

Intensive Bimanual Intervention for Children Who Have Undergone Hemispherectomy: A Pilot Study

Maxime T. Robert, PhD; Claudio L. Ferre, PhD; Karen Y. Chin, MSc; Marina B. Brandao, PhD; Jason Carmel, MD, PhD; Rodrigo Araneda, PhD; Yannick Bleyenheuft, PhD; Kathleen Friel, PhD; Andrew M. Gordon, PhD

Centre for Interdisciplinary Research in Rehabilitation and Social Integration (Dr Robert), Faculty of Medicine, Université Laval, Quebec, Canada; Department of Occupational Therapy (Dr Ferre), Boston University, Boston, Massachusetts; Department of Biobehavioral Sciences (Ms Chin and Dr Gordon), Teachers College, Columbia University, New York, New York; Burke Neurological Institute (Ms Chin and Dr Friel), Weill Cornell Medicine, New York, New York; Graduate Program in Rehabilitation Sciences (Dr Brandao), School of Physical Education, Physical Therapy and Occupational Therapy, Universidade Federal de Minas Gerais, Belo Horizonte, Minas Gerais, Brazil; Weinberg Family Cerebral Palsy Center (Dr Carmel), Vagelos College of Physicians and Surgeons, Columbia University, New York, New York; Institute of Neuroscience (Drs Araneda and Bleyenheuft), Université catholique de Louvain, Brussels, Belgium.

Purpose: To conduct a pilot study to assess the feasibility and effectiveness of an intensive bimanual intervention on upper limb function in children who have undergone hemispherectomy.

Methods: Thirteen children received 90 hours of intensive bimanual training (Hand-Arm Bimanual Intensive Therapy, HABIT). The Jebsen-Taylor Test of Hand Function (JTTHF), Box and Block Test (BBT), Assisting Hand Assessment (AHA), ABILHAND-Kids, and Canadian Occupational Performance Measure (COPM) were assessed by a masked clinician twice before, immediately, and 6 months after treatment.

Results: Significant improvements over time were found in the JTTHF, AHA, ABILHAND-Kids, and COPM.

Conclusion: Completion of HABIT was feasible for children with hemispherectomy. Improvement of bimanual function and functional goals can be related to the nature of the activities prioritized in HABIT training. (*Pediatr Phys Ther* 2021;33:120–127)

Key words: bimanual therapy, diffusion MRI, pediatric, rehabilitation

INTRODUCTION

Hemispherectomies are used for seizures emanating from one hemisphere that cannot be controlled with antiepilepsy medication. Hemispherectomy can be classified as either anatomical or functional and is performed on approximately 16% of individuals with intractable seizures.¹ Moreover, hemispherectomy results in complete severing of the descending motor tracts and the corpus callosum. These procedures generally result in a contralateral hemiparesis with a varying degree of sensorimotor impairments.² Some children with congenital hemiplegia due to cerebral palsy (CP) also develop intractable seizures requiring hemispherectomy.³ Despite a remarkable

ability for the brain to functionally “rewire” itself depending on the age at surgery,⁴ the resulting contralateral hemiparesis is often associated with difficulties in performing daily activities.²

Constraint-induced movement therapy (CIMT), in which the less-affected upper extremity is restrained, is considered one of most efficacious interventions for hand function in children with unilateral CP.^{5–8} CIMT is designed so that children are actively engaged in unimanual activities. Modest improvement in hand function has been reported in children following hemispherectomy.⁹ However, there are some limitations to this approach. For example, CIMT focuses on unimanual dexterity, which may greatly limit the possibility for children with hemispherectomy to be independent in their daily routines, as most activities require the simultaneous use of both hands. More importantly, after hemispherectomy, approximately one-third of children experience a near-complete loss of ability to grasp, release, and manipulate objects.² Thus, many CIMT activities may be difficult or frustrating.

