual hand performance were specified as primary outcome measures: 1) Box and Blocks Test (BBT), and 2) Assisting Hand Assessment (AHA). The secondary measure included the Canadian Occupational Performance Measure (COPM)—a standardized caregiver perception questionnaire about functional activities. The primary and secondary measures are measures of activity according to the domains of the International Classification System (WHO, 2001).

The Box and Block test assesses manual dexterity by counting the number of blocks that are transferred with a single hand from one compartment to another within 60 seconds. The test assesses coordination of grasp, hold, and release of a small objects; is easy to explain; and can readily be performed by many children. Normative scores for children between the ages of 3-10 years were reported (Jongbloed-Pereboom, Nijhuis-van der Sanden, & Steenbergen, 2013). The test takes only a minute to perform for each hand and has a high reliability (.85 and .99 intraclass correlation coefficients for test–retest and interrater reliability measures, respectively). Clinically meaningful differences for the Box and Blocks test have not been established for young children.

The AHA is a test designed for children with congenital hemiplegia and measures how effectively they use their affected hand during bimanual activities. It has excellent validity and reliability (interrater = .97, intrarrater = .99) (Holmefur, Krumlinde-Sundholm, & Eliasson, 2007; Krumlinde-Sundholm, Holmefur, Kottorp, & Eliasson, 2007). The AHA is scored from video recordings of 12-14 play activities. These activities are subsequently scored based on 22 predefined items using a 4-point rating

scale. The raw score sum ranges from 22 (low ability) to 88 (high ability). Rasch analysis provides measures of equal intervals in logits (log odds probability units) by converting ordinal rating scale observations to interval level data through a logarithmic transformation based on probabilities. Interval level measures in logits for the AHA range from -10.18 to 8.70 logits. In order to facilitate interpretation of results, the logit scale is converted to a user-friendly 0 to 100 AHA-unit scale that also presents the data on an interval level (Krumlinde-Sundholm, 2012). The smallest detectable difference on the AHA is a change of 4 AHA-units.

Caregivers were trained to perform and record a semi-structured play session in their home with guidance by a person certified in use of the AHA. The children were seated in an upright posture at a table that permitted sufficient movement of the upper limbs. The AHA is conducted in a 15-20 minute semi-structured play session, which is video recorded in a standardized manner. Children were asked to play with a range of selected toys that afforded bimanual activities and are challenging enough to engage them to act and elicit a variety of actions. For example, children were required to manipulate a music box that plays music when the lid opens (a lid that can only be opened with both hands). The caregiver sat across from the child while interacting playfully and encouraging the child to play with the toys. The project supervisor supervised the play session remotely. Scoring of the assessment was done from the video recording. A blind, certified observer scored the videos according to a set of predetermined, specific criteria.

An experienced tester conducted an initial baseline test during the training period. In order to avoid caregivers having to return to our center for follow-up testing, they were provided with the supplies and trained to administer the Box and Blocks Test and the

AHA in their own home. Caregivers received written step-by-step instructions on how to administer the tests and administered the tests immediately when they returned home from training (i.e., prior to beginning of intervention) in order to determine reliability with the baseline assessment performed by the experienced tester. The caregiver-administered tests were performed while an experienced tester supervised remotely. Caregivers were asked to wear headphones so that the experienced tester could direct them and so that children would not be influenced by overheard comments. The AHA sessions were video recorded and scored by the blind observer.

COPM was used to quantify caregiver's perception of performance on functional goals that they considered relevant to their child. Caregivers were asked to identify functional activities on which they wanted their child to improve. Then, they were asked to rate the importance of each activity on a 1-10 scale. Afterwards, parents were asked to rate child's performance and their satisfaction level, using the 1-10 scale, for the five most important goals. Functional goals typically include self-care and grooming items (i.e., dressing, tying shoes, holding bottle/cup with both hands, eating) or items that relate to other everyday activities (i.e., opening a jar with both hands, carrying an object with both hands) The COPM is a valid and reliable measure (Verkerk, Wolf, Louwers, Meester-Delver, & Nollet, 2006) for tracking flexible and consistent outcomes that are relevant to children and their families (Cusick, McIntyre, Novak, Lannin, & Lowe, 2006; Randall & McEwen, 2000). A change of 2 has been established as a clinically meaningful difference for the COPM.

### viii. Sample Size Calculation

Sample size calculations were based on estimates obtained from the first cohort recruited for this study. Many standard methods of estimating sample size are based on generic formulas that generate data from the relationship between effect size and power. Study designs that involve repeated measures however, exhibit patterns of dependencies across the measurements that complicate the estimation of sample sizes (Overall & Doyle, 1994). Fortunately, programs exist to account for the interdependency of treatment effects, patterns of correlation among the repeated measurements, and parameters commonly used in sample-sized estimation (i.e., alpha, beta). Documentation for the formulas used in such software is available (Faul, Erdfelder, Lang, & Buchner, 2007). Two approaches to estimating sample size were used for comparison purposes. Given the similar outcome across the two approaches, only one method is presented here and the other is presented in Appendix H.

The first approach was performed using G*Power (Version 3.1.7) to estimate the required sample size needed for an interaction. G*Power defines the effect size as the standard deviation of the effect being test divided by the common standard deviation within each of the groups. For the Box and Blocks Test, using an effect size $f$ =.27, identified by Cohen (2013) as a medium effect, $\alpha$ = .05, 1-$\beta$ =.8, number of groups = 2, number of measurements = 3 and a correlation among the repeated measurements = .9, a total number of 16 participants was required.

For the COPM, using an effect size $f$ =.19, $\alpha$ = .05, 1-$\beta$ =.8, number of groups = 2, number of measurements = 3 and a correlation among the repeated measurements = .9, a total number of 24 participants were required.

In order to be as conservative as possible, we used the COPM to identify our sample size requirement, as this required the highest number of participants. Based on the attrition rates from our feasibility study and our pilot cohort, we anticipated an attrition rate of 30% for this study. Thus, we attempt to recruit at least 15 children for each group to reach a total number of 12. Analyses were conducted once we recruited 12 children for each group.

### ix. Statistical Analysis

Analysis was based on intent-to-treat principles as specified by CONSORT guidelines (Moher, Schulz, & Altman, 2001). A 2 (group) x 2 (test session) ANOVA with repeated measures on test sessions were performed on AHA-units for the AHA, Box and Blocks data, and on raw and log-transformed COPM data using R. Separate paired t-tests and intraclass correlation were used to examine the consistency between caregiver- and supervisor-administered assessments.

## III. RESULTS

### i. Patient Flow

Participants were recruited in two separate phases: families recruited from the CHASA retreat in July 2013 and families recruited after the retreat (Post-CHASA), between January 2014 and January 2015. Collectively 150 individuals were screened across the two cohorts. Ultimately, 44 qualified individuals agreed to participate and were randomized into the H-HABIT or LIFT-control group. A total of 24 participants (12 in each group) completed the intervention. Patient flow for the two recruitment phases combined is shown in the CONSORT flow diagram (Figure 1). One child in each group could not complete the Box and Blocks test because of behavior problems (i.e., young

children who became frustrated and did not want to perform the task). Data for AHA and COPM were collected for all 24 participants at each time point. Table 1 describes participant characteristics. There were no baseline differences between the two groups.

As mentioned above, the total number of participants recruited was the result of two separate cohorts. In cohort 1, a total of 22 CHASA families were assessed for eligibility. All CHASA families except for one were screened remotely (i.e., screening videos). One family attending the retreat decided they wanted to participate in the study and were screened on-site using the same tasks. Eighteen families that met the inclusion criteria were randomized equally across the two groups. Four families (*n*=2 in each group) self-selected out after randomization and receiving training but prior to the pretest. An additional 5 families dropped out after home training began (*n*=2 in H-HABIT; *n*=3 in LIFT-Control). That is, for families who began the intervention, there was a 36% dropout rate. A total of 9 families (*n*=5 in H-HABIT; *n*=4 in LIFT-Control) from the CHASA retreat out of the 18 randomized completed the study.

Given the high percentage of dropouts, an additional screening tool was developed for participants screened Post-CHASA (Appendix I). The screening tool was created to assess whether caregivers could feasibly perform the activities according to the required schedule. Caregivers were provided with a template of a weekly schedule and were asked to list activities that were part of their typical routine (i.e., school hours, extracurricular activities, other therapy). They were then asked to specify the specific days and time they would complete the required hours of home-based activities within their typical schedules. Caregivers were also asked to indicate contingency plans in case

the person assisting with childcare for siblings became unavailable (in order to keep 1:1 caregiver-child ratio during daily activities).

In cohort 2, a total of 128 post-CHASA families were assessed for eligibility. A greater number of families from Cohort 2 were assessed for eligibility compared to Cohort 1 given the larger pool of families to screen from. All of the post-CHASA (Cohort 2) families aside from one local family were screened remotely. One local family preferred to be screened at our center. A total of 34 families met the inclusion criteria and were asked to complete the additional screening tool. Eight of the families that completed the screening tool decided they were unable to participate because the schedule was too demanding. That is, 8 of the families self-selected out upon completing the form. A total of 26 post-CHASA families were randomized evenly across the two groups. Five families randomized to the H-HABIT group did not attend the training session (none of the families provided a reason) and one family withdrew from the study after randomization and training but prior to the pretest. Two families from the LIFT-control group withdrew before the intervention began. Three families in the LIFT-control group withdrew from the study once the intervention phase began. The majority indicated the schedule was too demanding. For families who began the intervention, in the post-CHASA phase of recruitment, there was a 15% dropout rate. A total of 15 post-CHASA families ($n$=7 in H-HABIT; $n$=8 in LIFT-control) completed the study.

Figure 1. Patient Flow

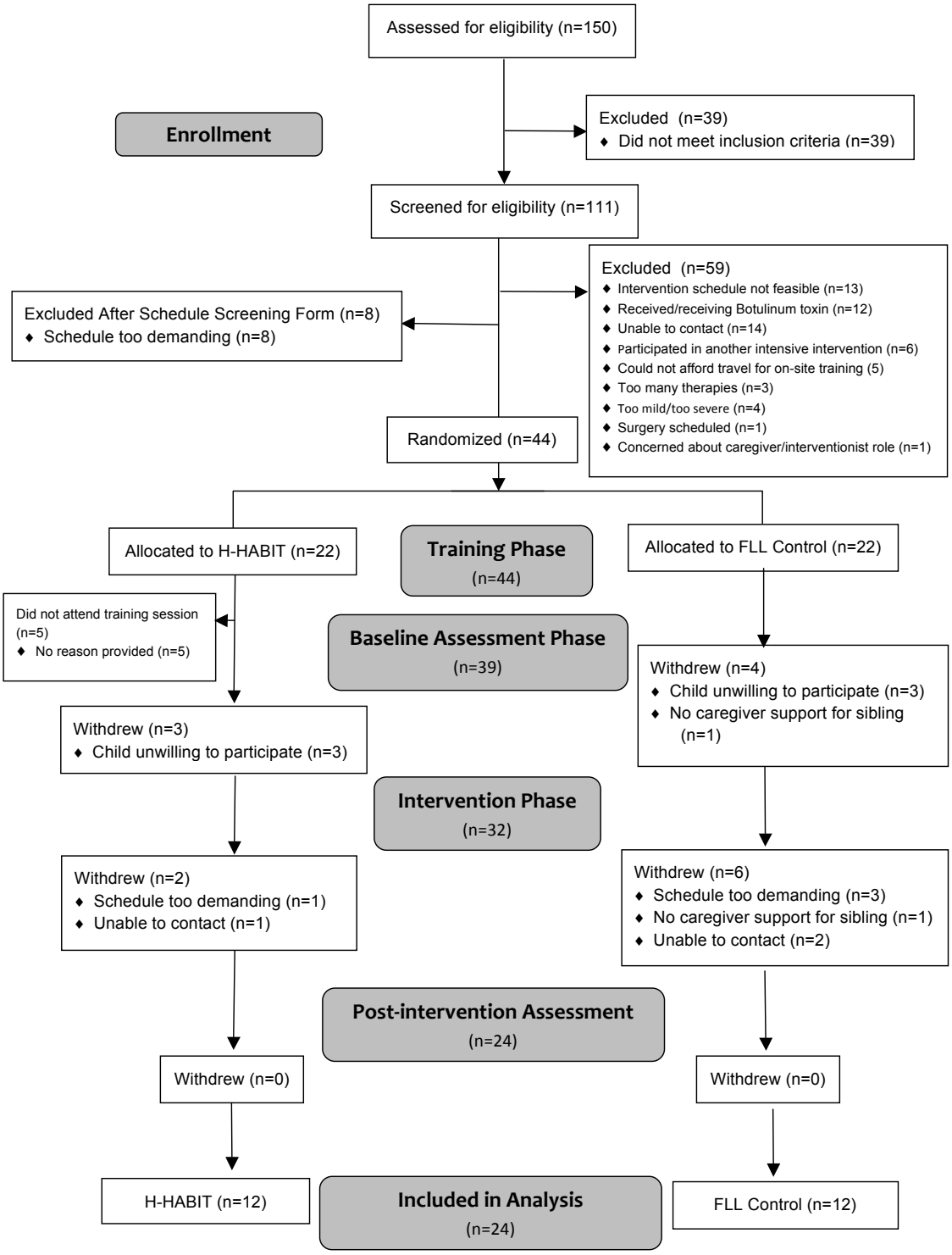


**Table 1.** Participant Characteristics

| Characteristics | | H-HABIT (n=12) | LIFT-Control (n=12) |
|---|---|---|---|
| **Mean Age in Months (SD)** | | 61.7 (31.5) | 69.6 (27.9) |
| **Gender** | | | |
| | Male | 5 | 5 |
| | Female | 7 | 7 |
| **Paretic UE** | | | |
| | Right | 7 | 8 |
| | Left | 5 | 4 |
| **MACS** | | | |
| | I | 2 | 1 |
| | II | 7 | 8 |
| | III | 3 | 3 |
| **GMFCS** | | | |
| | **I** | 2 | 3 |
| | **II** | 10 | 9 |
| **Baseline AHA (AHA Units)** | | 59.8 (11.7) | 50.7 (17.9) |
| **Baseline Box and Blocks Test** | | 9 (5.8) | 10.2 (7.5) |

Abbreviations: SD, Standard deviation; MACS, Manual Ability Classification System, GMFCS, Gross Motor Function Classification System

### ii. Treatment Characteristics

Two caregivers in the LIFT-control group did not submit logs and were for the purposes of calculating practice averages identified as missing data. The supervisor for the LIFT-Control group made frequent attempts when monitoring daily logs to have the caregivers to document their activities. These two caregivers verbally confirmed that they did do the required home training schedule, but did not have the time to fill out daily logs. Weekly supervision meetings continued throughout the 9 weeks for these two families. Participants in the H-HABIT and LIFT-control groups completed on average 82.9 hours (sd=12.7) and 76.7 hours (sd=7.29) of home training, respectively. Caregivers who did not reach the required hours indicated they were unable to finish all 90 hours even with the extra week allotted for illness/missed training days. There were no statistical differences between the two groups for total amount of home training completed ($p<.05$). Remote supervision of the activities by supervisors and monitoring of daily logs confirmed that caregivers adhered to the treatment protocols. No adverse events were reported. For caregivers who completed the study and reported their activities, daily logs generally indicated that they found the intervention feasible, although caregivers in LIFT-Control consistently indicated lower ratings of feasibility compared to H-HABIT group. According to caregivers in the H-HABIT group, 61% of the daily logs indicated it was very easy or easy to carry out the day's session, 78% indicated the child tolerated the session either very well or well, 73% indicated the child was very attentive or attentive, and 55% indicated it was very easy or easy to incorporate the activities into their schedule (Table 2). According to caregivers in the LIFT-Control

group, 48% of the daily logs indicated it was very easy or easy to carry out the day's session, 60% indicated the child tolerated the session either very well or well, 52% indicated the child was very attentive or attentive, and 43% indicated it was very easy or easy to incorporate the activities into their schedule. Participants continued their usual and customary care during the 9 weeks.

The majority of activities performed by the H-HABIT group included common, low cost supplies/toys appropriate for this age group (Table 3). The most common type of activity performed included manipulative games/tasks (38% of all logged activities) and functional tasks (16% of all logged activities). On average, families performed 7 activities per day that lasted about 19 minutes per activity.

During the course of the intervention, families were active on their respective social media groups. Ten out of 12 families participated in the upper-limb social media group whereas 9 out of 12 families participated in the lower-limb social media group. Reasons for not participating the in the social media groups included: did not have a social media account (*n*=1 in H-HABIT group, *n*=1 in LIFT-control group), and some families preferred not to participate but did not specify a reason (*n*=1 in H-HABIT group, *n*=2 in Lift-Control group). In general, families used the groups to post pictures or videos of achievements and ideas for activities. For example, one caregiver in the LIFT-Control group created an obstacle course for her child and posted a brief video of her child crossing a "finish line" tape. Another caregiver used the group to ask other caregivers who had been trained in the same cohort if they would like to receive trophies for their children as they finished the intervention. Caregivers also posted questions about suggestions for activities. Examples of caregiver comments and posts are demonstrated in

Appendix J. Supervisors in each group also posted motivational comments in order to keep families engaged. Caregivers who completed the intervention remained active with their posts and continued encouraging caregivers in subsequent cohorts.

Table 2. Responses to Daily Log Feasibility

| | **How Easy was it to carry out the session?** | | **How well did your child tolerate the session?** | | **How attentive was your child during the session?** | | **How easy was it to fit today's training session into your schedule?** | |
|---|---|---|---|---|---|---|---|---|
| | H-HABIT | LIFT-Control | H-HABIT | LIFT-Control | H-HABIT | LIFT-Control | H-HABIT | LIFT-Control |
| Very difficult | 0% | 1% | - | - | - | - | 5% | 4% |
| Difficult | 6% | 20% | - | - | - | - | 11% | 23% |
| Neutral | 33% | 31% | - | - | - | - | 29% | 31% |
| Easy | 51% | 40% | - | - | - | - | 47% | 32% |
| Very easy | 10% | 8% | - | - | - | - | 8% | 11% |
| | | | | | | | | |
| Disliked very much | - | - | 0% | 1% | - | - | - | - |
| Disliked | - | - | 3% | 12% | - | - | - | - |
| Neutral | - | - | 19% | 27% | - | - | - | - |
| Tolerated well | - | - | 54% | 44% | - | - | - | - |
| Tolerated very well | - | - | 24% | 16% | - | - | - | - |
| | | | | | | | | |
| Very distracted | - | - | - | - | 1% | 1% | - | - |
| Distracted | - | - | - | - | 7% | 18% | - | - |
| Neutral | - | - | - | - | 20% | 28% | - | - |
| Attentive | - | - | - | - | 52% | 39% | - | - |
| Very attentive | - | - | - | - | 21% | 13% | - | - |

**Table 3.** Categorization of Activities Performed During H-HABIT

| Activity Category | Percent of Total Activities | Activity Examples | Example of Affected Upper Extremity Use |
|---|---|---|---|
| Manipulative games/tasks | 38% | Molding clay, puzzles, board games, Legos, stacking cups | Stabilize construction piece while less-affected hand connects other pieces |
| Functional tasks | 16% | Dressing, undressing, cutting with scissors, eating | Hold and rotate paper during cutting with scissors |
| Miscellaneous | 10% | Book/page turning, games in bath tub, pretend play with figurines, bubbles | Supination of forearm while pouring water into cups during bath time |
| Fine Motor | 11% | Beading, stickers, magnets, coin bank, marbles | Placing beads on pipe cleaner |
| Gross motor | 6% | Two-handed ball play, scooter handles, putt-putt golf, baseball | Throw and catch large ball with both hands |
| Arts and Crafts | 9% | Painting, drawing, holiday-themed crafts | Stabilize/rotate paper while tracing shapes |
| Card Games | 8% | Card flipping during matching | Simultaneously flip two cards |
| Video games | 1% | Tablet, computer | Holding/rotating tablet less-affected swipes on screen |

### iii. Consistency Between Caregiver- and Supervisor-administered Assessments

There were no baseline differences between the two groups on any of the measures. Paired sample t-tests were used to analyze any potential differences between the assessments performed by the supervisor during training at our center and the pre-intervention assessments performed by the caregivers at home. The average AHA scores were 56.2 (sd=15.3) and 56.2 (sd=15.2) for the supervisor-administered baseline and caregiver-administered pretest respectively. The average Box and Blocks score were 9.9 (sd=5.6) and 9.8 (sd=6.3) for the supervisor-administered baseline and caregiver-administered pretest, respectively. For both the AHA and Box and Blocks test there was no significant difference ($p>.05$). The data for the two assessments were highly correlated with an ICC(2)=0.99 (n=24, $p<.001$) for the AHA and ICC(2)=0.92 (n=22, $p<.001$) for the Box and Blocks test.

### iv. Dexterity

Table 4 shows the means for all measures across each time point and the eta-squared ($\eta^2$) for the testing session and interaction effects. Shapiro-Wilk normality tests indicated the data were normally distributed. Repeated measures ANOVA revealed a main effect for the Box and Blocks $F(1,20)=32.19$, $p<.001$ (session). There was also a significant group x session interaction with children in the H-HABIT group showing greater improvement, $F(1,20)=18.53$, $p<.001$, (Figure 3). Separate post-hoc comparisons using the Holm adjustment method revealed no significant differences across the two sessions for both the H-HABIT group ($p>.05$) and LIFT-control group ($p>.05$) for the Box and Blocks.

### v. Quality of Spontaneously Using Affected Hand as an Assisting Hand

The repeated measures ANOVA revealed no main effects or interaction for the AHA, $F(1,22)=2.31$, $p>.05$ (group), $F(1,22)=1.19$, $p>.05$ (session), $F(1,22)=1.12$, $p>.05$ (group x session interaction). Two children in the H-HABIT group and none in the LIFT-control demonstrated a clinically meaningful improvement (i.e., change>4).

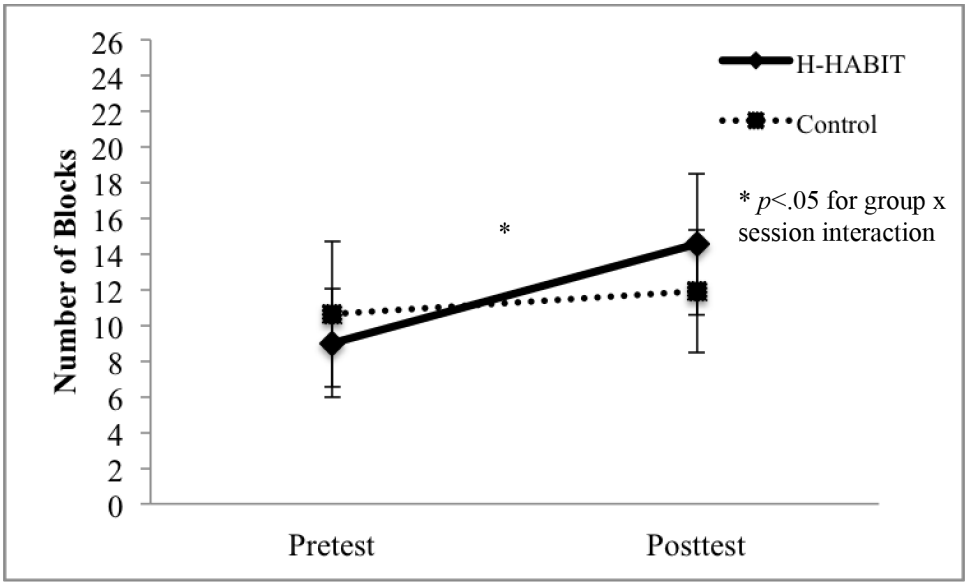
**Table 4.** Results of Hand Function Assessments by Testing Session

| | ***Baseline**** ***(95% CI)*** | **Pretest**** ***(95% CI)*** | **Posttest**** ***(95% CI)*** | **Test Session Effect *P* Value ($\eta^2$)** | **Interaction *P* Value ($\eta^2$)** |
|---|---|---|---|---|---|
| **Box and Blocks (no. of blocks)** | | | | | |
| HABIT | *8.3* (5.4, 12.7) | 9.0 (5.9, 13.3) | 14.5 (8.8, 18.3) | - | - |
| LIFT-Control | *11.8* (6.8, 16.2) | 10.6 (5.8, 15.5) | 11.9 (17.2, 15.9) | - | - |
| **Mean** | ***9.9*** ***(8.8, 14.8)*** | **9.8** **(8.8, 14.7)** | **13.2** **(8.1, 14.1)** | **P < .001** **(.060)** | **P < .001** **(.031)** |
| **AHA (0-100 Units)** | | | | | |
| HABIT | *59.4* *(51.9, 66.8)* | 59.8 (52.3, 67.2) | 61.25 (55.0, 67.5) | - | - |
| LIFT-Control | *51.2* *(42.9, 64.9)* | 50.7 (41.3, 64.1) | 51.1 (42.1, 63.8) | - | - |
| **Mean** | ***56.2*** ***(49.6, 62.8)*** | **56.2** **(49.8, 62.6)** | **57.1** **(51.1, 63.1)** | **P = .154** **(.001)** | **P = .303** **(.001)** |
| **COPM-Performance (0-10 rating)** | | | | | |
| HABIT | - | 2.9 (2.3, 3.5) | 6.8 (6.1, 7.5) | - | - |
| LIFT-Control | - | 2.7 (2.2, 3.2) | 4.5 (3.7, 5.8) | - | - |
| **Mean** | **-** | **2.8** **(2.4, 3.2)** | **5.7** **(5.0, 6.5)** | **P < .001** **(.546)** | **P < .01** **(.056)** |
| **COPM-Satisfaction (0-10 rating)** | | | | | |
| HABIT | - | 3.8 (2.6, 4.8) | 7.3 (6.6, 7.9) | - | - |
| LIFT-Control | - | 2.3 (1.7, 2.9) | 4.7 (3.8, 6.0) | - | - |
| **Mean** | **-** | **3.0** **(2.3, 3.6)** | **6.1** **(5.3, 6.9)** | **P < .001** **(.468)** | **P = .216** **(.011)** |

Abbreviations: AHA, Assiting Hand Assessment; COPM, Canadian Occupational Performance Measure

*Administered by supervisor; **Administered by caregiver

**Figure 2.** Box and Blocks Test by Testing Session



### vi. Caregiver-rated Functional Goals

Although upper- and lower-extremity goals were obtained from parents, only upper-extremity goals are reported. The majority (85%) of upper-limb functional goals established by caregivers for the COPM were bimanual and related to dressing (Table 5). For COPM-performance, repeated measures ANOVA revealed a main effect $F(1,22)=105.41$, $p<.001$ (session). There was also a significant group x session interaction children in the H-HABIT group showing greater improvement, $F(1,22)=10.82$, $p<.01$ (Figure 3). Separate post-hoc comparisons using the Holm adjustment method also revealed significant differences across the two sessions for both the H-HABIT group ($p<.001$) and LIFT-control group ($p<.001$). Eleven out of twelve children in the H-HABIT made group made clinically meaningful improvements (i.e., change>2). Only four out of twelve made clinically meaningful improvements in the LIFT-control group.

Repeated measures ANOVA revealed a main effect for the COPM-satisfaction, $F(1,22)=115.63$, $p<.001$ (session). There was no significant group x session interaction $F(1,22)=2.59$, $p>.05$ (Figure 4). Ten out of twelve children in the H-HABIT made group made clinically meaningful improvements (i.e., change>2). Six out of twelve made clinically meaningful improvements in the LIFT-control group.

