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ORIGINAL ARTICLE

Effectiveness of the home-based training program Tele-UPCAT (Tele-monitored UPper Limb Children Action Observation Training) in unilateral cerebral palsy: a randomized controlled trial

Elena BEANI [1, 2], Valentina MENICI [1, 3], Elisa SICOLA [1], Adriano FERRARI [4], Hilde FEYS [5], Katrijn KLINGELS [5, 6], Lisa MAILLEUX [5], Roslyn BOYD [7], Giovanni CIONI [1], Giuseppina SGANDURRA [1, 2] *

[1]Department of Developmental Neuroscience, IRCCS Stella Maris Foundation, Pisa, Italy; [2]Department of Clinical and Experimental Medicine, University of Pisa, Pisa, Italy; [3]University of Pisa, Pisa, Italy; [4]Unit of Children Rehabilitation, IRCCS S. Maria Nuova Hospital, Reggio Emilia, Italy; [5]Department of Rehabilitation Sciences, KU Leuven, University of Leuven, Leuven, Belgium; [6]Rehabilitation Research Center, Faculty of Rehabilitation Sciences, University of Hasselt, Hasselt, Belgium; [7]Queensland Cerebral Palsy and Rehabilitation Research Centre, Centre for Children's Health Research, Faculty of Medicine, University of Queensland, Brisbane, Australia

*Corresponding author: Giuseppina Sgandurra, Department of Developmental Neuroscience, IRCCS Stella Maris Foundation, Pisa, Italy.
E-mail: g.sgandurra@fsm.unipi.it

ABSTRACT

BACKGROUND: The effects of unilateral cerebral palsy (UCP) are largely observed in the upper limb (UL), which represents the main focus of rehabilitation for this disorder. Thanks to an increment in home training and progress in technology innovative systems have been created. The Tele-UPCAT (Tele-monitored UPper Limb Children Action Observation Training) platform is dedicated to the delivery at home of a program for UL rehabilitation, based on action observation therapy (AOT).

AIM: This study aimed to investigate the immediate effectiveness of Tele-UPCAT for promoting UL skills in children with UCP and to determine if immediate effects were retained in the medium and long term.

DESIGN: Tele-UPCAT was conducted on an intention-to-treat basis and was proposed as a randomized, allocation concealed (waitlist controlled) and evaluator-blinded clinical trial with two investigative arms: intensive in-home AOT program and standard care (SC).

SETTING: This is a home-based AOT program delivered with a customized ICT platform.

POPULATION: Thirty children (mean age 11.61±3.55 years) with confirmed diagnosis of spastic UCP with predominant UL involvement and cognitive level within or at normal limits were enrolled in this study. Orthopedic surgery or an intramuscular botulinum toxin A injection in the UL within 6 months prior to enrolment represented an exclusion criteria.

METHODS: Participants were randomized using concealed random allocation. They were assessed according to the study design with the Assisting Hand Assessment (AHA), the Box and Block Test (BBT) and the Melbourne Assessment 2 (MA2). Linear mixed models were used for statistical analysis.

RESULTS: A significant difference between the AOT and SC groups was identified immediately after the training on the AHA (6.406 [2.73] P=0.021) with an effect size (ES) of 1.99, and for the BBT of the less affected hand (9.826 [4.535] P=0.032) with an ES of 1.44. These effects were sustained at medium and long term.

CONCLUSIONS: This study supports the effectiveness of AOT home training in promoting UL skills in children with UCP, with immediate effects lasting for 6 months.

CLINICAL REHABILITATION IMPACT: This should encourage the use of technology for rehabilitative purposes and further applications of the AOT paradigm.

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KEY WORDS: Cerebral palsy; Information technology; Child; Upper extremity.

Cerebral palsy (CP) is the most common motor disorder in childhood and unilateral spastic CP (UCP, involving one side of the body, often the upper limb (UL) more than the lower limb) is the most common motor distribution.[1, 2] UCP results in limitations in independence and in activities and restricts participation.[3]

A systematic review and meta-analysis by Sakzewski *et al.*[4] highlighted the efficacy of nonsurgical UL therapies for children with UCP and indicated that programs of constraint-induced movement therapy (CIMT), bimanual training and botulinum-toxin A combined with occupation therapy (OT) are indicated for improving UL outcomes. There was strong evidence that goal-directed OT home programs are effective in increasing the dose of intervention.[5]

New approaches based on the recent discovery of the mirror neuron system (MNS) have fostered the development of action observation therapy (AOT), based on the observation of meaningful actions followed by their execution.[6] A recent systematic review by Novak and collaborators[5] applied a traffic light system to summarize evidence on interventions for preventing and treating children with CP, where AOT was indicated with a green light, which means highly recommended, for its role in being effective on the improvement of motor function in children with CP.

