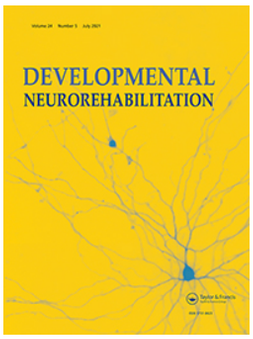

Developmental Neurorehabilitation

ISSN: (Print) (Online) Journal homepage: https://www.tandfonline.com/loi/ipdr20

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To cite this article: LWME Beckers, EAA Rameckers, PBM Aarts, JJW van der Burg, RJEM Smeets, COAD Author Group, MLAP Schnackers, B Steenbergen, IJM de Groot, ACH Geurts & YJM Janssen-Potten (2021) Effect of Home-based Bimanual Training in Children with Unilateral Cerebral Palsy (The COAD-study): A Case Series, Developmental Neurorehabilitation, 24:5, 311-322, DOI: 10.1080/17518423.2021.1886189

To link to this article: https://doi.org/10.1080/17518423.2021.1886189

Published online: 20 Feb 2021.

DEVELOPMENTAL NEUROREHABILITATION
2021, VOL. 24, NO. 5, 311–322
https://doi.org/10.1080/17518423.2021.1886189

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ARTICLE

Effect of Home-based Bimanual Training in Children with Unilateral Cerebral Palsy (The COAD-study): A Case Series

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ABSTRACT

Purpose: To explore the child- and parent-related effects of home-based bimanual training in children with unilateral cerebral palsy.

Methods: Case series of 14 children (2–7 years) who completed goal-oriented task-specific training for 3.5 hours/week for 12 weeks by a program adopting implicit (n = 5) or explicit (n = 9) motor learning. A therapist and remedial educationalist coached parents. Progression on bimanual goals (Canadian Occupational Performance Measure (COPM)) and therapy-related parental stress (interviews) were of primary interest. Data were collected at baseline (T0), halfway through and at the end of training (T1 and T2), and after 12 weeks (T3).

Results: On the COPM performance scale a clinically relevant change was seen in 50% (7/14), 86% (12/14), and 85% (11/13) of the children, at T1, T2, and T3, respectively. Some parents indicated that they had experienced stress because of the training intensity.

Conclusion: The child- and parent-related effects of the home-based bimanual training programs are encouraging.

ARTICLE HISTORY
Received May 18, 2020
Revised November 17, 2020
Accepted February 02, 2021

KEYWORDS
Cerebral palsy; upper extremity; home-based training; bimanual training; therapy-related parental stress

Introduction

Children with unilateral spastic cerebral palsy (uCP) often encounter problems in the development of their arm-hand function and skill performance. The ultimate aim of rehabilitation is to increase independence and participation. To achieve this, effective therapies are required to improve the performance of bimanual activities in everyday life. It is indicated that the practice and actual performance situation need to be comparable, and that the more similar the context, the better transfer of learning occurs. Considering this principle of practice specificity, bimanual training in the children's daily home environment seems to be the best approach to realize performance of acquired skills in the daily living environment.

Previous research has established the effectiveness of bimanual training,[1] and evidence for home-based programs is growing.[2] However, to our knowledge, only one effectiveness study has so far focused on parent-delivered bimanual training at home. In that randomized controlled trial (RCT), children performed home-based activities for 2 hours a day, 5 days a week, for 9 weeks. This included child-friendly games, aiming to improve reaching, grasping, releasing, in-hand manipulation, and using the affected hand as an assisting hand. Participants were remotely monitored through webcam-based software, weekly meetings, and logs. The training was more successful on dexterity (Box and Blocks Test (BBT)) and in making progress on functional goals (Canadian Occupational Performance Measure (COPM)) than was lower extremity home-based training, although there was no effect on bimanual performance (Assisting Hand Assessment (AHA)).[3] The clinical usefulness of the intervention may be limited because of the high intensity of the training.

According to Sakzewski et al., upper limb interventions in children with uCP should be goal-directed, adequately dosed, and based on motor-learning approaches that use activity-based therapy.[2] These guidelines also apply to home-based training. A remaining question to be addressed is the suitability of different motor-learning approaches in the specific context of parent-delivered programs. Explicit motor learning generates consciously accessible information about how to produce appropriate movements.[4,5] In the implicit approach, procedural learning about how a movement is performed occurs as a result of repeated practice, without active accumulation of declarative knowledge.[4,5] In order to evaluate the potential of these contrasting motor-learning principles in home-based training, two programs were developed: a home-based bimanual training program adopting implicit motor-learning principles, hereafter called 'implicit program,' and a home-based

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bimanual training program adopting explicit motor-learning principles, hereafter called 'explicit program.' In both programs parents give their child task-oriented and result-oriented instructions and feedback. In the explicit program parents also provide instructions and feedback on the motor performance of the bimanual activities. In the implicit program, the bimanual and appropriate motor performance of the activity is elicited through manipulation of the organization of the activities.

There are indications that home-based programs can induce parental stress.[6,7] It is imperative to take this into account in the development and evaluation of such interventions. However, it has not been established what strategies are effective in order to prevent or limit therapy-related parental stress and other unintended psychological and social effects. Consequently, coaching by a remedial educationalist was incorporated in the home-based training programs under study, and therapy-related parental stress was monitored throughout.

Despite the clinical need for home-based interventions, there remains a paucity of evidence on home-based bimanual training. Moreover, parental stress is often ignored in the design and evaluation of home-based interventions. In order to fill this gap, the main objective of our programs was to improve the bimanual performance of targeted activities by the child, while preventing an increase of (therapy-related) parental stress. Secondary effects were expected on manual capacity, general bimanual performance, and participation of the child, and on empowerment of the parents. The objective of this case series was to explore the child- and parent-related effects of the home-based bimanual training programs in children with uCP.