**Table 5.** Functional Goals Identified by Caregivers

| Occupational Areas | | Number of Goals Identified | | |
|---|---|---|---|---|
| | | **H-HABIT** | **Lift-Control** | **Goals** |
| **Personal Care** | Feeding | 9 | 9 | Holding a cup with both hands, stabilizing a bowl, cutting with fork/knife, holding yogurt while eating with spoon, drink from open cup, opening snack bags |
| | Dressing (Upper Body) | 5 | 9 | Zipping up jacket, putting shirt on/off buttoning shirt, putting on hat |
| | Dressing (Lower Body) | 13 | 10 | Tying shoe laces, putting sucks on/off, putting on pants, putting on shoes, buttoning pants, zipping up pants, putting on ski pants straps |
| | Hygeine | 6 | 5 | Washing hands, brushing hair, making ponytail, washing upper body, washing hair, burshing teeth, wiping self |
| **School** | Use of School Materials | 7 | 3 | Holding paper while cutting with scissors, stabilizing paper while writing, holding book with both hands |
| | Other | 2 | 1 | Carrying tray, holding backpack with both hands |
| **Play** | Ball Play | 2 | 0 | Catching ball with both hands |
| | Other | 2 | 1 | Holding raquet with both hands, picking up game pieces, holding bike with both hands |
| **Other** | Household | 1 | 0 | Pulling out chair with both hands |
| | Car | 1 | 0 | Buckling Seatbelt |

**Figure 3.** COPM-Performance by Testing Session

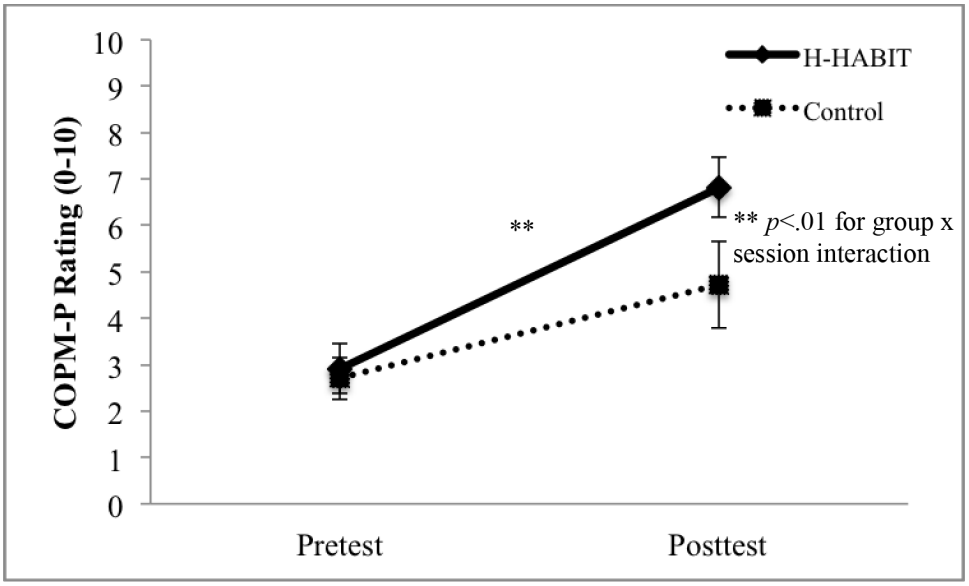
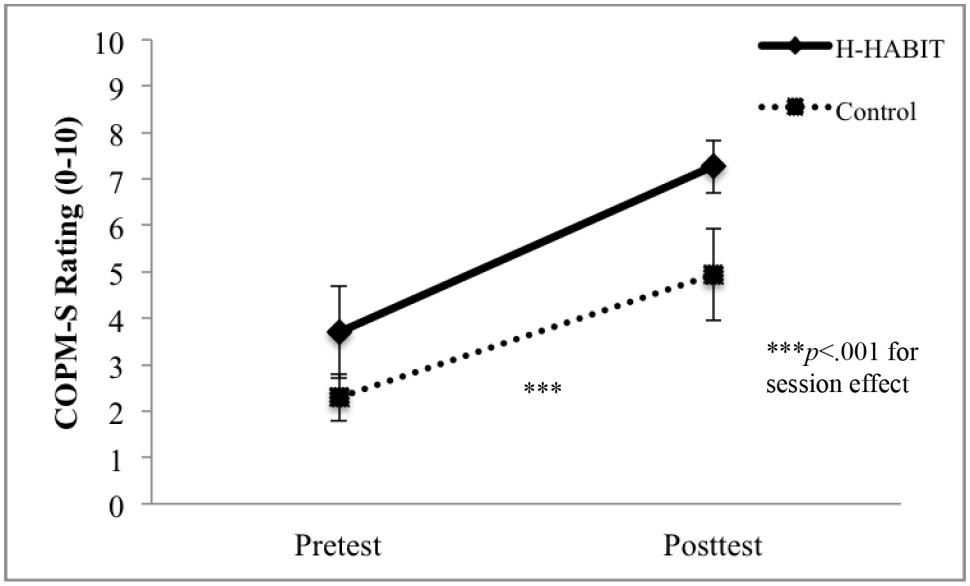


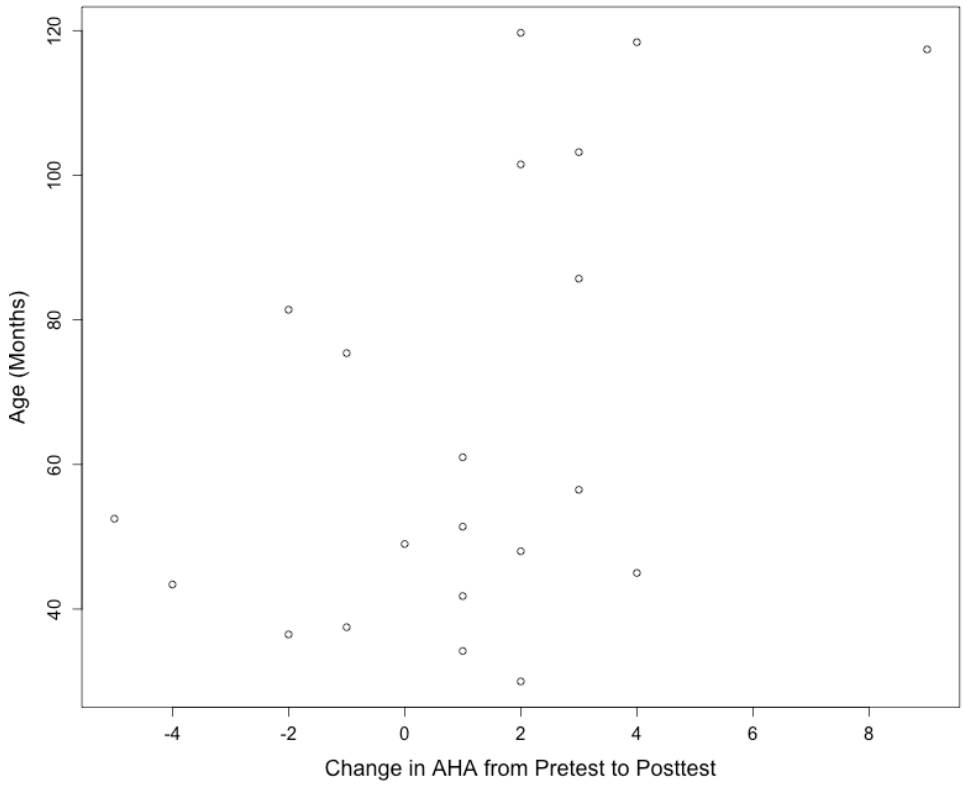
**Figure 4.** COPM-Satisfaction by Testing Session



### vii. Predictors of Improvement

Age was the only variable related to changes on the outcome measures. Age was positively correlated with the change between the pretest and posttest on the AHA ($r$=0.5, $p<.05$) (Figure 5), suggesting older children demonstrated greater change. Conversely, young children tended to show greater variability between the pre- and post-assessments. There was no relationship between feasibility according to the daily logs and changes on any of the outcome measures, between feasibility and the total number of minutes of the intervention completed, and between total time completed and changes on any of the outcome measures. Initial severity was not related to changes in the Box and Blocks test nor for the AHA. The MACS was also not related to any of the outcome measures. Finally, there were no differences between the two cohorts in the amount of change for any of the measures.

Figure 5. Relationship Between Change in AHA and Age. Older children demonstrate greater change on the AHA.



## IV. DISCUSSION

This study examined whether children receiving an intensive home-based bimanual intervention administered by a caregiver made greater improvements in upper extremity function relative to a control group. Children in the control group participated in an intervention of equal intensity and duration that also controlled for the amount of social interaction between caregiver-child dyads. The hypothesis that children in the H-HABIT group would make greater improvements in upper extremity function was only partially supported. Children in H-HABIT made greater improvements in dexterity relative to the LIFT-Control group. However, both groups showed no change in the quality with which they chose to use their affected hand as an assisting hand in the context of bimanual skills. Parents of children in the H-HABIT group perceived larger improvements in their children's performance of parent-specified functional goals. Parents in both groups showed equal improvements in satisfaction about how children performed such functional goals.

These findings have important implications given the increasing growth of home-based interventions. Importantly, this study adds another tool to the list of intensive, motor-learning based approaches that can be used to treat children with hemiplegia. However, the high number of dropouts and lack of improvements in quality of bimanual skills raise questions about the feasibility of home-based programs when supervision of caregivers is performed remotely.

### i. Home-based Interventions as a Tool for Intensive Rehabilitation

Caregiver-directed home-based interventions have emerged as a promising upper limb rehabilitation approach for children with unilateral spastic cerebral palsy (USCP).

Interestingly, up until about 15 years ago there were no evidence-based approaches for improving hand function in children with the disorder. Fortunately, neuroplasticity research with animals (Taub, 1980; Tower, 1940) and models of adult stroke rehabilitation (Taub et al., 1993; Wolf et al., 2006) demonstrated the effectiveness of intensive, motor learning-based approaches for eliciting improvements in manual skills following brain injury. These principles and techniques were adapted and incorporated into the development of treatment tools for pediatric populations (Charles, Lavinder, & Gordon, 2001; Charles, Wolf, Schneider, & Gordon, 2006). The result has been an emergence of studies consistently demonstrating the efficacy of intensive approaches such as constraint-induced movement therapy and bimanual training—although the latter has been studied less extensively. Clinicians now have a growing range of options for targeting upper-limb deficits in children with USCP. However, questions still remain about how these intensive therapies can be structured and delivered in such a way that they are an optimal fit for the individual characteristics of a child and his/her family. Not all families have access to intensive massed practice programs whereas some families are unable to meet the requirements of a demanding home program.

Despite the overwhelming evidence suggesting intensity as a critical component of a successful rehabilitation program (Gordon, 2011; Sakzewski, Gordon, & Eliasson, 2014), clinical decisions about how rehabilitation services are provided to children are often guided by service factors such as the available resources, health insurance policy and place of residence (Aaron et al., 2014). Decisions made on these factors contrast with approaches such as “family-centered care” which suggest that the structure of treatment should align with the needs of the child and family. Family participation and input should

be central to the decision-making process. Parents have identified home programs as one way for maximizing their child's potential and increasing intensity of services when presented with service factors that act as barriers. Studies suggest that if home programs are modeled using best available evidence and parent input, parents are more likely to use a home program and thus implement the intervention at higher dosages (Ferre et al., 2015; Novak & Berry, 2014; Novak et al., 2009).

The current study adds to a growing list of intensive interventions that have been embedded into naturalistic settings with caregivers playing an active role in both the planning and delivery of therapeutic activities. These programs include modified constraint-induced therapy (Al-Oraibi & Eliasson, 2011; Chen et al., 2013; Eliasson et al., 2005; Eliasson et al., 2011; Gelkop et al., 2015; Rostami & Malamiri, 2012b; Wallen et al., 2011) functional goal training or intensive occupational therapy (Novak et al., 2009; Sakzewski et al., 2015; Wallen et al., 2011), and intensive bimanual training (Ferre et al., 2015; Gelkop et al., 2015)—all of which have been delivered at home or at school. In general, these studies have used a distributed practice model (i.e., ~2 hrs. day for ~ 2 months) with schoolteachers or parents supplementing the activities performed during a weekly therapy session with a clinician. Our study used a similar dosing schedule but unlike past studies, activities were performed exclusively in the child's home with a caregiver and weekly supervision sessions were performed remotely.

**ii. Home-based Programs are Feasible, but for Whom?**

In our previous pilot study of H-HABIT, the majority of participants (~90%) completed the entire duration of the intervention (Ferre et al., 2015). In contrast, in the current study there was a ~25% dropout rate for families who began the intervention, and

a large number of families who self-selected out upon completing a screening tool (*n*=8). Even more concerning was the number of families that decided not to participate and who self-selected out after receiving training but prior to the intervention. Although the dropout rate is close to what is considered acceptable for randomized controlled trials (i.e, 20%), the large number of families unable to complete the intervention raises questions about factors that might influence adherence. One reason for the discrepancy in completion rates between the two studies might be related to differences in caregiver supervision. Previously, only local families were included in the study and an experienced supervisor made weekly home visits to the caregiver's home to provide feedback, support, and model activities. Here we expanded the sample to families spread over a large distance and supervised them remotely using telerehabilitation.

Increasing the range of families allowed to participate to include non-local families might have come at the cost of reduced quality of supervision and support. Supervisors still met with families on a weekly basis, however the interactions during those weekly meetings might have been affected by limitations associated with telerehabilitation techniques. These include delays in audio, children being distracted by the video feed, and inability of the supervisors to model activities using the same toys being employed by the family. It is also possible that in-person visits create accountability and thus encourage families to continue meeting the intervention requirements (Mayo, 1981). The retention rate in this study was much lower than those observed in previous randomized controlled trials of home-based interventions (Eliasson et al., 2011; Novak et al., 2009; Wallen et al., 2011) that incorporated weekly face-to-face visits with a therapist. The retention rate was also lower than a pilot study examining the

feasibility of a home-based computer training system (Bilde et al., 2011)—although the amount of daily practice and total dose required for the intervention was lower than the current study.

The reduced quality of supervision is unlikely to have accounted for all of the dropouts however, as the majority of families withdrew from the study because the "schedule was too demanding." Conversely, families that completed the study indicated in the daily logs that the schedule was feasible and that they were able to easily carry out the session. This raises the question of what type of family characteristics might predict success in an intensive home-based intervention. After our initial recruitment cohort yielded a 36% dropout rate, we developed a screening tool to potentially screen out families that had best intentions of completing the study but arguably would be unable to because of an already demanding schedule. With the introduction of the screening tool, only 25% of participants who began the intervention from Cohort 2 withdrew from the study. Thus, it appears that the additional screening tool might help further identify the type of selective population that would be able to meet the requirements of H-HABIT. However, given that the caregivers in cohort 2 were trained in smaller groups and were required to attend our center, the differences in training setting cannot be ruled out as contributing factor to participant retention. In addition, cohort 1 was a sample of convenience in that families did not have to go out of there way to get trained as the training occurred at a family retreat they were already attending. Families in cohort 2 were required to attend our center for training. That is, it required active decision and commitment (i.e., self-selection), and travel to our center. This may have attracted a slightly different subset of the population or influenced their commitment to the study.

### iii. Feasibility of Lower-limb Training as a Control Group

We observed a higher percentage of dropouts in the LIFT-control group (*n*=10) compared to the H-HABIT group (*n*=6). Daily logs also indicated that caregivers in the H-HABIT group perceived the intervention to be slightly more feasible than caregivers in the LIFT-Control group. Despite previous data indicating that caregivers are just as likely to participate in a home-based lower-limb training protocol as an upper-limb protocol (Appendix F), we still observed a disproportionate completion rate between the two groups. It is possible that the priority for caregivers is to see their children's hand-function improve. This assumption might be supported by the overwhelming upper-limb as opposed to lower-limb goals established by caregivers in the study—although the majority of activities of daily living involve the upper extremities, and parents are thus less likely to set as many lower-extremity goals. Unfortunately, the control group in this study was designed to control for attention and intensity of training while incorporating motor learning-based principles. Supervisors ensured that there was no overlap between the two training groups. This meant excluding activities such as very common child-friendly activities such as biking, riding a scooter, swimming, or climbing. A more "client-friendly" approach would be to perform whole-body body practice and train the upper- and lower-limb simultaneously (Bleyenheuft, Arnould, Brandao, Bleyenheuft, & Gordon, 2014). This would greatly increase the repertoire of activities for both the upper- and lower-limb groups, but for this purpose, "contaminate" the groups since some activities would overlap.

It is also possible that the asymmetrical dropout rate was influenced by the two-groups having different supervisors. Caregivers came up with a training program in coordination with their respective supervisors and also met on a weekly basis. In order to provide constant support, participants were encouraged to be in frequent communication with the supervisors. Thus, caregiver motivation might be dependent on interaction styles with their supervisors. Although the current study did not have any data to speak to such issues, previous studies have show that the collaborative bond between a physical therapist and patient, or "working alliance," might be related to treatment adherence (Hall, Ferreira, Maher, Latimer, & Ferreira, 2010).

Despite the asymmetry in withdrawals, caregivers who completed LIFT-control were highly satisfied. Some caregivers expressed their satisfaction on their group social media pages by sharing photos or posts of their children developing a new skill during the course of or shortly after the intervention (e.g., child started riding a bike). Determining whether the children made clinically meaningful changes on standardized measures of lower-limb function is currently being examined.

**iv. Children in H-HABIT Make Unimanual but not Bimanual Improvements**

In this study, children in the H-HABIT group made greater improvements in dexterity following the intervention. Despite the significant finding, the associated $\eta^2$ indicated that only a small percent of the variance in our statistical model (~3%) was accounted for by the interaction. The overwhelming majority of children in the H-HABIT group however, demonstrated improvements on the Box and Blocks test. It is possible that the study was slightly underpowered to accurately capture the differences over time between the two groups.

The advantage of bimanual training is that it affords the possibility to practice strategies that have direct influence on activities of daily living. The lack of changes on the AHA in our study is surprising given the improvements seen in other studies using an equal dosage of bimanual training (Brandao et al., 2013; Gordon et al., 2011) or even an equal dosage of home-based unimanual training (Eliasson et al., 2011). One possibility for the lack of changes might be due to the focus placed on unimanual skills by caregivers in the H-HABIT group. Despite training caregivers to embed practice in the context of bimanual activities, caregivers often focused on asking children to grasp and release with only the affected hand. This might also contribute to the consistent improvements seen in unimanual dexterity. The supervisor reminded caregivers to focus on bimanual activities during the weekly hour of supervision, however supervisors were unable to control how activities were structured the remainder of time. It is important to point out however, that children in the H-HABIT did improve on the performance of functional goals, 85% of which were bimanual. Thus, this approach does permit direct practice of these goals.

Another possibility for the lack of changes on the AHA is that children in this study on average scored higher at baseline compared to other studies demonstrating change on the AHA with this dosage of therapy (Eliasson et al., 2011; Gordon et al., 2011). In contrast, another home-based study of intensive therapy with children displaying similar baseline levels to our study also found little or no change after a 2-month intervention (Wallen et al., 2011). Children who score higher on the AHA at baseline might have already established patterns of how they spontaneously choose to use the affected hand in the context of bimanual skills by 3 years of age and thus have less

room for change as they get older (Holmefur, Krumlinde-Sundholm, Bergstrom, & Eliasson, 2010). It is interesting to note however that our data revealed a moderately strong correlation between age and improvements on the AHA. That is, older children were more likely to make gains. It is also conceivable that older children were more willing than younger children to follow specific instructions about how to use the hands during the activities, to be self-motivated, and to remain engaged throughout the entire 2 hours of training. Although the findings that younger children have greater room to improve and older children make greater gains seem contradictory, it may be the case that the relationship between age and improvements is not linear but rather best explained by an optimal fit curve.

Finally, in order to make our study cost-effective for caregivers, we trained them to administer our behavioral assessments, which relieved them from having to return to our center after completing the intervention for posttesting. Comparisons between a baseline test administered by an experienced tester and the pretest administered by the caregivers showed high correlations and no statistical differences between the two time points. However, closer examination of the individual data revealed differences between the two time points of up to 3 units for the AHA. Although this margin is still below the defined smallest detectable difference and within the range observed during a reliability study with trained AHA providers (Krumlinde-Sundholm, 2012), it does suggest that there might be additional measurement error introduced when caregivers administer the test. Despite the fact that the testing sessions used to assess change both came from caregiver-administered assessments, the additional noise created by this approach may mask individual changes. This is the first study to our knowledge to use caregivers to

administer the AHA. It is important to note that we trained caregivers to administer the assessments in order to increase access to the intervention. The issue of reliability and validity when using this approach merits further investigation—especially given that training caregivers to administer the assessment might increase feasibility of participation in research studies for caregivers.

### v. Does Participation in an Intervention Increase Parental Satisfaction?

Parents of children in the H-HABIT group perceived superior performance of specific functional goals the parents had selected prior to the intervention, but parents in both groups were equally satisfied with their children's performance. Most of the functional goals established by parents in both groups were related to bimanual activities of children's daily routine in the home or school environments. The finding that caregivers in H-HABIT perceive children making greater improvements on performance of functional goals lends further support to specificity of training and that the nature of bimanual training may favor achievement of goals that are important in relevant contexts (de Brito Brandao, Gordon, & Mancini, 2012). Equal satisfaction of their child's performance of upper-limb goals between parents in both groups, despite only the H-HABIT group receiving upper-limb training, might be explained by parent expectations to see improvements following participation in such an intensive program. That is, caregivers in the LIFT-control group may have wanted justification for the investment in time they made by dedicating themselves to a 9-week intervention. It also raises questions about the clinical utility of the COPM as a valid measure of behavioral improvements. Parent perception may be skewed when they are not blinded to treatment allocation.

### vi. Limitations and Future Considerations

Although the interventions in this study were designed based on previously established interventions and parent responses to a survey, there was still a high dropout rate. Future studies would benefit from a more comprehensive screening tool that might capture which families can feasibly participate in an intervention of this intensity. Many of the families that withdrew from the study claimed the schedule was too demanding. The schedule of daily hours of training chosen for this study was based on previous studies that have also used distributed models of intensive training (Al-Oraibi & Eliasson, 2011; Eliasson et al., 2005; Eliasson et al., 2011). Despite the requirements for families in the Eliasson et al. (2005) study amounting to two daily hours of CIMT for two months, the majority of families were only able to complete about 60 hours. Yet, children still demonstrated significant bimanual improvements. It is conceivable that immediate changes can be elicited at lower dosages, although maintenance of such changes might not be observed when children are assessed at a long-term follow-up (Gordon, 2011). It is also possible that caregivers find administering CIMT easier during home-based activities because the presence of a physical restraint reduces the amount of time they have to spend encouraging the child to avoid using compensation, as is the case with bimanual training.

Another limitation of this study is the lack of psychosocial measures or standardized descriptions of family characteristics. For example, parental stress levels were not measured for any of the participants. The increased caregiving for children with disabilities may adversely affect psychological health (Eker & Tuzun, 2004). Although

parental stress levels have been shown to remain stable throughout the course of a home-based intervention (Chen et al., 2013; Ferre et al., 2015), it is possible that caregivers with high stress levels are less likely to complete an intervention of this type. The level of motor abilities of a child might also relate to how parents perceive ease of caregiving for their child. Parent perception of ease of caregiving could potentially be used to match interventions with a family's needs (Ward et al., 2014).

It is also very likely that successful completion of the program is determined by interactions between characteristics of the caregiver including work status (i.e., employed vs. unemployed), number of children, age of siblings, motivation, and socioeconomic status with child characteristics such as age, severity, distractibility, and responsiveness to reward systems. Future studies of home-based interventions would benefit greatly from unmasking the family dynamics that might predict optimal fit for a home-based program. This is in accordance with behavioral change theoretical frameworks which stress the importance of identifying mediators that might predict optimal fit or adherence to an intervention (Sirur, Richardson, Wishart, & Hanna, 2009).

The higher number of families that withdrew from the study in lower-limb training group might also indicate that families prefer to be involved in an intervention that involves upper-limb training. Perhaps a more appropriate comparison group would be an upper-limb intervention of equal intensity and duration, but one that is not based on motor learning principles. Home-based CIMT has already been demonstrated to be an effective approach. Thus a more valuable comparison would be comparing H-HABIT to an equal dosage of some of the techniques used in usual and customary care (e.g., neurodevelopmental treatment, stretching, or repetition of behaviors). However,

although the comparison is methodologically interesting, it might not be practical for families given that such approaches do not lend themselves well to intensive paradigms (i.e., stretching for 2 hours daily).

The lack of bimanual improvements might suggest that greater caregiver guidance should be provided to ensure training activities are performed correctly. This might mean increasing the supervisor-caregiver ratio and increasing the amount of weekly supervision. It is also possible that when intensive bimanual training is distributed over a longer period of time, changes in how a child uses his/her affected upper extremity as an assisting hand in a bimanual context takes longer to consolidate (Ferre et al., 2015; Sakzewski et al., 2015). Although only immediate changes are reported here, a 6-month follow up is currently being collected. A separate consideration is that participation in intensive therapies is becoming increasingly common, and thus some children are less likely to show large gains because of "oversaturation" of therapy. Finally, null results may also provide valuable clinical information regarding treatment distribution (Eliasson, 2015). Finding the optimal dosing parameters for interventions remains a critical question in the rehabilitation of children with cerebral palsy (Kolobe et al., 2014). Future studies could examine the effects of decreasing the amount of daily hours of H-HABIT, but extending the period of time during which the activities are performed and increasing the total dosage of H-HABIT. The modified schedule might make activities more feasible for families yet still provide the required intensity needed for eliciting changes that are maintained over a long-term period.

Finally, in order to increase feasibility for families, the design of the study was such that we trained caregivers to administer the assessments. Although the AHA and

Box and Blocks are designed to be objective, it is conceivable that the manner in which caregivers administered the tests might have influenced their child's performance. Caregivers in this study were not blinded to their group allocation. Ideally, individuals blinded to the study would perform both the administration and scoring of the tests. Here, only the individual scoring the AHA was blinded.

Despite these limitations, children in the H-HABIT group made considerable improvements in measures of dexterity and goal performance. This suggests that H-HABIT still might serve as a useful tool for clinicians. Several studies in the past decade and a half have demonstrated intensive training leads to improvements in hand function, especially compared to usual and customary care (Sakzewski, Ziviani, & Boyd, 2014b). The home-based intervention examined in this study is not meant to replicate or even supplant previous interventions, but rather provide an additional dimension to how intensive rehabilitation paradigms can be implemented.

Despite the encouraging results of this study, caution is warranted. Implementing intensive home-based interventions can disrupt the psychosocial dynamics of the families receiving such services. Thus, it is important for researchers to be cognizant of the secondary negative effects home-based interventions might have. One way potentially disruptive effects can be avoided is by clearly delineating the sections of the day reserved for therapy. For example, in this study we trained caregivers to use subtle observable cues to separate "therapy time" from "family time". Options for cues could include wearing a "therapy" scarf or hat, consistently using a specific location in the home such as a table that is only used for therapy activities, or using games and activities only during daily therapy that are then hidden afterwards. The purpose is to create a context for therapy in

the home environment that can be discrete from the child and caregiver's typical routine. Researchers should also encourage caregivers to avoid using constant prodding and requests to perform certain behaviors of interest outside of the daily therapy schedule. Although this system might not feasible for language or behavioral interventions in which the goal is to change how caregivers interact with their child on a permanent basis, it might serve as a useful technique for motor-based interventions with clearly specified dosages. Restricting therapy to the required daily time and creating a distinct context for the activities might help serve to decrease stress in caregivers and children.

Using caregivers as a way to implement intensive interventions provides a cost-effective alternative to expensive clinic-based interventions. It also provides the opportunity for caregivers to increase their personal involvement in the rehabilitation of their own child. Caregivers are the person most likely to understand how a child responds in their typical environment and thus are a valuable source of input for clinicians. Home-based models provide a valuable intervention approach to add to the repertoire of options clinicians have to chose from when developing individualized treatment programs for children and their families.

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## APPENDIX A

## Literature Review

### I. Introduction

Early brain injury can lead to anomalous developmental trajectories that result in deficits in motor skills. Such is the case for children with cerebral palsy—a group of developmental disorders of movement and posture that arise from perinatal brain injury and cause activity limitations (Bax et al., 2005). The most common form of the disorder is unilateral spastic cerebral palsy (USCP), which is characterized by motor impairments predominantly affecting one side of the body (Stanley et al., 2000). Individuals with USCP have difficulty performing activities of daily living independently (Skold et al., 2004) and participating in social or school activities. Designing an intervention that directly improves independence and participation is challenging—especially given that they are broadly defined constructs, are difficult to measure, and are unique to each child. Furthermore, there is no available evidence of an effective intervention that addresses participation (Novak et al., 2013). A more achievable aim is focusing on the efficiency and effectiveness of children's motor performance in specific actions. This motor learning-based approach has also been coined as "task-oriented training" (Carr & Shepherd; Winstein & Wolf, 2008).

Evidence-based rehabilitation for children with USCP has only recently begun to expand. In many cases there exists a gap between clinical practice and the most current theoretical models grounded in scientific evidence. Novak et al. (Novak & McIntyre, 2010; Novak et al., 2013) systematically categorized empirical support for CP interventions and concluded that the majority of published intervention studies lack

strong support for their efficacy. Most of the models lacking support include therapy activities that involve the child acting as a passive participant in therapy (e.g., passive stretching). In contrast, interventions for upper-limb rehabilitation shown to be effective were intensive therapies based on principles of motor learning. Most importantly, these models involve active, self-generated movements by the child receiving the therapy—a characteristic of rehabilitation that has been shown as being critical for inducing neuroplastic changes (Friel, Williams, Serradj, Chakrabarty, & Martin, 2014).

Two motor learning-based approaches that have been used with considerable success are constraint-induced movement therapy (CIMT) and intensive bimanual training. CIMT involves restraint of the unaffected limb coupled with structured training of the affected limb (Taub & Wolf, 1997). Intensive bimanual training preserves the structured practice of CIMT but focuses on engaging the child in bimanual activities rather than relying on use of a restraint to promote affected hand-use (Charles & Gordon, 2006). There is sufficient evidence suggesting that CIMT, when provided at high levels of intensity, leads to improvements in amount and/or quality of unilateral upper extremity movement (Chen & Yang, 2007; Hoare, Imms, Carey, & Wasiak, 2007; Huang et al., 2009; Novak et al., 2013). Studies of intensive bimanual training have shown comparable efficacy (Dong, Tung, Siu, & Fong, 2013; Facchin et al., 2011; Gordon et al., 2011; Gordon et al., 2007; Green et al., 2013; Hoare et al., 2013; Hung et al., 2011; Sakzewski et al., 2011).

While promising, the results of intensive bimanual and CIMT interventions are often limited to studies that involve one-on-one supervision by an interventionist for several hours a day (i.e., massed practice). In some cases, in order to achieve the desired

intensity that is characteristic of functional improvements, the interventions require the child be engaged in structured activities for a significant portion of the day (~6 hrs. per day, for ~3weeks). Although this method has been shown to be effective, massed practice models might not be appropriate for younger children who are unable to sustain the attention and energy needed to be engaged in activities for long periods.

Massed practice models are also not available in many communities. In cases where they are available, there are often barriers that prevent families from attending.(Sakzewski, Ziviani, et al., 2014b) One study examining the barriers in delivering evidence-based rehabilitation to children found that therapists cited the availability of time, space, and materials as a major limitation (Sakzewski, Ziviani, & Boyd, 2014a). Direct one-on-one supervision by a therapist presents a burdensome financial challenge. The cost of staffing a camp, purchasing supplies for activities, and securing a location often results in interventions being offered as a for-fee service to families. In response to some of these limitations, interventions have been adapted and modified to suit the constraints of the family and child receiving the therapy. Specifically, interventions have been adapted for a home-based setting and have incorporated the use of caregivers in delivery of therapy activities.

This review examines the use of home-based models that were designed to augment schedules of usual and customary care and have been used successfully to deliver intensive interventions to children with USCP. Evidence regarding the effectiveness of adapted models will be presented along with what is known about treatment fidelity. The challenges associated with home-based models are discussed,

including how they might affect the psychosocial dynamic of participants, and are compared in relation to home–based models of other pediatric clinical populations.