Indeed, AOT has been used with promising results in some pilot studies mainly on adult patients with stroke,[7] with some recent studies carried out on children with CP,[8] indicating positive effects on the UL function.

Together with the development of new rehabilitative approaches, in recent years there has been increasing interest in promoting home based rehabilitation to optimize patient wellbeing and family involvement[9] and reduce burden and costs.[10] To date, studies have been carried out mainly on infants,[11] with some applications on older children.[12] Rehabilitation programs based on integration of Information and Communication Technologies (ICTs) into clinical and research practice, are increasingly delivered in the home situation. There are several examples of its application in childhood[13] and in infancy;[14] in particular in Italy, as highlighted by Maresca *et al.*[10]

In the panorama of the home-based approach by means of a technological device, we present the Tele-UPCAT (Telemonitored UPper Limb Children Action Observation Training) program, which combines AOT and home-based therapy in a system built by the BioRobotics Institute of Scuola Superiore Sant'Anna in collaboration with IRCCS Fondazione Stella Maris.[15] The Tele-UPCAT system represents the technological evolution of the UP-CAT approach[16] which applied the same AOT paradigm in the clinical environment. The Tele-UPCAT has been developed to provide AOT training directly at home. Prior to examining effectiveness,[17, 18] a feasibility study was performed,[19] with positive results. In fact, the Tele-UPCAT has already been assessed, in detail, for the acceptability and ease of use for providing AOT home-based training from the point of view of both developers (clinicians and engineers) and children and adolescents who attended the training program. The feasibility, described in the dedicated paper,[19] has been explored by analyzing the results of the adherence to the training and by means of an ad-hoc questionnaire. Standard items of usability and acceptability were used to create the questionnaire, in which the participants' opinion of the feasibility of the training (in terms of study design and procedures) was investigated. Data showed that all the feasibility criteria were met and the total scores were all above 82.15%, with high results in all of the four subscales. Since there were no differences related to the age and the sex, the Tele-UPCAT system showed to be a feasible home-based AOT training program both for children and adolescents with UCP.

With these data in mind the present work investigates the effectiveness of AOT, by means of Tele-UPCAT, to promote UL skills in children with UCP. The working hypothesis is that children receiving AOT in the home environment would have significant improvements in UL uni- and bimanual skills immediately and that these effects would be sustained for the medium- and long-term follow-up.

Materials and methods

Participants

Starting from 29 March 2017, children and adolescents with UCP ranging in age between 5 and 20 years, who had been referred to IRCCS Fondazione Stella Maris (Pisa) and Unit of Children Rehabilitation of S. Maria Nuova Hospital (Reggio Emilia), were enrolled according to the following inclusion criteria:

- confirmed diagnosis of spastic motor type UCP;
- predominant UL involvement with a House Functional Classification System (HFCS)[20] level ≥2, which means at least the ability to passively hold an object in the hand or better;
- cognitive level within or at normal limits, that is IQ≥70, as assessed in the last year prior to recruitment on the Wechsler Preschool and Primary Scale of Intelligence-Third Edition (WPPSI-III), Wechsler Intelligence Scale

for Children Fourth Edition (WISC-IV) or Wechsler Adult Intelligence Scale (WAIS);

• availability to commit to a home program of intensive therapy for 3 weeks.

Children were excluded if they had received orthopedic surgery or an intramuscular botulinum toxin A injection in the UL within 6 months prior to enrolment.

The study was completed on 29 November 2018.

Study design

This trial was approved by the Ethics Committees of the involved Clinical Units: Tuscan Pediatric Ethics Committee (number 169/2016) and Provincial Ethics Committee Reggio Emilia (number 2017/0112191). It was registered at http://www.clinicaltrials.gov (NCT03094455) on 16 March 2017. All parents provided written informed consent to participate in the trial.

