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Letter to the editor

Feasibility and effectiveness of HABIT-ILE in children aged 1 to 4 years with cerebral palsy: A pilot study

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Dear Editor,

Recent scientific literature supports the use of intensive, activity-based, goal-directed interventions, rather than usual care, to encourage significant motor improvements in children with cerebral palsy (CP) [1–3]. Among these interventions, the Hand-arm Bimanual Intensive Therapy Including Lower Extremities (HABIT-ILE) proposes the practice of voluntary movement control, with many movement repetitions and progressive shaping in a child-friendly context. In addition to bimanual coordination, it includes continuous stimulation of lower extremities (LEs) and trunk [4]. Few intensive interventions have targeted young children with CP, mainly focusing on the upper extremity (UE) of children with unilateral CP [5–8]. Therefore, we perceived a need for intensive early interventions targeting the whole spectrum of CP.

This pilot study aimed to investigate the feasibility and effectiveness of HABIT-ILE as an early intensive intervention in pre-school children aged 1 to 4 years with unilateral CP. We hypothesized that HABIT-ILE could be performed in this population, improving their manual abilities as well as gross motor function, including LEs and trunk.

Children were recruited from CP centres of Belgian university hospitals and via spontaneous applications from parents through our lab websites/Facebook page. Children aged 12 to 59 months with a diagnosis of unilateral CP were eligible if they did not have:

- unstable seizures;
- scheduled botulinum toxin injections or surgery 3 months before or during the study period, or;
- visual troubles interfering with the intensive treatment and/or evaluation, or;
- participate in another intensive therapy 3 months before or during the study period.

This pilot study was approved by the université catholique de Louvain Ethic's Review Board in Brussels, and all parents gave their written informed consent. We used a single-group self-controlled design.

Assessments were performed at 4 times: baseline (T0), pre-therapy (T1, 2 weeks after T0), post-therapy (T2, 3 or 4 days after the conclusion of therapy) and follow-up (T3, 3 months later). Between T0 and T1, children followed their conventional therapy: physiotherapy (60–135 min per week), occupational therapy (30–135 min per week) and psychomotor therapy (20–60 min per week).

Feasibility was monitored by using therapy registers on a "time-task" log. Interventionists were instructed to record the time and type of activity performed by each child. The primary outcome was the Assisting Hand Assessment (AHA [9]) for children $>$ 18 months old ($n = 9$) and the Mini-AHA [10] for those $<$ 18 months old ($n = 3$), to measure how the child used the paretic hand collaboratively with the non-paretic hand in bimanual play [11]. The test performance was video-recorded for subsequent blinded scoring by a certified examiner.

Five secondary outcomes were assessed: the Melbourne Assessment 2 (MA2 [12,13]) to evaluate the quality of UE movements and the Gross Motor Function Measure-66 (GMFM-66 [14]) to evaluate gross motor function. Performances were video-recorded for subsequent blinded scoring. The Pediatric Evaluation of Disability Inventory (PEDI [15]) was used to evaluate functional skills in the self-care and mobility domains. The ACTIVLIM-CP [16] questionnaire measured global activity performance in everyday life activities. Parents completed these questionnaires. The Canadian Occupational Performance Measure (COPM [17]) was used to establish and evaluate functional goals (defined by parents) regarding the child's performance and parent's satisfaction.

The intervention was similar to previous HABIT-ILE procedures [18,19], children participating 5 days a week over 2 weeks in a day camp-like therapy. To match the specificity of children $<$ 5 years old, we modified the original HABIT-ILE protocol as required. Daily sessions consisted of 5 hr of HABIT-ILE per day, with 3 hr in the morning, 2.5 h off (nap time) and 2 h later in the afternoon, for a total of 50 h of HABIT-ILE within the 2-week schedule. All activities/games were age-suitable. For children with lower levels of motor abilities, activities were directed toward promoting the gross motor level appropriate for the age. For example, to achieve passing from floor to sitting position, children were first stimulated with toys to sit from a semi-lying supine position supported by a 45° triangle cushion; the angle decreased progressively toward the goal of unassisted sitting. Regarding bimanual manipulation stimulation, children with complete non-use of the paretic UE were offered toys, presenting them to both hands in a bimanual game context. If no bimanual grasping occurred after several trials,

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the toy was placed in the more affected hand to induce participation in the bimanual game.