## II. Modified Interventions

Adapted models of intensive upper-limb interventions are typically designed by modifying 3 main components of intensive camp-based or clinic-based models: 1) the setting in which the therapy is delivered, 2) the dosing schedule of activities, and 3) the person responsible for engaging the child in the activities. The interventions have been designed in such a way to make treatment more accessible and child-focused or family-centered. The activities are typically delivered in the environment of the child's daily routine.

Eliasson et al. (2011) and Morgan, Novak, and Badawi (2013) outline theoretical frameworks that highlight the important role the environment plays in rehabilitation. Using Brofenbrenner's ecological model of child development (Bronfenbrenner & Morris, 1998), Eliasson et al. (2011) argues that delivering training in settings that are part of the child's typical routine (i.e., at home or in preschool setting) may be an important determinant in the efficacy of a treatment. Morgan et al. (2013) borrows the notion of "enriched environments" from animal neuroplasticity literature and applies it to early intervention for infants at-risk of developing CP. The authors attempt to develop a rationale for "enriching" various aspects of the child's motor, cognitive, sensory, or social milieu to promote positive motor outcomes. Despite being based on different lines of evidence, both papers suggest that providing treatment outside the standard hospital or clinical setting might be advantageous for rehabilitation and permits intervention at

younger ages—particularly at younger ages in which the developing nervous system may exhibit greater plasticity (Rosenbaum et al., 2002; Spittle et al., 2009).

The majority of studies that have embedded upper-limb therapy into a more naturalistic setting have been adaptations to CIMT delivered in the context of the child's routine at home or at school (Al-Oraibi & Eliasson, 2011; Chen et al., 2013; Eliasson et al., 2011; Gelkop et al., 2015; Klingels et al., 2013; Novak et al., 2009; Novak et al., 2007; Rostami & Malamiri, 2012a; Taub et al., 2011; Wallen et al., 2011). The outcomes of adapted CIMT studies have been overwhelmingly positive—children receiving CIMT in home and/or preschool settings consistently demonstrate gains in upper-limb function. One small study that directly compared home-based and clinic-based CIMT found greater improvement in the home program (Rostami & Malamiri, 2012b). A recent systematic review found that an overwhelming majority of CIMT studies involve at least some form of a home component(Myrhaug, Østensjø, Larun, Odgaard-Jensen, & Jahnsen, 2014). Although the models have not been examined as extensively as adapted CIMT, intensive bimanual approaches and functional goal training have shown similar promise when provided at home or school (Al-Oraibi & Eliasson, 2011; Eliasson et al., 2005; Ferre et al., 2015; Gelkop et al., 2015; Novak et al., 2009; Novak et al., 2007; Wallen et al., 2011).

In order to feasibly provide training outside of the clinic, researchers have modified the dosing schedule of therapy. For example, instead of providing 6 hours of daily massed practice for a period of 3 weeks, distributed practiced models have been developed in which the daily amount of training is typically reduced to ~2 hours and the duration of the entire intervention is increased to ~9 weeks (Eliasson et al., 2011).

Unfortunately, there is a wide range of variability in the total dosage of therapy provided for distributed practice models. Interventions have ranged in duration from 5 to 10 weeks with the total expected home practice suggested ranging from 28-168 hours. Despite the variability in intervention designs, studies that have used distributed practice models have consistently shown gains in upper-limb function and parent satisfaction of goal performance (Al-Oraibi & Eliasson, 2011; Eliasson et al., 2005; Eliasson et al., 2011; Ferre et al., 2015; Gelkop et al., 2015; Hoare et al., 2013; Novak et al., 2009; Wallen et al., 2011).

Reducing the amount of time children receiving training spend in the clinic has increased the responsibility of caregivers by placing them in the role of primary interventionist. Thus, in addition to adapting the setting and dosing schedule, the studies described above have also incorporated the use of schoolteachers and parents in delivery of therapy to the child. Caregivers are typically trained prior to the onset of interventions and are consulted as part of the development of a treatment for their children. The inclusion of caregivers in the planning of activities is in line with service approaches such a family-centered practice that argue caregivers are apt to play the role of "expert" as a result of their ability to recognize the health care needs of their own child (Dunst, Johanson, Trivette, & Hamby, 1991; Winton & Bailey, 1997) Furthermore, the increased participation of the caregiver also shifts the role of the child's therapist from directly treating the child to promoting caregiver competency—that is, providing caregivers with the tools needed to help address some of the daily problems associated with CP (Novak & Cusick, 2006).

There has been very little consistency in how home-based interventions for children with CP have been delivered. The dose of direct therapy has ranged from 26-168 hours. The number of visits from a therapist has ranged from weekly supervision to several sessions per week. This is particularly significant since there is some indication that compliance might be directly related to the number of home visits received by a therapist (Mayo, 1981). Finally, the actual person carrying out the therapy has varied across and even within studies with activities being led by teachers at school while others used family members at home (Eliasson et al., 2005). Despite being unclear whether the interventions have been delivered as intended, reported results match the outcomes of more controlled interventions in clinical settings.

Relying on caregivers in adapted settings can also provide challenges. The nature of home-based interventions makes tracking parents' performance in the delivery of activities difficult. Researchers are limited to caregivers' unstandardized reports for tracking compliance. Caregivers are also not trained therapists. Thus, one has to question whether the fidelity of a treatment has been compromised when caregivers are primarily responsible for the delivery of activities. There is also the potential for the implementation of a home-based program to disrupt the psychosocial dynamics between parents, the child, and the entire family by increasing parental stress (Novak et al., 2009). Although home-based interventions for children with USCP have only relatively recently received attention, several home-based models of other pediatric clinical populations have been studied quite extensively. Understanding how these paradigms have handled the challenges described above might provide valuable insight for designing home-based interventions for children with USCP.

## III. Caregiver-directed Home-based Pediatric Rehabilitation Paradigms

The involvement of caregivers in implementing intervention strategies for children with developmental disabilities has a relatively long history. For example, using caregivers to help with the treatment of children with Autism has been studied for at least four decades (Schopler & Reichler, 1971). As with the treatment of many developmental disabilities, the inclusion of caregivers as part of a rehabilitation program has been a result of evidence suggesting that introducing intervention at earlier stages has a greater impact than when provided at later stages in development (Rogers, 1996). Very young children receiving therapy are more likely to fatigue, have difficulty sustaining attention, or experience separation anxiety. Providing intervention at earlier ages thus necessitates the involvement of caregivers in adapted settings.

Despite a long history with the use of caregivers in intervention delivery, methodologically sound research is still lacking. Most home-based intervention studies are designed in a manner to improve access to therapy without consideration of how the mode of delivery might impact factors other than the behavior being targeted. For example, several studies have successfully used home-based strength training or supported treadmill walking to target lower-limb deficits in children with cerebral palsy (Damiano & Abel, 1998; Damiano, Vaughan, & Abel, 1995; Dodd, Taylor, & Graham, 2003; Gates et al., 2012; Johnston et al., 2011). However, the majority of these studies have neglected any of the potential negative effects on the psychosocial behavior of the child receiving the intervention. This is particularly troubling given that one study found

that even a short, home-based strength training program can inhibit self-concept in children with cerebral palsy(Dodd, Taylor, & Graham, 2004).

Potential negative effects of a home-based intervention are not limited to the child. Implementing an intensive intervention increases the burden of the caregiver. Unfortunately, very little is understood the relationship between intensive home-based interventions and parental stress as the overwhelming majority of studies do not systematically monitor stress levels (McConachie & Diggle, 2007). One study that surveyed parents of children with autism following participation in an intensive home-based behavioral intervention reported correlations between both the child's age at initiation of the intervention and the length of the intervention with parental stress (Hastings & Johnson, 2001). Surprisingly, mothers report lower stress levels when intervention begins at younger ages and is implemented for relatively longer durations. However, reductions in perceived parental stress also occur when improvements are found in communication skills and social interaction following a short duration intervention (Wong & Kwan, 2010). Thus, it appears that either the intensity of a home-based intervention or improvements in behavioral outcomes can have positive effects on parental stress levels.

The lack of monitoring is a limitation that is not only related to the construct of caregiver stress. Few studies address treatment fidelity beyond the participants' adherence to the time requirements of a study. Shifting interventions from the clinic to the home setting reduces the amount of experimental control researchers have given the relatively limited amount of supervision. Little is known about the quality with which caregivers deliver intervention when performing the activities unsupervised. As evidence

for the efficacy of parent-mediated intervention grows, there is a strong need to identify the characteristics within treatment programs that might help document the consistency of application of treatment protocols (Drew et al., 2002). A recent review of studies on parent training for young children with autism spectrum disorder (ASD) found 17 studies performed in the context of a randomized clinical trial (Oono, Honey, & McConachie, 2013). Yet, the authors found a high risk of methodological bias in the majority of the studies as well as no evidence of statistical gains for most of the designated primary outcomes. The lack of improvement in such cases raises questions about the relationship between maintaining treatment fidelity and efficacy of home-based interventions.

Measuring fidelity is a major challenge for researchers as evidenced by the dearth of home-based studies that have examined the construct. The literature on speech rehabilitation is another example of a field that has used the home as a context for intervention with little attention to the quality of the treatment. It is widely accepted that the early social milieu and communicative context can be structured to increase the rate and maximum limit of future language development (Snow, 2013; Tannock, Girolametto, & Siegel, 1992). Training caregivers to provide the necessary structure and supportive language environment provides an optimal opportunity to intervene at early stages. The success of parent-directed language programs depends on parents learning strategies and using them with sufficient frequency and accuracy to shape their child's development. According to Roberts and Kaiser (2011), proper examination of such interventions require procedures for teaching parents specific strategies, measuring the extent to which parents' implement these strategies (i.e., treatment fidelity), and measuring child language outcomes. In terms of language outcomes, there is strong evidence suggesting

parent-directed interventions are an effective approach for eliciting improvements (Roberts & Kaiser, 2011). However in their review, the authors found that only 28% of the identified studies measured treatment fidelity and only 50% of provided an adequate description of parent training procedures.

One study measured fidelity by providing a schedule detailing the course of treatment, recording attendance data, and eliciting caregiver perception of their ability to provide the correct treatment approach (Girolametto, Pearce, & Weitzman, 1995). The authors based their model on the one described by LeLaurin and Wolery (1992) in which 3 levels of fidelity should be clearly outlined: 1) the schedule describing the sequence of treatment implementation, 2) data indication the intervention occurred as scheduled, and 3) measurement on the parents' interaction with the intervention techniques. Others have used coding of video-recorded therapy sessions to ensure treatment protocols were adhered to—although the outcomes or behaviors coded were not precisely described (Green et al., 2010). In contrast, Kaiser and Roberts (2013) specified three distinct aspects of parent training (preteaching, coaching, feedback) that were evaluated for sessions in the clinic and at home and provided a checklist that was used to assess proper implementation of intervention strategies. The study stands alone as one of the few caregiver-directed home-based language interventions to clearly specify a protocol for assessing fidelity. Kaiser and Roberts (2013) also assessed parental stress using the parental stress index (Abidin, 1995), but only as a descriptive characteristic for their sample. That is, parental stress was not monitored throughout the course of the intervention.

One way researchers have managed to improve monitoring of treatment fidelity is via telerehabilitation. Telerehabilitation involves the use of communication and information technologies to increase accessibility and promote continuity of treatment for individuals with disabilities(Kairy et al., 2009). Providing in-person supervision of activities provides a logistical challenge when interventions include a large number of children spread over a large geographical are. Telerehabilitation offers a time-and cost-saving technique to overcome such challenges (Cooper et al., 2001; Torsney, 2003).

Although the majority of studies using telerehabilitation have been developed in adult clinical populations, the technique has also been used effectively with children. Cason(Cason, 2009) successfully piloted an early intervention program with a group of rural families over a 12-week period during which an occupational therapist remotely provided consultative services to a caregiver and facilitated the caregiver's interactions with their child. Caregivers expressed a high level of satisfaction with the program as a result of the increased intensity of services afforded by the technique. Feasibility and effectiveness of telerehabilitation paradigms have been also been used for delivering speech therapy to improve fluency in children with stuttering problems(Sicotte et al., 2003), coaching parents of children diagnosed with autism spectrum to provide intervention(Baharav & Reiser, 2010; Gibbs & Toth-Cohen, 2011), and even guiding parents to assist with the treatment of a swallowing disorder(Malandraki, Roth, & Sheppard, 2014). Despite the paucity of large, well-designed studies using telerehabilitation in pediatric populations, the delivery model appears to offer increased accessibility of services with parents reporting high levels of satisfaction.

## IV. Conclusions and Future Considerations

A survey of the literature indicates a great deal of variability with regards to monitoring treatment fidelity, parent training procedures, and the effects of home-based treatment programs on the psychosocial dynamic in the family. Most concerning is the lack of description of training procedures for parents. No study to date has been published on the effectiveness of individual training components in relation to parent and child outcomes. Of the few studies that do describe training, the dosage of training procedures is rarely reported. Thus, questions remain about the relationship between parent fidelity of implementation of intervention strategies and child outcomes. In order to increase reproducibility, future studies should clearly designate training procedures and direct measures used to observe proper implementation of the intervention. Proper documentation of such procedures would help reduce methodological bias and would permit more robust conclusions about the efficacy of home-based training programs. Promising results from studies that have incorporated the use of technology to monitor home programs offer a potential solution for overcoming some of the obstacles associated with tracking treatment fidelity. Telerehabilitation reduces travel time and increases access to professional consultation for remote communities. Increased access provides healthcare professionals with greater opportunity to ensure families adhere to treatment protocols.

Implementing intensive home-based interventions can disrupt the psychosocial dynamics of the families receiving such services. Thus, it is important for researchers to be cognizant of the secondary negative effects home-based interventions might have. One way potentially disruptive effects can be avoided is by clearly delineating the sections of

the day reserved for therapy. For example, using subtle observable cues might be one way to separate “therapy time” from “family time”. Options for cues could include wearing a “therapy” scarf or hat, consistently using a specific location in the home such as a table that is only used for therapy activities, or using games and activities only during daily therapy that are then hidden afterwards. The purpose is to create a context for therapy in the home environment that can be discrete from the child and caregiver’s typical routine. Researchers should also encourage caregivers to avoid using constant prodding and requests to perform certain behaviors of interest outside of the daily therapy schedule. Although this system might not feasible for language or behavioral interventions in which the goal is to change how caregivers interact with their child on a permanent basis, it might serve as a useful technique for motor-based interventions with clearly specified dosages. Restricting therapy to the required daily time and creating a distinct context for the activities might help serve to decrease stress in caregivers and children.

Using caregivers as a way to implement intensive interventions provides a cost-effective alternative to expensive clinic-based interventions. It also provides the opportunity for caregivers to increase their personal involvement in the rehabilitation of their own child. Caregivers are the person most likely to understand how a child responds in their typical environment and thus are a valuable source of input for clinicians. Home-based models provide a valuable intervention approach to add to the repertoire of options clinicians have to chose from when developing individualized treatment programs for children and their families.

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# APPENDIX B

DEVELOPMENTAL NEUROREHABILITATION

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BRIEF REPORT

## Feasibility of caregiver-directed home-based hand-arm bimanual intensive training: A brief report

Claudio L. Ferre[1], Marina B. Brandão[2], Ya-Ching Hung[3], Jason B. Carmel[4,5], & Andrew M. Gordon[1]

[1]*Department of Biobehavioral Sciences, Teachers College, Columbia University, New York, NY, USA,* [2]*Faculdade de Ciências Médicas de Minas Gerais, Belo Horizonte, Brazil,* [3]*Queens College, City University of New York, Flushing, NY, USA,* [4]*Brain and Mind Research Institute, Weill Cornell Medical College, New York, USA, and* [5]*Departments of Neurology and Pediatrics, Weill Cornell Medical College, Burke Medical Research Institute, New York, USA*

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**Abstract**

*Objective*: To determine feasibility of a home-based, intensive bimanual intervention with children with unilateral spastic cerebral palsy. *Methods*: Eleven children (aged 29–54 months) received 90 hours of home hand-arm bimanual intensive therapy (H-HABIT) provided by their trained caregivers. Parenting stress levels and compliance were monitored using the Parenting Stress Index and daily logs. Quality of bimanual performance and changes in performance/satisfaction of functional goals were assessed using the Assisting Hand Assessment (AHA) and Canadian Occupational Performance Measure (COPM), respectively, at two pretreatment baseline sessions and two posttreatment sessions (immediate and six months). *Results*: Ten children completed the study with caregivers completing on average 85.6 hours of H-HABIT. Daily logs indicated high caregiver compliance. Stress levels remained stable across the intervention. Children demonstrated significant improvements in the AHA and COPM. *Conclusion*: H-HABIT is a feasible intervention for improving hand function and merits further investigation in a randomized-control trial.

**Keywords**

Bimanual training, family focused, hand rehabilitation, hemiplegia, home-based therapy, unilateral cerebral palsy, upper-limb function

**History**

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### Introduction

Children with unilateral spastic cerebral palsy (USCP) experience unilateral deficits in the execution of movement [1] that arise from early prenatal or neonatal brain injury. Children with USCP also have sensory impairments [2], impairments in movement planning [3] and deficits in the quality of bimanual performance; however, longitudinal studies of children with USCP suggest that performance improves over time when children are provided with access to therapy [4]. The goal of interventions designed for children with USCP is to improve affected limb-use in order to promote functional independence and participation.

One evident trend in treatment approaches for children with USCP is an increased involvement of the caregiver in intervention delivery [5–7]. Increased involvement on behalf of the child's family is a notion consistent with theoretical frameworks such as family centered care [8], which suggests that caregivers and professionals are equally responsible in the promotion and delivery of health care to the child. Studies that have adapted interventions by modifying dosing schedule and training teachers or caregivers to lead manual activities have demonstrated to be an effective approach for improving hand function in children with USCP [5–7].

One approach that has yet to be formally tested when provided exclusively in the home is intensive bimanual training. Hand-arm bimanual intensive therapy (HABIT) is a motor-learning-based training focused on improving the amount and quality of involved hand-use in the context of bimanual tasks [9]. Camp-based studies of HABIT in children with USCP age four years and above have shown the therapy to improve the quality of bimanual performance [10]. This study examined the feasibility of home-based HABIT (H-HABIT) with caregivers as the primary interventionists. The aims of this study were to determine the feasibility of H-HABIT in terms of compliance, caregiver perception of difficulty in completing the activities and the impact of the intervention on the psychosocial dynamic between caregivers and their child (i.e. caregiver stress); and to collect preliminary outcome data on the effectiveness of H-HABIT to inform a power analysis for a future randomized control trial.

### Methods

#### Study design

A single-group design was chosen to examine feasibility of the intervention. Measures were taken during a one-week baseline period, during the treatment phase and immediately after and six-months after a 90-hour home-based bimanual

Correspondence: Claudio L. Ferre, Biobehavioral Sciences, Teachers College, Columbia University, 525 West 120th St., Box 93, New York, NY 10027, USA. E-mail: clf2124@tc.columbia.edu

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intervention with caregivers as the primary interventionists. Caregivers were trained to administer H-HABIT immediately following the baseline period.

### Participants

Participants were recruited from referrals from clinicians, our website (http://www.tc.edu/centers/cit/), and online support groups. Eligible participants between the ages of 30 and 54 smonths with USCP were invited to participate if they met the following inclusion criteria: (1) ability to grasp and release blocks from affected hand, (2) ability to follow two-step instructions and complete testing and (3) ability of the caregiver to provide one-to-one attention to the child during the daily activities. Children were excluded from the study if they received an intensive upper extremities intervention or botulinum toxin therapy in the upper extremity within the past six months. All children continued to receive their customary individual therapy during the course of the study. Informed consent was obtained from caregivers. This study was approved by the Teachers College, Columbia University Institutional Review Board.

### Caregiver training

Caregiver training was standardized and adapted from an established manual of procedures for HABIT [11]. All caregivers received training from the same experienced supervisor over a series of three sessions each lasting about 1.5–2 hours. The first training session involved only the supervisor and caregiver. Caregivers were instructed on the general intervention procedures, discussed reinforcement strategies and were guided through videos of other caregivers performing H-HABIT. We were particularly concerned about not disrupting the complex psychosocial dynamic between caregivers and children. In order to avoid disruptions, caregivers were encouraged to use a subtle cue to the child (e.g. H-HABIT hats or scarves) to indicate that H-HABIT hours were separate from the child's typical routine. For the second session, caregivers returned with their child and watched as the supervisor modeled how to administer H-HABIT. Then, caregivers were asked to model with the child for the supervisor and were provided with feedback. The final training session occurred in the family's home and also marked the beginning of the 90-hour intervention. The supervisor visited the family's home and provided feedback to the caregiver as they performed activities with the child. Home visits occurred weekly throughout the intervention (1 hour/visit).

### H-HABIT intervention procedures

Children completed an intervention in which they received 2 hours/day of H-HABIT, for five days/week that lasted nine consecutive weeks (90 hours total). The intervention supervisor helped caregivers design an individualized program for the child. Activities were chosen based on the ability of the child's affected hand and focused on using the hand as an assisting hand during increasingly complex bimanual coordination. Task demands were graded by varying the constraints of the task or providing activities that required progressive skilled use as performance improved. Caregivers were instructed to place emphasis on making the intervention enjoyable and intrinsically motivating for their children. Age-appropriate reward systems (e.g. sticker charts) and age-specific knowledge of results was provided as encouragement and to ensure the activities were highly motivating and fun. Caregivers incorporated functional (e.g. cleaning and eating) and play activities (i.e. building blocks and magnets) using child-friendly games. Caregivers recorded daily activities and time spent on each activity using an online log. The supervisor monitored daily logs remotely and provided ideas for skill progression.

### Measures

*Participant characteristics*. The Manual Ability Classification System was used to classify the children's typical manual performance [12].

*Feasibility*. Aspects of feasibility included compliance, caregiver perception of difficulty in completing the activities and the caregiver stress levels. Caregiver stress levels were monitored with the Parenting Stress Index-Short Form (PSI-SF) [13] at each of the assessment time points. The PSI-SF is a 36-item self-report measure used to evaluate stress related to family interactions and has established reliability and validity [14].

Compliance was tracked using online daily logs. Participation rate was calculated as the percentage of the required 90 hours completed by the participants. The daily logs also included questions adapted from Wallen et al. [7] and were used to elicit perceptions of the difficulty in completing the day's activities (Figure 1).

*Hand function*. The Assisting Hand Assessment (AHA, version 4.3) is an instrument that quantifies how effectively a child with unilateral disability uses his/her affected hand in bimanual activities. It has excellent validity and reliability [15, 16]. A change of five AHA units is regarded as the smallest reliable difference for the AHA [17]. The tests were scored by an occupational therapist certified to score the AHA and that was blinded to the study. The AHA was administered twice before the intervention (separated by one week), immediately after and six months after the intervention.

The Canadian Occupational Performance Measure (COPM) was used with parents to establish and evaluate children's functional goals [18], in terms of performance and caregiver satisfaction levels. It is a valid and reliable measure [19] with a change of two points indicating a clinically meaningful difference. The COPM was used to assess goal performance at the second baseline, immediate posttest and six-month follow-up.

### Statistical design

A repeated-measures ANOVA on test sessions was performed on the PSI raw data, AHA-units for the AHA and on raw and log-transformed COPM data using R (R Core Team, 2012). *Post hoc* comparisons using Tukey HSD test were used between the first baseline and each subsequent measure and between the first posttest and second posttest. For the AHA, which included two baseline assessments, Tukey HSD test

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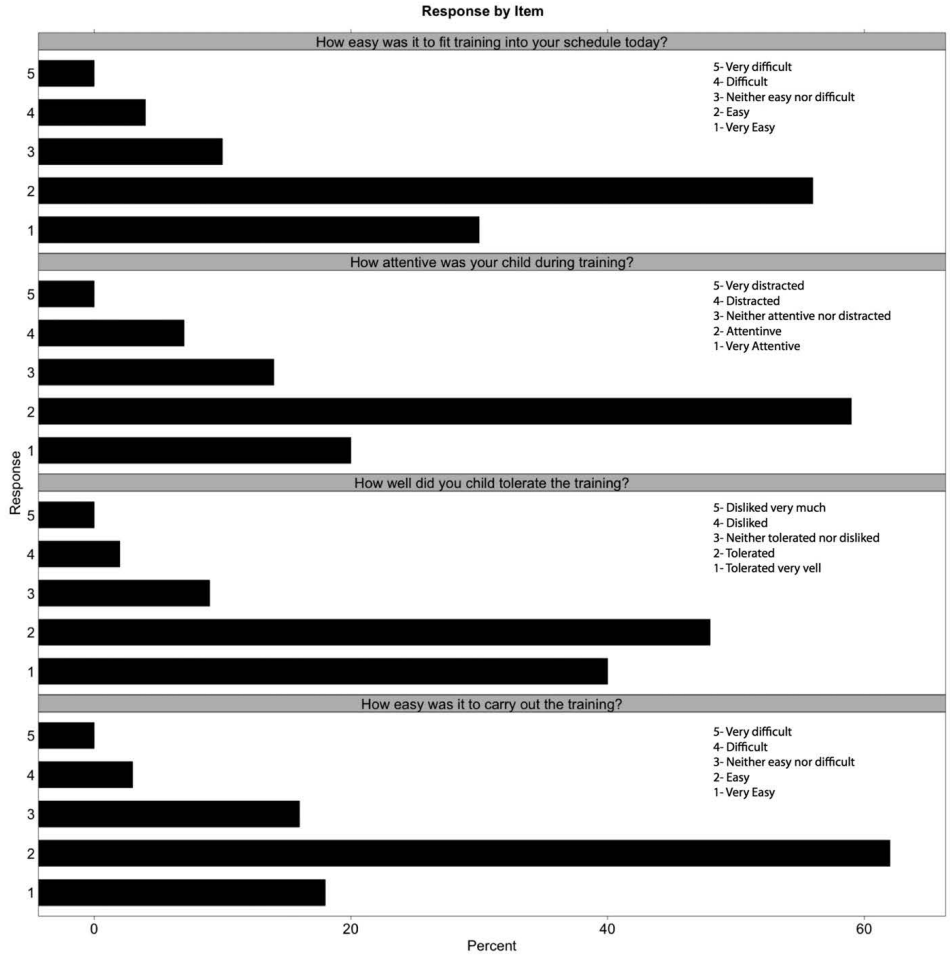


Figure 1. Responses to daily log questionnaire regarding feasibility.

was used between the average of the two baseline measures and each subsequent measure.

The magnitude of change attributed to the intervention was also determined using net changes scores as calculated by subtracting baseline change from treatment change and comparing to published smallest detectable differences for the AHA. Change scores from baseline to immediate posttest were also calculated for the COPM to identify the number of kids achieving a minimal clinically important difference.

## Results and discussion

Fifteen children were invited for pre-screening. Of those 15 children, 11 enrolled in the study. Reasons for not enrolling included too mildly impaired ($n = 1$), parents deciding to have child receive botulinum toxin therapy instead ($n = 1$), schedule of intervention was too demanding for caregiver work schedule/child's school schedule ($n = 1$) and unspecified ($n = 1$). One family enrolled in the study dropped out after four weeks with the caregiver indicating that the intervention schedule was too demanding. A total of 10 children completed the study. Demographic data for children that enrolled are listed in Table I.

## Feasibility

Ten families completed the entire nine weeks of intervention without any report of adverse events. On average, caregivers

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demonstrated high compliance completing 86.5 hours of H-HABIT with their children. The majority of activities included common, low cost supplies/toys appropriate for this age group (Table II). The most common type of activity performed included manipulative games/tasks (39% of all logged activities) and functional daily living tasks (22% of all logged activities). On average, families performed about 7.5 activities per day that lasted about 18.2 minutes per activity.

Home observations by the supervisor and monitoring of daily logs confirmed that treatment protocols were adhered to. Responses to the daily questionnaires were consistent across the sample with the majority of logs indicating that 80% of the time caregivers found it either very easy or easy to fit the training into their daily schedule, 86% the child was very attentive or attentive during the activities, 88% of the time the child tolerated the training either very well or well and that 79% of the time it was very easy or easy to carry out the training (Figure 1).

Parenting stress as measured by the PSI-SF showed no significant differences ($p > 0.05$) across the five assessments for either the total score or the three subscales of parental distress, parent–child dysfunctional interaction and difficult child (Table III). That is, there was no increase in parental stress during the intervention. All caregivers scored within one standard deviation of the normative range for this measure [13].

Table I. Participant characteristics.

| Child | Sex | Age (months) | Affected side | MACS | Gestation period | Enrolled in preschool |
|---|---|---|---|---|---|---|
| 1 | F | 45 | Right | II | Term | Yes |
| 2 | F | 43 | Right | III | Term | Yes |
| 3 | M | 34 | Right | II | Term | No |
| 4* | M | 45 | Right | I | Term | Yes |
| 5 | M | 52 | Left | III | Preterm | Yes |
| 6 | M | 45 | Right | II | Preterm | Yes |
| 7 | M | 47 | Right | II | Term | No |
| 8 | F | 29 | Right | IV | Term | No |
| 9 | F | 29 | Right | II | Term | Yes |
| 10 | M | 54 | Left | I | Term | Yes |
| 11 | F | 38 | Right | III | Term | Yes |

*Did not complete study.
MACS: Manual Ability Classification System.