A more realistic therapy goal for children with hemispherectomy may be to improve function of the affected upper extremity as a nondominant assist for tasks that require 2 hands. Hand-Arm Bimanual Intensive Therapy (HABIT) is a structured form of bimanual training that requires use of both hands to complete activities developed for children with CP.^{5,10} HABIT has shown similar unimanual and bimanual hand improvements for children with CP regardless of the corticospinal tract (CST)

0898-5669/110/3303-0120

Pediatric Physical Therapy

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Correspondence: Maxime T. Robert, PhD, Teachers College, Columbia University, 525 W. 120th St, Box 93, New York, NY 10027 (maxime.robert@fmed.ulaval.ca).

Grant Support: This work was supported by a grant from the Brain Recovery Project. M.T.R. was supported by a postdoctoral training award from Fonds de Recherche en Santé du Québec.

The authors declare no conflicts of interest.

DOI: 10.1097/PEP.0000000000000804

organization (ipsilateral vs contralateral) following an intensive bimanual intervention.¹¹ More recently, HABIT was modified to include the lower extremities (HABIT-ILE)¹² where improvements similar to the upper extremity are observed in children with CP.¹³ The extent to which HABIT may benefit children following hemispherectomy including when hand function is severely affected is unknown.

The purpose of this pilot study was to determine the feasibility and preliminary effectiveness of HABIT to improve upper extremity function in children with hemispherectomy. We hypothesized that bimanual function would improve following HABIT.

METHODS

Design

A longitudinal study design was used. Informed assent and consent were obtained from participants and caregivers, respectively. This study was approved by the Institutional Review Boards (IRBs) of Teachers College, Columbia University, Burke Neurological Institute, Weill Cornell Medical College, and Université catholique de Louvain.

Participants

Thirteen children participated (9 males, 4 females, age 7.516.5 years, with hemispherectomy; n = 10, functional, n = 3 anatomical; n = 3 ipsilateral, n = 3 contralateral projections; n = 8 right, n = 5 left hemisphere; n = 8, congenital; Table). Eleven children participated in New York HABIT summer camps and 2 in Brussels HABIT-ILE summer camps. Participants were recruited from our Web site (<http://www.tc.edu/centers/cit/>), online communities, or Belgian university hospitals. Prior the intervention, potential participants received an on-site physical examination or instructions were sent to their caregiver or physical or occupational therapist on videoing the motoric inclusion criteria described next. This allowed us to verify testing compliance. Inclusion criteria included: (1) age 6 to 17 years, (2) undergone hemispherectomy, (3) capable of participating in a 6-hour long-day camp based on the parent's perception (note that all participants attended school for a similar duration), (4) capable of following directions regarding hand use and testing, (5) capable of communicating needs, (6) nonactive seizure within 6 months, (7) active movement of proximal parts of the limb, and (8) able to use the more affected arm as a stabilizer. Exclusion criteria included: (1) unwillingness to comply with instructions or other behavioral issues making delivery of an intensive therapy unfeasible, (2) health problems unassociated with hemiplegia, (3) visual problems interfering with treatment/testing (ie, participants had to be able to see and interact with objects although moving them to allow this was permitted), (4) orthopedic surgery on the more-affected hand within 1 year, and (5) botulinum toxin within the past 6 months.

Procedure

For the New York program, assessments were collected twice before, immediately after, and 6 months after training. For the Brussels program, assessments were collected once before

TABLE
Demographic and Clinical Characteristics of Participants

Participants	Gender	Ethnicity	Affected Hemisphere	MACS Score	Age at Camp	Age at Surgery	Congenital Hemiplegia	Medical History	Diffusion MRI	Lateralization	Surgery
HX1	Female	White	Left	II	15.4	9.3	Yes	Perinatal stroke	No	Not available	Functional
HX2	Male	White	Left	II	14.3	0.5	No	Hydrocephalus	No	Not available	Anatomical
HX3	Male	White	Right	III	10.3	6.6	Yes	In utero stroke	No	Not available	Functional
HX4	Male	White	Right	II	15.5	11.3	No	Porencephaly	No	Not available	Functional
HX5	Male	Hispanic	Right	III	7.5	2.5	No	Epilepsy	No	Not available	Functional
HX6	Male	White	Right	III	11.3	9.5	Yes	Rasmussen's encephalitis	Yes	Ipsilateral	Functional
HX7	Male	White	Right	III	7.5	1.2	Yes	In utero stroke	Yes	Contralateral	Functional
HX8	Female	White	Right	III	9.1	0.9	Yes	Perinatal stroke	Yes	Ipsilateral	Anatomical
HX9	Male	White	Right	II	8.0	0.9	Yes	Perinatal stroke	Yes	Contralateral	Functional
HX10	Female	White	Right	II	16.5	12.2	No	Rasmussen's encephalitis	No	Not available	Anatomical
HX11	Male	White	Left	III	9.86	12.5	Yes	In utero stroke	No	Not available	Functional
HX12	Male	Hispanic	Left	II	10.6	10.2	Yes	Perinatal stroke	Yes	Ipsilateral	Functional
HX13	Female	Hispanic	Left	III	10.3	3.1	No	Not available	Yes	Contralateral	Functional