One-way repeated measure ANOVA (RM-ANOVA) was used for the Mini-AHA/AHA, MA2, GMFM and ACTIVLIM-CP measures, then Tukey's HDS post-hoc analyses. Friedman Repeated Measures Analysis of Variance on Ranks was used for the COPM and PEDI scores. The Student-Neuwman–Keuls method was used for post-hoc analyses. Effects were considered significant at $P < 0.05$. We estimated the effect size for each assessment.

Among the 12 children, 2 dropped out during the study: one had a seizure the day before the first assessment and the second was excluded because the impairment proved to be bilateral on clinical examination at T0. One child's MA2 video was lost because of recording problems during the baseline evaluation. Because of behavioural issues, 2 children did not finish the GMFM-66 at T1. Finally, one child was unable to attend the 3-month follow-up owing to visa constraints.

Ten of the 12 children completed the 10 days of HABIT-ILE, representing 50 h of intervention. Children were engaged for a mean (SD) of 44.82 (3.10) h, representing 90% (range 81% to 99%) of the total time. The activities performed with the UE were classified as gross dexterity (53%), manipulative activities (10%) and functional goals (27%). For the LE, 32% of the intervention was spent sitting on a chair, 28% standing, 10% sitting on a mat, 10% walking/running, 4% cycling, 3% sitting on a ball, 2% lying on a mat, and 1% standing on the balance board. Two activities were performed only by non-ambulatory children: dragging/crawling (8%) and standing while holding (2%).

Children demonstrated significant improvements in bimanual hand function on the Mini-AHA/AHA ($F = 14.61$, $P < 0.001$, effect size [η_P^2] = 0.65). Post-hoc analysis revealed significant differences after therapy (Fig. 1A and Table 1).

They showed significant improvements in unimanual UE function assessed by the MA2 in both the nonparetic UE ($F = 14.79$, $P < 0.001$, $\eta_P^2 = 0.63$) and paretic UE ($F = 48.81$, $P < 0.001$, $\eta_P^2 = 0.66$). Regarding the paretic UE, post-hoc analysis revealed significant differences after therapy as compared with both baseline assessments (Fig. 1C and Table 1). Scores were higher for the non-paretic UE after therapy and at follow-up (Fig. 1D and Table 1). Gross motor function assessment with the GMFM-66 showed significant improvements ($F = 6.01$, $P = 0.003$, $\eta_P^2 = 0.52$). Post-hoc analysis revealed significant differences at follow-up as compared with both pre-intervention assessments (Fig. 1B and Table).

PEDI responses revealed significant improvements in both domains (daily activities: $F = 18.27$, $P < 0.001$, $\eta_P^2 = 0.73$; mobility: $F = 21.25$, $P < 0.001$, $\eta_P^2 = 0.70$). Post-hoc analysis also revealed significant improvements in both domains after therapy (Fig. 2A and B; Table 1). Global activity performance in daily living assessed with the ACTIVLIM-CP questionnaire demonstrated significant improvements ($F = 3.51$, $P = 0.029$, $\eta_P^2 = 0.29$). Post-hoc analysis revealed significant differences at follow-up (Fig. 2C and Table 1). Functional goals determined with the COPM showed high scores for performance ($F = 59.34$, $P < 0.001$, $\eta_P^2 = 0.92$) and parent's satisfaction ($F = 48.56$, $P < 0.001$, $\eta_P^2 = 0.93$). Post-hoc analysis revealed significant differences for performance and parent's satisfaction after therapy as compared with both pre-camp assessments (all $P < 0.001$; Fig. 2D–E, Table 1).