## Bimanual and functional goal performance

Table III shows the means for the AHA and COPM at each time point. Children demonstrated significant improvements in the AHA, $F(3,27) = 4.64$, $p < 0.01$, $\eta^2 = 0.34$. Tukey's HSD *post-hoc* tests revealed no difference between the two baseline measures and significant differences between the six-month follow-up and the baseline period ($p < 0.05$). There was also no difference between the immediate post-test and the six-month follow-up. Net change scores revealed that 4 of 10 children improved the smallest detectable difference between the baseline period and the immediate follow-up,

Table II. Description and categorization of bimanual activities performed during H-HABIT.

| Activity category | Percent of total activities | Activity examples | Example of affected upper extremity use |
|---|---|---|---|
| Manipulative games/tasks | 39% | Molding clay, puzzles, board games, Legos, stacking cups | Stabilize construction piece while less-affected hand connects other pieces |
| Functional tasks | 22% | Dressing, undressing, cutting with scissors, eating | Hold and rotate paper during cutting with scissors |
| Miscellaneous | 11% | Book/page turning, games in bath tub, pretend play with figurines, bubbles | Supination of forearm while pouring water into cups during bath time |
| Fine motor | 10% | Beading, stickers, magnets, coin bank, marbles | Placing beads on pipe cleaner |
| Gross motor | 7% | Two-handed ball play, scooter handles, putt-putt golf, baseball | Throw and catch large ball with both hands |
| Arts and crafts | 6% | Painting, drawing, holiday-themed crafts | Stabilize/rotate paper while tracing shapes |
| Card games | 4% | Card flipping during matching | Simultaneously flip two cards |
| Video games | 1% | Tablet, computer | Holding/rotating tablet less-affected swipes on screen |

Table III. Means and 95% confidence intervals for assessments at each time point.

| | Baseline 1 | Baseline 2 | Midway | Posttest 1 | Posttest 2 | Test session effect $p$ value ($\eta^2$) |
|---|---|---|---|---|---|---|
| PSI-SF total | 58.6 (49.7–69.3) | 61.4 (48.5–69.3) | 57.3 (46.5–68.3) | 56.8 (44.9–66.5) | 58.5 (45.5–65.1) | >0.05 (0.01) |
| PD | 19.9 (13.8–24.3) | 19.8 (11.7–22.3) | 18.6 (11.6–22.8) | 16.2 (12.0–22.5) | 19.8 (11.8–22.4) | >0.05 (0.00) |
| PCDI | 18.0 (10.1–24.3) | 18.8 (9.6–25.2) | 17.6 (9.2–25.6) | 19.6 (10.0–26.06) | 22.3 (12.1–28.07) | >0.05 (0.02) |
| DC | 20.1 (10.8–34.8) | 22.2 (10.6–35.0) | 20.3 (8.8–33.6) | 20.3 (10.4–35.6) | 20.6 (9.0–36.2) | >0.05 (0.00) |
| AHA | | | | | | |
| AHA units (0–100) | 56.2 (45.8–66.6) | 56.4 (47.6–65.2) | – | 59.2 (47.5–67.7) | 61.7 (51.7–71.7) | <0.01 (0.34) |
| COPM | | | | | | |
| Performance | – | 2.9 (2.1–3.7) | – | 5.6 (4.6–7.3) | 6.5 (5.33–7.6) | <0.05 (0.26) |
| Satisfaction | – | 3.00 (2.1–3.9) | – | 6.0 (4.6– 7.4) | 6.6 (5.8–7.4) | <0.05 (0.24) |

PSI-SF: parental stress index-short form; PD: Parental Distress Subscale; PCDI: Parent–Child Dysfunctional Interaction Subscale; DC: Difficult Child Subscale; AHA: Assisting Hand Assessment; and COPM: Canadian Occupational Performance Measure.

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and 5 of 10 between the baseline period and six-month follow-up.

The majority of functional goals established for the COPM were bimanual and related to dressing (e.g. putting on socks). Children showed a significant improvement in the performance domain of COPM $F(2,27) = 4.43$, $p < 0.05$, $\eta^2 = 0.26$. The *post-hoc* test indicated a significant difference between the baseline period and immediate posttest which was maintained at six-month follow-up, $p < 0.001$. Children also showed a significant improvement in the satisfaction domain of COPM, $F(2,27) = 3.88$, $p < 0.05$, $\eta^2 = 0.24$. The *post-hoc* test indicated a significant difference between the baseline period and immediate posttest, which was maintained at six-month follow-up, $p < 0.01$. For both domains of the COPM, 7 of 10 children achieved a minimal clinically important difference (i.e. > 2).

The aim of this study was to examine the feasibility of caregiver-directed, home-based HABIT (H-HABIT) and to collect preliminary data in preparation for a randomized trial. Caregivers demonstrated a high rate of compliance and commitment to completing the entire duration of the intervention and indicated that the children tolerated the activities. Importantly, they were able to complete the intervention without increasing their levels of parental stress. Improvements in bimanual performance were evident and the majority of children exceeded the minimal clinically important difference for functional goal performance. These results suggest that H-HABIT is a feasible model that warrants further exploration to examine its efficacy for improving hand-function in children with USCP.

One risk of including caregivers in the delivery component of an intervention is the potential burden created by the demands of the schedule. In order to reduce potential stress, we trained caregivers to delineate the daily hours of the intervention from the child's typical daily routine and to avoid verbal prodding to use the affected hand outside of the daily two hours. The stability of parental stress throughout the intervention period and feasibility indicated in the daily logs suggest that this may have been an effective strategy for avoiding disruption of the psychosocial family dynamic. It has previously been shown that the increased caregiving demands resulting from having a child with CP directly affects caregiver health [20] and that caregivers of children with hemiparesis are up to twice as likely to report increased levels of stress relative to parents of typically developing children [21]. Caregivers in this study demonstrated parental stress levels that are similar to caregivers of typically developing children [13]. Thus, it should be noted that the sample of caregivers participating in this study might come from a highly selective population of caregivers of children with CP. Information about parental stress could serve as a useful screening tool to identify families that might be good candidates for the model of home-based intervention used in this study. However, future studies are needed to address this question directly.

Definitive conclusions about the efficacy of H-HABIT cannot be drawn from the data in this study given that it lacked a comparison control group. Regardless, the results are encouraging as about half of the children improved the smallest detectable difference for the AHA and the majority achieved a minimal clinically important difference for the COPM. Although the children as a group demonstrated a linear trend for improvement on the AHA, the *post-hoc* tests only revealed significant differences between the baseline and six-month follow-up assessment. It is conceivable that the improvements might be related to development of hand function associated with age as opposed to the intervention itself. However, almost all of the children in this study were beyond the age (i.e. 3 years) at which higher functioning children (i.e. can grasp with affected hand by 18 months of age) tend to reach their maximum limit on the AHA [4]. Thus, the rates of change between the immediate post-test and the 6-month follow-up appear to exceed what one would expect on average for a high functioning group. Moreover, a larger sample may have revealed significant differences between the baseline and immediate post-test.

In summary, this study demonstrated that a home-based model of bimanual training using caregivers as interventionists is a feasible approach for improving hand function in children with USCP. A randomized trial examining this intervention model could provide an invaluable clinical intervention tool that could be used to target the deficits in children with USCP.

## Declaration of interest

The authors report no conflicts of interest.

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# APPENDIX C



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*ARTICLE*

# Efficacy of Constraint-Induced Movement Therapy and Bimanual Training in Children with Hemiplegic Cerebral Palsy in an Educational Setting

N. Gelkop[1], D. Gol Burshtein[1], A. Lahav[1], A. Brezner[2], S. AL-Oraibi[3], C. L. Ferre[4], & Andrew M. Gordon[4]

*[1]Meshi Kindergarten, Jerusalem, Israel, [2]Sheba Medical Centre, Jerusalem, Israel, [3]El Ashmite University, Amman, Jordan, [4]Department of Biobehavioral Science, Teachers College, Columbia University, New York, USA*

**ABSTRACT.** We examined the efficacy of modified constraint-induced movement therapy (CIMT) and hand-arm bimanual intensive therapy (HABIT) in a special education preschool/kindergarten in Israel. Twelve children (1.5–7 years) with congenital hemiplegic cerebral palsy were randomized to receive modified CIMT ($n = 6$) or HABIT ($n = 6$). Occupational and physical therapists administered usual and customary care for 8 weeks; children then crossed over to receive CIMT or HABIT 2 hr/day, 6 days/week for 8 weeks from their occupational therapist. The Assisting Hand Assessment and Quality of Upper Extremity Skills Test were administered 2 months prior to the intervention, immediately before, immediately after intervention, and 6 months after the first baseline assessment. Both groups demonstrated no change during baseline and comparable improvement following CIMT and HABIT ($p < .001$), which was maintained at 6-month follow-up. Results suggest that modified CIMT and HABIT provided in school-based settings can lead to improvements in quality of bimanual skill and movement patterns.

**KEYWORDS:** Bimanual training, cerebral palsy, constraint-induced movement therapy, hand function, hemiplegia, intervention studies

Hemiplegic cerebral palsy is characterized by motor impairments largely confined to one side of the body. Nevertheless, even the "less-affected" upper extremity (UE) has subtle motor impairments (e.g., Brown et al., 1987; Gordon et al., 1999, 2003), and importantly, bimanual deficits can be seen when children with

Address correspondence to: Andrew M. Gordon, PhD, Department of Biobehavioral Science, Teachers College, Columbia University, 525 West 120th Street, Box 93, New York, NY 10027, USA (E-mail: ag275@tc.columbia.edu). AQ2

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hemiplegia are asked to perform tasks that involve specific spatial-temporal demands between the two hands (e.g., Hung et al., 2004, 2010).

For a little over a decade, constraint-induced movement therapy (CIMT) has been used for targeting impairments in hand function in children with hemiplegia. CIMT involves restraint of the less-affected hand and intensive practice with the more affected hand. There is a growing body of evidence demonstrating the effectiveness of CIMT when used with children with hemiplegia (for reviews, see Gordon, 2011; Gordon, in Press; Hoare et al., 2007; Sakzewski et al., 2009). Another approach used by clinicians treating impaired hand function in children with hemiplegia involves bimanual training. Hand-arm intensive bimanual therapy (HABIT) is a form of bimanual intervention that maintains the intensity and progressive task-specific practice of CIMT, but is performed in the context of bimanual activities (Charles & Gordon, 2006). Randomized clinical trials have demonstrated that HABIT results in improved quality and quantity of bimanual upper extremity use (Gordon et al., 2007, 2008) and is equally effective as CIMT in improving unimanual hand function and spontaneous bimanual hand-use (Gordon et al., 2011).

The emerging evidence demonstrating the efficacy of HABIT and CIMT, while promising, has been in most part restricted to children 3.5 years of age and older in day-camp or intensive one-on-one settings (6 hr/day of therapy). These settings permit CIMT and HABIT to be administered intensively and typically require one-on-one supervision by an interventionist for several hours per day (DeLuca et al., 2012). However, 6-hr sessions might not be appropriate for younger children, are not available in many communities, and may create caregiver burden associated with scheduling and commuting (Wallen et al., 2008). In response to some of these limitations, interventions with modified schedules have been developed.

Eliasson et al. (2005, 2011) modified the amount of time a constraint is worn to 2 hr each day and used parents and/or preschool teachers in the children's home or preschool setting to perform therapy. These studies demonstrated positive effects of modified CIMT (mCIMT) for improving hand function. Using a home-based program involving parents, Wallen et al. (2011) compared mCIMT with a form of intensive goal-directed training. Children in both groups showed equal improvement following an 8-week period on parent-rated measures of occupational performance. Despite the positive changes, a majority of parents (75%) did not find it easy to carry out mCIMT (Wallen et al., 2011).

Although there is growing support for interventions with modified schedules as an effective rehabilitation approach, it is unclear whether the models that have been studied are sensitive to the challenges faced by families in need of rehabilitation services. Services for families of children with disability should address the diverse needs of the child and family (King et al., 2002). The interventions described by Eliasson et al. (2005, 2011) and Wallen et al. (2011) involved significant participation by the family in a home setting when the desired intensity of therapy could not be met in the school alone. In many cases, logistical and cultural barriers prevent families from being active participants in therapy. Regardless of these barriers, service approaches for young children with CP tend to be dictated by the severity of disability while ignoring the degree of family difficulties (Parkes et al., 2004; Weindling et al., 2007). While there is support for the efficacy of intensive rehabilitation when provided in intensive 6 hr/day settings or with the active involvement of the

AQ3

child's family, these approaches may not provide the optimal structure given that many children do not receive or have access to services in such a context.

A recent study attempted to describe the pattern of use of rehabilitation services in children with CP in Canada (Majnemer et al., 2013) and found that an overwhelming majority of children received physical therapy or occupational therapy at the school setting in the form of weekly interventions. More than half of the children were in regular schools (53%) and just under half were in special (segregated) schools (45%). Interestingly, only 10% of the children in the study received therapy in rehabilitation centers and less than 5% through private services. Despite this observation, intensive, adapted models of intervention (i.e., CIMT or HABIT) have yet to be tested when provided exclusively in a school setting.

In the United States, children with CP are often mainstreamed in regular schools and intensive interventions such as CIMT or HABIT are not usually provided. Therapy in this context tends to focus on educational issues or strategies to increase school participation. In Israel, however, therapy is embedded within the daily school activities. This is particularly the case for children who attend special rehabilitation schools that combine an educational and rehabilitation curriculum. One advantage of this type of setting is that it offers an opportunity to provide rehabilitation at a high intensity within the context that the more affected hand will be used.

The purpose of this study was to examine the efficacy of intensive upper extremity training (CIMT and HABIT) when provided exclusively in a special education preschool/kindergarten using the child's usual and customary care provider. By modifying the schedule of the children's typical therapy and providing a very specific technique, the study attempted to make a comparison of a baseline period of usual and customary care with a period of intensive therapy using the same therapists in the school setting. A second, exploratory aim of the study was to compare the efficacy of CIMT versus HABIT in this particular setting.

## METHOD

### Design

The intervention was administered during the children's regular preschool or kindergarten hours. Participants were evaluated on four separate occasions including two baseline assessments and two post-intervention assessments (Figure 1). Children were evaluated 2 months before the start of the intervention (pre-baseline) and immediately before the start of the intervention (post-baseline). During this baseline period, children received their usual and customary care therapy before being crossed over to receive CIMT or HABIT. Children were then tested immediately after the study intervention (immediate post-intervention), and 2 months after the completion of the intervention (2-month post-intervention). All children were assessed by physical therapists blinded to group allocation. Two primary outcome measures were used to (1) quantify bimanual performance under the International Classification and Functioning and Health (ICF) "activity and performance" domain (i.e., Assisting Hand Assessment [AHA]) and (2) to

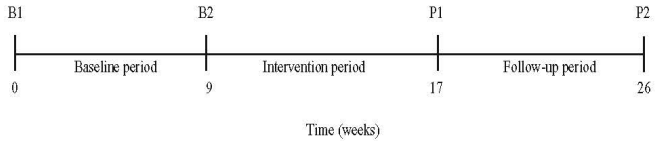


**FIGURE 1.** Timeline of study design. B1 – pre-baseline, B2 – post-baseline, P1 – immediate post-intervention, P2 – 2-month post-intervention.

measure upper extremity function related to the "body function" domain (i.e., Quality of Upper Extremity Skills Test [QUEST]) of the ICF.

### *Participants*

Participants were recruited from classrooms at the Meshi Children's Rehabilitation Center in Jerusalem, a preschool/kindergarten for children with physical disabilities. All children between the ages of 1.5 to 7 years with congenital hemiplegia were invited to participate. The inclusion criteria included: (1) ability to extend wrist 20°, (2) ability to release objects from the hand, and (3) age-appropriate cognitive ability as identified by child's placement in age-appropriate classes and based on evaluations by school psychologists. Children were excluded from the study if they received an intensive therapeutic intervention involving the upper extremities or botulinum toxin therapy in the upper extremity within the past 6 months or intended any new treatment within the study period. Two groups were created by matching children according to age, cognitive level (class level), and initial hand function as determined by AHA and QUEST scores. The two groups as a whole were then randomized using concealed allocation to receive either CIMT or HABIT.

A total of 16 children were invited to participate. One child was excluded because of recent participation in an intensive intervention; one child was excluded because the lack of the ability to extend the wrist actively; and two children were excluded because they were unable to actively release objects from the hand. A total of 12 children completed the study. Informed consent was obtained from all the children's parents and an age-appropriate assent script was used to inform the children that they would be participating in a study. All children continued to receive their customary individual therapy during the course of the study. The study was approved by the local ethics committee. Table 1 describes the participant characteristics.

Sample size calculations were performed based on AHA scores derived from an earlier HABIT study (Gordon et al., 2007). A mean improvement of $0.94 \pm 0.54$ logits for the HABIT group was reported. With $\alpha = 0.05$ and $1 - \beta$ (statistical power) $= 0.67$, 12 participants total were required.

### *Measures*

The *AHA* (*version 4.3*) is a validated and reliable measure that quantifies the effectiveness with which a child with unilateral disability uses his/her affected (assisting) hand in a bimanual activity (Krumlinde-Sundholm et al., 2007; Holmefur et al.,

**TABLE 1. Characteristics of Child Participants by Intervention Group**

| Characteristics | CIMT ($n = 6$) | HABIT ($n = 6$) |
|---|---|---|
| Mean age (SD), years | 4.25 (1.58) | 4.33 (1.86) |
| Gender | | |
| Male | 1 | 1 |
| Female | 5 | 5 |
| Paretic hand | | |
| Right | 3 | 3 |
| Left | 3 | 3 |
| MACS # (Only for <4 years) | | |
| I | 1 | 1 |
| II | 1 | 3 |
| III | 2 | 1 |

CIMT = constraint-induced movement therapy, HABIT = hand-arm bimanual intensive training, MACS = manual ability classification system.

2007). The AHA is scored from video recordings of 12–14 play activities. These activities are subsequently scored based on 22 predefined items using a 4-point rating scale. The raw score sum ranges from 22 (low ability) to 88 (high ability). Rasch analysis provides measures of equal intervals in logits (log odds probability units) by converting ordinal rating scale observations to interval level. Interval level measures for the AHA range from –10.18 to 8.70 logits. In order to facilitate interpretation of results, the logit scale is converted to a user-friendly 0 to 100 scale that is still Rasch-based and presents interval level data (i.e., AHA units) (Krumlinde-Sundholm, 2012). In our study, administration of the AHA was videotaped and then scored by one of two therapists certified to score the AHA. Each therapist scored half of the videotapes.

The *QUEST* (Law et al., 1991) was used to characterize upper extremity function in four domains: dissociated movement, grasp, protective extension, and weight bearing. The assessment is validated in children with cerebral palsy ages 18 months to 8 years, is sensitive to change, and has been shown to be reliable (0.92 = test–retest total score reliability and 0.84–0.94 = test–retest reliability for subcategories) (Haga et al., 2007). Two participants (one from each group) were unable to complete the assessment due to lack of cooperation).

The *Manual Ability Classification System (MACS)* (Eliasson et al., 2006) was used by an experienced occupational therapist (who also served as the project supervisor) to classify the children into one of five levels based on how they use their hands when handling objects in daily activities. The performance-based classification system is designed for children between the ages of 4 and 18 years. The MACS has established high between-therapists reliability (0.97 = intraclass correlation coefficient) and validity (Eliasson et al., 2006).

### *Procedure*

#### *Usual and Customary Care*

During the baseline period of usual and customary care (UCC), children received 2–3 sessions of occupational and 2–3 sessions of physical therapy per week, with each session lasting 45–60 min. The activities during these sessions were based on

principles of neurodevelopmental theory (NDT) and goals defined by the parent and therapist at the beginning of the school year. The focus of the activities was to improve strength, range of motion, and awareness of the hand while using guided movements to influence the overall pattern of the child's movements. Stretching was also included as part of the child's activities. The therapy during the baseline period was provided with a 1:1 interventionist–child ratio.

*Constraint-Induced Movement Therapy and Hand-Arm Bimanual Intensive*

CIMT and HABIT are comprised of intensive, progressive task practice based on motor learning approaches. CIMT involves restraint of the less-affected upper extremity and progression of unimanual tasks. HABIT is performed in the absence of a restraint and tasks are progressed bimanually. The intervention was administered during the children's regular preschool or kindergarten hours. Each child randomized to the CIMT or HABIT group received 2 hr of therapy 6 days a week for a period of 8 weeks. The 2 hr included a 1-hr individual session and a 1-hr group session. During the individual sessions, children worked one-on-one with their occupational therapist. Group sessions were carried out by either two or three interventionists and resulted in a 1:2 or 1:1 interventionist to child ratio. The interventionist team for group sessions consisted of occupational therapists (OTs) and therapist assistants. Prior to the intervention, an experienced supervisor instructed all of the OTs and interventionists in the principles of motor learning and the specifics of both CIMT and HABIT using a standardized manual of procedures. All interventionists were monitored by an experienced supervisor and participated in group meetings three times a week in which they received feedback about how to administer the intervention, ideas for activities, and strategies for increasing cooperation from the children.

For both interventions, children were engaged in an individualized program according to his/her specific ability. Interventionists were instructed to place emphasis on making the intervention enjoyable and intrinsically motivating. Task difficulty was graded by changing the task constraints, which required progressive skill in hand use and increasingly providing tasks involving greater difficulty. The grading of activities by interventionists was dependent on the individual abilities of each child. Strategies for grading activities and changing the constraints of the tasks were discussed in group meetings. Children participated in whole- and part-task practice. Whole-task practice involved executing a sequence of movements within the context of tasks (e.g., puzzles) and was performed continuously during a play session lasting for at least 15 to 20 min. The focus of the task was targeted movements and spatial and temporal coordination within the context of completing an activity. Part-task practice was used to break down motor skills into smaller components (e.g., turning puzzle pieces to promote forearm supination) while increasing repetitions and skill. Part-task practice (Schmidt & Lee, 1999; Winstein, 1991) was used to provide repetitions of movement components embedded in tasks (see Winstein et al., 2003). For example, during a card game, we could ask the participant to turn over as many cards as possible in a 30-s period. The movement, which is part of the game of cards, would focus on supination, which is limited in many participants. The

task could be initially adapted to make grasping easier (e.g., beveling the cards) and progressively made more difficult by removing the adaptations.

Age-specific encouragement was provided to ensure activities were highly motivating and fun and age-specific instruction was provided to improve performance on subsequent trials. Interventionists tracked compliance by recording daily task performance using a daily log in which they indicated the activity performed and time spent on each activity. Compliance reports were reported daily to the project supervisors. Interventionists were told to avoid using manual guidance in order to encourage spontaneous movements by the child. Activities included activities of daily living (e.g., cleaning, eating) and playing with an assortment of child-friendly games performed indoors and outdoors.

*Procedures Specific to Constraint-Induced Movement Therapy*

Based on previous studies (see Gordon et al., 2005), CIMT was modified to ensure that the therapy was child-friendly. Children wore custom-made gloves on the less-affected hand, and unimanual activities were performed with the affected hand. The glove was only worn during the 2 hr of CIMT. Children performed fine-motor and gross-motor activities that elicit general movements of interest and included functional unimanual and play activities that were tailored to the children's age range. Interventionists assisted when necessary to complete activities. For example, when cutting fruit, the interventionist stabilized the fruit, while the child cut with a toy knife.

*Procedures Specific to Hand-Arm Bimanual Intensive Therapy*

HABIT was performed without a restraint, but rather, participants were engaged in age-appropriate fine and gross motor bimanual activities (Charles & Gordon, 2006). Activities were chosen based on the ability of the child's paretic hand and focused on using the assisting hand for tasks requiring increasingly complex bimanual coordination. Task demands were graded and the children were encouraged to be active in identifying movements to complete an action (i.e., problem solving). Interventionists avoided using verbal requests to use the paretic hand as much as possible, and instead modified the environment by providing tasks that required the use of both hands to elicit desired movements.

Participants in the CIMT and HABIT groups completed an average of 70 out of 80 hr (88%) of structured practice. 60% was spent in group sessions and 40% was spent in individual sessions. No adverse events were reported. According to the daily reports and group meetings, there was full compliance in both groups.

### *Data Analysis*

A 2 (Group) $\times$ 4 (Test sessions) ANOVA with repeated measures on test sessions was performed on the logit-based AHA units for the AHA and on raw and log-transformed QUEST data using SPSS 15. Data for the QUEST were transformed to ensure that the distribution of the sample did not affect the results. Mauchly's test of sphericity indicated that the assumption of sphericity had not been violated for either the AHA ($\chi^2_{(5)} = 3.35, p = .647$) or QUEST ($\chi^2_{(5)} = 7.85, p = .169$). Given the qualitatively similar findings between raw and transformed data for the QUEST, the raw data are reported. The Group $\times$ Test session interaction

examined whether there was a difference over time between the treatment groups. Eta-squared values ($\eta^2$), an effect size estimate, are reported and can be interpreted as the proportion of total variability in the outcome measures that is accounted for by variation in the predictor (Levine & Hullett, 2002). Given that main effects for the ANOVA only indicate that there is difference between the means, planned comparisons were used to determine which differences in means at each time point were significantly different from each other. Bonferroni corrections were used with a significance level of $p < .01$. Intent to treat principles were used for the analysis.

## *RESULTS*

Table 2 includes descriptive statistics for the HABIT and CIMT groups at each time point and the change score (i.e., post-baseline to immediate post-intervention) as well as the corresponding ANOVA details. There were no significant baseline differences between the participant characteristics in the CIMT and HABIT groups for any of the measures ($p > .05$ for all measures). The AHA data revealed a main effect of testing session with the means of the two groups improving over time ($p < .05$). There was no main effect of group ($p > .05$) indicating no average difference between the two groups. Planned comparisons revealed that the main effect of test session was due to significant differences between post-baseline and immediate post-intervention with the average of the groups improving from 45.2 to 55.8 AHA units ($p < .05$). The difference was maintained at 2-month post-intervention with the groups scoring 9.5 AHA units higher than the baseline period. AHA-unit scores improved 11.7 and 9.5 for the CIMT and HABIT groups, respectively (Figure 2), although there was no Group $\times$ Test session interaction. On an individual level, 10 out of the 12 children (5 out of 6 in each group) improved $\geq$5 AHA units, thereby exceeding the smallest detectable difference (Krumlinde-Sundholm, 2012).

A main effect of testing session was also present for the QUEST total score with AQ5 scores improving over time ($p < .05$) (Table 2). Planned comparisons revealed significant differences between post-baseline and immediate post-intervention with the average of the groups improving from 55.9 to 72.2. The difference was maintained at 2-month post-intervention with a mean of 71.5. There was no main effect for group ($p > .05$). Children improved 19 and 13.6 standardized scores for the CIMT and HABIT groups, respectively (Figure 3). There was no Group $\times$ Test session interaction ($p > .05$) suggesting that both groups improved equally over time. On average, children in the CIMT and HABIT groups improved 35% and 24%, respectively, with both groups, thereby exceeding the smallest detectable difference (i.e., 14%) for the QUEST (Klingels et al., 2008). Separate analyses were also conducted on subtest scores for the QUEST. A main effect of testing session was seen for all of the scores ($p < .05$) indicating improvement over time. For all of the subscores except the dissociated movements test, there was no Group $\times$ Test interactions. The interaction for the dissociated movements subscore indicated children in the CIMT group demonstrated a greater change ($p < 0.05$) between post-baseline and immediate post-intervention compared with children in the HABIT group.