Tele-UPCAT was conducted on an intention-to-treat basis and was proposed as a randomized, allocation concealed (waitlist controlled) and evaluator-blinded clinical trial with two investigative arms including an intensive in-home AOT rehabilitation program and a Standard Care (SC) group.

Pediatric neurologists and physiatrists were in charge of the identification of eligible children, of their recruitment and of the clinical trial management.

The primary outcome measure was the Assisting Hand Assessment test (AHA);[21-23] while other secondary clinical outcome measures included: Melbourne Assessment 2 (MA2),[24] and Box and Block Test (BBT).[25] Further questionnaires (such as ABILHAND-Kids,[26] Participation and Environment Measure (PEM-CY)[27] and Children and Youth and Cerebral Palsy Quality of Life, (CPQoL)[28] and the questionnaire regarding usability and acceptability[19]) were administered. Moreover, quantitative data from a sensorized toy and wearable sensors (Actigraphs) were used, as described in the study protocol[15] but their results are outside the scope of the current paper. The primary and secondary clinical outcome measures were scored from video recordings of the assessment by trained therapists masked to group assignment of the child and also to the time point assessment of the recording.

The assessments were carried out by two therapists whose years of experience in children assessment and rehabilitation differs (one senior and one junior). Both are certified AHA raters with a high interobserver agreement.

After the completion of the baseline assessment, participants were block randomized in one of two investigative arms, immediate AOT or SC group using concealed random allocation. This was carried out by a blind researcher who was not involved in the trial, with the use of a computer-generated code of random numbers. Participants were assessed at baseline (T0) and subsequently after the first intervention period of 3 weeks, which was AOT for the experimental group and SC for the control group (T1). The control group which started from the SC, defined as the usual care of participants, mainly consisted of one or two sessions of physical or occupational therapy per week. After this 3-week period (*i.e.* at T1) they had the opportunity to carry out AOT training, so an additional assessment was carried out after the end of the AOT training sessions (T1 plus). Both groups were then assessed at 8 weeks (T2), and 24 weeks (T3) after the end of the training program. In order to monitor the other therapies potentially carried out during the period of the study, all families were required to keep diary with a daily writing all the details of their routine.

The AOT home program was composed of a set of 15-day sessions of three sequential goal-directed actions, with increasing complexity within the training days, to be performed uni-manually for the first 8 sessions and bi-manually for the final 7 sessions. Each daily session lasted approximately 1 hour a day: it was organized in alternate periods of observation and action lasting 3 minutes and was repeated twice for each of the three daily actions. Training was provided thanks to the Tele-UPCAT system, composed of an all-in-one computer with an *ad-hoc* software, a kit of toys and a pair of Actigraphs which were delivered directly to the children's houses, together with a detailed user's manual. Each system was set up with the chosen activities for the specific child. The content of the rehabilitative activities was based on some typical everyday actions, such as, filling a glass with water for a unimanual action (action 1: opening the bottle; action 2: filling a glass with the water; action 3: dropping a little stone in the glass), while, cooking with a toy oven for a bimanual action (action 1: bringing the Play-Doh closer; action 2: manipulating the Play-Doh and dividing it into pieces and placing them in a saucepan; action 3: opening the play oven, inserting the saucepan, closing). All the details of the system have been described in the study protocol.[15]

Activities were customized on the basis of the child's level of ability while maintaining the same general goals: children used the same type of objects and the customization consisted of differences in the range of the required movement, the type of grasp, the shape and size of the material in order to facilitate the affordance and to guarantee the feasibility of the proposed activity for each child both in terms of movements and goals. For this reason, starting from the exercises according to the overall library of

Table I.—*Template for Intervention Description and Replication (TIDieR) checklist.*[18]