Materials and Methods

Participant Information

Figure 1 illustrates the participant flow. Initially, 12 participants were enrolled in an RCT,[8] of whom seven were allocated to either the implicit or explicit training approach, and five to the control group. After early closure of the RCT (Stage 1) due to recruitment problems,[9] 11 additional participants were recruited for the purpose of this case series (Stage 2). These participants received either the implicit or explicit program, according to the parents' preference. Of the total of 18 children who started a home-based training program, 14 completed the training and are reported on in this case series. Five families performed the implicit program (two randomized and three self-chosen) and nine families the explicit program (three randomized and six self-chosen). Reasons for drop-out of participants were related to personal circumstances and/or the training intensity.

Participants were recruited from five rehabilitation centers in the Netherlands between April 2016 and March 2018. Children were eligible if they had a clinically confirmed diagnosis of uCP, were aged 2 through 7 years, and had Manual

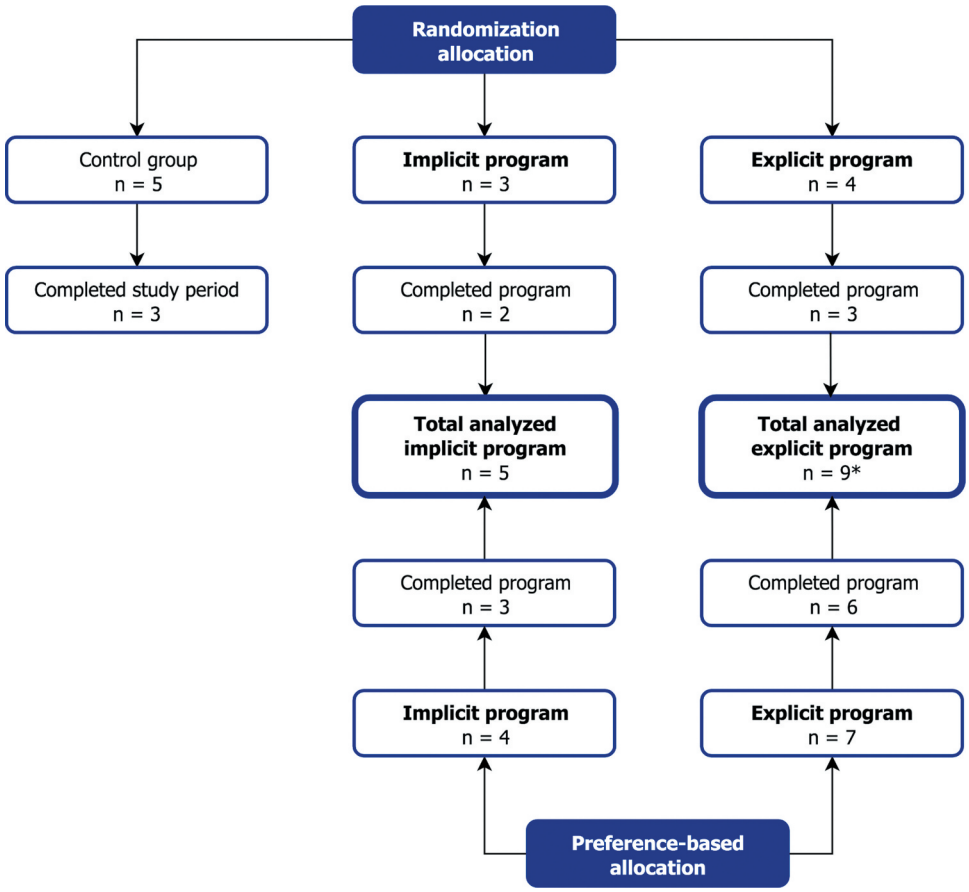

Figure 1. Participant flow.
* One participant that was included in the analyses dropped out at follow-up (T3).

Ability Classification System (MACS), as well as Gross Motor Function Classification System (GMFCS), Levels I to III.[10,11] Potential participants were excluded if 1) the child had a co-morbidity affecting arm-hand function; 2) the child had received surgery or other medical intervention within the nine months prior to or during the study that might affect motor function; 3) the child was to participate in an intensive therapy program focusing on the upper limbs during the study; 4) parents were unable to respond to interviews and questionnaires in Dutch; 5) the rehabilitation team expected parents to be unable to adhere to the programs (e.g. as parents both work full-time and do not have the time to supervise the home-based training); or 6) there were any other medical or psychosocial contraindications.

Ethical approval was granted for both study stages by the medical ethical committee 'Commissie Mensgebonden Onderzoek regio Arnhem – Nijmegen' (Protocol Number NL53670.091.15). Written informed consent was obtained for each participant.

Demographic Information

Table 1 provides characteristics of participants and baseline findings. Eleven boys and three girls participated. Their median age was 4.0 years (IQR 1.5). The majority of children were functioning at MACS Level II, and GMFCS Level I. Supplemental File 1 presents the children's motor impairments (range of motion and strength).

All children had problems in performance of bimanual activities. The rehabilitation needs defined by parents at the start of the program included five categories of activities: (un) dressing (26 needs), eating and drinking (18 needs), crafts (10 needs), play (7 needs), and gross motor (outdoor) activities (9 needs). The most common needs were related to putting on, taking off, and zipping a jacket (9 children), pulling lower garments down and up (6 children), using cutlery while eating (6 children), and holding paper while cutting, drawing, coloring, or erasing (9 children).

Therapeutic Interventions

The implicit and explicit programs both incorporated goal-oriented and task-specific training principles. For each child, five bimanual needs were identified collaboratively with parents, providing the input for the intervention goals. Therapists analyzed the children's performance for each identified activity and prepared individual training programs. Exclusively, in the explicit program, a movement analysis by the therapist informed the content of the training. Parents were provided with instructional videos and a manual before the start of the training. Intended training intensity was 3.5 hours per week for 12 weeks. Parents in the implicit program elicited the desired bimanual performance by organization of the training activities, complemented with task-oriented instructions and feedback; parents in the explicit program gave instructions and feedback focused on motor performance, in addition to task-oriented instructions and feedback. A therapist coached parents in applying the training, and a remedial educationalist supported parents by concentrating on parent–child interaction and child behavior (i.e. interdisciplinary coaching). The therapist had a face-to-face meeting with parents at the start of the program and had weekly contact with them throughout the training, primarily by telephone and (three times) through home visits. The remedial educationalist met parents before the start of the training and phoned them two weeks afterward. Support during the remaining weeks of the program was available on request of either the parents themselves or the therapist, if training was impeded by parent–child interaction or child behavior. The coaching was provided by professionals from the child's rehabilitation center. All professionals had attended an instructional course and received a manual. A detailed protocol of the programs is published elsewhere.[12]

After finishing the home-based training, children received usual care for 12 weeks before the follow-up. This entailed care according to an individual approach determined by the treating physician. Except for the treatment options mentioned in the exclusion criteria, no restrictions in treatment options were used.