**TABLE 2.** Mean (95% Confidence Interval) for all Measures by Intervention Group at Each Time Point

| | Pre-baseline (95% CI) | Post-baseline (95% CI) | Immediate post-intervention (95% CI) | 2-month post-intervention (95% CI) | Change score: Post-baseline to immediate post-intervention (95% CI) | Test session effect: $p$ value ($\eta^2$), $F$ value (df) | Interaction (Test session × Group): $p$ value ($\eta^2$), $F$ value (df) |
|---|---|---|---|---|---|---|---|
| **AHA (AHA units)** | | | | | | | |
| CIMT | 43.5 (28.5, 58.5) | 47.3 (32.4, 62.3) | 59.0 (46.7, 71.3) | 57.2 (45.7, 68.7) | 11.7 (4.3, 19.1) | – | – |
| HABIT | 43.7 (28.7, 58.6) | 43.0 (28.0, 58.0) | 52.5 (40.2, 64.8) | 52.2 (40.7, 63.7) | 09.5 (2.1, 16.9) | – | – |
| Mean | 43.6 (33.0, 54.2) | 45.2 (34.6, 55.8) | 55.8 (47.0, 64.5) | 54.7 (46.5, 62.8) | 10.6 (5.6, 15.6) | **$p < .001$ (.68)** **$F(3,30) = 23.20$** | **$p = .48$ (.02)** **$F(3,30) = .82$** |
| **QUEST total** | | | | | | | |
| CIMT | 55.6 (37.2, 74.0) | 55.0 (35.4, 74. 6) | 74.0 (56.9, 91.2) | 73.0 (57.9, 88.1) | 19.0 (8.4, 29.6) | – | – |
| HABIT | 54.8 (36.4, 73.2) | 56.8 (37.2, 76. 4) | 70.4 (53.3, 87. 6) | 70.0 (54.9, 85.1) | 13.6 (3.0, 24.2) | – | – |
| Mean | 55.2 (42.2, 68.2) | 55.9 (42.0, 69.8) | 72.2 (60.1, 84.3) | 71.5 (60.8, 82.2) | 16.3 (9.1, 23.6) | **$p < .001$ (.78)** **$F(3,24) = 30.92$** | **$p = .671$ (.01)** **$F(3,24) = .52$** |
| **QUEST prot.** | | | | | | | |
| CIMT | 50.8 (22.0, 79.6) | 44.4 (17.5, 71.3) | 60.7 (40.1, 81.2) | 58.3 (35.0, 81.7) | 16.2 (2.6, 29.9) | – | – |
| HABIT | 46.7 (17.9, 75.5) | 49.7 (22.8, 76.6) | 57.2 (36.7, 77.7) | 58.8 (35.4, 82.2) | 07.6 (–6.1, 21.2) | – | – |
| Mean | 48.8 (28.4, 69.1) | 47.0 (28.0, 66.1) | 58.9 (44.4, 73.5) | 58.6 (42.1, 75.1) | 12.0 (2.4, 21.4) | **$p < .005$ (.41)** **$F(3,24) = 5.95$** | **$p = .564$ (.05)** **$F(3,24) = .70$** |
| **QUEST diss.** | | | | | | | |
| CIMT | 59.7 (45.1, 74.3) | 58.4 (46.2, 70.6) | 79.4 (65.9, 92.9) | 73.1 (62.8, 83.5) | 20.9 (14.1, 27.8) | - | - |
| HABIT | 64.4 (49.8, 79.0) | 65.6 (53.4, 77.8) | 70.0 (56.5, 83.5) | 73.8 (63.5, 84.1) | 04.4 (–2.5, 11.3) | - | - |
| Mean | 62.0 (51.7, 72.4) | 62.0 (53.4, 70.7) | 74.7 (65.1, 84.3) | 73.5 (66.2, 80.8) | 12.7 (5.00, 20.4) | **$p < .001$ (.57)** **$F(3,24) = 17.01$** | **$p < .05$ (.16)** **$F(3,24) = 4.67$** |
| **QUEST weight.** | | | | | | | |
| CIMT | 56.2 (26.1, 86.3) | 60.7 (27.2, 94.2) | 86.8 (56.8, 116.9) | 86.4 (62.1, 110.7) | 26.2 (–6.5, 58.7) | – | – |
| HABIT | 48.4 (18.3, 78.5) | 55.6 (22.1, 89.1) | 84.4 (54.4, 114.5) | 73.2 (49.0, 97.5) | 28.82 (–3.7, 61.4) | – | – |
| Mean | 52.3 (31.1, 73.6) | 58.1 (34.4, 81.8) | 85.6 (64.4, 106.9) | 79.8 (62.7, 97.0) | 27.5 (6.2, 48.8) | **$p < .001$ (.57)** **$F(3,24) = 10.85$** | **$p = .883$ (.01)** **$F(3,24) = .22$** |
| **QUEST grasp.** | | | | | | | |
| CIMT | 54.8 (42.0, 67.6) | 54.8 (38.3, 71.3) | 68.9 (54.3, 83.5) | 73.3 (57.7, 89.0) | 20.9 (14.1, 27.8) | – | – |
| HABIT | 60.7 (47.9, 73.6) | 56.6 (40.1, 73.5) | 68.9 (54.3, 83.5) | 71.9 (56.2, 87.5) | 04.4 (–2.5, 11.3) | – | – |
| Mean | 57.8 (48.7, 66.8) | 55.7 (44.0, 67.4) | 68.9 (58.6, 79.2) | 72.7 (61.5, 83.6) | 13.2 (6.2, 20.1) | **$p < .001$ (.68)** **$F(3,24) = 18.46$** | **$p = .566$ (.03)** **$F(3,24) = .69$** |

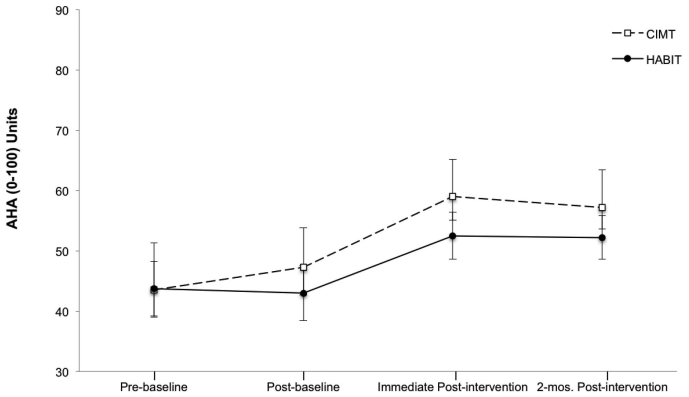


**FIGURE 2.** Mean ± standard error of the mean for AHA at four time points by intervention group.

## *DISCUSSION*

Constraint-induced movement therapy and hand-arm bimanual intensive therapy resulted in improvements in the quality of bimanual hand-use and upper extremity function when administered 2 hr a day over a period of 8 weeks by the child's usual

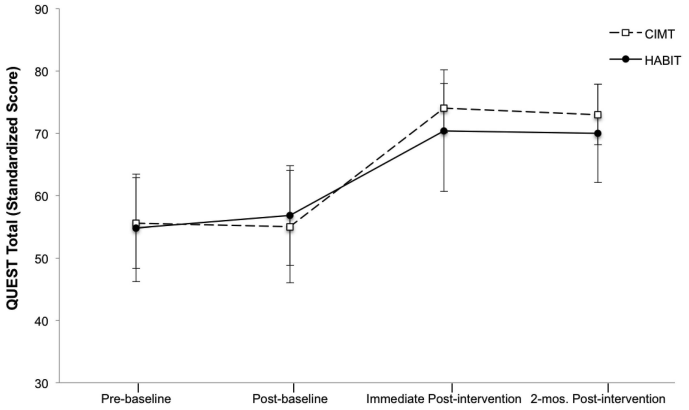


**FIGURE 3.** Mean ± standard error of the mean for QUEST at four time points by intervention group.

and customary care provider. These results suggest that improvements in hand function can be attained even when modifying the amount of daily therapy and having the therapy performed by clinicians solely in a special rehabilitation school setting. The setting for the therapy in this study is of particular significance given that in some countries, children are most likely to receive services through special education schools. The study illustrates how children's typical therapy schedules can be modified and provided at a higher intensity in a specific educational setting. The improvements associated with the interventions were maintained at a 2-month follow-up. The gains achieved in this study were comparable to previous studies of adapted models of intervention. This is also the first study to our knowledge to test bimanual training in a special rehabilitation school setting. These findings have important implications, given the ability to provide therapy in cost-effective settings in which active participation by families is not feasible.

Similar to the results found in previous studies of modified forms of CIMT training in young children (Eliasson, 2005; Eliasson et al., 2011), participants in this study improved significantly following an intensive period of training. However, unlike other studies of modified schedules of therapy which have used a combination of parents, school teachers, and/or therapists across different environments to fulfill the intervention requirements, the intervention in this study examined how children responded when their usual and customary care provider from a specific special education setting was trained to provide the intensive therapy.

The eta-squared ($\eta^2$) indicated that about 68% and 78% of the variance in AHA and the QUEST, respectively, could be attributed to the treatments (Table 2). The comparable efficacy between the two groups in this study is also consistent with studies in older children comparing CIMT with intensive bimanual therapy (e.g., Gordon et al., 2011; Sakzewski et al., 2011). Despite differences in baseline levels across individuals in our sample, an overwhelming majority of the children exceeded the smallest detectable difference for improvement in the AHA. AQ6

Our findings add to a growing body of evidence that suggests CIMT and bimanual training are effective in improving hand function in children with hemiplegia. The data also suggest that improvements in hand function can be attained without extensive one-to-one supervision by an interventionist. By providing young children with access to intensive therapy, interventions can be administered during a developmental period in which the nervous system might exhibit greater plasticity.

The method of implementing CIMT and HABIT examined in this study is in accordance with the approach described by Eliasson et al. (2011) and is based on principles of motor learning (Schmidt & Wrisberg, 2001; Shea & Wright, 1995) and theoretical frameworks that place emphasis on the importance of the child in relation to the developmental context and interactions at multiple levels of analysis (Brofenbrenner & Morris, 1998; Gottlieb, 1991). The latter frameworks highlight the importance of the settings (e.g., preschool, home, health care programs) in which the child spends most of his/her time and how development is highly contingent upon the experiences encountered in these environments. Providing therapy in the child's typical environment can have important positive effects on his/her motor development. The motor learning framework suggests adapting the environment as a way to elicit changes in motor behaviors (e.g., handing an object at higher location in order to elicit wrist extension).

The comparable improvements between children in the HABIT and CIMT group provide clinicians with different sets of tools that can be used when designing interventions. For example, in situations where extensive one-to-one supervision is not always possible, CIMT might be more appropriate given that children are less likely to compensate with the less-affected hand when performing activities while wearing a restraint. If the goal of the therapy is to achieve better performance of functional goals or bimanual coordination, HABIT might serve as a more practical approach. It is important to note, however, that it remains to be determined which type of therapy might best suit different ages or level of severity (Gordon, 2011).

Finally, the data indicated that there were significant changes following the treatment period, and no change during the baseline period during which children received their NDT-based usual and customary care, suggesting that it may be important for therapists to reconsider treatment in school environments. Specifically, perhaps therapy should be given intensively in bursts (e.g., 2 months a year), with no treatment until the next intensive burst (i.e., consolidate the normal therapy hours into a shorter time period). However, it is important to point out that this consideration depends on what treatment is given and the environment in which it is provided. It is also important to consider that following the intensive intervention, children continued to receive ongoing therapy that may serve to reinforce the gains for retention. Further work is needed to determine how best to adjust dosing schedules across different types of school settings.

### *Limitations*

Although participants in both groups made significant improvements in their hand function, this study did not include a no-intervention group, thus making it difficult to determine whether the improvements would have also been seen in a group of children that did not receive an intervention. However, no difference was found between the two baseline measurements across a period equal to the intervention period (2 months), a time during which children in both groups were receiving an equal amount of their usual and customary care, indicating that the measures are stable and any changes are likely to reflect improvements associated with the intervention. There was a slight trend for improvement in the AHA for children in the CIMT group during the baseline period, but this difference was not significant and was below the smallest detectable difference for this particular measure.

It is conceivable that the lack of differences between the two groups is a result of the small sample size in each group, and that a single-group pre–post study would have been sufficient to address the efficacy of one of these adapted models of therapy in this setting. However, both groups were included to provide exploratory data that could be used to generate hypotheses for subsequent studies. Interestingly enough, pairwise comparisons revealed significant pre–post changes for both groups, suggesting the relatively robust finding of change across time even with a small sample size. There was little suggestion of greater improvements of a given group, but this would need to be more rigorously tested in a larger, multisite, trial to determine whether this is the case. It is important to point out however, that the eta-squared ($\eta^2$) for the interaction parameters (Table 2) suggest that a very small proportion of the total variance is accounted for by any difference between the two groups over time. That is, there was little suggestion of a trend for favorable

improvements for one group over the other. The lack of interaction is also consistent with other studies involving direct comparisons between CIMT and bimanual training (e.g., Facchin et al., 2011; Gordon et al., 2011; Sakzewski et al., 2011).

It is also possible that the measures used in the study might lack the sensitivity or specificity needed to capture differences between the two groups. For example, an earlier RCT found similar improvements in the AHA and Jebsen-Taylor Test of Hand Function for children that participated in either HABIT or CIMT, however, children in the HABIT group made greater progress on goals related to activities of daily living (Brandao et al., 2012; Gordon et al., 2011), and movement kinematics improved more for the HABIT group (Hung et al., 2011). AQ7

Finally, although the participants in this group were followed up to 2 months past the intervention period, a 6-month follow-up period would be optimal, but was not possible because of the school year schedule.

## CONCLUSION

This study demonstrated the effectiveness of CIMT and HABIT for improving the quality of bimanual hand-use and movement in children with hemiplegia when provided in a modified schedule of 2 hr/day for 8 weeks. Importantly, the improvements were achieved by their usual and customary care therapist in a special education preschool/kindergarten. These results suggest that a feasible alternative to traditional day camp interventions can be implemented when performed solely by clinicians in a school setting.

***Declaration of Interest:*** The authors report no conflict of interest. The authors alone are responsible for the content and writing of this article. AQ8

## ABOUT THE AUTHORS

AQ9

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# APPENDIX D

*Clinical Research Article*

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## Comparison of Structured Skill and Unstructured Practice During Intensive Bimanual Training in Children With Unilateral Spastic Cerebral Palsy

**Marina B. Brandão, PhD[1], Claudio Ferre, MA[2], Hsing-Ching Kuo, MS[2], Eugene A. A. Rameckers, PhD[3-5], Yannick Bleyenheuft, PhD[6], Ya-Ching Hung, EdD[7], Kathleen Friel, PhD[8], and Andrew M. Gordon, PhD[2]**

**Abstract**

*Background.* High-intensity training aims to improve hand function in children with unilateral spastic cerebral palsy (USCP). However, the extent to which skill training is required is not known. *Objectives.* To compare the effects of intensive bimanual training with and without structured progression of skill difficulty, on manual dexterity, bimanual hand use, daily functioning, and functional goals in children with USCP. *Method.* Twenty-two children were randomized to structured practice group (SPG) or unstructured practice group (UPG), and received 6 h/d training during 15 days. Children from the SPG were engaged in fine and gross motor bimanual activities, with skill progression and goal training. Children from UPG performed the same activities without skill progression or goal training. Participants were evaluated before, immediately and 6 months after training by a physical therapist blinded to group allocation. The primary outcomes were the Jebsen-Taylor Test of Hand Function (JTTHF) and Assisting Hand Assessment (AHA). Secondary outcomes included the Canadian Occupational Performance Measure (COPM), Pediatric Evaluation of Disability Inventory (PEDI), and ABILHAND-Kids. *Results.* Both groups showed similar improvements in the JTTHF, AHA, ABILHAND-Kids, COPM-satisfaction, and PEDI ($P < .05$). A significant interaction in the COPM-performance scale ($P = .03$) showed superior improvements of the SPG immediately, but not 6 months, after the intervention. Conclusions: Children from both groups demonstrated improvements in dexterity and functional hand use. This suggests that for intensive bimanual approaches, intensive training at such high doses may not require structured practice to elicit improvements. However, there may be immediate added benefit of including goal training.

**Keywords**

upper extremity, hand, hemiplegia, cerebral palsy, bimanual training, intensive rehabilitation, training ingredients, constraint-induced movement therapy, goal practice, hand-arm bimanual intensive training (HABIT)

Unilateral spastic cerebral palsy (USCP) is characterized by motor impairments mainly lateralized to one side of the body, with the resulting impaired hand function affecting functional activity and participation. Increasing evidence supports the use of 2 treatment approaches based on motor learning principles, constraint-induced movement therapy (CIMT) and intensive bimanual training. CIMT involves physical restraint of the less-affected upper extremity (UE) along with intensive, skilled practice of the more-affected hand.[1-10] Bimanual training involves practice of functional and play activities requiring the use of both hands.[10-20] Hand-arm bimanual intensive training (HABIT) is a highly structured form of bimanual training.[11,13,19,21] Both CIMT and HABIT were designed with the idea that *intensity of training*[22,23] and *progression of skill* difficulty[24] are essential to improve motor function. They include part and whole practice, and modifying tasks to elicit desired movements and to ensure successful use of the more-affected hand.[21,24,25]

[1]Faculdade de Ciências Médicas de Minas Gerais, Belo Horizonte, Brazil
[2]Teachers College, Columbia University, New York, NY, USA
[3]Maastricht University, Maastricht, Netherlands
[4]Adelante Center of Expertise in Rehabilitation and Audiology, Hoensbroek, Netherlands
[5]University for Professionals for Pediatric Physical Therapy, AVANSplus, Breda, Netherlands
[6]Université Catholique de Louvain, Brussels, Belgium
[7]Queens College, City University of New York, Flushing, NY, USA
[8]Burke-Cornell Medical Research Institute, White Plains, NY, USA

**Corresponding Author:**
Andrew M. Gordon, Department of Biobehavioral Sciences, Teachers College, Columbia University, Box 93, New York, NY 10027, USA.
Email: ag275@columbia.edu

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Recently, several randomized trials have shown both approaches result in improvements in unimanual and bimanual measures.[12-18,20,26,27]

There is evidence to suggest that the behavioral demands of the tasks and motor skill training may elicit cortical reorganization[28] underlying concurrent functional outcomes. For optimal efficacy, it is believed that training must be challenging, involve progressive increase of behavioral demands and active participation. Studies of skilled training in animals show plasticity of UE cortical representation, whereas those animals receiving unskilled training (repetition only) did not.[28,29] Friel et al[30] investigated mechanisms for restoring motor function after unilateral corticospinal system damage in cats during early postnatal development. They compared skilled practice of the affected limb in a reaching task along with restraint of the unaffected forelimb limb in one group of cats (analogous to CIMT) with restraint only (analogous to forced use[31]) in another group. The skilled training group had improvements in motor skills of the affected limb along with expansion of the motor map, whereas the restraint-only group did not demonstrate plastic changes. Thus, skilled training is an important element in animal models of rehabilitation.

In humans, the extent to which skill training is required during intensive training is not well-understood. Uswatte et al[32] found similar outcomes of shaping and task-practice without skill progression during CIMT in adult stroke patients' hand function. The extent to which skilled practice is required for pediatric UE rehabilitation approaches is not known. One precision grip study in children with USCP demonstrated that task repetitions alone could lead to better motor performance of this task.[22] It is conceivable that intensity alone may be the key element to improvements in hand function; that is, at such high doses, specific components may not be important.

In the present study, we compare the effects of intensive bimanual training with and without structured progression of skill difficulty, on manual dexterity (Jebsen-Taylor Test of Hand Function, JTTHF) bimanual hand use (Assisting Hand Assessment, AHA), daily functioning (ABILHAND-Kids, Pediatric Evaluation of Disability Inventory, PEDI) and functional goals (Canadian Occupational Performance Measure, COPM) in children with USCP. We hypothesized that children in the structured practice group (SPG) would have greater improvements compared to children in the unstructured practice group (UPG) on all measures.

## Method

### Participants

Participants were recruited from area clinics, our Web site (http://www.tc.edu/centers/cit/), and online support groups. Potential participants were first screened via e-mail/telephone. Those between the ages of 6 and 13 years with congenital hemiplegia were invited to receive an on-site physical examination or an examination videotaped by their physical/occupational therapist. The inclusion criteria were established based on prior HABIT trials[11-13]: (1) the ability to lift the more-affected arm 15 cm above a table and grasp light objects, (2) mainstreamed in school, and (3) demonstrated ability to follow instructions during screening/testing. Exclusion criteria included (1) health problems unassociated with cerebral palsy, (2) current/untreated seizures, (3) visual problems interfering with treatment/testing, (4) severe muscle tone at any joint (Modified Ashworth score >3.5), (5) orthopedic surgery on the more-affected hand within 1 year, and (6) botulinum toxin in the more-affected UE within the past 6 months or intended treatment within the study period. Informed consent was obtained from participants and caregivers. The study was approved by the Teachers College Institutional Review Board.

### Procedures

*General Procedures.* Three bimanual training day-camps were conducted at the university from July 2010 to July 2012. Participants (6-8 each camp) were randomized offsite using concealed allocation stratified by age and JTTHF screening score, into an SPG and UPG. Each camp had 3 to 4 children per group, each group separated in different rooms.

Participants in each group were engaged in treatment 6 h/d for 15 consecutive weekdays (90 hours) during school recess by trained interventionists. These included graduate students in kinesiology/neuroscience, speech pathology, and psychology, and undergraduates. The interventionists were not aware of the study hypotheses or that different procedures would be carried out in each room. The pretreatment training, administered by the supervisors, was standardized during a 2-hour session based on established manual of procedures, before interventionists were assigned to a group and child. Group training focused on procedures common to the 2 groups, strategies to engage children in use of hands, safety and data logging procedures. Additional training was provided specific to each group during the interventions and daily team meetings. Interventionists and children in each room were kept partitioned at all times, and interventionists were instructed not to discuss the camp with each other outside of treatment hours. The 2 treatment rooms had supervisors, who were not blinded to the study hypotheses, and were responsible for ensuring treatment fidelity. The SPG was supervised by a physical therapist who modeled and ensured uniformity of treatment. The UPG was supervised by a nonclinician who ensured that interventionists did not provide structured practice or graduation of task complexity. Emphasis in this group was to engage children to use the more-affected hand in play

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activities. Both groups were monitored by an occupational therapist and a nonclinician who followed all the activities, to avoid possible confounds in the specificities of each training modality (ie, skill progression).

Room design permitted participants to work individually with their interventionist or in groups (1:1 interventionist:participant ratio). Interventionists were paired with children prior to randomization based on age, gender, and caregiver input. Interventionists avoided verbal prodding to use the more-affected hand, and instead provided tasks necessitating the use of both hands and established rules prior to each activity, allowing the child to choose which hand to use for different components of a bimanual activity. Caregivers were instructed to engage participants in bimanual activities for 1 h/d during and for 6 months following the intervention and document practice using home activity logs. No information regarding training of functional goals at home or skill progression was provided for either group.

*Structured Practice Procedures.* Children from the SPG were engaged in age-appropriate fine and gross motor bimanual activities using motor learning approaches consistent with HABIT.[23] Activities were selected by considering the role of the more-affected hand increasing in complexity from a nondominant passive assist (eg, stabilizing paper while drawing) to active manipulator (eg, reorienting paper while cutting) using increasingly complex bimanual coordination and participants' interests. Task demands were graded and participants were engaged in active problem solving.

Children participated in whole and part task practice. Whole task practice involved sequencing successive movements within the context of tasks (eg, card games). The activities were performed continuously for at least 15 to 20 minutes. The spatial and temporal coordination of targeted movements were practiced within the context of completing the task. Part task practice[21,25] required breaking down motor skills into smaller components (eg, playing-card turning to promote forearm supination), while increasing repetitions and skill requirements. This served to provide specificity/intensity of treatment by requiring as many targeted repetitions as possible over repeated 30-second intervals. Part practice included both bilateral symmetrical (eg, reaching toward object(s) with both hands) and asymmetrical (eg, pulling apart objects) movements. Task difficulty was graded by varying the spatial and temporal constraints, or by providing tasks that require progressive skilled use of the more-affected hand as performance improved. Task- and age-specific knowledge of results were provided for encouragement.[23] Supervisors instructed interventionists regarding the main focus of the treatment (eg, reaching) for each child based on his or her pronounced movement deficits, interests, and potential for improvement. Activity logs were collected, which included activities performed, time on task including part/whole practice, the number of repetitions, task performance and skill progressions.

Practice of functional goals established by caregivers was based on children's interests and abilities. Goal training was performed up to 30 min/d during camp. With the exception of goals, tested task items were never trained.

*Unstructured Practice Procedures.* Children from the UPG were engaged in age-appropriate fine and gross motor bimanual play activities (with the exception of functional goal practice) *with the same activities* without any adaptations or progression. As in the SPG, rules were established requiring use of their more-affected hand. Activities were selected from the same battery of choices as that of the SPG, according to child's interest and willingness to use the more-affected hand. Interventionists were trained only to provide activities that require the use of both hands in a playful and enjoyable context. Thus, the aim of the unstructured training was to provide children opportunities to use their more-affected hand in bimanual play activities, without considering how this hand was used or how use could be improved or challenged. Interventionists were specifically told "these children receive excessive therapy during the year, and that our purpose was to provide a fun, supportive environment to avoid frustration or perception that activities were therapeutic." Thus, the focus of training was only to keep the more-affected hand involved in the performance of the activities. Supervisors ensured that no increase in task complexity, verbal prompts how to use the more-affected hand or gradation of tasks demands were provided. Such supervision happened throughout the intervention and in daily meetings with interventionists. Activity logs were collected, which included activities performed and the time spent in these activities.

## Measures

Participants were evaluated directly prior to treatment (pretest), within 2 days ("immediate") and 6 months after treatment by a physical therapist blinded to group allocation (verified following testing). Two primary outcome measures were used to quantify bimanual and unimanual outcomes under the International Classification of Functioning and Health (ICF) "activity and participation" domain.[33]

The AHA version 4.3[34] quantifies the effectiveness with which a child with unilateral disability uses his or her affected (assisting) hand in *bimanual activity*. The AHA has excellent validity/reliability.[35] The test was videotaped and scored offsite by an experienced evaluator blind to group assignment. Data were reported in logit-based units (AHA-units).

The JTTHF is a standardized test of simulated functional tasks quantifying the time to complete a battery of unimanual activities.[36] The activities include flipping index cards,

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object placement, simulated eating, stacking checkers, and manipulating empty and full cans. Reliability for children with nonprogressive hand disabilities is high.[37]

Four secondary measures were also used (ICF "activity and participation domain").

To establish/evaluate children's functional goals we conducted the COPM with caregivers. The COPM identifies and measures changes in functional problems considered relevant by clients through interview, and is valid/reliable.[38-40] The most relevant functional goals to be accomplished are defined, ranked in importance, and rated on performance and satisfaction.[38-40] In this study, caregivers selected the goals and rated the child's performance and level of satisfaction since these are abstract concepts for children of this age.

The ABILHAND-Kids is a valid/reliable questionnaire assessing manual ability of children.[41] The test comprises a list of manual activities in which the caregivers score the amount of difficulty children with cerebral palsy may experience during their performance in activities of daily living that required hand use. Data were reported in logit-based units.

To assess children's daily functioning, caregivers were interviewed using the PEDI, a valid/reliable test[42] focusing on child's functioning in daily living activities at home.[43] Children's self-care functional skills and caregiver assistance were assessed.

A subset of 13 children (7 in SPG, 6 in UPG) wore an activity monitor (Manufacturing Technology Inc, Fort Walton Beach, FL, #7164; 5.1 cm × 2.6 cm × 1.5 cm, 42.9 g) on each wrist during a continuous 3-hour period in camp on the seventh or eighth intervention day to determine how much the children in each group moved. The units sample (10 Hz) and store summed values in memory, and data are downloaded to a personal computer. The number of accelerations is measured as activity counts (0.01664 g for an acceleration of 2.13 g directed parallel to the *x*-axis with a frequency of 0.75 Hz), which were used to determine the percentage of time each hand was used.[34]

### Statistical Design

Sample size calculations were performed based on JTTHF scores derived from an earlier HABIT trial.[13] A mean decrease from 380.84 to 249.58 seconds (131.25 ± 121.86 seconds) for the HABIT group was reported. With significance level = .05, $1 - \beta = 0.80$, $\mu 1 - \mu 2 = 131.25$, and $\sigma = 121.86$, and estimated 10% dropout, 11 participants in each group were required. Intention-to-treat principles were employed.

A 2 (group) × 3 (test sessions) analysis of variance with repeated measures on test sessions was performed using SPSS 15 for all measures except the COPM. Since goal performance may be influenced by development and practice in ongoing care (follow-up), a 2 (group) × 2 (pre/post) analysis of performance was performed on the COPM. The overall group-by-test session *interaction* tested whether the pattern of change between sessions varied across groups. Newman-Keuls post hoc tests were used to compare pretest and each of the posttests.

## Results

### Patient Flow

Patient flow is shown in the CONSORT diagram (Figure 1). During recruitment (June 2010-2012), 86 individuals were screened. Ultimately, 22 qualified individuals agreed to participate and were randomized into the SPG and UPG (see Figure 1 legend for details). One child from the UPG proved to have developmental dyspraxia and was not provided the intended treatment (ie, requiring sequential instructions, analogous to part practice) and the AHA was not usable. The child and the matched pair from the SPG were excluded. Thus, 20 participants (10 per group) completed the study. Table 1 describes participant characteristics. There were no significant group differences in baseline scores for any measure.

### Treatment Characteristics

Activity logs showed both groups spent more than 90% of time engaged in activities that required the use of the more-affected hand (SPG, 94.3%; UPG, 98.9%). In the SPG, whole task practice accounted for 87.8% of the time, and the remaining 12.2% comprised part task practice. Only whole practice occurred for the UPG. The *accelerometry data* for the subset of children who wore it indicated that on average the less-affected hand moved 79% and the more-affected hand moved 74% of the time for the SPG, and the less-affected hand moved 82% and the more-affected hand moved 68% for the UPG ($P > .05$, paired *t* test between the SPG and UPG). Thus, there was no difference in the amount of more-affected UE use between groups.

On average, activities for the SPG progressed in difficulty 55.4 times (standard deviation [SD] = 35.4, range = 11-117) throughout the intervention. Skill progression was not reported or observed for the UPG.

### Manual Dexterity and More-Affected Hand Use in Bimanual Activities

For the JTTHF, there was a 65.9-second (28.1%) and a 77.4-second (33.2%) decrease in time for the SPG and UPG, respectively (Table 2, Figure 2A). Newman-Keuls post hoc tests revealed a significant improvement between the pretest and immediate posttest that was maintained at 6 months. For the AHA, there was an increase of 2.3 and 2.8

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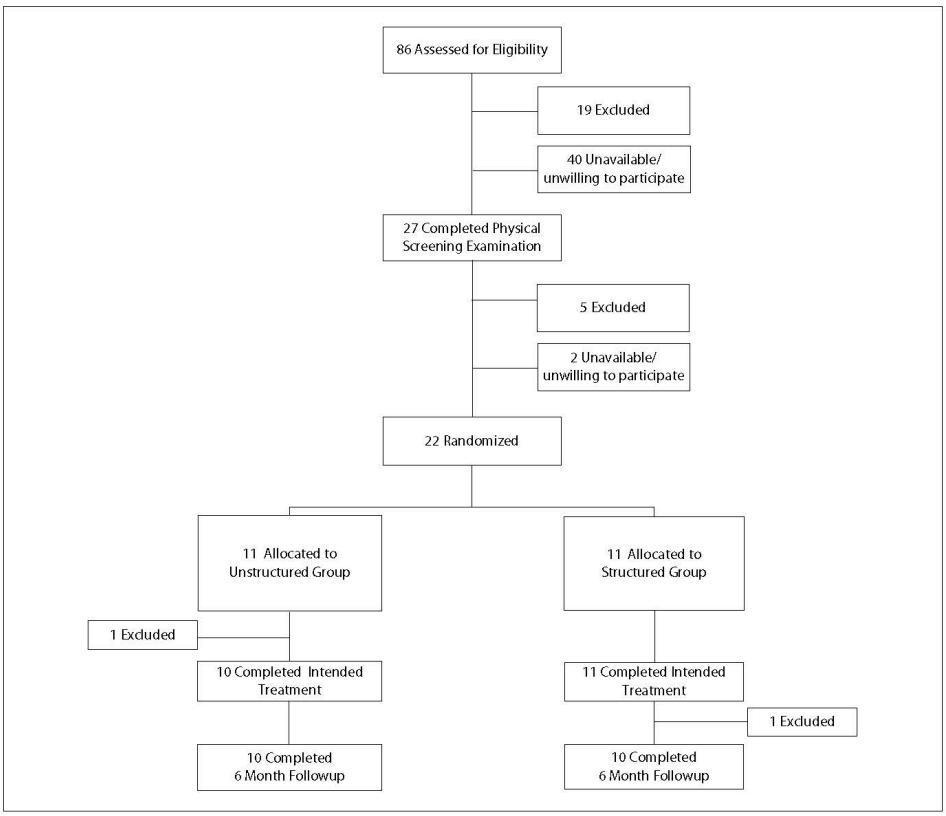


**Figure 1.** CONSORT flow diagram showing progress through the stages of the study, including flow of participants, withdrawals, and inclusion in analyses. A total of 86 individuals potentially met the study criteria and were invited to undergo physical screening. Twenty-two children qualified for the study and were randomized into the structured practice group (SPG) or unstructured practice group (UPG). One child from the UPG proved to have apraxia and was not provided the intended treatment. The child and the matched pair from the SPG were excluded.