Abbreviations: MACS, Manual Ability Classification System; MRI, magnetic resonance imaging.

(due to IRB constraints), immediately after, and 3 months after training. A total of 6 bimanual training day-camps were conducted from 2013 to 2016 (4 at Columbia University and 2 at Université catholique de Louvain). Each camp had between 1 and 4 children with hemispherectomy together in a room with other children with congenital hemiplegia participating in a separate study.^{11,14,15} For both programs, the room had between 4 and 6 children in total and participants were engaged in treatment for 90 hours during 15 (New York) or 10 days (Brussels). Because of the relatively small population of children with hemispherectomy and an absence of difference between 6 hours a day or 9 hours a day previously reported,¹³ we combined the data from both sites. Both programs had an interventionist for each child and individualized the therapy based on the child's functional level and goals. Interventionists consisted of physical and occupational therapists, graduate students in kinesiology/neuroscience, speech pathology, psychology, and undergraduates. Each interventionist received a 2-hour training, which focused on strategies to engage children in use of hands and safety with additional ongoing training during the interventions and daily team meetings. The camp room had experienced supervisors responsible for ensuring treatment fidelity and uniformity. Children were engaged in fine and gross motor activities individually chosen according to the child's abilities to use the affected hand as a manipulator or stabilizer. Selected activities took into consideration possible cognitive, behavioral, motor, and visual impairments. In case of visual impairments, the task environment was modified accordingly to ensure the success of the task. Visually they had to be able to see and interact with objects. For one child, we had to place objects relatively close and within his visual field. Other modifications included providing either 1-step commands or guiding children step-by-step through the activity as needed. Most children however were able to participate without these additional modifications.

Task difficulty was progressively graded based on the child's motor capacities. The procedures mainly included the use of whole task practice (sequencing successive movements in the context of activities, such as self-care or play activities), and individualized functional goal training. Whenever possible, we included part task practice (practice of specific components of the task in a repetitive sequence of 30 seconds). The activities chosen for goal training were selected according to the children's and parents' priorities, reported using the Canadian Occupational Performance Measure (COPM) at baseline. For additional details about HABIT, see Charles and Gordon,⁵ Gordon,⁶ and Gordon et al.¹⁰

Behavioral Measures

For the New York program, participants were evaluated approximately 1 week prior (pre1), directly prior (pre2), immediately after (post), and 6 months (follow-up) after treatment. For the Brussels program, participants were evaluated before, 2 days after, and 3 months after treatment. Every assessment was done by the same experienced physical therapist who was masked to the training. Five outcome measures were used to quantify bimanual performance, functional goals, and unimanual capacity.

The Assisting Hand Assessment (AHA, version 5.0) quantifies the effectiveness with which a child with unilateral disabilities uses his or her affected (assisting) hand in bimanual activities.¹⁶ The AHA has excellent validity/reliability. The test was videotaped and scored offsite by a certified occupational therapist who is an experienced AHA instructor. The occupational therapist was also masked to the training. Data were reported in logit-based units (AHA-units). An improvement of 5 units is considered the smallest detectable difference.¹⁷

The ABILHAND-Kids is a valid/reliable questionnaire assessing the perceived manual ability of children.¹⁸ The test comprises a list of manual activities in which the caregivers score the amount of difficulty children may experience during their performance in activities of daily living that require hand use. Data were reported in logit-based units.