Outcome Measures

In selecting the outcome measures, four aspects were taken into account: 1) the robustness of psychometric properties (e.g. sensitivity to change); 2) the ability to compare the results with existing and future studies; 3) the intention to capture potential changes across the activity and participation domains of the International Classification of Functioning, Disability, and Health (ICF); and 4) the importance of assessing constructs of the motor functioning of the child and of parental stress that are of specific interest in the implementation of home-based bimanual training.

Parent-rated Outcomes at Child Level

Canadian Occupational Performance Measure (COPM): Parents identified and ranked five needs involving bimanual activities of self-care, play and/or leisure, using the child-adapted version of the COPM. For each activity, parents rated the performance and their satisfaction with this performance, generating an averaged performance and satisfaction score. As the COPM performance scale corresponds to the goal-oriented and family-centered approach of the interventions, this was the primary outcome. The COPM has good psychometric properties, which also hold in the child-adapted version.[13]

Hand-Use-at-Home questionnaire (HUH): The amount of spontaneous use of the child's affected hand at home was assessed using the HUH, which consists of 18 age-related play and self-care activities for which bimanual execution is required or preferred. Parents rated the frequency of spontaneous use of the affected hand in these activities on a 5-point scale. Test–retest reliability and construct validity of this Dutch questionnaire in children with CP are very good and good, respectively.[14,15]

Lifestyle Assessment Questionnaire for CP (LAQ-CP): The child's participation was assessed through the LAQ-CP, a parent-reported questionnaire. The LAQ-CP consists of 46 items and examines the impact of the disabilities on six dimensions. Weighted aggregation of the domain scores is used to

Table 1. Participant characteristics at study enrollment.

Case	Home program	Allocation	Age (years)	Gender	Affected side	MACS level	GMFCS level	House Functional Classification type	BRIEF (-P)[a] total score	Education	PS[b]	NEO-FFI-3[c]
1	Implicit	Random	3	M	L	N.A.[d]	I	4	#	Mainstream nursery school/day-care	3	#; 38; 34; 35; 37
2	Implicit	Preferred	5	F	R	II	I	6	75	Mainstream primary school	#	29; 46; 39; 37; 37
3	Implicit	Preferred	3	M	R	N.A.[d]	II	4	#	Mainstream nursery school/day-care	#	28; 40; 36; 31; #
4	Implicit	Random	4	M	R	II	I	5	115	Mainstream primary school	3	27; 36; 35; 34; 40
5	Implicit	Preferred	4	M	R	I	I	3	#	#	#	29; 43; 36; 36; 42
6	Explicit	Preferred	6	M	R	II	I	3	126	Mainstream primary school	#	30; 41; 38; 38; 42
7	Explicit	Random	4	M	L	II	I	5	104	Mainstream primary school	3	#; #; #; #; #
8	Explicit	Preferred	2	M	R	N.A.[d]	I	7	#	N.A.	#	34; 39; 35; 31; 38
9	Explicit	Random	4	M	R	II	I	5	#	School for physically disabled children	3	25; 43; 35; 27; 35
10	Explicit	Preferred	3	F	R	N.A.[d]	I	7	#	Mainstream nursery school/day-care	#	33; 38; 41; 36; 41
11	Explicit	Random	2	M	R	N.A.[d]	I	7	#	Therapeutic toddler group	2	31; 37; 39; 34; 39
12	Explicit	Preferred	2	M	R	N.A.[d]	I	5	#	Mainstream nursery school/day-care	#	36; 36; 34; 32; 39
13	Explicit	Preferred	7	F	L	II	I	5	110	School for physically disabled children	#	29; #; 36; 31; 41
14	Explicit	Preferred	4	M	R	III	I	3	83	Mainstream primary school	#	26; 44; 30; 31; 39

[a]Behavior Rating Inventory of Executive Function (BRIEF) for children aged 5 through 7 years, and BRIEF Preschool Version (BRIEF-P) for children aged up to 5 years, to assess executive functioning of the child
[b]Parenting Scale (PS), to assess parenting style
[c]Neuroticism-Extraversion-Openness Five-Factor Inventory (NEO-FFI-3), to assess the personality of the parent (neuroticism; extraversion; openness; agreeableness; conscientiousness)
[d]N.A: not applicable; MACS is not validated for children <4 years of age. For younger children, the rehabilitation team judged that manual ability was comparable to MACS Levels I–III
M: male; F: female; L: left; R: right
Missing value

calculate a total Lifestyle Assessment Score (LAS).[16] The translated Dutch version has not yet been validated.[17]

Observer-rated Outcomes at Child Level

Goal Attainment Scaling (GAS): The highest prioritized rehabilitation goal, identified with the COPM, was scaled using GAS. The progress toward the rehabilitation goal was videotaped and scored in relation to predetermined criteria as formulated in the GAS scale. As knowledge about the capacities of the child was needed, the personal GAS scale was formulated by the coaching therapist. GAS is responsive to measurements of individual change over time in children with CP and has good inter-rater reliability and acceptable validity.[18,19]

Assisting Hand Assessment (AHA): Bimanual performance of the child was assessed using the AHA version 5.0 (either Small Kids or School Kids version). The AHA measures the typical application of a child's assisting hand in a range of standardized bimanual activities. The AHA has been reported to have good validity, reliability and responsiveness to change in children with uCP.[20,21]