AHA-units for the SPG and UPG, respectively (Figure 2B, Table 2). Post hoc tests revealed a significant improvement between the pretest and immediate posttest, but a return to baseline levels at 6 months, which was uncorrelated with age. There were no group × test session interactions for either measure.

### *Functional Goals and Daily Functioning*

The majority of goals were bimanual (remaining goals were unimanual with the more-affected hand). Most of the goals comprised self-care activities (eg, dressing, grooming, and eating), followed by play (eg, ball activities). At camp, the SPG spent on average 370 minutes practicing goals whereas the UPG did not practice goals. At home, children spent 165 and 184 minutes for the SPG and UPG, respectively, practicing goals. Both groups presented significant improvements after the intervention on satisfaction and on performance, but there was a significant group × test session interaction, with greater improvement for the SPG (Table 2). This difference was no longer present at 6 months (paired *t* test). There was no correlation (Pearson) between the amount of home practice of goals and COPM improvement.

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**Table 1.** Baseline Participant Characteristics.

| Characteristics | SPG (n = 10) | UPG (n = 10) |
|---|---|---|
| Mean age (SD) years, months | 8, 6 (1, 5) | 8, 3 (1, 5) |
| Gender, n (%) | | |
| Male | 6 (60) | 6 (60) |
| Female | 4 (40) | 4 (40) |
| More affected hand, n (%) | | |
| Right | 7 (70) | 7 (70) |
| Left | 3 (30) | 3 (30) |
| Lesion location (type) | | |
| Right | 3 ($0^a$, $2^b$, $1^c$) | 3 ($0^a$, $1^b$, $2^c$) |
| Left | 7 ($1^a$, $5^b$, $1^c$) | 7 ($0^a$, $6^b$, $1^c$) |
| Race, n (%) | | |
| Caucasian | 8 (80) | 7 (70) |
| African American | 1 (10) | 0 (0) |
| Hispanic | 1 (10) | 2 (20) |
| Mixed | 0 (0) | 1 (10) |
| MACS | | |
| I | 2 | 1 |
| II | 6 | 7 |
| III | 2 | 2 |
| Baseline JTTHF, mean (SD), seconds | 233 (159) | 234 (168) |
| Therapy | | |
| PT | 10 | 7 |
| OT | 10 | 9 |

Abbreviations: SPG, structured practice group; UPG, unstructured practice group; SD, standard deviation; MACS, Manual Ability Classification System; JTTHF, Jebsen-Taylor Test of Hand Function; PT, physical therapy received off-site (number of individuals); OT, occupational therapy received off-site (number of individuals).
[a]Disorder of cellular migration.
[b]Abnormality of periventricular white matter.
[c]Cortical/subcortical lesion.

For the ABILHAND-Kids, both groups presented significant improvement with no group × test session interactions. Both groups improved on the functional self-care skills and caregiver assistance scale of the PEDI, with no significant interaction. Post hoc tests indicated significant improvements for the functional self-care skills at immediate posttest that were maintained at 6 months. However, the caregiver assistance scale was only significantly higher at the 6-month follow-up.

### *Control Group*

It is conceivable that the similar improvements were simply because of repeating the tests over a 3-week period. To determine whether that was likely the case, we subsequently conducted the tests twice, 3 weeks apart, in a group of 10 (nonrandomized) children with USCP (mean age = 8.9 years, 5 males, 5 females) who did not receive treatment. There were no significant changes in the AHA (test 1 = 65.5 AHA-units [sd = 12.4]; test 2 = 64.8 AHA-units [SD = 14.3]), JTTHF (test 1 = 496.1 seconds [SD = 306.6]; test 2 = 465.8 seconds [SD = 315.9]), ABILHAND-Kids (test 1 = 1.6 logits [SD = 1.3]; test 2 = 1.6 logits [SD = 1.1]), PEDI functional skills self-care (test 1 = 61 [SD = 7.57]; test 2 = 63.3 [SD = 6.93]), or PEDI caregiver assistance (test 1 = 35.7 [SD = 3.2], test 2 = 36.7 [sd = 5.7]).

## Discussion

To our knowledge, this study is the first to compare components of pediatric intensive bimanual rehabilitation protocols. Children from both the SPG and UPG demonstrated improvements in dexterity and functional use of the hands. This suggests that at least for such intensive bimanual approaches, training may not require structured practice to elicit improvements in clinical measures, and that the emphasis can be placed on fun activities that require use of both hands. However, considering that the SPG showed superior improvements in functional goals, there may be added benefit of including goal training. These findings are discussed in relation to other treatment approaches and neuroplasticity in animal and humans.

Similar dexterity improvements for both groups did not support our hypothesis of greater improvements for the SPG, and are not in agreement with animal model studies that point to the importance of skilled training in the acquisition of motor skills.[28-30] However, in a study in which squirrel monkeys were exposed to a reaching task that did not involve skilled training, the animals showed improvements in the number of pellet retrievals and increase in speed to retrieve them.[28] Thus, it is possible that even without skilled training, intensive use of the more-affected hand leads to improvements in the speed to perform unimanual tasks.[28] Improvements in both groups are analogous to the similar improvements in the amount and quality of more-affected hand use between a group of adults with hemiparetic stroke submitted to CIMT regardless of whether shaping was used.[28] However, as acknowledged by the authors, it is not possible to conclude that skilled training is not relevant as the task practice group was submitted to some elements of shaping, such as verbal feedback and information related to the individual's performance.[32] In the present study, we carefully monitored children's activities during play activities from both groups. However, it was not possible to monitor every interaction or to avoid verbal feedback and encouragement.

This study suggests that bimanual structured practice may not be essential for improvements. The structure of intensive protocols is not well described in the literature. Some authors describe protocols comprising 1 interventionist to 2 children,[14-16] or combining caregivers' and therapists actions,[44] which may reduce the amount of skilled training. Nevertheless, these studies report improvements in hand

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**Table 2.** Results.

| | Pretest | Immediate Follow-up | 6-Month Follow-up | Change Score | Test Session Effect *P* Value ($\eta^2$) | Interaction *P* Value ($\eta^2$) |
|---|---|---|---|---|---|---|
| AHA (AHA-units) | | | | | | |
| SPG | 61.9 (55.4, 68.4) | 64.2 (56.0, 72.4) | 62.4 (55.4, 69.4) | 2.3 (−0.9, 5.5) | — | — |
| UPG | 63.8 (57.3, 70.3) | 66.6 (58.4, 74.8) | 63.0 (56.0, 70.0) | 2.8 (−0.4, 6.0) | — | — |
| Mean | 62.9 (58.2, 67.5) | 65.4 (59.6, 71.2) | 62.7 (57.7, 67.7) | 2.5 (0.4, 4.7) | *P* < .05 (.217) | *P* = .48 (.020) |
| JTTHF | | | | | | |
| SPG | 234.8 (126.0, 343.6) | 168.9 (82.7, 255.1) | 182.6 (94.7, 270.4) | −65.9 (−108.5, −23.3) | — | — |
| UPG | 233.4 (124.7, 342.2) | 156.0 (69.8, 242.2) | 162.4 (74.5, 250.2) | −77.4 (−120.0, −34.8) | — | — |
| Mean | 234.2 (157.2, 311.0) | 162.5 (101.5, 223.4) | 172.5 (110.4, 234.6) | −71.7 (−101.0, −42.3) | *P* < .001 (.462) | *P* = .792 (.007) |
| COPM-Performance | | | | | | |
| SPG | 3.6 (2.7, 4.4) | 6.9 (6.1, 7.7) | 6.8 (6.0, 7.5) | 3.3 (2.4, 4.2) | — | — |
| UPG | 3.2 (2.4, 4.1) | 5.1 (4.3, 5.9) | 6 (5.3, 6.8) | 1.9 (1.0, 2.8) | — | — |
| Mean | 3.4 (22.8, 4.0) | 6.0 (5.4, 6.6) | 6.4 (5.9, 6.9) | 2.6 (1.9, 3.3) | *P* < .001 (.928) | *P* = .031 (.067) |
| COPM-Satisfaction | | | | | | |
| SPG | 4.7 (3.4, 6.0) | 8.2 (7.3, 9.1) | 7 (6.2, 7.7) | 3.5 (2.2, 4.8) | — | — |
| UPG | 3.8 (2.5, 5.0) | 5.9 (4.9, 6.8) | 6.2 (5.5, 7.0) | 2.1 (0.8, 3.4) | — | — |
| Mean | 4.2 (3.3, 5.1) | 7 (6.4, 7.7) | 6.6 (6.1, 7.1) | 2.8 (1.9, 3.7) | *P* < .001 (.928) | *P* = .131 (.022) |
| ABILHAND-Kids | | | | | | |
| SPG | 1.7 (0.8, 2.5) | 2.5 (11.5, 3.5) | 3.0 (22.0, 3.9) | 0.8 (0.0, 1.6) | — | — |
| UPG | 1.4 (0.5, 2.2) | 1.9 (0.9, 2.9) | 2.1 (11.2, 3.1) | 0.5 (−0.3, 1.3) | — | — |
| Mean | 1.5 (0.9, 2.1) | 2.2 (11.5,2.9) | 2.6 (11.9, 3.2) | 0.7 (0.1, 1.2) | *P* < .01 (.246) | *P* = .667 (.017) |
| PEDI: Self-care | | | | | | |
| SPG | 65 (61.2, 68.8) | 69.5 (66.2, 72.8) | 70.3 (67.0, 73.6) | 4.5 (2.78, 6.2) | — | — |
| UPG | 61.0 (57.2, 64.8) | 63.9 (60.6, 67.2) | 67.2 (64.0, 70.5) | 2.9 (1.2, 4.6) | — | — |
| Mean | 63.0 (60.3, 65.7) | 66.7 (64.3, 69.1) | 68.8 (66.4, 71.1) | 3.7 (2.5, 4.9) | *P* < .001 (.540) | *P* = .358 (.025) |
| PEDI: Caregiver assistance | | | | | | |
| SPG | 33.9 (30.2, 37.6) | 33.7 (30.0, 37.4) | 37.1 (34.3, 39.9) | −0.2 (−2.5, 2.1) | — | — |
| UPG | 28.9 (25.2, 32.6) | 31.2 (27.5, 3439) | 34.4 (31.6, 37.2) | 2.3 (0.0, 4.6) | — | — |
| Mean | 31.4 (28.8, 34.0) | 32.5 (29.8, 35.1) | 35.8 (33.8, 37.8) | 0.9 (−0.7, 2.8) | *P* < .001 (.449) | *P* = .240 (.042) |

Abbreviations: SPG, structured practice group; UPG, unstructured practice group; JTTHF, Jebsen-Taylor Test of Hand Function; AHA, Assisting Hand Assessment; COPM, Canadian Occupational Performance Measure (performance and satisfaction scales); PEDI, Pediatric Evaluation of Disability Inventory (functional skills and caregiver assistance scales).

function.[15,44] The high intensity in both groups seems to be the main contributor to the similar improvements in dexterity as the (albeit nonrandomized) post hoc control group did not demonstrate changes in the same measures. Only part practice, skill progression, and goal training differed. Intensity is considered an important ingredient in motor learning, as it leads to repetitions of movements and the development of new motor strategies.[23] Thus, in intensive protocols the amount of practice is seemingly more important than what is practiced. However, these results may be dose dependent, as it is possible that at lower intensity, other components (eg, specificity of training) may be more important.[10] The low number of movement repetitions resulting from usual and customary schedules of physical/occupational therapy may partly explain the lack of evidence for these approaches.[45] Thus, it is possible that skilled practice is important in such low dose interventions.

One unexpected result was the lack of maintenance of AHA increases at the 6-month follow-up. Although it seems contrary to previous studies documenting persistent changes on this outcome following CIMT or HABIT,[3,13,15] the lack of retention may be because of the age of the participants, who were generally older than those from other studies. Eliasson et al[46] reported no change in AHA following CIMT in 8- to 17-year-old children. We concur with their suggestion that changes in bimanual function are harder to achieve in older children.[46] It is possible that at older ages, dexterity (eg, JTTHF) and functional use (eg, COPM, PEDI, ABILHAND-Kids) may improve and be maintained, but children may have well established their own strategies to use the more-affected hand in the performance of bimanual activities, and that this strategy is more difficult to change. Goal performance was the only measure to improve differentially, that is, greater initial gains for the SPG. These

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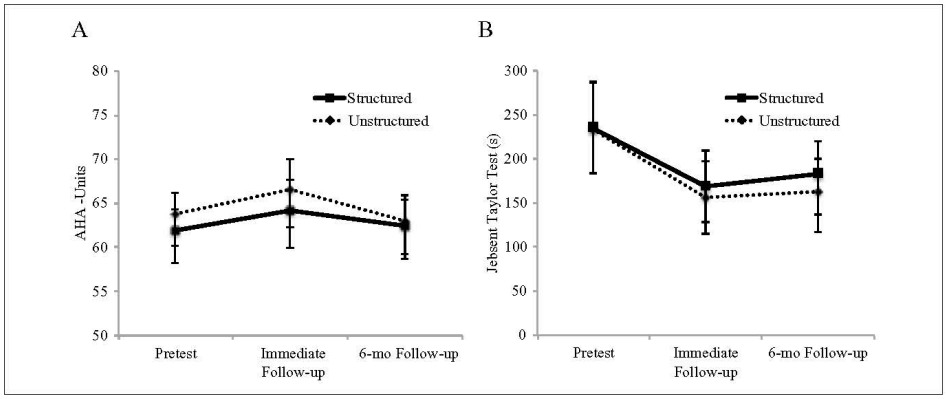


**Figure 2.** (A) Mean ± standard error of the mean (SEM) scaled logit scores on the Assisting Hand Assessment (AHA); higher scores represent better performance. (B) Mean ± SEM time to complete the 6 timed items (writing excluded) of the Jebsen-Taylor Test of Hand Function. Faster times correspond to better performance. The potential maximum allowable time to complete each item was 180 seconds, resulting in a maximum score of 1080 seconds.

results corroborate studies that found superior improvements on goal performance when goals are practiced.[13,47] If the 6-month follow-up data are compared, the differences were no longer significant. As children from the SPG had the opportunity to practice some of the goals and this practice involved skill progression, it is possible that structured training contributed to the development of strategies to perform relevant functional goals.[47] Such information confirms the need of specific and direct training for the accomplishment of goals related to the use of hands by children with USCP, but the end result over a longer period may not differ.

## Limitations

Although the interventions were administered in separate rooms and the groups and interventionists were blinded to the study questions, as mentioned above it is not possible to ensure that skill progression did not occur in the UPG. It is possible that children from either group could have been self-motivated to challenge themselves. However, the children, interventionists and families were not aware of the differences in the protocols provided for each group.

It is also possible that when caregivers selected goals for the COPM, they might be aware of the functional activities that are relevant to their children. This may have motivated goal practice despite that caregivers from both groups were not instructed to practice these goals at home and no information in how to progress performance of these goals was offered during the intervention period. On average, goal practice totaled ~3 hours, which is a small fraction of the amount of treatment time. Moreover, the children and interventionists from the USG were not aware of the goals. Furthermore, it is possible that there were indeed differences between groups that either was not measured (eg, ICF body structure and function level) or that the employed measures were not sensitive enough to detect. Finally, the study may have been underpowered to detect group differences. However, as indicated by the $\eta^2$ values (Table 2), the differences in changes were extremely small (favoring the UPG) and would require an extremely large sample size for this difference to be statistically significant, which would bring the clinical significance of such changes into question.

## Clinical Implications

The present study shows that the intensive nature of bimanual practice seems to be the main contributor for improvements in manual dexterity. It is not known whether these findings would hold true at lower dosages. It may be possible to combine interventions that involve bimanual play with specific functional training. This may reduce the costs of the provision of high-intensive interventions without compromising the benefits. This possibility needs to be tested in a larger cohort of children.

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# APPENDIX E

*Research Articles*

# Bimanual Training and Constraint-Induced Movement Therapy in Children With Hemiplegic Cerebral Palsy: A Randomized Trial

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**Andrew M. Gordon, PhD[1,2], Ya-Ching Hung, Ed.D.[3], Marina Brandao[4], Claudio L. Ferre, MA[1], Hsing-Ching Kuo, MS[1], Kathleen Friel, PhD[5], Electra Petra[1], Ashley Chinnan[1], and Jeanne R. Charles, PhD[6]**

**Abstract**

*Background.* Constraint-induced movement therapy (CIMT) promotes hand function using intensive unimanual practice along with restraint of the less-affected hand. CIMT has not been compared with a treatment with equivalent dosing frequency and intensity in children with cerebral palsy (CP). *Objectives.* The authors report a randomized trial comparing CIMT and a bimanual intervention (hand-arm intensive bimanual therapy; HABIT) that maintains the intensity of practice associated with CIMT but where children are engaged in functional bimanual tasks. *Methods.* A total of 42 participants with hemiplegic CP between the ages of 3.5 and 10 years (matched for age and hand function) were randomized to receive 90 hours of CIMT or an equivalent dosage of functional bimanual training (HABIT) conducted in day-camp environments. A physical therapist blinded to treatment allocation tested hand function before and after treatment. The primary outcomes were changes in Jebsen-Taylor Test of Hand Function (JTTHF) and Assisting Hand Assessment (AHA) scores. Secondary measures included the Goal Attainment Scale (GAS). *Results.* Both the CIMT and HABIT groups demonstrated comparable improvement from the pretest to immediate posttest in the JTTHF and AHA ($P < .0001$), which were maintained at 6 months. GAS, however, revealed greater progress toward goals for the HABIT group ($P < .0001$), with continued improvement across test sessions for both groups ($P < .0001$). *Conclusions.* Both CIMT and bimanual training lead to similar improvements in hand function. A potential benefit of bimanual training is that participants may improve more on self-determined goals.

**Keywords**

hand, upper-extremity rehabilitation, hemiplegic cerebral palsy, bimanual training, grasp, constraint-induced movement therapy

## Introduction

Cerebral palsy (CP) is the most common pediatric physical disability, with congenital hemiparesis comprising one of the most common forms.[1] Strong evidence supporting any upper-extremity treatment approach is lacking.[2,3] Yet there is increasing evidence that hand function in individuals with CP does improve during development[4-6] and with intensive practice.[7,8]

One means to achieve intensive practice is constraint-induced movement therapy (CIMT), involving concurrent physical restraint of the less-affected hand and unilateral training of the hemiplegic hand.[9-11] There is increasing evidence supporting the efficacy of pediatric CIMT,[2,12,13] and its clinical use is proliferating.

Nevertheless, CIMT is potentially invasive,[14] and it is uncertain whether improved hemiparetic hand use results in improved function. Children with hemiplegic CP have impaired bimanual coordination above and beyond their unimanual impairments,[15-17] which may underlie some functional limitations. Furthermore, studies of CIMT (including our own)[18,19] have not compared it with other, equally

[1]Teachers College, Columbia University, New York, NY, USA
[2]College of Physicians and Surgeons, Columbia University, New York, NY, USA
[3]Queens College, CUNY, Flushing, NY, USA
[4]Universidade Federal de Minas Gerais, Brazil
[5]Columbia University Medical Center, New York, NY, USA
[6]Emory University School of Medicine, Atlanta, GA, USA

**Corresponding Author:**
Andrew M. Gordon, Department of Biobehavioral Sciences, Box 199, Teachers College, Columbia University, 525 West 120th Street, New York, NY 10027, USA
Email: ag275@columbia.edu

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intense treatments. Thus, it is unknown whether similar improvements in hand function can be achieved with an equally intensive bimanual treatment.

We report a randomized clinical trial comparing CIMT and a bimanual intervention (hand-arm intensive bimanual therapy, HABIT)[20,21] that maintains the same dosing frequency and intensity and the progressive task-specific practice associated with CIMT, but children are engaged in functional bimanual rather than unimanual tasks. HABIT has a proposed neuroscientific rationale[13,22] and is a formalized approach that has an equal amount of personal interaction and intensity as CIMT, encourages functional gains,[23] and has preliminary evidence suggesting efficacy.[15,21] Yet bimanual training is different from CIMT because of the type of activities practiced. Based on theories of motor learning (practice specificity),[24,25] we hypothesized that the unimanual training associated with CIMT would lead to greater improvements in unimanual dexterity, whereas bimanual training would lead to greater improvements in bimanual hand use and goal attainment.

## Method

### Participants

Participants were recruited from clinics in the NYC area, our Web site (http://www.tc.edu/centers/cit/), ClinicalTrials.gov (NCT00305006), and online support groups. Potential participants were first screened via e-mail and/or telephone. Those between the ages of 3.5 and 10 years with congenital hemiplegia were invited to receive an on-site physical examination or an examination videotaped by their physical or occupational therapist. The inclusion criteria were established based on prior CIMT and HABIT trials[19,20]: (1) ability to extend wrist >20° and fingers at the metacarpophalangeal joints >10° from full flexion, (2) the ability to lift the more affected arm 15 cm above a table surface and grasp light objects, (3) >50% difference in the Jebsen-Taylor Test of Hand Function (JTTHF) score between the 2 hands and a time below the maximum possible (1080 s) of the paretic hand, (4) mainstreamed in school and a Kaufman Brief Intelligence test score >70, and (5) demonstrated ability to follow instructions during screening and complete the testing. Exclusion criteria included the following: (1) health problems unassociated with CP, (2) current/untreated seizures, (3) visual problems interfering with treatment/testing, (4) severe muscle tone (Modified Ashworth score > 3.5), (5) orthopedic surgery on the paretic hand within 1 year, (6) botulinum toxin therapy in the upper extremity within the past 6 months or intended treatment within the study period, and (7) balance problems precluding wearing a sling. Informed consent was obtained from participants and caregivers. The study was approved by the University Institutional Review Board.

### Procedures

*General intervention procedures.* CIMT and HABIT procedures share in common intensive progressive task practice based on motor learning approaches. They differ in that CIMT requires hand restraint and progression of unimanual tasks, whereas in HABIT, there is absence of restraint, and tasks are progressed bimanually. We first describe the common procedures followed by procedures specific to CIMT and HABIT.

Six HABIT and CIMT day camps were conducted at the University from July 2007 to 2009. The day camps were held in 2 separate rooms. Participants in 1 room received CIMT, and the other received HABIT; 2 to 5 children participated in each treatment at each camp. Participants were engaged in treatment 6 h/d for 15 consecutive weekdays (adjusted for holidays; 90 hours, making up any missed hours) during school recess by trained interventionists. Eight of the interventionists (out of 42 interventionist/child pairs) were physical or occupational therapists (PT/OT; 3 in the CIMT group and 5 in the HABIT group). The remaining interventionists were graduate students in Kinesiology, Neuroscience, Speech Pathology, or Psychology programs and undergraduates. Despite a higher level of knowledge and awareness of how to interact in a therapeutic manner, the PT/OT interventionists were required to provide only the specific procedures related to CIMT/HABIT and were restrained from using other treatment modalities. The expected outcomes were not discussed with interventionists. The pretreatment training, administered by the supervisors, was standardized based on the established manual of procedures for each treatment and reinforced by supervisors and during daily meetings. Each room was always supervised by additional experienced PTs/OTs, who modeled and ensured uniformity of treatment.

Participants worked individually with their interventionist or in groups (1:1 interventionist to participant ratio always maintained). Interventionists were paired with children prior to randomization using family-centered approaches considering caregiver and supervisors' best judgment based on the child's age and gender. Emphasis was placed on making participation enjoyable. With the exception of goals (see the following), tested task items were never trained. Children participated in whole and part task practice. Whole task practice involved sequencing successive movements within the context of tasks (eg, card games). The activities were performed continuously for at least 15 to 20 minutes. Targeted movements and spatial and temporal coordination were practiced within the context of completing the task. Part task practice (analogous to "shaping")[19,20] required breaking down motor skills into smaller components (eg, playing-card turning to promote forearm supination) while increasing repetitions and skill requirements. It also increased intensity of treatment by requiring as many repetitions as

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possible over repeated 30-s intervals (typically a minimum of 5 intervals).

Task difficulty was graded by varying the spatial/temporal constraints or by providing tasks that required progressive skilled use as performance improved. Task difficulty was increased when the participant was successful on 7 of 10 repetitions. Task performance was recorded, and task- and age-specific knowledge of results was provided for encouragement.[26-28]

Supervisors instructed interventionists regarding the main focus of the treatment (eg, supination) for each child, based on his/her pronounced movement deficits, interests, and potential for improvement. Data activity logs were collected, which included activities performed, time spent in part/whole practice, and the number of repetitions.

Caregivers were instructed to engage participants in home practice (unimanual without restraint for CIMT and bimanual for HABIT) for 1 h/d during and for 6 months following the intervention and document practice using activity logs.

*CIMT procedures*. CIMT was modified to be child focused (for details see Gordon et al[19]). Participants' less-affected hands were restrained with slings, and unimanual activities were performed with the paretic hands. The sling was strapped to participants' trunks with the distal end sewn shut and was continuously worn except when toileting or during breaks (not >15 min/d).

Participants performed fine-motor and manipulative gross-motor activities that elicit general movements of interest and that included a range of age-appropriate, unimanual functional and play activities. The interventionist provided assistance where appropriate. For example, during scissor use, the child cut paper while the interventionist held and rotated the paper.

*HABIT procedures*. HABIT[20] did not use a physical restraint, but instead, participants were engaged in age-appropriate fine- and gross-motor bimanual activities using motor learning approaches.[28] Activities were selected by considering the role of the paretic hand, increasing in complexity from a non-dominant passive assist (eg, stabilizing paper while drawing) to active manipulator (eg, reorienting paper while cutting) using increasingly complex bimanual coordination and participants' interests. Task demands were graded, and participants were engaged in active problem solving.

Interventionists avoided verbal prodding to use the paretic hand and instead constrained the environment by providing tasks necessitating the use of both hands to elicit desired movements. Part practice included both bilateral symmetrical (eg, reaching toward object[s] with both hands) and asymmetrical (eg, pulling apart objects) movements.

## Measures

Participants were evaluated directly prior to treatment (pre-test), within 2 days ("immediate"), and 1 and 6 months after treatment by a physical therapist blinded to group allocation (verified following testing). Two primary outcome measures were used to quantify unimanual capacity and bimanual performance under the International Classification of Functioning and Health (ICF) "activity and performance" domain.[29]

The Assisting Hand Assessment (AHA, version 4.3,[30,31]) quantifies the effectiveness with which a child with unilateral disability uses his/her affected (assisting) hand in bimanual activity. It has excellent validity and reliability (interrater = .97, intrarater = .99).[31] The test was videotaped and scored off-site by an experienced evaluator blinded to group allocation.

The JTTHF is a standardized test of simulated functional tasks quantifying the time to complete a battery of unimanual activities.[32] The activities performed with the paretic hand include flipping index cards, object placement, simulated eating, stacking checkers, and manipulating empty and full cans. Reliability for children with stable hand disabilities is high (.95-.99).[33] To determine whether the less-affected hand was affected by the restraint (CIMT) or use (HABIT), we also measured the JTTHF for this hand before and immediately after CIMT/HABIT.

Three secondary measures were also used. The dissociated movements (ICF "body function") and grasp (ICF "body function" and "activity and performance") subtests of the Quality of Upper Extremity Skills Test (QUEST)[34] were used to characterize dissociation of distal and proximal upper-extremity movements and attainment of specific grasp patterns of the 2 hands (ICF activity and performance and body function/structure). The sum score for both hands is converted to a standardized score (maximum = 100).

The Goal Attainment Scale (GAS; ICF "activity and performance" and "participation") was used to quantify progress on established goals before group assignment (1 functional and 1 play goal defined by caregivers and/or older participants). Goals were assessed for appropriateness based on age and current abilities and scaled off-site by a physical therapist. The goals were practiced up to 30 min/d. However, because emphasis was placed on embedding practiced movements into fun activities rather than providing "goal-training" as is often done in traditional occupational/physical therapy, interventionists were given latitude as to how much training was provided (if at all) within the 30-minute limit. The CIMT group was unable to practice bimanual goals and, instead, practiced unimanual movement components comprising the goal. Goal achievement[35] was rated by the caregiver and verified by a physical therapist. Scores were transformed to standardized $T$ scores (mean = 50; standard deviation = 10)[35]:

$$T = 50 + \frac{10\sum x_j}{\sqrt{n - np + n^2 p}},$$

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where $x_j$ is the attainment score, $n$ the number of scales, and $P$ the expected correlation of scales ($P = .3$). A $T$ score of 50 indicates that goals were attained.