To establish and measure children's functional goals, we conducted the COPM with the children and caregivers.¹⁹ The COPM identifies and measures changes in functional problems considered relevant by clients through interview, and is valid/reliable. The most relevant functional goals to be accomplished are defined and ranked in importance. Then, they rate child's performance and their satisfaction with child's performance in the 5 most important goals. 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. A change of 2 points or more is considered clinically meaningful.

Unimanual dexterity was assessed using (1) the Jebsen-Taylor Test of Hand Function (JTTHF) and (2) the Box and Block Test (BBT). The JTTHF is a standardized timed test of simulated functional tasks quantifying the time to complete a battery of unimanual activities.²⁰ The activities include flipping index cards, object placement, simulated eating, stacking checkers, and manipulating empty and full cans. Reliability for children with nonprogressive hand disabilities is high.²¹ The BBT measures the number of blocks moved between the 2 boxes in 60 seconds.²² Both unimanual dexterity tests were performed on each hand separately starting with the more affected hand.

MRI Data Acquisition

Three participants (HX5, HX10, and HX11) were excluded due to Medtronic plates that were not certified safe in a 3T magnetic resonance imaging (MRI). Neuroimaging was performed on 10 participants. Due to excessive head movements and noise during the MRI, diffusion tensor imaging (DTI) was not performed on 4 participants. Thus, DTI was performed in a total of 6 participants, before training. The DTI was used to reconstruct the CST to identify the absence or presence of fibers. The MRI protocol was performed on a 3T scanner (Siemens Magnetom Trio, Citigroup Biomedical Imaging Center, Weill Cornell Medical College). A total of 75 slices were acquired (matrix 112×112 , field of view [FOV] = 224 mm, 65 directions, b-value = 1000 s/mm^2 , repetition time [TR] = 9000 ms, echo time [TE] = 83 ms). Participants from the Brussels program had T1-weighted MRI performed with a 3T scanner (Philips, Eindhoven, the Netherlands). A total of 70 slices were acquired (matrix 112×112 , FOV = 224 mm, 55 directions, b-value = 800 s/mm^2 , TR = 6422 ms, TE = 83 ms). The

participants were positioned in a supine position with padding to minimize head movements. The participants did not receive sedation.

MRI Data Analysis

For the New York program, DTI analysis was performed using DTI Studio (John Hopkins University, Baltimore, Maryland, whereas for the Brussels program, DTI analysis was performed using BrainVoyager (Brain Innovation B.V., Maastricht, the Netherlands). An image was first created to mask the background noise at the threshold of 30 dB, using standard linear regression for tensor calculation. Images containing movement artifacts were excluded by visually inspecting the original images using the apparent diffusion constant function.²³ Reconstruction of the CST was performed using the Continuous Tracking method.²⁴ Fiber tracking started less than 0.15 and was terminated if the tract turning angle was more than 70 (for a similar procedure, see Kuo et al¹⁴). The region of interest to identify the CST was determined using anatomical location.²⁵ Two investigators (M.T.R. and C.L.F.) independently established the lateralization of the CST with complete agreement between them.

Data Analysis

Statistical analysis was performed using SPSS (version 25, Statistical Production and Service Solutions, Chicago, Illinois). Gaussian distribution was verified using a test of goodness of fit. Logs transformation was performed in case of an absence of normal distribution. A mixed linear model on test sessions was performed for clinical outcomes with time as a fixed factorial factor.²⁶ Identification of the participants was a random effect. An unstructured covariance structure model was prioritized, as it requires no assumption in the error structure and the most commonly used in longitudinal data.²⁶ Mixed linear models allow the estimation of interindividual variability and intraindividual patterns of change over time and take the different time points between Brussels and New York and missing data into account. The time factor was the independent variable and the clinical measures were the dependent variables.

RESULTS

Patient Flow

During recruitment, 39 individuals were screened (Figure 1). Ultimately, 13 qualified individuals agreed to participate. Among those, 4 children were unable to attend the follow-up assessment because of testing burden associated with not being local and/or no response to subsequent contact. Technical difficulties accounted for 1 missing AHA video and 1 missed AHA scoring (HX12).