Observational Skills Assessment Score (OSAS): The OSAS was used to measure the quality of use of the affected hand in age-appropriate, standardized bimanual motor tasks in which the use of both hands is required. The OSAS tasks involve many repetitions of actions to reliably assess quality of use. Quality of use is scored every second in order to make the OSAS sensitive to subtle differences. There is evidence that OSAS has satisfactory reliability.[22]

Parent-related Outcomes

Parental interviews: We explored parental stress experienced throughout the programs with a qualitative approach. Parents participated in interviews during and after the intervention as part of a mixed-methods process evaluation that was embedded in the effect study. The specific procedures have been reported elsewhere.[23]

Therapy-related Parental Stress Questionnaire (in Dutch: Vragenlijst Ouderlijke Stress als gevolg van Therapie van het kind (VOST)): The project team has developed the Dutch VOST to measure therapy-related parental stress and the factors that contribute to this stress. The questionnaire consists of three parts. In Part I, parents rate their perceived overall parental stress due to therapy of their child on a Visual Analogue Scale (VAS) and describe in their own words what caused the stress. In Part II, 34 potential stress-inducing factors are presented, covering six domains. For each domain, parents select those factors which have caused them stress and have the opportunity to define additional factors. Moreover, parents rate the amount of stress for each domain separately on a VAS. In Part III of the questionnaire, parents identify the

three factors selected in Part II which led to most stress. The psychometric properties of VASs are considered to be satisfactory,[24] while those of the VOST are still under investigation.

Parenting Stress Questionnaire (in Dutch: Opvoedingsbelastingvragenlijst (OBVL)): The OBVL was used to assess generic stress related to parenting. This Dutch questionnaire consists of five scales (total of 34 items). The reliability and validity of the OBVL are good.[25]

Empowerment questionnaire (EMPO): The EMPO Version 3.1 was used to assess empowerment of parents. This is a Dutch 27-item questionnaire divided into three subscales. Reliability and validity of the EMPO have been confirmed to be sufficient to good.[26]

Data Collection

Data were collected at four time points: at baseline (T0), halfway through the training period (T1, after 6 weeks of training), at the end of the program (T2, after 12 weeks of training), and at follow-up (T3, 12 weeks after the end of the program). The COPM and therapy-related parental stress evaluated by parental interviews were of primary interest. To limit the burden on families during the training period, secondary outcome measures were not assessed at T1.

The parent most involved in the training rated the parent-reported outcome measures. COPM was thrice assessed face-to-face and once by telephone (T1). Parents did not have access to previous COPM-scores. All questionnaires were completed online using Castor Electronic Data Capture.

Assessments were completed by one of two research assistants who were trained and experienced in the use of the outcome measures and were blinded as to intervention. The GAS (i.e. performance of the activity set by the rehabilitation goal), AHA, and OSAS were video-recorded and scored on subsequent viewing by trained assessors blinded as to treatment and time point. The AHA evaluator was qualified to administer this measure.

Results

Parent-rated Outcomes at Child Level

Canadian Occupational Performance Measure (COPM)

On the COPM performance scale, 86% (12/14) of the children exceeded the clinically relevant change of two points after the program. The scores between T0 and T1 already showed an improvement of at least two points in half of the children (7/14). At follow-up, 85% (11/13) of the children showed a clinically relevant increase compared to baseline. The COPM satisfaction scores improved by clinically relevant amounts during the first half of the program in 43% (6/14) and during the total program in 64% (9/14) of the children. In 88% (7/8) of the improved cases, the T3 score was still >2 points higher than T0 (Table 2).

As can be seen from Figure 2, clinically relevant change scores on the COPM performance and satisfaction scales were achieved with both the implicit and explicit programs.

Hand-Use-at-Home Questionnaire (HUH)

For individual children, the minimal detectable change of the HUH is 1.66 logits.[15] For none of the children was an effect of this magnitude found (Table 2).

Lifestyle Assessment Questionnaire for CP (LAQ-CP)

No values for statistically significant or clinically relevant change scores for LAQ-CP have been reported in the literature, so we chose an arbitrary difference of 20 points to be regarded as relevant. During the program, one child (8%) improved and one child (8%) deteriorated on the physical independence domain. The physical independence of three other children (27%) was better at follow-up than at baseline. An unintended change was an increase of economic burden in two families (15%) at T2 and in three other families at T3 (27%). The other domain scores and the LAS indicated no changes in participation (Table 2).

Observer-rated Outcomes at Child Level

Goal Attainment Scaling (GAS)

At T2, two children (17%) scored −3 (deterioration), four children (33%) scored −2 (equal to starting level), three children (25%) scored 0 (expected goal), one child (8%) scored 1 (somewhat more than expected), and two children (17%) scored 2 (much more than expected). Thus, after the program, 50% of the children (6/12) reached the expected goal or more. At T3, two children (18%) scored −2, four children (36%) scored −1 (less than expected), two children (18%) scored 0, and three children (27%) scored 1, meaning that 45% of the children (5/11) reached or maintained the expected goal or more at follow-up.

Assisting Hand Assessment (AHA)

In two children, the AHA score showed a clinically relevant (at least 5 units) increase at T2 (14%), as well as T3 (15%), compared to baseline. In two other children (14%), this change was only found at T2 and, in another two children (15%), this change only occurred at T3. The scores of one child (7%) showed a clinically relevant decrease at both T2 and T3 (Table 3).

Observational Skills Assessment Score (OSAS)

When compared to the smallest detectable difference as reported by Speth et al.,[22] the individual data showed no significant effects (Table 3).

Parent-related Outcomes

Parental Interviews

During the interviews, some parents indicated that they felt no stress at all, but for others, the home-based training led to varying degrees of stress. Generally, parents mentioned feelings like frustration, irritation, and disquiet rather than stress, which in their opinion was too strong a word. The most stress-inducing factor was the prescribed intensity of the training, i.e. to find a time for the training as well as the need to meet the required training hours. As one parent explained:

Table 2. Parent-rated outcomes at child level.