Item	Description
Why	Rationale: The evidence-based treatment with AOT in promoting UL functional skills, could be delivered also in the home environment thanks to the technology. Core elements: 1. The UL as a main goal of the rehabilitative intervention of children with UCP 2. The application of technology in delivering AOT at home 3. The customization of rehabilitation activities 4. The duration of the effect of the AOT
Materials	• Tele-UPCAT system composed by an all-in-one computer equipped with a camera; which contained the rehabilitative activities calibrated on functional UL abilities and organized increasingly challenging, presented with a dedicated age-related software (“observation module”) • A box containing a kit of toys to be used for the activities (“motor performance module”) • A pair of Actigraphs realized as bracelets with Velcro straps
Who	Pediatric neurologists and physiatrists for identification of eligible children, recruitment and clinical trial management. Child therapists with experience in the assessment and rehabilitation of children with UCP and certified raters of AHA for the evaluation, the set up and the supervision of the home training. Parents/caregivers coached by therapists to carry out the home training.
How	Clinical setting for assessments and home environment for training and follow-up assessments
How much	15 sessions in 3 weeks, 5 days a week with a daily duration of about 1 hour
Tailoring	Tailored to the child individually manual abilities (HFCS defined at baseline) and age (less or more than 12 years)
How well	To record the feasibility of the training, participants completed an ad-hoc questionnaire which highlighted a good rate of acceptance and usability of the Tele-UPCAT.[18]

UL: upper limb; AOT: action observation therapy.

activities, the rehabilitative staff selected the more suitable kit of exercises basing on the overall level of manual skills of the child; this was driven by the established HFCS level and, in case of doubt, on the observed behavior during the T0 assessment.

The Tele-UPCAT intervention is summarized according to the Template for Intervention Description and Replication (TIDieR) Checklist in Table I.[18]

Statistical analysis

Means and standard deviations of the baseline (T0) demographic and clinical data for all selected outcome measures for both groups were calculated to identify the potential baseline differences between groups. Due to the normal distribution of clinical outcome measures, assessed with Shapiro-Wilk’s Test, the independent sample t test was carried out to verify that the 2 groups had the same baseline.

Linear mixed models were fitted, with fixed effects TIMING and GROUP (along with their interaction) and random effects AGE and HFCS. The primary endpoint analysis was related to the AOT Immediate training effect, *i.e.* the difference between the two groups in the time before and immediately after the training session: [Right After Training] - [Just Before Training], *i.e.* (AOT at T1+SC at T1 plus) - (AOT at T0+SC at T1). Cohen’s d effect sizes (ES) were also calculated to evaluate the strength of the AOT Immediate training effects. According to Cohen[29] we have defined the substantive significance between groups as small (<0.2), medium (<0.5) or large (≥0.8), respectively.

If the effects were significant, two further post-hoc analyses were conducted. The first was related to the all-time effects *i.e.* the AOT effects at each time point with respect to baseline and SC, measuring the difference in the scores [Overall After Training] - [Overall Before Training] with the following formula: (AOT at T1 and following timings+SC at T1 plus and following timings) - (AOT at T0+SC at T0+SC at T1). The second *post-hoc* analysis was the within AOT medium-long term effects, *i.e.* the difference in the timings when both groups were 8 (T2) and 24 (T3) weeks after AOT training using the following formula: (AOT at T0+AOT at T2+AOT at T3) - (SC at T0+SC at T2+SC at T3).

The all-time effects analysis was conducted in order to assess if the effects were retained during the follow-ups. The AOT medium-long term effects analysis may not be significant, as it describes differences between groups when no differences in training are in place. Finally, a further analysis was carried out to analyze the long term effects comparing the delta scores of T3-T2.

All analyses were conducted in R (version 4.2.1, 2022), supported by external statistical expertise. Significance was set for P value<0.05.

Data availability

The data associated with the paper are not publicly available but are available from the corresponding author on reasonable request.

Results

Participants

The minimum sample size was 10 children per group, but when a possible 20% of drop-outs was considered, 12 children per group were needed to be recruited, to reach a minimum number of 24 participants. As described in the study protocol,[15] it was estimated to detect a 1.40 effect size (value based on our preliminary data[16, 30]) at a significant level of 0.05% and 80% power. However, when the minimum number was reached, we continued with the enrolment, thus obtaining a total of 30 children and adolescents (Figure 1), living in 10 different Italian regions. The total group was composed of 14 females and 16 males, with a mean age of 11.61±3.55 years and an age range between 6.47 and 18.68 years. The affected side was the right side in 10 cases and the left side in 20 cases. The mean AHA score was 53±11.30 AHA units.