Participant characteristics are shown in the Table. Six of 13 participants completed the diffusion MRI at baseline. Three participants had contralateral (crossed) projections and 3 had ipsilateral (same side) projections. Lateralization was not an indicator of baseline hand function nor changes overtime for any measure (Table). There was a Gaussian distribution for all measures with the exception of the JTTHF, as 5 children received

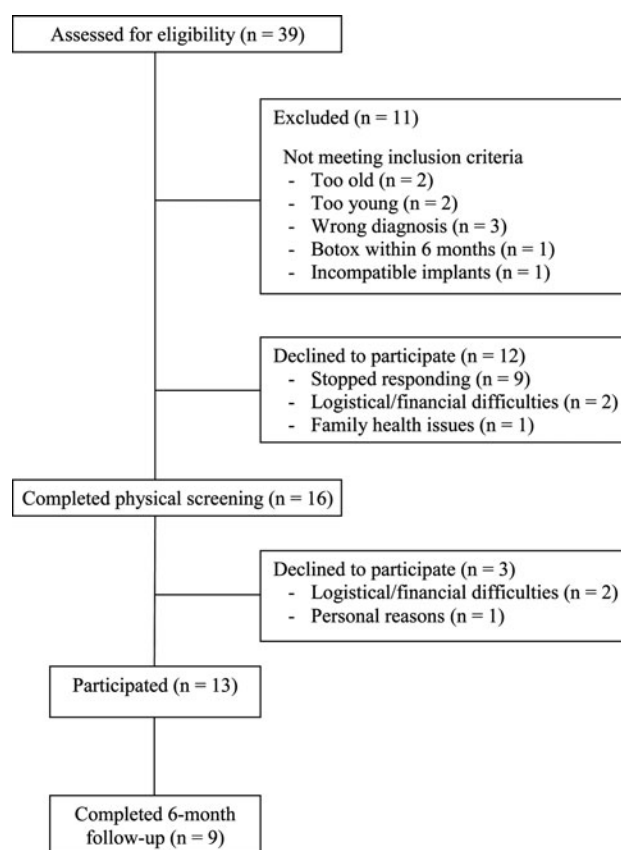


Fig. 1. CONSORT flow diagram showing progress through the stages of the study, including flow of participants, withdrawals, and inclusion in analyses. A total of 39 children were screened via telephone/email, and 11 of these were excluded for the following reasons: too old ($n = 2$), too young ($n = 2$), wrong diagnosis ($n = 3$), botulinum toxin treatment within 6 months ($n = 1$), incompatible implants ($n = 1$), and poor cognition ($n = 1$). A total of 28 children potentially met the study criteria and were invited to undergo physical screening; 12 children chose not to undergo physical screening. Of the remaining 16 children, 3 declined to participate: logistical/financial difficulties ($n = 2$) and personal reasons ($n = 1$). Ultimately, 13 of the remaining children agreed to participate. A total of 9 children completed the 6-month follow-up assessment.

maximum scores (ie, being unable to complete any of the tasks within the allocated time). Thus, log transformation was performed on this measure. For the clinical measurements, congenital hemiplegia and time from surgery were added as factors in the mixed linear model. These were not significant ($P > .05$). No change was observed for any measure for the less-affected hand ($P > .05$). No significant changes were observed between the 2 preassessments for the clinical measures except where noted. Similar results were obtained when the 2 children from the Brussels program were excluded in the analysis.

Treatment Characteristics

All children completed the 90 hours of training. On average, children spent 96.9% ($SD = 3.1\%$) of their time in whole task practice and 3.1% ($SD = 3.0\%$) on part task practice.

Clinical Measures

Reliability. For all clinical measures, the intrarater reliability between the 2 baselines was good to excellent.

Specifically, it was 0.848 (confidence interval [CI] = 0.389-0.962) for the AHA, 0.792 (CI = 0.226-0.944) for the ABILHAND-Kids, 0.862 (CI = 0.443-0.966) for the COPM performance, 0.921 (CI = 0.680-0.980) for the COPM satisfaction, 0.951 (CI = 0.819-0.987) for the BBT, and 0.814 (CI = 0.309-0.950) for the JTTHF.