					LAQ-CP						
					Dimensional scores						
Case	Time point	COPM performance (0–10)	COPM satisfaction (0–10)	HUH logits (−4.69 – +5.17)	Physical independence (0–100)	Mobility (0–100)	Clinical burden (0–100)	Schooling (0–100)	Economic burden (0–100)	Social integration (0–100)	Overall score (0–100)
1[a]	**T0**	4.2	4.4	−1.022	64.59	32.14	25.00	.00	29.17	18.75	43.20
	T1	6.8*	6.8*								
	T2	6.8*	7.0*	−1.431	#	#	#	#	#	#	#
	T3	7.4*	6.6*	−.838	45.83	14.29	22.73	.00	25.00	15.63	30.57
2[a]	**T0**	5.8	5.8	.376	16.67	14.29	20.46	.00	.00	18.75	18.73
	T1	7.0	7.4								
	T2	6.8	7.0	.238	12.50	17.86	11.36	.00	.00	12.50	16.51
	T3	7.0	7.2	1.066	16.67	14.29	6.82	.00	.00	18.75	18.28
3[a]	**T0**	5.2	5.6	−1.218	62.50	28.57	20.46	.00	.00	12.50	37.55
	T1	7.0	7.0								
	T2	7.8*	8.4*	−.665	43.75	32.14	18.18	.00	.00	12.50	31.75
	T3	7.8*	8.4*	−.500	41.67	25.00	20.46	.00	.00	12.50	29.15
4[a]	**T0**	2.8	4.6	.786	#	#	#	#	#	#	#
	T1	6.2*	7.2*								
	T2	8.0*	9.0*	1.684	18.75	28.57	31.82	.00	45.83	28.13	29.57
	T3	7.8*	7.8*	1.684	#	#	#	#	#	#	#
5[a]	**T0**	5.2	5.6	−.343	37.50	25.00	27.27	.00	16.67	21.88	31.35
	T1	6.2	7.0								
	T2	7.8*	8.0*	.238	25.00	25.00	34.09	.00	25.00	15.63	26.40
	T3	7.0	8.0*	−.044	12.50	21.43	27.27	.00	16.67	12.50	19.37
6[b]	**T0**	5.6	6.2	.098	18.75	50.00	22.73	.00	16.67	15.63	29.86
	T1	7.6*	8.2*								
	T2	7.4	6.8	1.066	16.67	53.57	29.55	.00	25.00	28.13	33.79
	T3	7.7*	7.8	.925	16.67	46.43	27.27	.00	41.67	34.38	34.55
7[b]	**T0**	5.0	10	−1.022	14.58	17.86	27.27	.00	8.33	25.00	21.26
	T1	6.0	10								
	T2	7.2*	10	.376	20.83	17.86	25.00	.00	8.33	6.25	19.22
	T3	7.4*	10	.238	18.75	10.71	20.46	.00	8.33	15.63	18.50
8[b]	**T0**	6.0	6.8	−1.927	45.83	25.00	27.27	.00	4.17	40.63	37.50
	T1	4.2	4.4[†]								
	T2	8.0*	8.2	−.665	56.25	32.14	27.27	.00	45.83	50.00	48.67
	T3	8.4*	8.6	−.343	41.67	21.43	31.82	.00	12.50	46.88	37.29
9[b]	**T0**	2.2	3.2	−2.597	39.58	32.14	31.82	50.00	8.33	43.75	39.19
	T1	6.0*	6.4*								
	T2	6.8*	7.4*	−1.431	39.58	32.14	31.82	37.50	25.00	68.75	45.96
	T3	6.6*	6.6*	−1.665	31.25	25.00	31.82	37.50	33.33	62.50	40.34
10[b]	**T0**	4.8	8.0	.098	62.50	35.71	18.18	.00	8.33	25.00	42.89
	T1	6.4	6.4								
	T2	8.0*	9.0	.376	52.09	25.00	25.00	.00	4.17	21.88	35.46
	T3	7.8*	8.4	1.066	39.58	28.57	13.64	.00	8.33	15.63	30.53
11[b]	**T0**	3.0	3.0	.925	50.00	17.86	13.64	12.50	.00	12.50	30.17
	T1	7.6*	8.0*								
	T2	8.8*	8.8*	1.211	20.83	10.71	13.64	12.50	.00	12.50	17.83
	T3	8.0*	8.0*	2.265	33.33	35.71	15.91	12.50	20.83	25.00	33.63
12[b]	**T0**	4.2	5.0	−1.927	56.25	32.14	18.18	.00	25.00	25.00	41.06
	T1	6.4*	6.8								
	T2	7.4*	7.6*	−.500	47.92	28.57	18.18	.00	16.67	25.00	36.44
	T3	7.0*	7.2*	−1.431	47.92	28.57	20.46	.00	16.67	18.75	35.11
13[b]	**T0**	4.4	5.0	2.053	12.50	21.43	34.09	62.50	16.67	37.50	26.19
	T1	5.6	5.6								
	T2	6.6*	7.0*	1.211	10.42	21.43	20.46	50.00	25.00	28.13	23.38
	T3	6.6*	6.8	3.134	6.25	7.14	20.46	62.50	8.33	40.63	19.67
14[b]	**T0**	4.2	3.6	−1.218	12.50	21.43	27.27	.00	8.33	21.88	20.78
	T1	6.6*	6.0*								
	T2	6.4*	6.6*	−1.022	25.00	21.43	38.64	.00	29.17	21.88	27.33
	T3	#	#	#	#	#	#	#	#	#	#

* Clinically relevant improvement compared to baseline
[†]Clinically relevant decline compared to baseline
[a]Implicit program
[b]Explicit program
Missing value
For COPM and HUH, higher score indicates better performance; for LAQ-CP, higher score indicates lower participation

"... I think that if we, for example, could determine ourselves how much I practice, that that would give me less stress." [parent]

Parents experienced most stress at the beginning of the program, when they were still figuring out how to implement the training.

Figure 2. COPM change scores.