All children participating in this pilot study were able to complete the HABIT-ILE, so the principles on which the therapy is

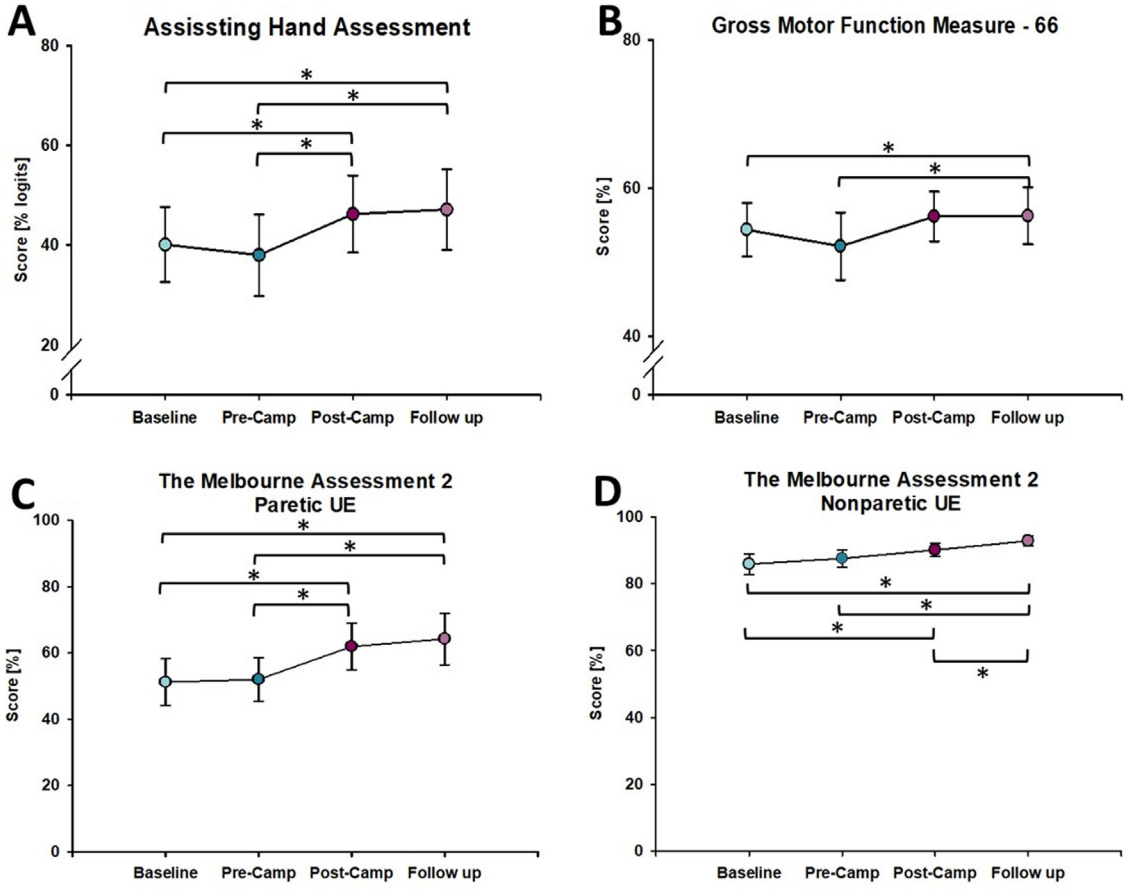

Fig. 1. Improvements in manual abilities and gross motor function during HABIT-ILE. Data are mean (SD). * $P < 0.05$.

Table 1
Results of changes in each assessment.