Children wore activity monitors (Manufacturing Technology Inc, Fort Walton Beach, Florida, #7164; 5.1 cm × 2.6 cm × 1.5 cm, 42.9 g)[36] on their wrists during the AHA test sessions. The units sample (10 Hz) and store summed values in memory and data were downloaded to a PC. The number of accelerations is measured as activity counts (0.01664 g for an acceleration of 2.13 g directed parallel to the $x$-axis at 0.75 Hz), which were used to determine the percentage of time each hand was used while performing AHA activities.[21]

### Statistical Design

Sample size calculations were performed based on AHA scores derived from an earlier HABIT trial.[21] A mean improvement of 0.94 ± 0.54 logits for the HABIT group was reported. With $\alpha = .05$, $1 - \beta = 0.80$, and an estimated 20% dropout, 21 participants in each group were required. Intention-to-treat principles were used.

A 2 (Group) × 4 (Test Sessions) ANOVA with repeated measures on test sessions was performed on raw and log-transformed AHA, JTTHF, QUEST, and accelerometry data using SPSS 15. Since the findings were qualitatively similar, only analyses of raw data are reported. The overall Group × Test Session interaction tested whether the time course differed between treatment groups. Planned comparisons were used between the pretest and each of the posttests and the immediate posttest with the 1- and 6-month posttest. Bonferroni corrections were used, resulting in a significance level of $P < .01$. A 2 (Group) × 3 (Posttests) ANOVA with repeated measures on posttests was performed on the GAS $T$ scores, with the group factor indicating whether one group had greater goal attainment.

## Results

### Patient Flow

Patient flow is shown in the CONSORT flow diagram (Figure 1). During recruitment (June 2007-2009), 183 individuals were screened. Ultimately, 44 qualified individuals agreed to participate and were randomized into the HABIT or CIMT group (see Figure 1 legend for details). Participants (4-10 in each camp) were randomized offsite using concealed allocation stratified by age and JTTHF screening score. A total of 42 participants (21 in each group) completed the study. Table 1 describes participant characteristics. There were no significant group differences in baseline scores for any measure (Table 2).

### Treatment Characteristics

Participants in the HABIT and CIMT groups completed all 90 hours of treatment and averaged 79% and 81% of the time in structured practice, respectively (remaining time was spent in transitioning between tasks, toileting, etc). Children spent 17% and 16% of the practice time, respectively, in part practice, with the remainder in whole practice. Direct observation by the supervisors and monitoring of daily logs confirmed that both treatment protocols were completely adhered to and only the intended treatments were received. Home logs indicated that children averaged 286 minutes of the requested 360 min/wk engaging in home practice during the 6 months following the intervention.

No adverse events were reported. Participants stopped usual and customary care (UCC) during the treatment but resumed afterward (proportion receiving therapy was similar between groups: $P > .05$, Table 1). There were no changes to preexisting therapy.

### Quality and Amount of Movement

Table 2 shows the means for the HABIT and CIMT groups at each time point and the pretest to immediate posttest difference for all measures. For the JTTHF, there was a 141.7 s (37.8%) and a 131.2 s (34.5%) decrease for the CIMT and HABIT groups, respectively (Figure 2A). Similarly, the AHA scaled logit scores improved 2.24 and 3.0 points for the CIMT and HABIT groups, respectively (Figure 2B). Planned comparisons revealed that the changes were attributed to differences between the pretest and immediate posttest that were maintained over the 6 months. There were no Group × Test Session interactions for either primary measure.

There was no difference in the less-affected hand JTTHF for either group (CIMT change score = 6.1, 95% confidence interval [CI] = −3.5, 15.7; HABIT change score = 5.2, 95% CI = −0.8, 11.3; $P > .05$ in both cases).

To account for the asymmetric distribution of right- and left-side hemiparesis across the 2 groups, we repeated the analyses adding side as an additional factor. There was no effect of side or group interaction for either measure.

A main effect of testing session was also seen for secondary measures (QUEST dissociated movements and grasp subtests, and the accelerometry, Table 2) for both treatments. QUEST changes were largely a result of increased scores for the paretic hand ($P < .001$, not shown separately in Table 2). However, scores for the less-affected hand increased slightly (raw score pretest to immediate posttest across both groups, 23.3 to 23.9) for the grasp ($P < .001$) but not for the dissociated movements subtest. Accelerometry results indicated that the less-affected hand moved ~91% and the paretic hand ~64% of

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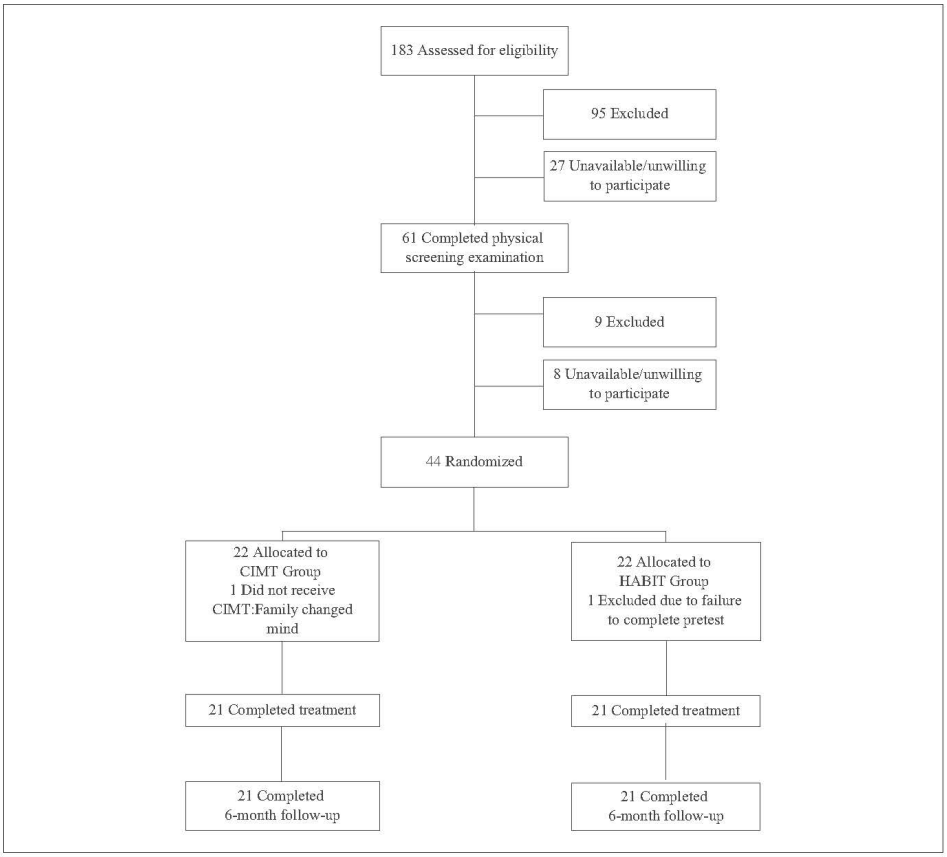


**Figure 1.** CONSORT flow diagram showing progress through the stages of the study, including flow of participants, withdrawals, and inclusion in analyses. A total of 183 individuals were screened via telephone/e-mail, and 95 of these were excluded for the following reasons: too old (n = 11), too young (n = 35), poor cognition (n = 11), wrong diagnosis (n = 22), botulinum toxin treatment within prior 6 months or planned within study period (n = 2), uncontrollable seizures (n = 7), prior CIMT within 6 months (n = 5), surgery (n = 1), and disease too severe (n = 1). A total of 88 individuals potentially met the study criteria and were invited to undergo physical screening; 27 individuals chose not to undergo physical screening. Of the remaining 61 individuals, 9 were excluded on the basis of the physical exam: too severe (n = 8) and too mild (n = 1). Of these individuals who qualified, 8 chose not to participate: not available during day-camp period (n = 5) and hardship associated with participating (n = 3). Ultimately, 44 of the remaining individuals agreed to participate and were randomized into the HABIT or CIMT group. One participant dropped out after randomization (unaware of group allocation), and another was excluded after the intervention for inability to comply with testing procedures. A total of 21 individuals from each group completed the intervention and subsequent posttests. Abbreviations: CIMT, constraint-induced movement therapy; HABIT, Hand-Arm Intensive Bimanual Therapy.

the time for each group at baseline. Hand use increased ~14% for both groups (ie, making up half of the baseline difference) for the hemiparetic hand, whereas it remained unchanged for the less-affected hand. The changes did not correlate with AHA scores changes ($r = -0.041$) but did correlate with JTTHF changes ($r = 0.48$; $P < .001$).

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**Table 1.** Baseline Participant Characteristics

| Characteristics | CIMT (n = 21) | HABIT (n = 21) |
|---|---|---|
| Mean age (SD), years, months | 6, 3 (2, 2) | 6, 4 (1, 11) |
| Gender | | |
| Male | 9 (43%) | 11 (52%) |
| Female | 12 (57%) | 10 (48%) |
| Paretic hand | | |
| Right | 15 (71%) | 9 (43%) |
| Left | 6 (29%) | 12 (57%) |
| Lesion location[a] (type) | | |
| Right | 5(1,[b] 2,[c] 2,[d] 0[e]) | 8 (2,[b] 2,[c] 4,[d] 0[e]) |
| Left | 8 (3,[b] 2,[c] 3,[d] 0[e]) | 7 (1,[b] 3,[c] 2,[d] 1[e]) |
| Bilateral | 1 (0,[b] 0,[c] 1,[d] 0[e]) | 1 (0,[b] 1,[c] 0,[d] 0[e]) |
| Race | | |
| White | 15 (71%) | 12 (57%) |
| African American | 3 (14%) | 4 (19%) |
| Hispanic | 2 (10%) | 4 (19%) |
| Asian | 1 (5%) | 1 (5%) |
| MACS | | |
| I | 2 (10%) | 3 (14%) |
| II | 18 (85%) | 17 (81%) |
| III | 1 (5%) | 1 (5%) |
| Baseline JTTHF, mean (SD), s | 375 (254) | 381 (226) |
| KBIT, mean (SD) | 96.7 (20.7) | 99.3 (16.5) |
| 2PD[f] | 3.1 (0.83) | 3.4 (1.5) |
| Controlled seizures | 3 (14%) | 7 (33%) |
| Therapy | | |
| PT | 20 (95%) | 21(100%) |
| OT | 20 (95%) | 18 (85%) |

Abbreviations: CIMT, constraint-induced movement therapy; HABIT, Hand-Arm Intensive Bimanual Therapy; SD, standard deviation; MACS, Manual Ability Classification System; JTTHF, Jebsen-Taylor Test of Hand Function; KBIT, Kaufman Brief Intelligence Test; 2PD, 2 point discrimination (average of thumb and index); PT, physical therapy received off-site (number of individuals); OT, occupational therapy received off-site (number of individuals).
[a]MRI from 12 children were not available.
[b]Brain malformation.
[c]Abnormality of periventricular white matter.
[d]Cortical/subcortical lesion.
[e]Nonprogressive postnatal brain injury (CIMT child, left precentral gyrus, right corona radiata; HABIT child, bilateral swelling, more pronounced on right, minimal hyperdensity along body of left ventricle).
[f]Out of 30 participants, 12 were undetermined because of age.

### Goal Attainment

Goal characteristics and attainment are described in Table 3. The majority of goals were bimanual (remaining goals were unimanual with paretic hand). All goals were related to the ICF "activity and performance" domain. The HABIT group spent more intervention time practicing goals than the CIMT group, but home time practice of goals did not significantly differ between groups.

Both groups achieved ($T$ score of 50) or exceeded their expected level of goal performance and continued to improve across test sessions. However, the HABIT group made greater progress than the CIMT group, as indicated by significant group differences in their $T$ scores across all test sessions (Table 3). The combined amount of camp and home practice time did not correlate with GAS $T$ scores ($r = 0.06$) at immediate posttest.

Approximately 25% of the identified goals were not practiced in either group during the interventions or at home. However, the $T$ scores for these unpracticed goals were significantly higher for the HABIT group (Table 3). These scores appeared to improve across test session, but the improvement and interaction with group was not statistically significant.

### Predictors of Improvement

Participants with higher initial JTTHF scores had greater absolute ($r = 0.69$), but not percentage change in, JTTHF scores for CIMT and HABIT. Neither age nor any other variable was related to primary measure improvement for either treatment.

Finally, in the present study, only 8 of the 42 participants were assigned a physical or occupational therapist for the intervention. There was little difference in AHA or JTTHF changes (as evidenced by small mean differences and overlapping distributions) regardless of whether the interventionist was a PT/OT or not (PT/OT AHA change score = 1.37, 95% CI = −1.11, 3.86; non-PT/OT AHA change score = 2.91, 95% CI = 1.47, 4.35; PT/OT JTTHF change score = 118.9, 95% CI = 48.8, 189.0; non-PT/OT JTTHF change score = 140.6, 95% CI = 98.4, 182.9). Similar absences of differences were observed for the secondary measures.

## Discussion

Bimanual training and CIMT resulted in similar improvements in the primary measures, which did not support our hypothesis of specificity of training. This suggests that improvements in hand function associated with CIMT can be achieved with an equally intensive bimanual approach. However, there was specificity of training for goal attainment, whereby the HABIT group made better progress on established goals and transfer to unpracticed goals. These findings have important implications, given the increasing popularity and potential invasiveness of CIMT. It is important to note that our results suggest that intensive progressive task-specific training improves hand function.

### Both CIMT and Bimanual Training Improve Hand Function

In agreement with our quasirandomized CIMT/HABIT trial,[15] there were significant changes in all measures following CIMT and HABIT. The partial $\eta^2$ (Tables 2 and 3) indicated that 26% and 48% of the variance in AHA and

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**Table 2.** Results

| | Pretest (95% CI) | Immediate Posttest (95% CI) | 1-Month Posttest (95% CI) | 6-Month Posttest (95% CI) | Change Score (Pretest to Immediate Posttest) (95% CI) | Test Session Effect *P* Value (Partial $\eta^2$) | Interaction *P* Value (Partial $\eta^2$) |
|---|---|---|---|---|---|---|---|
| AHA (Logits) | | | | | | | |
| CIMT | 0.38 (−0.42, 1.17) | 0.80 (0.01, 1.58) | 0.90 (0.12, 1.67) | 1.05 (0.35, 1.74) | 0.42 (0.08, 0.76) | — | — |
| HABIT | 0.38 (−0.41, 1.17) | 0.94 (0.16, 1.72) | 0.98 (0.21, 1.76) | 0.99 (0.30, 1.69) | 0.56 (0.23, 0.90) | — | — |
| Mean | 0.38 (−0.20, 0.90) | 0.87 (0.30, 1.40) | 0.94 (0.40, 1.50) | 1.02 (0.60, 1.50) | 0.49 (0.25, 0.73) | $P < .0001$ (.260) | $P = .806$ (.008) |
| JTTHF (s) | | | | | | | |
| CIMT | 374.8 (268.9, 480.6) | 233.1 (154.0, 312.2) | 207.1 (146.1, 268.1) | 221.0 (150.1, 291.8) | −141.7 (−195.4, −88.0) | — | — |
| HABIT | 380.8 (275.0, 486.7) | 249.6 (170.5, 328.7) | 236.9 (176.0, 297.9) | 222.7 (151.9, 293.5) | −131.2 (−185.0, −77.5) | — | — |
| Mean | 377.8 (306.1, 449.5) | 241.4 (187.7, 295.0) | 222.0 (180.5, 263.6) | 221.8 (173.9, 269.8) | −136.5 (−174.0, −98.9) | $P < .0001$ (.480) | $P = .857$ (.006) |
| Accelerometry (%) | | | | | | | |
| CIMT | 65.3 (62, 69) | 77.6 (75, 81) | 77.8 (75, 81) | 79.0 (76, 82) | 12.3 (9, 15) | — | — |
| HABIT | 63.1 (60, 67) | 78.3 (75, 81) | 76.4 (74, 79) | 77.8 (75, 81) | 15.2 (13, 18) | — | — |
| Mean | 64.2 (60, 70) | 78.0 (76, 80) | 77.1 (76, 79) | 78.4 (77, 80) | 13.8 (12, 16) | $P < .0001$ (.774) | $P = .346$ (.027) |
| QUEST—Dissociated Movement (standardized scores) | | | | | | | |
| CIMT | 85.2 (81.6, 88.8) | 90.3 (88.1, 92.6) | 91.3 (89.0, 93.5) | 89.1 (86.4, 91.8) | 5.2 (1.7, 8.7) | — | — |
| HABIT | 87.7 (84.1, 91.3) | 91.2 (89.0, 93.4) | 90.8 (88.5, 93.1) | 90.9 (88.2, 93.6) | 3.4 (0.2, 6.9) | — | — |
| Mean | 86.5 (84.1, 89.0) | 90.8 (89.2, 92.3) | 91.0 (89.5, 92.5) | 90.0 (88.2, 91.8) | 4.3 (1.9, 6.8) | $P < .0001$ (.200) | $P = .369$ (.026) |
| QUEST—Grasp (standardized scores) | | | | | | | |
| CIMT | 69.5 (63.6, 75.4) | 80.6 (75.8, 85.4) | 81.2 (76.8, 85.7) | 78.8 (72.8, 85.0) | 11.1 (6.1, 16.1) | — | — |
| HABIT | 68.6 (62.7, 74.5) | 79.4 (74.6, 84.2) | 79.9 (75.5, 84.4) | 76.2 (70.1, 82.3) | 10.8 (5.8, 15.8) | — | — |
| Mean | 69.0 (65.0, 73.0) | 80.0 (76.8, 83.3) | 80.6 (77.6, 83.6) | 77.5 (73.3, 81.7) | 11.0 (7.5, 14.5) | $P < .0001$ (.359) | $P = .948$ (.005) |

Abbreviations: CIMT, constraint-induced movement therapy; HABIT, Hand-Arm Intensive Bimanual Therapy; JTTHF, Jebsen-Taylor Test of Hand Function; AHA, Assisting Hand Assessment; QUEST, Quality of Upper Extremity Skills Test; Mean, represents the average for the CIMT and HABIT groups.

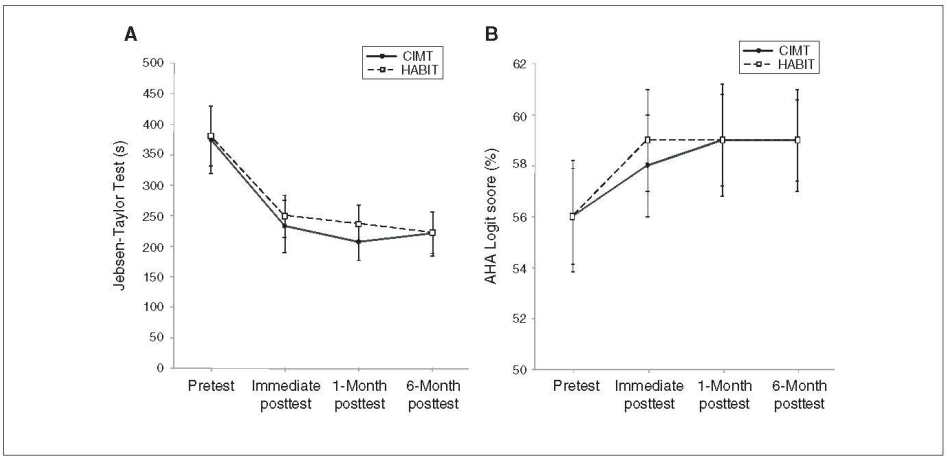


**Figure 2.** A. Mean ± standard error of the mean (SEM) time to complete the 6 timed items (writing excluded) of the Jebsen-Taylor Test of Hand Function. Faster times correspond to better performance. The maximum allowable time to complete each item was 180 s, resulting in a maximum score of 1080 s. B. Mean ± SEM scaled logit scores on the AHA; higher scores represent better performance. Abbreviations: CIMT, constraint-induced movement therapy; HABIT, Hand-Arm Intensive Bimanual Therapy; AHA, Assisting Hand Assessment.

JTTHF, respectively, and up to 77% for secondary measures, is attributable to the treatments. Similar findings have been reported following adult stroke cerebral vascular accident (CVA) interventions.[37,38] The robust JTTHF changes for both treatments (137 s) were nearly twice those reported in a 13-year follow-up study of hand function

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**Table 3.** Goal Attainment Scale

| Goal Category | Number of Goals | | | | | |
|---|---|---|---|---|---|---|
| Unimanual | 6 (17.0%) | | | | | |
| Bimanual | 36 (83.0%) | | | | | |
| Functional goal type | | | | | | |
| Dressing | 26 (62.0%) | | | | | |
| Eating | 12 (28.6%) | | | | | |
| Improve object manipulation | 3 (7.1%) | | | | | |
| Other | 1 (2.3%) | | | | | |
| Play goal type | | | | | | |
| Ball play | 19 (45.2%) | | | | | |
| Computer/video game | 6 (14.2%) | | | | | |
| Scissors | 5 (11.9%) | | | | | |
| Other[a] | 12 (28.7%) | | | | | |
| Goal Practice (minutes) | Intervention | Home | | | | |
| CIMT | 70.8 (78.7) | 12.1 (22.1) | | | | |
| HABIT | 138.0 (98.2) | 25.9 (32.3) | | | | |
| *t* Test | $t = 2.45, P < .05$ | $t = 1.6, P = .11$ | | | | |
| | Immediate Posttest (95% CI) | 1-Month Posttest (95% CI) | 6-Month Posttest (95% CI) | Group Effect *P* Value (Partial $\eta^2$) | Test Session Effect *P* Value (Partial $\eta^2$) | Interaction *P* Value (Partial $\eta^2$) |
| GAS (*T* score—all goals)[b] | | | | | | |
| CIMT | 51.0 (47.5, 54.4) | 54.5 (51.5, 57.6) | 59.0 (55.8, 62.3) | — | — | — |
| HABIT | 59.1 (55.6, 62.7) | 61.3 (58.1, 64.4) | 63.8 (60.5, 67.0) | — | — | — |
| Mean | 55.0 (52.6, 57.5) | 57.9 (55.7, 60.1) | 61.3 (58.9, 63.7) | $P < .001$ (.264) | $P < .001$ (.235) | $P = .412$ (.022) |
| GAS (*T* score—unpracticed) | | | | | | |
| CIMT | 47.5 (39.4, 55.6) | 49.5 (42.9, 56.1) | 59.5 (53.5, 65.5) | — | — | — |
| HABIT | 60.0 (50.3, 69.7) | 60.7 (52.8, 68.6) | 61.4 (54.2, 68.7) | — | — | — |
| Mean | 53.8 (47.4, 60.1) | 55.1 (50.0, 60.3) | 60.5 (55.8, 65.2) | $P < .05$ (.258) | $P = .076$ (.158) | $P = .174$ (.110) |

Abbreviations: CIMT, constraint-induced movement therapy; HABIT, Hand-Arm Intensive Bimanual Therapy; CI, confidence interval; GAS, Goal Attainment Scale.
[a]Other play activities included braiding hair, holding a book or cards, playing with Legos, dressing a doll, and using a remote car.
[b]Average of functional and play *T* scores; Mean, refers to the mean of the CIMT and HABIT groups.

development in CP.[4] Yet these improvements were achieved in just 3 weeks and were maintained at the 6-month follow-up.

Since early development of corticospinal tract (CST) is activity dependent, prolonged movement restraint in the developing infant could conceivably affect the restrained hand.[13,39] Treatments that potentially promote rebalancing M1 excitability, such as bimanual practice or inhibiting contralesional M1 using transcranial magnetic stimulation,[40] may facilitate recovery. Our findings indicate that bimanual training is an effective alternative to CIMT.

Intensive approaches such as CIMT may be additive over repeated exposures.[41] Thus, emphasis should be placed on providing treatment in a child-friendly manner. There could also be an additive effect of combined (CIMT/bimanual) ingredients. Combined treatment showed improvement compared with a group of participants who received the same cumulative amount of an alternative treatment, although the dosing schedules were different.[42] Our findings of similar improvements in hand function for both treatments may indicate that both dosing schedule and intensity may be important.

### *Bimanual Training and Goal Attainment*

Bimanual training affords learning strategies that have direct impact on daily routines. Although both groups achieved expected goal performance, the attainment was higher for the HABIT group at all test sessions. Goal attainment continued to improve for both groups across test sessions, with the CIMT group appearing to make greater continued gains at the 6-month posttest (but $P > .05$). Not surprisingly, most goals were bimanual because

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participants had a well-functioning dominant hand. Performance of bimanual activities in hemiplegic CP is a complex process influenced by internal and external factors, with strategies chosen based on the least negative alternative.[43] Thus, improved bimanual performance is functionally important, and bimanual training allows direct practice of these goals.

It would not be surprising that goals that are practiced more improve more. However, despite the greater goal practice in the HABIT group, this was not the case because the amount of practice did not correlate with GAS improvements. It is particularly interesting that there was greater improvement even for unpracticed goals for the HABIT group. Thus, there was better transfer of practice from other bimanual skills to goal achievement. This transfer could be a result of improvements in the ability to coordinate movements between the 2 hands and/or increased problem solving (identified as important for plasticity)[44] required during bimanual performance, given the increased degrees of freedom. Accordingly, action planning in children with hemiplegia may improve following a combined CIMT/bimanual training.[45]

### Therapeutic Considerations

One potential advantage of CIMT is that the restraint allows the interventionist to focus solely on the more affected hand. Our protocol relies on a 1-to-1 interventionist to child ratio. In the absence of this possibility, the restraint may result in greater intensity because participants would have no choice but to use their more-affected hand (ie, forced use). However, the restraint would preclude opportunity to practice functionally meaningful (bimanual) movements.

The similar improvements regardless of whether or not interventionists were clinicians suggest that the 1-to-1 ratio can be maintained in an economically feasible manner. The similarity is likely a result of the fact that preintervention training is standardized, and clinicians were required to closely adhere to standardized procedures and not administer therapeutic techniques incompatible with motor learning approaches used by HABIT and CIMT.[28] Both treatments were supervised by PT/OTs, who maintained the integrity of the standardized training, which may have further reduced differences.

### Limitations

There may be individual differences that make 1 treatment more effective for a given individual or yield a response difference that depends on the hemiparetic side. There was an unequal distribution of side of hemiparesis in each group. However, when this was taken into account, the results did not change.

It is conceivable that the measures used are not sensitive enough to detect subtle differences between treatment outcomes. For example, there could be differential changes in the spatial–temporal coordination of the hands that can only be determined kinematically. Furthermore, although the sample size was fairly large for a physical rehabilitation study, differences may emerge with a larger and more diverse sample.

We did not randomize interventionist assignment or include a no-treatment/UCC group. However, for the latter, nearly all the improvement occurred at the immediate posttest. Although we only had 1 pretest, the JTTHF during screening (~1 month prior) and at pretest did not significantly differ. Furthermore, earlier studies of CIMT[4,18] and HABIT[21] indicate that these measures do not change over 6 months in a UCC group.

Finally, treatment efficacy may differ at lower dosages (ie, the 90 hours may wash out differences). In fact, an earlier 60-hour HABIT study found that hand function began to return toward baseline at 1-month posttest,[21] whereas we did not find this following 60 hours of CIMT[18] (albeit using different measures) or for either 90-hour treatment here.[46] Further research is required to establish both optimal ingredients, dosage responses, and feasibility in real-world settings.