Therapy-related Parental Stress Questionnaire (VOST)

In their answers to the open-ended question at baseline, three parents expressed feelings of stress during preparation for the home-based training program. This indicates that the reported stress related to the programs was already affecting the baseline VOST score. Five parents did not consider the parent-delivered training as therapy and therefore did not fill out the questionnaire at T2. In addition, there were many missing data, particularly for the VASs for the six domains. Consequently, only six cases had sufficient data to evaluate therapy-related parental stress throughout the study by means of this questionnaire. Five of these six cases (83%) showed a decline of overall parental stress during the program. The results of the stress levels for the separate domains were inconsistent, although the data revealed a slight decreasing tendency on the child, parent, and family domains, and a minor increase of stress on the domain 'characteristics of the therapy', at T2 and T3 (Table 4). At baseline, parents indicated 'incorporating the therapy into the planning and/or organization of my family' and 'my personal condition' most frequently as the factors most contributing to their stress (both 83%, 5/6), while at the end of the home-based training most frequently given was 'the time needed to exercise at home' (83%, 5/6).

Parenting Stress Questionnaire (OBVL)

At baseline, nine parents (64%) had a score outside of the normal range regarding one or more subscores (T-score ≥60) and/or the total score (T-score ≥65). The change scores of the OBVL were analyzed in light of the smallest detectable difference given in the questionnaire manual.[25] Change scores from baseline to the end of the program (T2) showed a reduction of parental stress on one or more scales in eight cases (57%), an increase in three parents (21%), and no change in three cases (21%) (Table 4).

Empowerment Questionnaire (EMPO)

At baseline, three parents (21%) had a score outside of the 'non-concerned range' for the total score (T-score >40) and one or more subscores (T-score >35). Based on the reliable change index,[27] the scores on the intrapersonal subscale significantly increased between T0 and T2 in nine parents (69%) and declined in one (8%). At T3, the scores of five parents (42%) were improved and of four parents (33%) worsened. For the interactional subscale, an approximately equal number of parents improved and worsened at T2 (5/13 (38%) and 7/13 (54%), respectively), as well as at T3 (3/12 (25%) and 4/12 (33%), respectively). The same applies to the behavioral control subscale at T2 (four (31%) improved

Table 3. Observer-rated outcomes at child level.

				OSAS[a]					
				Quality of use affected hand					
Case	Time point	GAS (−3 – +2)	AHA-units (0–100)	Reach (0/1)	Grasp fingers (1–3)	Grasp wrist (1–5)	Hold fingers (1–4)	Hold wrist (1–5)	Release (1–4)
1[b]	**T0**	−2	50	0	2	1	2	1	2.5
				0	2	1	1	1	1
	T2	−2	50	0	2	1	2	1	3*
				0	2	1	2*	1	2.0*
	T3	−2	49	1	2	1	2	1	2.0†
				0	1†	1	1	1	1
2[b]	**T0**	−2	55	0	2	1	2	1	3.0
				0	2	1	2	1	3
	T2	−2	57	0	2	1	2	1	3
				0	2	1	2	1	2.0†
	T3	1	57	0	2	1	2	1	3.0
				0	2	3*	2	3*	3
3[b]	**T0**	−2	48	0	2	1	2	1	2.0
				0	2	1	2	1	2
	T2	#	62*	0	2	1	2	1	3*
				0	2	1	2	1	2.0
	T3	#	52	0	2	1	2	1	2.0
				0	2	1	2	1	2
4[b]	**T0**	−2	63	0	2	3	2	1	2.5
				0	2	1	3	1	2
	T2	−2	59	0	2	1†	2	1	3*
				1	2	1	3	1	3.0*
	T3	1	61	0	1†	2†	3*	2*	1.0†
				1	2	1	2†	1	3*
5[b]	**T0**	−2	52	0	2	3	2	3	3.0
				0	2	3	3	3	2
	T2	0	63*	0	2	3	2	3	3
				0	2	1†	3	3	2.0
	T3	0	58*	0	2	3	2	3	2.0†
				0	2	1†	2†	1†	2
6[c]	**T0**	−2	50	0	2	1	2	1	2.0
				0	2	1	2	1	2
	T2	0	49	0	2	1	2	1	2
				0	2	1	2	1	2.0
	T3	0	58*	0	2	1	2	1	2.0
				0	2	1	2	1	2
7[c]	**T0**	−2	63	1	2	1	2	1	2.0
				0	2	1	2	1	1
	T2	2	64	0	2	1	2	1	3*
				0	2	1	2	1	2.5*
	T3	−1	65	0	2	1	2	1	3.0*
				0	2	1	2	1	3*
8[c]	**T0**	−2	71	0	2	1	2	1	3.0
				0	2	1	3	1	2
	T2	0	73	0	2	1	2	1	3
				0	2	3*	3	1	3.0*
	T3	−2	79*	0	2	1	2	1	3.0
				0	2	1	3	1	3*
9[c]	**T0**	−2	41	0	2	1	2	1	2.0
				0	2	3	1	3	2
	T2	−2	46*	1	2	1	2	1	2
				0	2	1†	2*	3	2.0
	T3	−1	46*	0	2	1	2	1	2.5*
				0	2	3	2*	1†	2
10[c]	**T0**	−2	73	0	2	3	2	1	3.0
				0	2	1	3	1	2
	T2	−3	67†	0	2	1†	2	1	3
				0	2	1	3	1	3.0*
	T3	−1	63†	0	2	1†	2	1	3.0
				0	2	3*	3	1	3*
11[c]	**T0**	−2	66	1	2	1	2	3	3.0
				0	2	3	3	1	3
	T2	#	71*	0	#	#	#	#	#
				0	#	#	#	#	#
	T3	#	69	0	#	#	#	#	#
				0	#	#	#	#	#

(*Continued*)

Table 3. (Continued).