The children were randomly allocated to the immediate AOT group (N=15, mean age 11.45±3.70 years, age range 7.12-18.68 years, M:F=8:7, right UCP:left UCP=6:9, mean AHA score 55±10.09 AHA units) or to the SC group (N=15, mean age 11.77±3.53 years, age range 7.17-17.85 years, M:F=8:7, right UCP:left UCP=4:11, mean AHA score 51±12.37 AHA units). At T0 the two groups did not show any statistically significant difference for any parameter of interest: sex, affected side, AHA units at T0.

All the children underwent the first 3 week period (15 sessions of AOT or SC) according to their allocation. Subsequently, all the AOT training sessions were fully completed by all the children, both in the AOT group and in the SC group also.

Regarding the assessments, all the children performed the required timepoint assessments (baselines and further follow-ups) with no missed sessions and no adverse events detection. This high retention reconfirms the feasibility of the program.[19]

Outcome measures

Primary outcome measure

AHA

The AOT group showed a higher significant difference in the AHA scores than the SC group both for the immediate results (at T1 or T1 plus). Moreover, the within-group analysis showed significant all time effects (Table II, Figure 2). This effect was then retained both at medium and long term and also between T2 and T3 (long lasting effects, Table III).

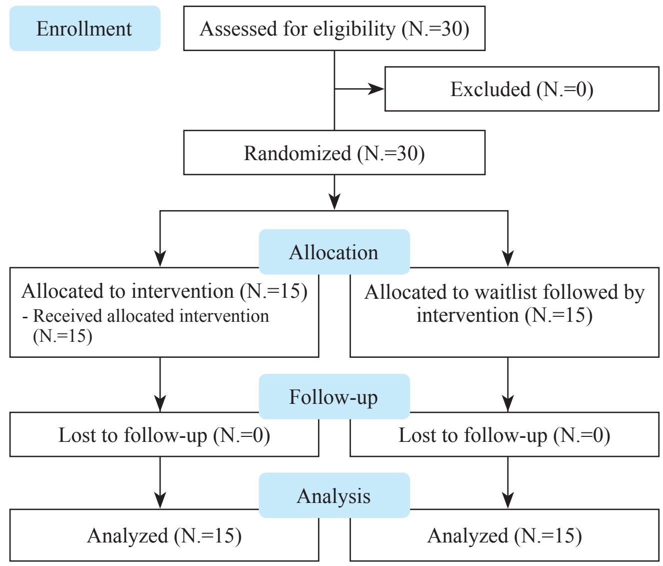

Figure 1.—The CONSORT flow diagram.

Moreover, we added a deeper analysis of the AHA delta scores in the AOT group before and after the training by calculating the percentage of children who showed a change in each single item (Figure 3).

Secondary outcomes

BBT

At the immediate post training assessment, the BBT of the less affected hand demonstrated significantly larger improvements in the AOT group compared to the SC group, whereas in the BBT of the affected hand data were not significant (Table II). Moreover, the within-group analysis showed significant results both at medium and long term and between T2 and T3 (long lasting effects, Table III).

MA2

At the immediate assessment, no differences were found in the AOT group with respect to the SC (Table II).

Discussion

To our knowledge this is the first study to provide evidence of the effects of a customized home-based AOT program, which is a recent approach supported by RCTs. Systematic reviews have also indicated intervention effectiveness in children with CP[8, 31] sustained also by evidence detected

TABLE II.—*Immediate effect of the AOT on primary and secondary outcome measures.*

			Immediate effect				
			AOT group (mean±SD)	SC group (mean±SD)	Estimate (SE)	df* (sign)	Effect size (95% Confidence Interval)
Primary outcome measure							
Assisting Hand Assessment (AHA)		T0	55.07±10.09	50.87±12.37	6.41 (2.73)	113.02 (0.02)	1.99 (1.06-2.93)
		T1	58.00±10.49	50.60±11.66			
		T1 plus	-	55.21±11.49			
Secondary outcome measure							
Box and Block Test	Less affected hand	T0	57.27±12.01	56.27±8.05	9.83 (4.54)	110.03 (0.03)	1.44 (0.55-2.33)
		T1	63.80±12.53	57.93±9.89			
		T1 plus	-	61.62±10.20			
	Affected hand	T0	26.40±10.43	23.40±11.73	4.78 (3.73)	110.17 (0.20)	1.16 (0.30-2.01)
		T1	29.13±9.17	26.79±11.03			
		T1 plus	-	29.92±11.59			
Melbourne Assessment 2	ROM	T0	18.00±5.88	15.38±5.25	2.87 (1.54)	105.51 (0.07)	1.48 (0.54-2.41)
		T1	19.47±5.81	16.08±5.14			
		T1 plus	-	18.00±5.20			
	Precision	T0	21.73±4.57	21.62±4.07	2.51 (1.29)	105.51 (0.05)	1.00 (0.13-1.88)
		T1	23.07±2.99	21.46±4.05			
		T1 plus	-	23.08±1.98			
	Dexterity	T0	9.27±3.30	9.39±2.43	1.99 (1.05)	105.51 (0.06)	2.53 (1.42-3.64)
		T1	10.40±3.20	9.31±2.46			
		T1 plus	-	10.50±3.24			
	Fluency	T0	11.67±2.94	10.39±2.90	1.44 (1.08)	105.51 (0.19)	0.98 (0.09-1.85)
		T1	12.60±2.95	10.54±3.10			
		T1 plus	-	11.33±3.29			