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## Declaration of Conflicting Interests

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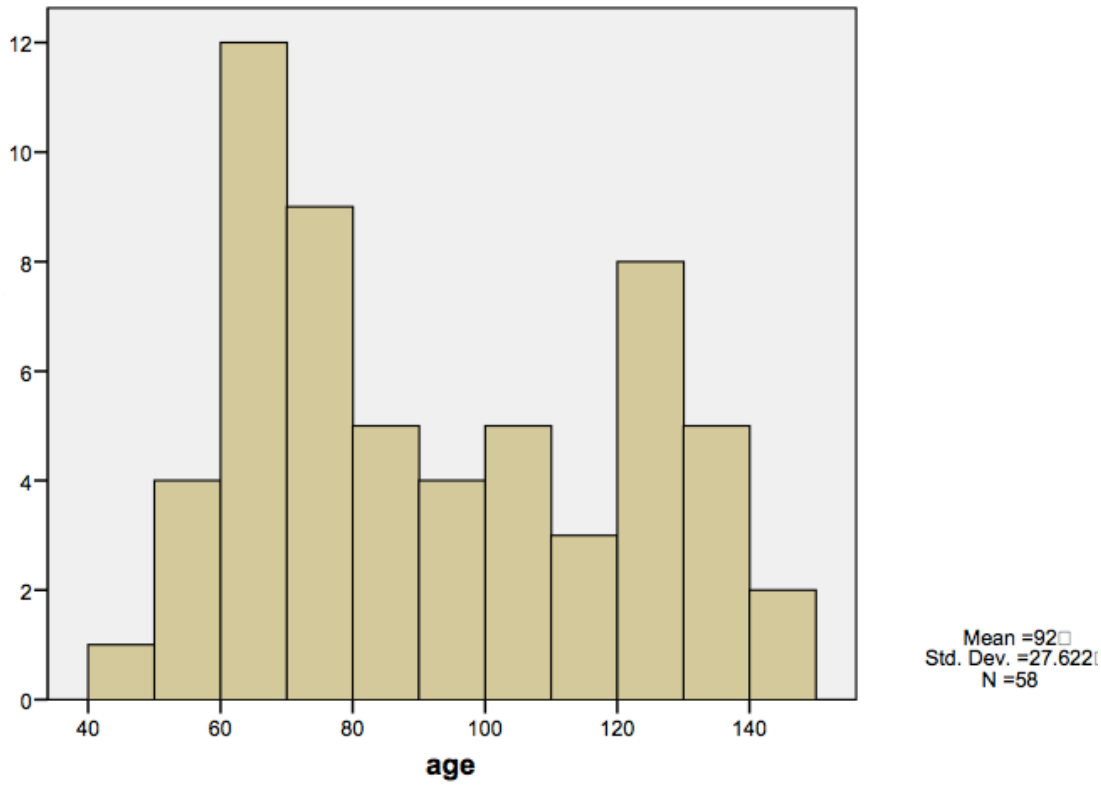
**APPENDIX F**

# Caregivers Responses to Survey

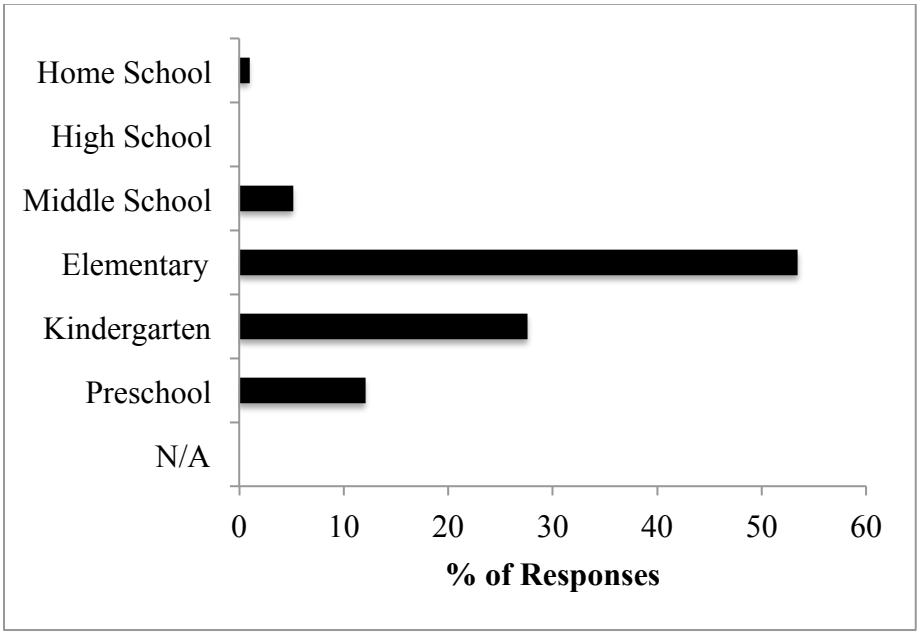
**Children's Information:**

N = 58 (36 female: 22 male)

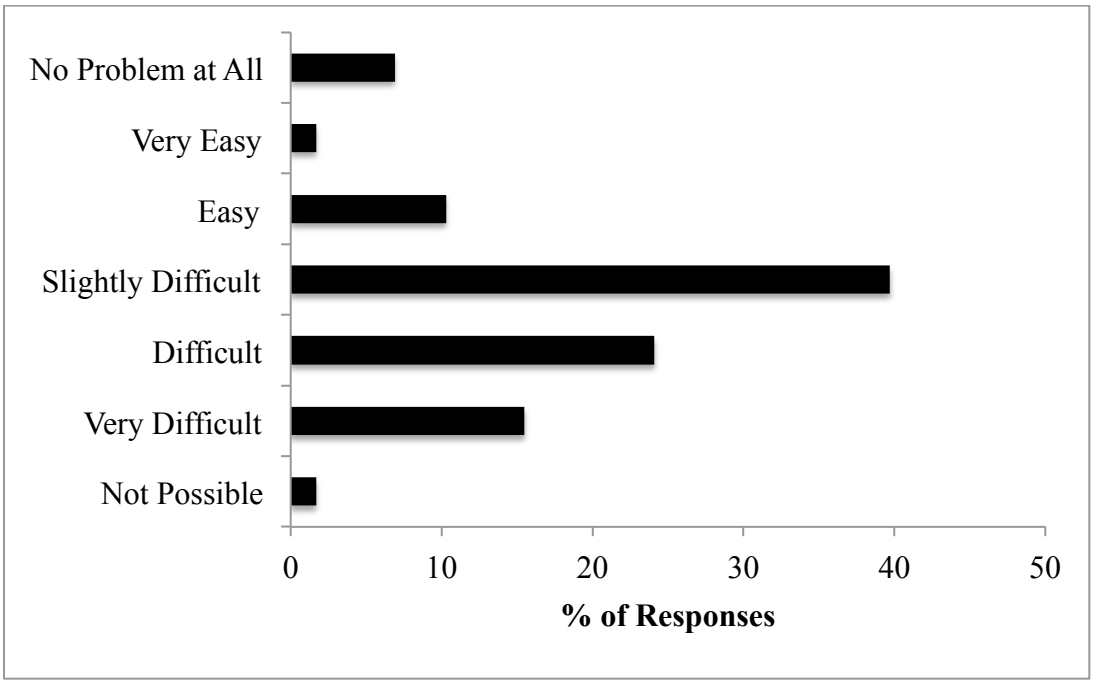
- Frequency Distribution of Ages (in months)



- Distribution of grade/level in school:



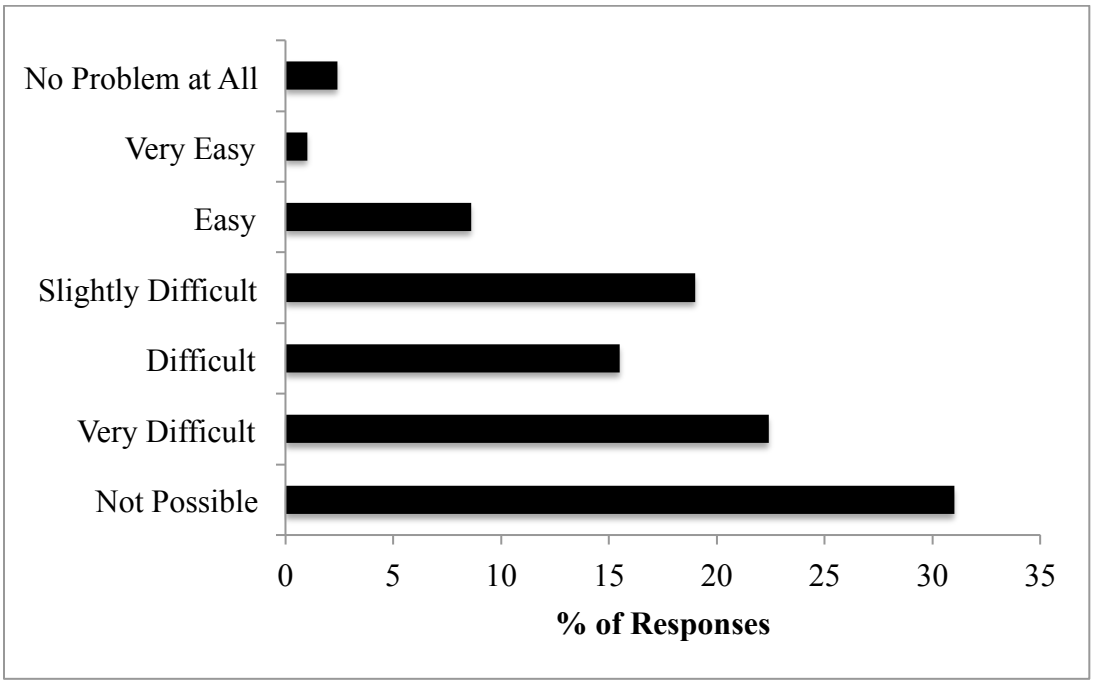

**Q1)** If we provided you with the necessary training and supervision, how difficult would it be for you to participate in a home-based intervention that involved you doing **bimanual training** with your child **2 hrs./day total, 5 days/week, for 9 weeks (total = 90 hours)**?



**Q2)** Would you choose to participate in such a program if available?

**Yes = 75.9 %, No = 24.1%**

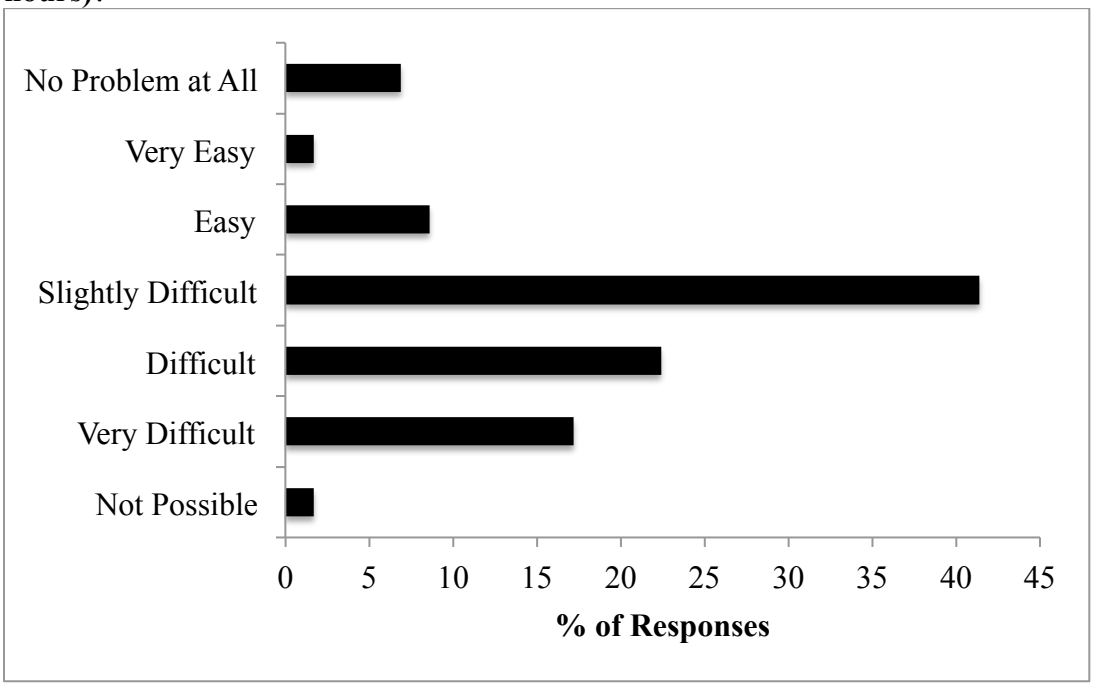
**Q3)** If we provided you with the necessary training and supervision, how difficult would it be for you to participate in a home-based intervention that involved you doing **bimanual training** with your child **6 hrs/day, 5 days/week, for 3 weeks (total = 90 hours)**? Note: the program would occur in summer.




**Q4)** Would you choose to participate in such a program if available?

**Yes = 39.7 %, No = 60.3%**

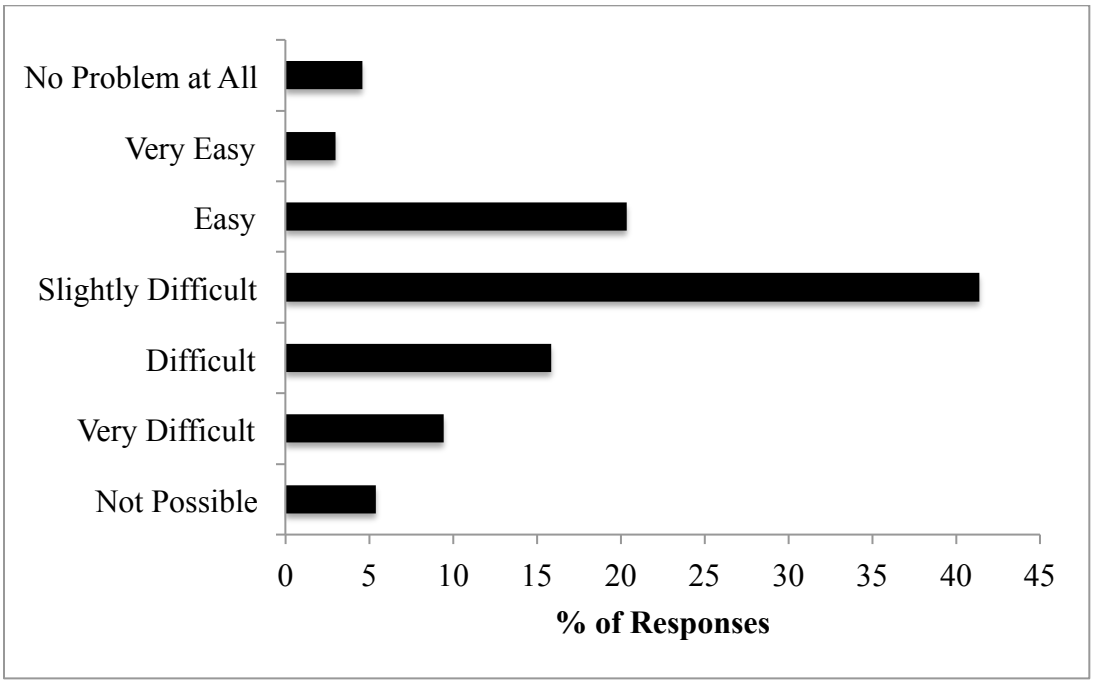
**Q5)** If we provided you with the necessary training and supervision, how difficult would it be for you to participate in a home-based intervention that involved you doing **lower limb training** with your child **2 hrs./day total, 5 days/week, for 9 weeks (total = 90 hours)**?



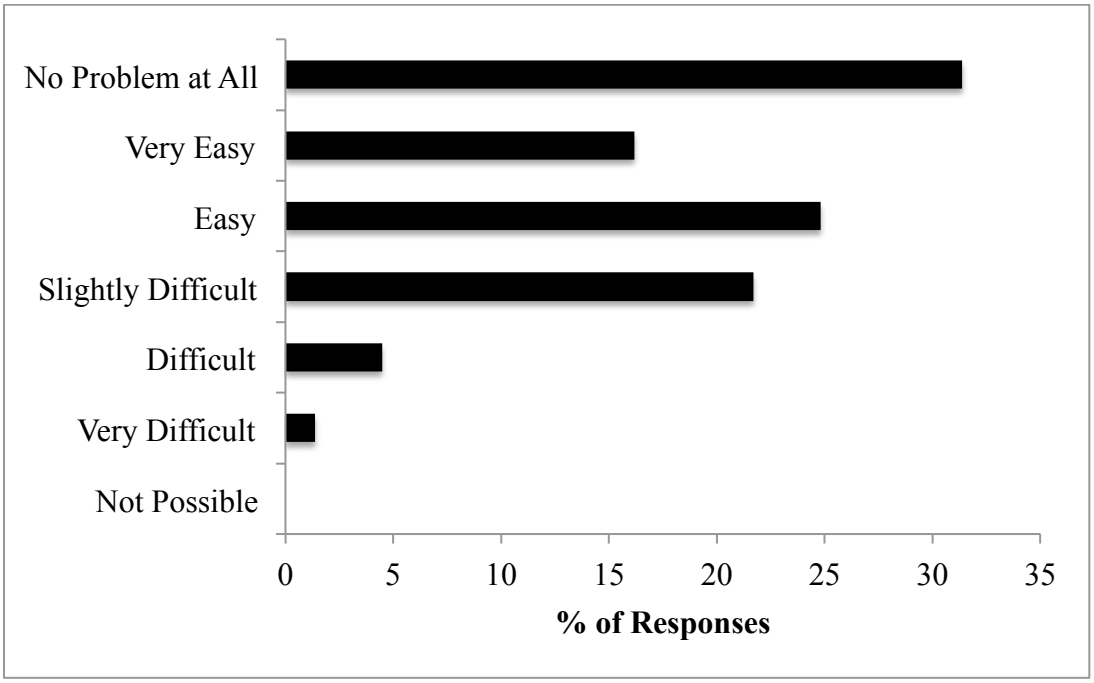

**Q6)** Would you choose to participate in such a program if available?

**Yes = 72.4 %, No = 27.6%**

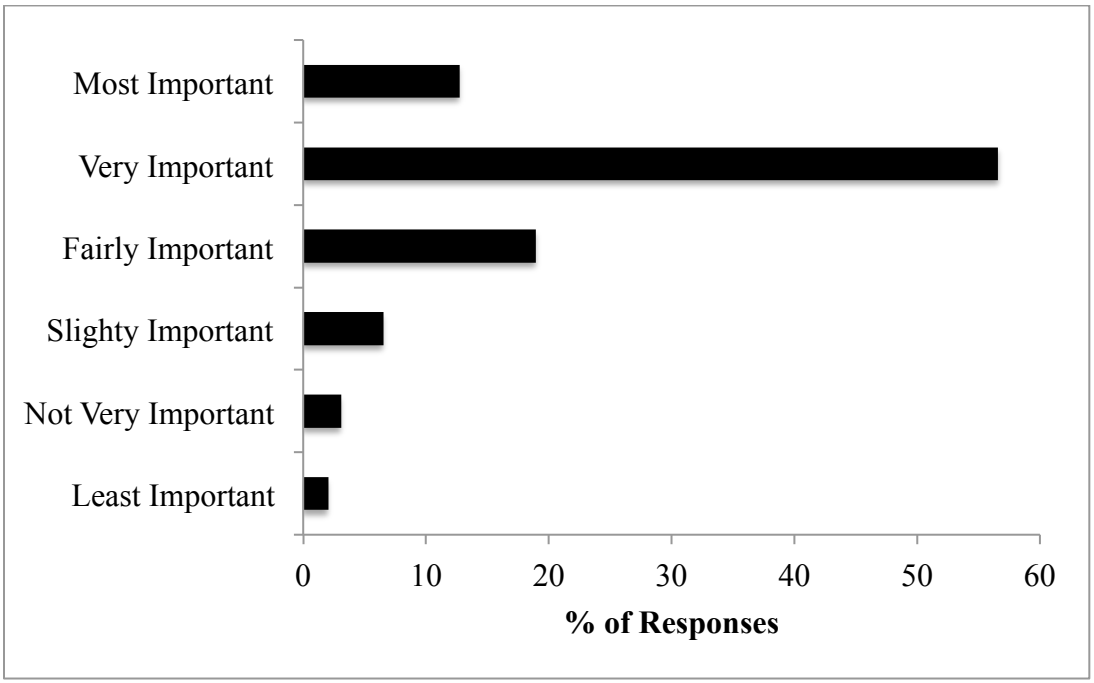
**Q7-11)** How difficult are the following bimanual activities? (Putting on/off shirt, putting on/off pant, putting on footwear, cutting with scissors, taking a cap off marker):



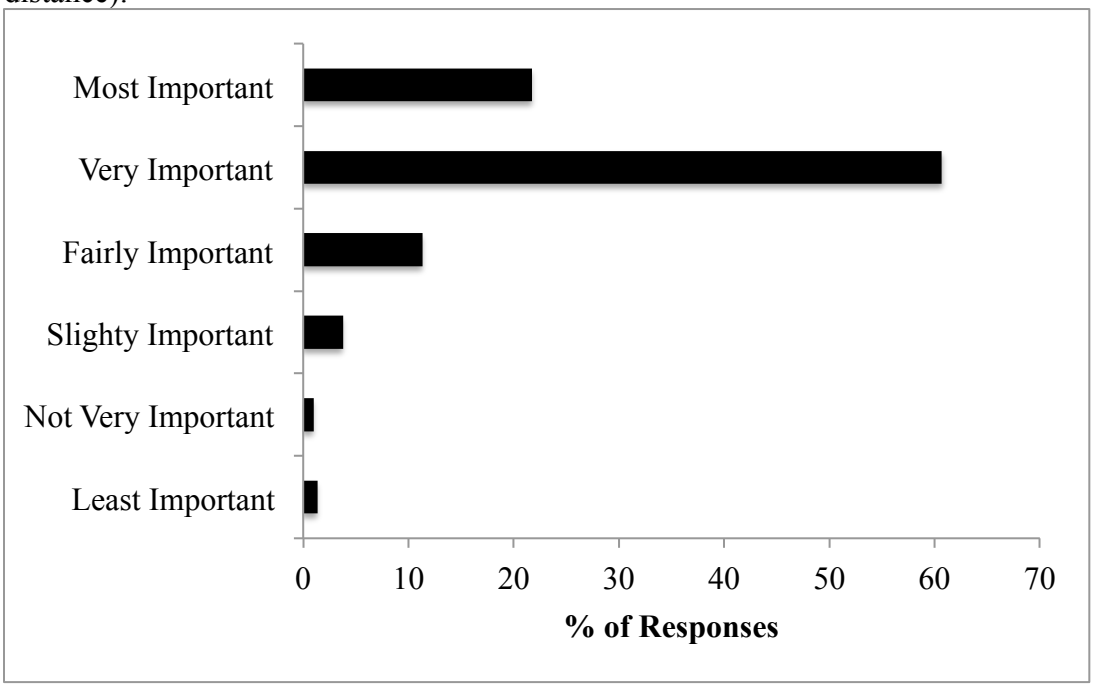

**Q12-16)** How difficult are the following lower limb activities? (Going up and down stairs, standing up from a chair, moving about in public places, getting in and out of a vehicle/train/bus, running over a short distance):



**Q17-21)** How important are the following bimanual activities to your child's quality of life? (Putting on/off shirt, putting on/off pant, putting on footwear, cutting with scissors, taking a cap off marker):




**Q22-26)** How important are the following lower limb activities to your child's quality of life? (Going up and down stairs, standing up from a chair, moving about in public places, getting in and out of a vehicle/train/bus, running over a short distance):



- Parents were asked to rank order of preferred interventions. Listed below are the **frequency of responses** for each intervention ranking:

Home-Based Bimanual – 90hrs in 9 weeks

**1st – 33**
**2nd – 12**
**3rd – 3**

Home-Based Bimanual – 90hrs in 3 weeks

**1st – 6**
**2nd – 8**
**3rd – 29**

Home-Based Lower Limb –90hrs in 9 weeks

**1st – 11**
**2nd – 26**
**3rd – 9**

# APPENDIX G



What is your child's last name?

What is your child's first name?

Activity 1
Please list the name of the activity

Activity 1 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

Activity 2
Please list the name of the activity

Activity 2 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

Activity 3
Please list the name of the activity

Activity 3 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes

○ No

Activity 4
Please list the name of the activity

Activity 4 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

○ Yes
○ No

Activity 5
Please list the name of the activity

Activity 5 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

○ Yes
○ No

Activity 6
Please list the name of the activity

Activity 6 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

○ Yes
○ No

Activity 7
Please list the name of the activity

Activity 7 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

Activity 8
Please list the name of the activity

Activity 8 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

Activity 9
Please list the name of the activity

Activity 9 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

Activity 10
Please list the name of the activity

Activity 10 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes

No

Activity 11
Please list the name of the activity

Activity 11 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

Yes
No

Activity 12
Please list the name of the activity

Activity 12 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

Yes
No

Activity 13
Please list the name of the activity

Activity 13 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

Yes
No

Activity 14
Please list the name of the activity

Activity 14 Time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

Activity 15
Please list the name of the activity

Activity 15 time
Please enter the amount of time (in minutes) spent on this activity

Did you increase the difficulty or complexity of the task?

- Yes
- No

How easy was it to carry out the training session?

| | Very Difficult | Difficult | Neutral | Easy | Very Easy |
|---|---|---|---|---|---|
| Please select one | | | | | |

How well did your child tolerate the training session?

| | Disliked very much | Disliked | Neutral | Tolerated | Tolerated very well |
|---|---|---|---|---|---|
| Please select one | | | | | |

How attentive was your child during the training session?

| | Very distracted | Distracted | Neutral | Attentive | Very attentive |
|---|---|---|---|---|---|
| Please select one | | | | | |

How easy was it to fit today's training session into your schedule?

| | Very Difficult | Difficult | Neutral | Easy | Very Easy |
|---|---|---|---|---|---|
| Please select one | | | | | |

Have you noticed any changes in how your child is using his/her hands?

Do you have any questions or comments?

**APPENDIX H**

R Code for Running Sample Size Simulation*

```
nPerGroup <- 15
nTime     <- 3
muTreat   <- c(57.6, 61.6, 60.6)
muSham    <- c(54.4, 55.3, 55.9)
stdevs    <- c(13.7, 13.9, 13.3)
stdiff    <- 2.45
nSim      <- 1000


Subject <- factor(1:(nPerGroup*2))
Time <- factor(1:nTime, labels = c("Pre", "Post", "Followup"))


theData <- expand.grid(Time, Subject)
names(theData) <- c("Time", "Subject")


tmp <- rep(c("HABIT", "Control"), each = nPerGroup * nTime)
theData$Method <- factor(tmp)


# to set up variance-covariance matrix
ones <- rep(1, nTime)
A <- stdevs^2 %o% ones
B <- (A + t(A) + (stdiff^2)*(diag(nTime) - ones %o% ones))/2


# run it through once to check that it works
library(MASS)
tmp1 <- mvrnorm(nPerGroup, mu = muTreat, Sigma = B)
tmp2 <- mvrnorm(nPerGroup, mu = muSham, Sigma = B)
theData$AHA <- c(as.vector(t(tmp1)), as.vector(t(tmp2)))
aovComp <- aov(AHA ~ Time*Method + Error(Subject/Time), theData)
summary(aovComp)


# descriptive statistics and graphs
print(model.tables(aovComp, "means"), digits = 3)
with(theData, interaction.plot(Time, Method, AHA))
```

```
runTest <- function(){
  tmp1 <- mvrnorm(nPerGroup, mu = muTreat, Sigma = B)
  tmp2 <- mvrnorm(nPerGroup, mu = muSham, Sigma = B)
  theData$AHA <- c(as.vector(t(tmp1)), as.vector(t(tmp2)))
  aovComp <- aov(AHA ~ Time*Method + Error(Subject/Time),
theData)
  b <- summary(aovComp)$'Error: Subject:Time'[[1]][2,5]
  b < 0.05}

  mean(replicate(nSim, runTest()))
```

* Adapted from G.J. Kearns, http://gjkerns.github.io/R/2012/01/20/power-sample-size.html

# APPENDIX I

**Child's Name:**

**For each day of the week in the schedule at the end of this form, please list the times your child:**

- leaves for and arrives from preschool/school
- is at daycare or with a babysitter
- does school homework
- is at physical/occupational therapy
- is at any other therapy (e.g. speech, school tutoring, etc.)
- has any extracurricular activities (e.g. swimming class, dance, gymnastics, etc.)
- is involved in any regularly occurring weekend activities (e.g. play dates)

**For each day of the week, please list the times you:**

- leave for and arrive from work
- have to take you child's sibling to any extracurricular activities
- have any of your own extracurricular activities (e.g. gym)

**Please list the 5 days, and specify the 2 hours in which you will do the activities:**

**Do you have any travel plans scheduled? If so, please indicate the dates:**

**If your child has any siblings, please indicate who will provide childcare during the daily 2 hours of activities:**

**In the event that the above person becomes ill or becomes unavailable, please indicate who will serve as an alternative for helping with childcare:**

## Monday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## Tuesday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## Wednesday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## Thursday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## Friday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## Saturday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## Sunday

| TIME | Activity | TIME | Activity |
|---|---|---|---|
| 7:00-7:30 | | 2:00-2:30 | |
| 7:30-8:00 | | 2:30-3:00 | |
| 8:00:8:30 | | 3:00-3:30 | |
| 8:30-9:00 | | 3:30-4:00 | |
| 9:00-9:30 | | 4:00-4:30 | |
| 9:30-10:00 | | 4:30-5:00 | |
| 10:00-10:30 | | 5:00-5:30 | |
| 10:30-11:00 | | 5:30-6:00 | |
| 11:00-11:30 | | 6:00-6:30 | |
| 11:30-12:00 | | 6:30:-7:00 | |
| 12:00-12:30 | | 7:00-7:30 | |
| 12:30-1:00 | | 7:30-8:00 | |
| 1:00-1:30 | | 8:00-8:30 | |
| 1:30-2:00 | | 8:30-9:00 | |

## APPENDIX J

### 1. Inquiring about ideas for activities:

**Participant A**
January 19
I am just starting HABIT training with my 3-year-old. Did anyone find particular toys/activities that worked particularly well for toddlers?
Unlike · Comment

- Seen by 10

You like this.

-

**Participant B** My daughter enjoyed Legos, play dough, beads, cutting paper, and unwrapping candy.
January 19 at 4:24pm · Like · 2

-

**Supervisor 3** There is an IOS app called CPToys that has a good battery to choose from. We know the OT researcher from Australia who developed it, and the nominal cost supports the large amount of time he put into developing the app. We re not affiliated in any way. Let us know if you have questions.
January 20 at 9:16am · Like · 2
Here is the link: http://www.cptoys.org/

CPtoys
Matching toys to upper limb treatment goals
CPTOYS.ORGIBY DR BRIAN HOARE
January 20 at 9:16am · Like · 1 · Remove Preview

-

**Participant A**
Great ideas! Thank you.
January 20 at 12:56pm · Like

## 2. Supervisor engaging participants to share ideas for reward systems:

**Supervisor 1**

September 22, 2014

Does anyone want to share some of the reward systems they used for keeping kids motivated during the 9 weeks? I know many of you were extremely creative. Obviously each child is unique and what works for one might not work for another, but its always fun to share some of these ideas.

Like · Comment

- Seen by 14

Participant C and 2 others like this.

- **Participant B** My daughter was three years old, so she required immediate rewards. Candy, opportunity to pick out activities that she found enjoyable in between activities that were more frustrating for her, and lots of encouragement.

  September 22, 2014 at 2:25pm · Unlike · 4

- **Participant C** Morsels of candy like chocolate chips or butterscotch morsels. They are small so they go a long way when you need extra bribes. Getting to play outside as the big end of the day reward. Watching a favorite cartoon after an hour or 30 minutes of play - just depending on the mood. Setting the timer so they know when it's reward time. Lots of clapping and praise too.

  September 22, 2014 at 2:43pm · Unlike · 3

### 3. Caregiver sharing achievement with other caregivers:

**Participant D** shared her video.

May 14, 2014

I never thought I would see this day. Even two days earlier, he could not do this (I saw him try). This video is of his second pull-up--after I scraped my jaw off the ground and grabbed my camera. The first one, he went up even higher. He is always exceeding my expectations, proving that anything is possible.

00:13

Unlike · Comment

- Seen by 15

You, Supervisor 2 and 3 others like this.

4. **Caregiver sharing photo of trophy purchased by another caregiver after completing intervention:**

**Participant E**
October 3, 2013
Child C celebrating the end of 9 weeks and super excited about the awesome trophy! (Thank you Participant F!)

Unlike · Comment

- Seen by 14