				OSAS[a]					
				Quality of use affected hand					
Case	Time point	GAS (−3 – +2)	AHA-units (0–100)	Reach (0/1)	Grasp fingers (1–3)	Grasp wrist (1–5)	Hold fingers (1–4)	Hold wrist (1–5)	Release (1–4)
12[c]	**T0**	−2	50	0	2	1	2	3	3.0
				0	2	3	3	3	2
	T2	1	47	0	2	4*	2	3	3
				0	2	3	3	3	3.0*
	T3	1	48	0	2	1	2	3	2.0†
				0	2	2†	3	3	2
13[c]	**T0**	−2	64	1	2	1	2	1	3
				0	2	1	3	1	3
				0	2	1	2	1	3
	T2	2	68	0	2	1	2	1	3
				1	2	1	3	1	3
				0	2	1	2	1	3
	T3	−1	63	0	2	1	2	1	3
				0	2	1	3	1	3
				0	2	1	2	1	3
14[c]	**T0**	−2	49	0	2	1	2	1	2.0
				0	2	1	2	1	2.0
	T2	−3	46	0	2	1	2	1	2
				0	2	1	1†	1	1.0†
	T3	#	#	0	#	#	#	#	#
				0	#	#	#	#	#

* Clinically relevant/statistically significant improvement compared to baseline
† Clinically relevant/statistically significant decline compared to baseline
[a] OSAS tasks are (from top to bottom) threading beads, and building with 'Pop-Onz' (Fisher Price®) for children aged 2.5 to 6 years, and buttering and cutting bread, small screw and nut construction, and large screw and nut construction for children from 7 years of age
[b] Implicit program
[c] Explicit program
Missing value
For GAS, AHA, and OSAS, higher score indicates higher ability

and five (38%) worsened), while at follow-up the score of eight parents (67%) had increased and of only two (17%) decreased. The results of the total score indicate that the empowerment of four parents (31%) had increased at T2 and of one (8%) decreased. At T3, five parents (42%) had a higher empowerment than baseline, and two (17%) a lower one (Table 4).

Intervention Adherence and Tolerability

Intervention adherence and tolerability, among others, were assessed in a process evaluation, to be published separately. The median of the average hours each child trained weekly is 2.8 (IQR 1.1). No adverse events were reported, except for one hospital admission due to an epileptic seizure, considered unrelated to the home-based training.

Discussion

This case series study aimed to investigate the effect of implicit and explicit home-based bimanual training programs on children with uCP and on their parents. The results suggest that improvements in performance of targeted bimanual activities can be achieved with both approaches when delivered by parents, and suggest that parental stress may decrease during these programs.

The most important clinically relevant finding was an improvement of the performance of, and satisfaction with, targeted activities. A retention of these effects 12 weeks after the end of the program was observed with the COPM. These findings are consistent with Ferre et al.,[3] who found a similar effect of home-based intensive bimanual training with the COPM, although their total training dose was twice as high as in our study. Although a few children in our case series improved on several other outcome measures, both the parent-rated and observer-rated outcomes did not provide apparent evidence for transfer of treatment effect to general manual capacity and/or performance of non-practiced bimanual tasks. The results also suggest that the participation of the children did not increase. Parents experienced some stress because of the training intensity. However, earlier literature findings of stable or increased stress levels were not confirmed.[6,28] Rather, the parent-related outcomes showed a tendency toward reduction of both therapy-related and generic parental stress during home-based training. This may be attributed to the novel role of the remedial educationalist in the programs, who coached the parents in addressing difficult behavior and problems in emotion regulation, motivation, and (sustained) attention of the child. The increased empowerment of some parents, reflected in their higher scores on the intrapersonal subscale of the EMPO, implies that their feelings of competence and self-efficacy grew.[26]

Study Limitations

The original research questions, particularly concerning the comparison of implicit and explicit motor learning, could not be answered as intended, due to recruitment problems during

Table 4. Parent-related outcomes.