Bimanual Hand Use. For the AHA, a significant difference was observed from pre1 to pre2 ($F = 15.684$, $df = 9$, $P = .003$). AHA changes showed significant improvement across time points (mean difference = 34.90; 95% CI = 31.499-

38.302; $P < .001$). Nine children had clinical meaningful improvement from pre to post and 7 of them from pre to follow-up (Figure 2A).

Functional Goals and Daily Functioning. Significant improvement of the ABILHAND-Kids was observed across time points (mean difference = 2.03; 95% CI = 1.383-2.671; $P < .001$; Figure 2B).

The majority of goals (80%) for the COPM indicated by the caregiver and the child were bimanual. Most of the goals composed of self-care activities (eg, dressing, grooming, and eating),

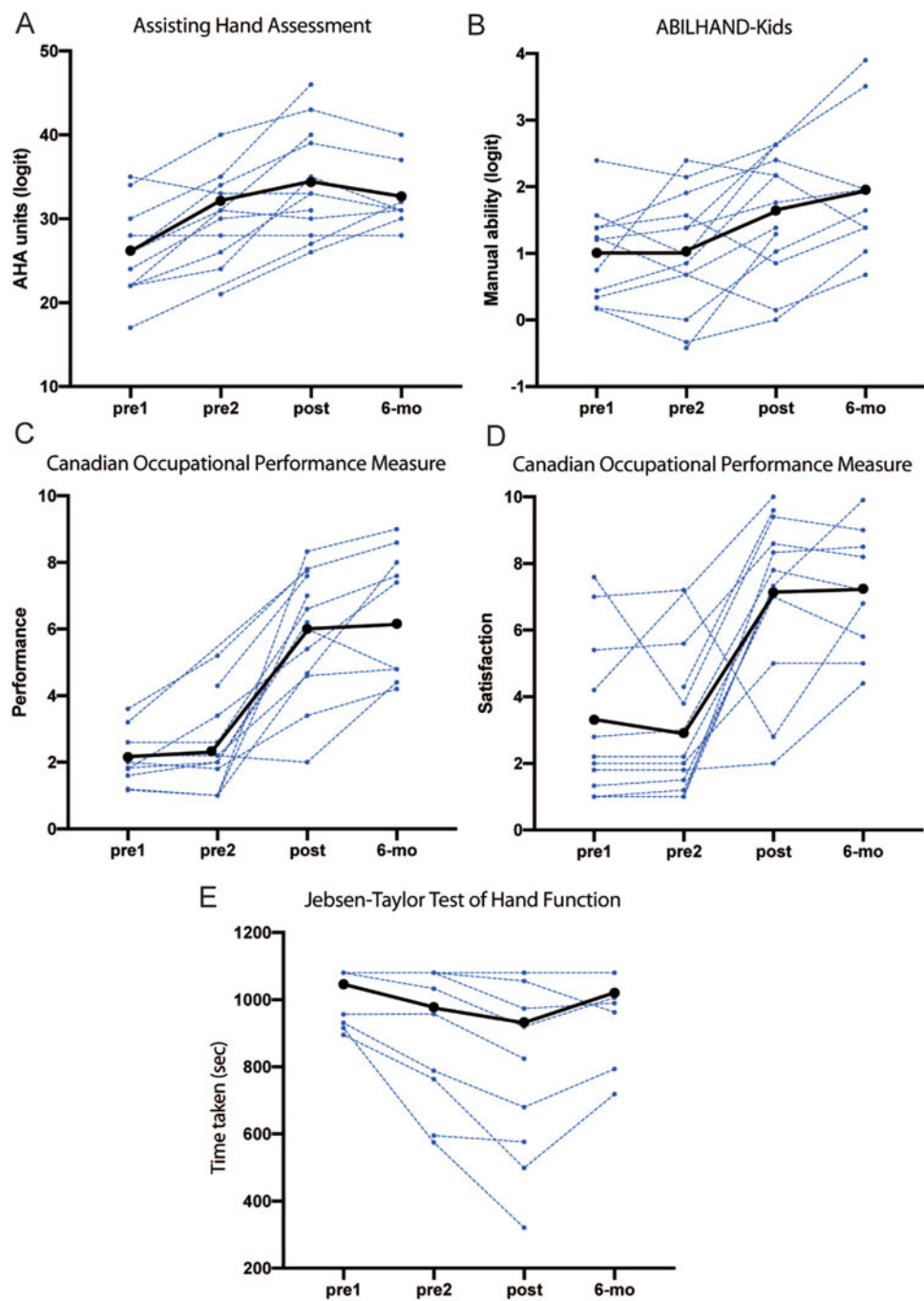


Fig. 2. Individual scores for the (A) Assisting Hand Assessment, (B) ABILHAND-Kids, (C) Canadian Occupational Performance Measure—Performance, (D) Canadian Occupational Performance Measure—Satisfaction, and (E) Jebsen-Taylor Test of Hand Function (JTTHF) for the more affected hand. Note that 5 children had a maximum score of 1080 on the JTTHF, which did not change during testing. This figure is available in color online (www.pedpt.com).

followed by play (eg, ball activities). The linear mixed model analysis showed significant improvement of COPM across time points for performance (mean difference = 6.594; 95% CI = 5.284-7.806; $P < .001$; Figure 2C) and satisfaction (mean difference = 7.11; 95% CI = 4.643-9.584; $P = .004$; Figure 2D). Clinical meaningful improvements from pre1 to post were observed in 10 children for performance and 11 children for satisfaction. Clinical meaningful improvements from pre1 to follow-up were observed in 7 children for performance and 9 children for satisfaction out of the 9 tested at follow-up.

Manual Dexterity. There were no significant improvements of the BBT across time points ($P > .05$). Only one child had a clinically meaningful improvement from pretest to follow-up.

Significant improvement across time points was observed for the JTTHF (mean difference = 2.96; 95% CI = 4.643-9.584; $P = .004$; Figure 2E). Five of 7 children who had initially achieved maximum scores (1080s, reflecting an inability of the test to measure above that) had no improvements.