Evaluation	Mean (SD)/median (Q1-Q3)				RM-ANOVA	
	T0	T1	T2	T3	Main effect	Multiple comparison
Mini-AHA/AHA	40.10 (23.68)	38.00 (25.83)	46.20 (24.50)	47.11 (24.24)	$F = 14.61$; $P < 0.001$	T0 vs T2 $P = 0.012$; T0 vs T3 $P < 0.001$ T1 vs T2 $P < 0.001$; T1 vs T3 $P < 0.001$
MA2 (paretic UE)	51.23 (21.13)	52.02 (20.96)	61.97 (22.43)	64.25 (23.43)	$F = 48.81$; $P < 0.001$	T0 vs T2 $P < 0.001$; T0 vs T3 $P < 0.001$ T1 vs T2 $P < 0.001$; T1 vs T3 $P < 0.001$
MA2 (nonparetic UE)	85.86 (9.31)	87.55 (8.28)	90.08 (6.20)	92.78 (4.62)	$F = 14.79$; $P < 0.001$	T0 vs T2 $P = 0.011$; T0 vs T3 $P < 0.001$ T1 vs T3 $P < 0.001$; T2 vs T3 $P = 0.037$
GMFM-66	53.56 (10.59)	52.19 (12.88)	56.12 (10.51)	56.28 (11.53)	$F = 6.01$; $P < 0.003$	T0 vs T3 $P = 0.006$ T1 vs T3 $P = 0.033$
PEDI (daily activities)	26.00 (13.75–33.00)	26.00 (16.75–32.75)	31.50 (17.50–40.25)	37.00 (23.00–38.00)	$\chi^2 = 21.00$; $P < 0.001$	T0 vs T2 $P < 0.050$; T0 vs T3 $P < 0.050$ T1 vs T2 $P = 0.050$; T1 vs T3 $P < 0.050$ T2 vs T3 $P < 0.050$
PEDI (mobility)	36.00 (9.50–45.00)	37.50 (14.25–45.50)	43.00 (17.25–49.25)	46.00 (16.50–49.50)	$\chi^2 = 21.49$; $P < 0.001$	T0 vs T2 $P < 0.050$; T0 vs T3 $P < 0.050$ T1 vs T2 $P = 0.050$; T1 vs T3 $P < 0.050$ T2 vs T3 $P < 0.050$; T1 vs T0 $P < 0.050$
ACTIVLIM-CP	-0.06 (1.45)	0.05 (1.69)	0.17 (1.66)	0.39 (1.41)	$F = 3.51$; $P = 0.029$	T0 vs T3 $P = 0.024$
COPM (performance)	2.10 (1.60–2.90)	2.20 (1.98–3.18)	6.65 (5.75–7.53)	8.00 (6.30–8.10)	$\chi^2 = 23.23$; $P < 0.001$	T0 vs T2 $P < 0.050$; T0 vs T3 $P < 0.050$ T1 vs T2 $P < 0.050$; T1 vs T3 $P < 0.050$
COPM (satisfaction)	1.60 (1.00–2.75)	2.20 (1.70–3.13)	7.10 (5.90–8.26)	8.20 (6.70–8.90)	$\chi^2 = 22.20$; $P < 0.001$	T0 vs T2 $P < 0.050$; T0 vs T3 $P < 0.050$ T1 vs T2 $P < 0.050$; T1 vs T3 $P < 0.050$

AHA: Assisting Hand Assessment; MA2: Melbourne Assessment 2; UE: upper extremity; GMFM-66: Gross Motor Function Measure-66; PEDI: Pediatric Evaluation of Disability Inventory; COPM: Canadian Occupational Performance Measure; T0: baseline; T1: pre-therapy 2 weeks after T0; T2: post-therapy; T3: follow-up 3 months after intervention; RM-ANOVA: repeated measures analysis of variance.