		VOST							OBVL[a] (t-scores)						EMPO[b] (t-scores)			
Case	**Time point**	**Overall (0–100)**	**Child (0–100)**	**Parent (0–100)**	**Family (0–100)**	**Environment (0–100)**	**Therapy (0–100)**	**Therapist (0–100)**	**Pcr (45–80)**	**Par (35–80)**	**Dep (46–80)**	**Rol (42–80)**	**Phy (46–80)**	**Total (30–80)**	**IP (20–80)**	**IA (20–80)**	**BC (20–80)**	**Total (20–80)**
1[c]	**T0**	*21*	*35*	*11*	*#*	*5*	*5*	*#*	57	63	54	46	48	52	57	34	33	36
	T2	*2*	*20*	*20*	*20*	*30*	*20*	*10*	57	63	56	50	57†	55	#	#	#	#
	T3	*10*	*17*	*17*	*16*	*16*	*14*	*#*	53	63	60†	50	57†	55	50†	39*	43*	40*
2[c]	**T0**	NT	NT	NT	NT	NT	NT	NT	45	35	46	42	46	30	80	80	66	78
	T2	NT	#	#	#	#	#	#	45	35	46	42	46	30	80	80	48†	80
	T3	#	#	#	#	#	#	#	45	35	46	42	46	30	80	80	75*	80
3[c]	**T0**	0	#	#	#	#	#	#	57	57	48	46	52	48	45	45	48	43
	T2	NT	#	#	#	#	#	#	46*	45*	48	46	48	41	57*	52*	43†	48*
	T3	#	#	#	#	#	#	#	46*	53	48	46	43	43	37†	45	56*	40
4[c]	**T0**	NT	NT	NT	NT	NT	NT	NT	61	66	66	60	73	70	33	45	43	34
	T2	NT	#	#	#	#	#	#	61	56*	66	49*	67	63	40*	52*	38†	38
	T3	19	0	#	29	34	36	#	51*	52*	66	52*	56	55	45*	45	48*	43*
5[c]	**T0**	0	#	#	#	#	#	#	51	50	50	60	61	53	45	72	48	57
	T2	4	#	#	#	#	#	#	45*	35*	46	56	46*	39	50*	62†	56*	59
	T3	#	#	#	#	#	#	#	45*	41*	46	52*	46*	39	#	#	#	#
6[d]	**T0**	*13*	*13*	*16*	*#*	*16*	*#*	*#*	56	47	60	73	62	62	40	61	38	43
	T2	*91*	*0*	*0*	*1*	*0*	*2*	*1*	56	50	53*	68	59	58	45*	52†	48*	45
	T3	*0*	*0*	*0*	*0*	*0*	*7*	*0*	56	50	50*	75	61	61	40	58†	48*	45
7[d]	**T0**	NT	NT	NT	NT	NT	NT	NT	51	50	60	49	59	53	45	52	33	38
	T2	NT	#	#	#	#	#	#	45*	35*	46*	42	46*	30	73*	62*	66*	71*
	T3	#	#	#	#	#	#	#	45*	50	56	42	71†	54	62*	61*	56*	61*
8[d]	**T0**	*60*	*56*	*16*	*44*	*#*	*12*	*#*	53	67	52	50	60	55	50	58	38	45
	T2	*13*	*19*	*18*	*22*	*12*	*#*	*#*	53	63	48	60*	62	56	57*	52†	33†	43
	T3	*28*	*46*	*10*	*16*	*#*	*#*	*#*	53	72	56	56	60	61	50	52†	33†	40†
9[d]	**T0**	0	30	#	#	#	#	#	45	35	60	66	56	51	57	72	48	61
	T2	#	30	30	#	#	#	#	56†	47†	60	70	61	61	45†	62†	62*	59
	T3	#	40	#	#	#	#	#	67†	52†	56	79†	61	68	45†	62†	43†	51†
10[d]	**T0**	*30*	*0*	*45*	*44*	*#*	*50*	*#*	60	50	56	53	48	52	57	80	62	67
	T2	*10*	*10*	*31*	*33*	*#*	*30*	*#*	53*	48	52	50	48	45	57	61†	56†	59†
	T3	*9*	*10*	*9*	*0*	*0*	*0*	*0*	53*	45	54	50	43	44	66*	72†	66*	71
11[d]	**T0**	*0*	*0*	*0*	*0*	*0*	*0*	*0*	46	41	52	46	62	45	66	52	48	57
	T2	*0*	*0*	*0*	*4*	*#*	*3*	*0*	46	41	48	46	52	41	80*	80*	48	71*
	T3	*#*	*#*	*#*	*#*	*#*	*#*	*#*	46	41	48	46	48	39	73*	62*	48	64*
12[d]	**T0**	*43*	*17*	*31*	*#*	*0*	*0*	*#*	57	45	52	46	52	45	50	52	48	48
	T2	*30*	*20*	*26*	*24*	*0*	*20*	*#*	53	50	48	53	57	48	50	45†	48	45
	T3	*12*	*#*	*#*	*20*	*#*	*20*	*#*	46*	57†	48	50	48	46	45†	52	48	45
13[d]	**T0**	0	0	0	#	0	0	0	56	44	56	49	56	49	50	62	56	59
	T2	2	#	#	#	#	#	#	45*	44	50	46	56	42	62*	58†	56	59
	T3	0	0	0	0	0	0	0	56	41	46*	49	56	44	57*	62	66*	64*
14[d]	**T0**	NT	NT	NT	NT	NT	NT	NT	45	50	46	49	46	42	45	39	62	45
	T2	10	41	40	41	#	#	#	56†	62†	46	49	51	50	50*	52*	62	54*
	T3	#	#	#	#	#	#	#	#	#	#	#	#	#	#	#	#	#

* Statistically significant improvement compared to baseline
†Statistically significant decline compared to baseline
[a]Pcr: Parent–child relationship problems; Par: Parenting problems; Dep: Depressive Mood; Rol: Parental role restriction; Phy: Physical health problems
[b]IP: Intrapersonal; IA: Interactional; BC: Behavioral control
[c]Implicit program
[d]Explicit program
Missing value
Italics represent participant included in the analysis of the VOST
For VOST and OBVL, higher score indicates higher stress; for EMPO, higher score indicates higher empowerment

the RCT.[9] As causality cannot be assumed and improvements may have been affected by maturation, the Hawthorne effect, or other mediating or moderating factors, the results should be interpreted with caution. Moreover, participants who discontinued the home-based training also withdrew from the assessments and could therefore not be analyzed. This fact should be considered in order to avoid an overestimation of the presented findings. Last, the lack of psychometrically sound instruments to assess some constructs of specific interest to home-based training left no other option but to use new measures that have not yet, or only to a limited extent, been validated.

Future Research

The combination of findings for child- and parent-related outcomes, though preliminary, provides support for the potential effectiveness of goal-directed task-specific bimanual home-based training programs. Interdisciplinary coaching, involving a remedial educationalist or health-care psychologist, is recommended to prevent therapy-related parental stress and may even produce reduction of generic parental stress. High-quality effect studies are warranted to confirm the hypotheses that can be generated from this study.

Conclusion

This study has been the first attempt to examine bimanual home-based training programs adopting interdisciplinary coaching for children with uCP. The programs seem to positively affect children's bimanual performance. A decrease of therapy-related and generic parental stress levels occurred more often than did an increase. These encouraging results require continued efforts to establish the effectiveness of interdisciplinary home-based training programs, both on child- and parent-related variables, in children with uCP.

Acknowledgments

We sincerely thank the children and their parents who generously gave their time and effort to participate in this study. We also greatly appreciate the dedicated commitment of the therapists, remedial educationalists, and their colleagues. We would like to acknowledge the research assistants who conducted and scored the assessments. We highly esteem the contribution of the COAD Author Group: Nicole Brouwers, Anke Defesche, Hanneke Denissen, Rosemarie Derickx, Yvonne Geerdink, Ruth Heijsen-van den Heuvel, Bregtje Janssen, Anne Jensen, Marjon Kissels, Martijn Klem, Nanne Land, Denise Martens, Judith van Munster, Bianca Olive, Marleen Philippines, Caroline Scheijmans, Lucianne Speth, Ingrid van den Tillaar, and Eef Vennix. Finally, we thank Les Hearn for proofreading the manuscript.

Funding

This work was supported by HandicapNL (former Revalidatiefonds), Johanna Kinderfonds, and Stichting Rotterdams Kinderrevalidatie Fonds Adriaanstichting in the 3rd Program Rehabilitation Research of ZonMw (the Netherlands Organization for Health Research and Development) under grant number 630000001; HandicapNL (former Revalidatiefonds) under grant number R2016006; and Stichting Vooruit under grant number 18–05/YvH/NS.

Declaration Of Interest

The authors report no conflict of interest.

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