DISCUSSION

The objective of this pilot study was to determine the feasibility and preliminary effectiveness of HABIT on upper limb function in children with hemispherectomy. Our results demonstrated significant improvements in bimanual use, unimanual dexterity (JTTHF), and functional goals and daily functioning.

Only one other study measured the effect of an intensive intervention of 30-hour CIMT on upper limb function in 4 individuals with hemispherectomy.⁹ Based on their BBT assessments, there were several differences compared with our results.⁹ Among those, unimanual grasping significantly improved in the former but the BBT score did not improve in the present study. A possible explanation may be the difference in impairment levels. In the CIMT study,⁹ the individuals had mild to moderate impairments, whereas in our group, more than half of the children were unable to transfer one block in the BBT test, hypothetically due to a lower integrity of the sensorimotor pathways as compared with their peers without motor problems.²⁷ Moreover, lack of improvement in unimanual grasping may be explained by the children's difficulty to manipulate objects, as reflected by the percent spent in part task practice. While our study results showed only 3% of the time spent was in part task practice, previous studies reported up to 21% on average in children with CP.^{10,28} Furthermore, while children with a Manual Ability Classification System (MACS) score of II were able to find strategies to overcome the difficulties, those with an MACS score of III usually had mild cognitive impairments, were unable to do so. In our study, despite the severity of the impairments, 2 children demonstrated improvement on the BBT score at post and follow-up, possibly resulting from the reasons listed earlier and a more intensive intervention as compared with de Bode et al's⁹ study. Our lack of BBT improvement may have reflected the inability of BBT to capture intraindividual changes overtime. Improvements of unimanual grasping in de Bode et al's⁹ study may also be explained by the fact that most activities of CIMT focus on unilateral activities involving grasping, whereas in HABIT, the focus is on improving bimanual