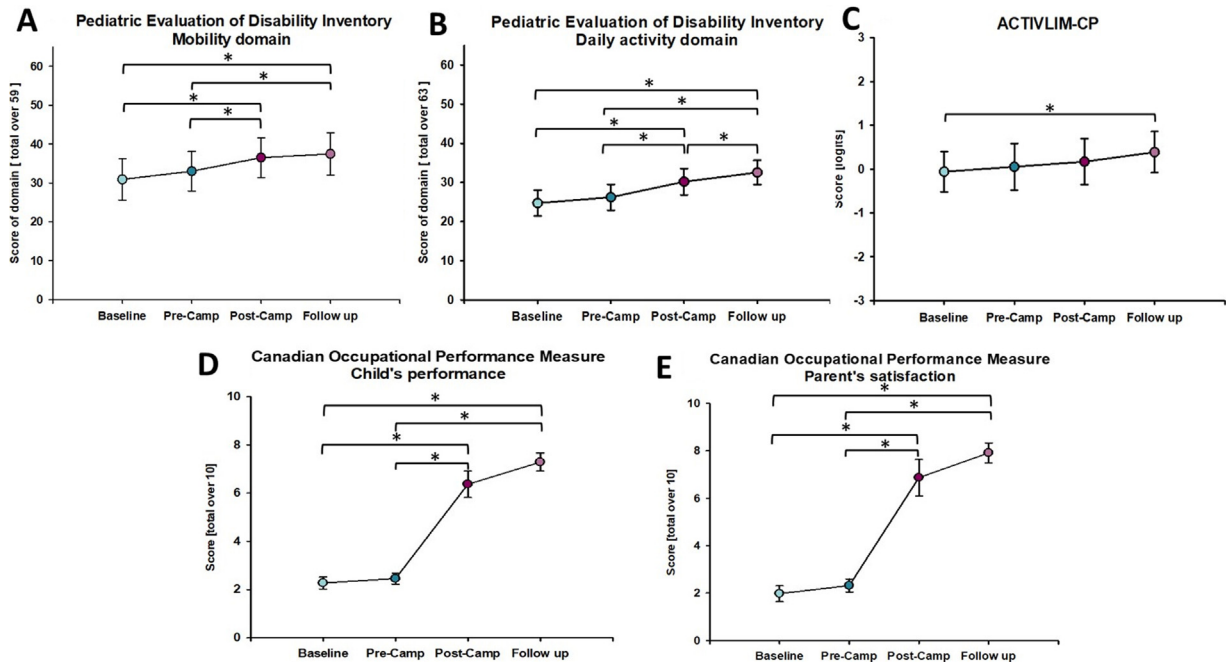

Fig. 2. Improvements in parents' reported questionnaire answers during HABIT-ILE. Data are mean (SD). * $P < 0.05$.

based (i.e., the use of shaping, positive reinforcement, performed without guidance of movement and high intensity with repetitive practice), can be applied in children of preschool age. In addition, the improvement magnitudes we found matched with those observed in school-age children completing the HABIT-ILE [18]. Regarding the paretic UE use in bimanual performance, the improvements we observed matched with previous studies of intensive interventions in young children with unilateral CP [5,7]. In addition, we observed significant differences in unimanual performance of both hands, gross motor function and functional goals trained during the therapy. These results highlight the potential benefits of applying this therapy to young children with unilateral and potentially bilateral impairments in the future.

A key consideration in the implementation of such intensive therapy is the tolerability of the necessary dosage. Different studies of the optimal dosage for intensive therapies in school-age children indicate that at least 50 to 60 h of intervention is needed to produce relevant motor improvements [20,21]. In the present study, we performed the therapy for 5 h/day (3 h in the morning, 2 h in the afternoon), 5 days a week, for 2 weeks, following the typical preschool child schedule.

The results observed in this pilot study support the feasibility and effectiveness of HABIT-ILE as an early intensive intervention for pre-school children with unilateral CP. The outcomes confirmed our hypothesis. An urgent next step will be to perform large randomized trials controlling for some critical factors such as the precise age, lesion type and impairment side.

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Disclosure of interest

The authors declare that they have no competing interest.

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