SE: Standard Error, df: difference; sign: significancy; ROM: Range of Movement, AOT: action observation therapy.

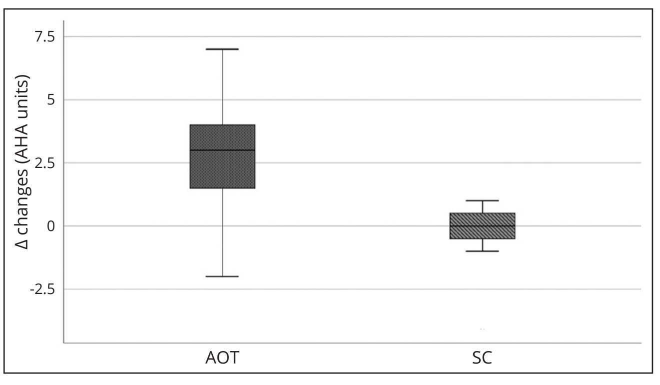

Figure 2.—The effect of the Action Observation Training (on the left) and of the Standard Care (on the right) at the Assisting Hand Assessment.

at the functional magnetic resonance imaging.[32, 33] This home based program was delivered by means of an ICT system, called Tele-UPCAT, in children and adolescents with UCP across broad developmental ages. The effects of the AOT training were related to the use of the assisting hand in bimanual tasks (AHA) and unimanual dexterity of the less affected hand (BBT) with large ES, while there were no significant effects in the unimanual skills (BBT and MA2) of the affected hand even if there were high values of ES. Moreover, for the first time, we showed the effects not only after the end of AOT home training but also their retention at 6 months of follow-up.

This study reports the evolution of the UP-CAT, an approach tested in a previous RCT study of AOT carried out

TABLE III.—*All time effects, medium and long term effects and long lasting effects of the AOT.*

	All time effect		AOT medium-long term effects		T3-T2 Long lasting effects					
					AOT group		SC group			
	Estimate (SE)	df* (sign)	Estimate (SE)	df* (sign)	T2	T3	T2	T3	Estimate (SE)	df* (sign)
Assisting Hand Assessment	2.25 (0.98)	113.10 (0.02)	-6.47 (3.73)	114.77 (0.09)	58.20±11.26	58.07±11.95	52.54±12.09	52.44±13.97	-0.05 (2.97)	113.10 (0.99)
Box and Block-Less affected hand	7.06 (1.62)	110.10 (0.00)	- 1.39 (6.13)	111.93 (0.09)	62.73±11.36	63.20±13.79	64.08±10.96	67.22±7.97	2.98 (4.90)	110.08 (0.55)

SE: standard error; df: difference; sign: significancy; AOT: action observation therapy.