coordination through active stabilisation.⁵ Nonetheless, children were able to decrease JTTHF scores of the more affected hand over time, demonstrating improvements in hand dexterity. A possible explanation of improvement in the JTTHF scores is the similarity between the activities (ie, object placement, simulated eating, stacking checkers, and manipulating empty, and full cans) practiced during training and the battery of unimanual activities within the JTTHF. Additionally, the size of some of the objects used in the JTTHF is considerably larger than the blocks used for the BBT, making it easier to manipulate. Lastly, to ensure the feasibility of delivering HABIT in children with hemispherectomy, several minor modifications were necessary such as the adaptation of tasks to consider participants' functional, visual, and cognitive abilities (1-step commands). For example, in the potential scenario where games were too complex for the participants, only the practice of specific components of the task was conducted. Another adaptation was the simplification (ie, simple forms and smaller scenes) of a drawing task. Lastly, the objects were moved relatively close to the participant and within his/her visual field as needed. The activities were mainly similar to that of the children with congenital hemiparesis, although the severity of hand impairments was often greater in children with hemispherectomy, and thus the way the hand was used during the activities may have differed (eg, using the hand more as a stabilizer or passive assist rather than an active manipulator).

In our study, improvement in bimanual function may be a direct result of HABIT, which uses motivation to promote neuroplasticity,²⁹ task specificity optimizing motor learning,³⁰ active learning,⁵ and diversified functional exercises, allowing individuals to practice specific spatial-temporal coordination skills.⁵ Despite the significant improvement of AHA changes from pre1 to pre2 suggesting that performance of the first test may have stimulated greater awareness of the affected hand, an improvement was still observed following the intensive intervention reflecting the advantages of HABIT. Improvement in children's bimanual functions was reflected by the improvement in COPM and ABILHAND-Kids measures. Furthermore, minimal clinical differences in the COPM occurred in the majority of participants. Improvement of performance and satisfaction in the functional bimanual goals set by the parents could be attributed to the practiced bimanual tasks during HABIT. The enhancement of these skills may have occurred due to better bilateral coordination⁶ and/or problem-solving³⁰ as practiced during the HABIT intervention. The improvement and maintenance of COPM scores following HABIT could therefore have a significant effect on their participation level.

Improvement in motor function after hemispherectomy differs for reasons such as the etiology, underlying pathology, onset of the underlying neurological disorder, and lateralization of the CST prior to the surgery.³¹ More recently, brain circuitries, particularly the sensorimotor pathways, have been a main focus in children with neurological disorders to quantify motor recovery.^{2,11} For example, studies performed in children who have undergone hemispherectomy have showed that contralateral projection before a surgery leads to a total loss of dexterity, which can limit recovery.^{2,32} The integrity of those projections may also have great implications in predicting changes in upper limb function.^{15,32} The prioritization of the type of

intervention may also rely on the lateralization of the CST. Although a recent study with children with unilateral CP suggested that lateralization of the CST may not have an effect on recovery following HABIT,¹¹ other studies have suggested the opposite with CIMT.^{7,33} However, results from these studies may not be directly applicable to children with hemispherectomy. In children with hemispherectomy, ipsilateral tracts often do better possibly because of reduced inhibitory interhemispheric activity. This disinhibition of the interhemispheric connections may also help the recovery of upper limb, particularly in bimanual function through the use of the affected/assisting hand.³⁴

A limitation of the study is the results cannot be generalized to all children with hemispherectomy due to the small sample size and the different severity of the impairments. Furthermore, there was not a control group receiving usual and customary care due to the limited number of children having undergone hemispherectomy. Therefore, we chose a within-subject design. Nonetheless, the results are an important step toward better understanding of how an intensive bimanual training may improve motor function in this population.

CONCLUSION

Children with hemispherectomy were able to complete HABIT as well as improving bimanual function and functional goals. Further studies should take into consideration the etiology and organization of CST to potentially be used as a biomarker to predict upper limb improvements.

What This Adds to the Evidence

This pilot study describes the effectiveness of an intensive bimanual training to improve bimanual function and functional goals in children who have undergone hemispherectomy.

ACKNOWLEDGMENTS

The authors thank the children and the families who participated in this study. We also thank the volunteers in their assistance with data collection.

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