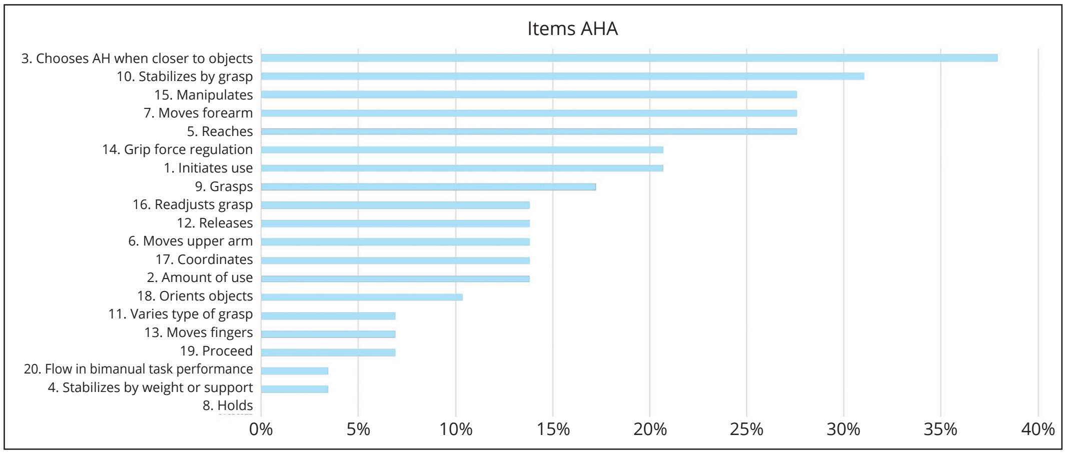

Figure 3.—Percentage of children who showed a change in each single item AHA delta scores, ranked on the basis of the Rasch analysis, in the AOT group before and after the training.

in hospital. In the current study, the main innovation was the use of technology and the delivery of AOT at home.[16]

The most important implication of the present study is that it suggests that the home-based training program seems to have an effect on UL skills in children with UCP.

We must consider that there are some differences in the study design of these two projects, regarding the setting (clinic *vs.* home), the control group (since in the first study there was a comparison between the effect of the AOT group and an active group, while in the present study we compare with the SC group) and the selected outcome measures (AHA version 4.4 and MUUL *vs.* AHA version 5.0 and MA2). For these reasons, although a direct comparison of the effectiveness of UP-CAT and Tele-UPCAT is not possible, a way to estimate the magnitude of findings could be made by considering the effect size of AHA, which is higher (1.40 in the UP-CAT and 1.99 in the Tele-UPCAT, respectively).

This indicates that delivering the AOT directly at home preserves the feasibility and the effects.

In facts as in UP-CAT, in the Tele-UPCAT the effect of AOT on UL and hand abilities was found across all HFCS groups (these were enlarged to 2-3 HFCS levels in the Tele-UPCAT) and age (evaluated as random factors). This means that even if there are specific differences in the motor execution addressed to obtain the same goal, the improvements were significant across all levels of abilities.

Another crucial difference with other studies involving the AOT (as the UP-CAT) is represented by the use of a technological system, which made the delivery of the AOT treatment at home possible.

These results appear to promote the development and the use of technologies for home rehabilitation, as confirmed by the growing use of tele-rehabilitation programs in childhood. These approaches, no doubt prompted by the emergency related to the COVID-19 pandemic,[34] could represent the future of rehabilitation.[35] Nevertheless, during the pandemic the tele-rehabilitation in children was almost exclusively delivered by means of video-calls[36] without detailed informative material for families. Furthermore, the majority of the current systems used for home rehabilitation are designed primarily for adults and then later adapted to children. However, there are many strengths in the use of technology for delivering rehabilitation at home.[37] Specifically, children with CP report the hand/arm use and use of assistive technologies as priorities when considering treatment outcomes, confirming their preference in using ICT for rehabilitation intervention.

Tele-UPCAT is the first system built for providing the AOT program directly at home. It was designed to guide children and their parents, step by step, in the sequence of observation and action, within an age-related framework.[15, 19] Kirkpatrick *et al.*[38] were the first to deliver AOT at home by instructing the parents on how to show the activities to their children. They found an equal improvement of UL function in the AOT group as after repeated practice. Recently, Molinaro and collaborators conducted a pilot study[39] reporting that AOT home training, provided by online sessions, permitted an immediate improvement in uni- and bimanual skills (assessed by AHA and Melbourne Assessment of Unilateral Upper Limb Function, MUUL), which was retained for 2 months.

Another recent study by Nuara *et al.*[40] provided AOT home training for children with UCP with both videos and child-to-child interaction. In this last study, they reported higher UL and manual abilities (measured with Besta Scale) immediately after the training session, but no changes were reported in the neurological impairment evaluated with Fugl-Meyer Assessment.

In both UP-CAT and Tele-UPCAT studies and according to the literature in this field, an improvement has been found in the spontaneous use of the affected hand during bimanual activities (as shown at AHA assessment).

In fact, if we consider the analysis of the single AHA items (Figure 3), some interesting details in the changes in the behavior of the children after the AOT training can be observed. The item which has changed most is "chooses the assisting hand when closer to the object." This is explained by the fact that the behavior of the children changed in that they no longer favored the less affected hand to reach all objects but they reached for the object with the affected hand and started using the affected hand more frequently to spontaneously handle some objects.

Furthermore, the item "reaches" also changed, since they used the movement of the more affected arm more often to reach objects with respect to the baseline assessment, in which they used the less affected side to bring objects close to the more affected hand, which they kept close to the trunk. Presumably the training, in particular the unimanual actions, promoted the movement of the more affected arm and worked as a reminder to use it, stimulating some actions that are not so often prompted in daily life, such as the supination of the forearm (item "moves forearm"). Nevertheless, this is not confirmed by an improvement in the dexterity and a reduced motor impairment of the more affected hand, as shown, respectively, by the results of the BBT and all the scores of the MA2, that is the ROM, the precision, the dexterity and the fluency of the UL movement. The hypothesis formulated is that the training acted on the global use of the more affected side, expressed in the improvement of the arm skills and the use of the hand for the bimanual actions, so not related to the hand dexterity alone, but in relation to the less affected side.

In fact, these children could have adopted a strategy for compensating their difficulties in bimanual tasks, *i.e.* the use of their less affected hand for fostering the integration of the affected hand.

These findings also support the hypothesis that the content of our training, focused on goal-directed activities which are customized for different motor impairments, directly urge the children to maintain the goal of the action, rather than the action kinematics.[41] In other words, children who are trained with our program, could tend to be more focused on the success of the action thus maintaining its goal, rather than executing a faithful reproduction of the observed movement. For this reason, the improvement in the AHA could mean that the effects of the training are related to the initiative of use of the affected side. The content of the AOT training, *i.e.* goal-directed uni- and bimanual actions, could prompt a sort of reminder for a higher use of the affected side.

This is confirmed by two other items of the AHA that have changed in many children: "stabilizes by grasp" and "manipulates". In this case, these items showed us that the more affected hand is used in a more effective way for keeping the object in the hand, by using the grasp and reducing its slipping or involuntary dropping, in particular when the less affected hand acts on it. Moreover, this skill opens up the possibility to move the object in the hand to better organize the bimanual function. Knowing that the AHA items are ordered on the Rasch analysis, it may sound odd that one of the most difficult items of the AHA changed ("manipulates") and the easiest one ("holds") did not change for any participant. This can be due to the fact that from one perspective the higher items may be related to the best performance of the child ("manipulates") or that they moved from a score which indicates that the performance was ineffective, (or there were no attempts to act with the hand), to how they initially used their hand in association with the other hand (*e.g.* "chooses the assisting hand when closer to the object"). However, it must be remembered that the easiest items need to be performed in the whole play session if they are to obtain a high score. In addition to this, we must consider sensitivity to a ceiling effect which is the case of the item "holds". This item required the ability to hold an object in the hand and was related also to the inclusion criteria of the sample (basing on HFCS).

In addition to the effect on the use of the more affected hand in the bimanual activities raising at the AHA, the very new result of the increased dexterity of the less affected hand opens interesting considerations regarding the role of this hand in children with UCP. This has already been described as important and should be considered for the rehabilitation process[42] and for what is included in the training, referring in particular to the bimanual actions.

Limitations of the study

Although this study offers important insights for the use of ICT solutions for providing AOT programs at home, some limitations need to be underlined. First of all, the number of enrolled participants, although beyond the minimum sample size, is still quite limited and would benefit from further studies for confirmation. Secondly, the waitlist study design with the training of the control group starting at T1, did not allow us to assess the between group differences at all time points. In order to confirm the effectiveness of in-home AOT, it could be also appropriate to compare the effect of AOT not exclusively with the SC, but with a group who carried out AOT training in a clinical setting. A further study could benefit from the addition of this third investigative arm, by using the same outcome measures. Moreover, even though different sets of exercises were provided according to the HFCS level, there was not a customization in relation to individual characteristics and interests or a further customization in relation to the children's progress. Furthermore, even if we did use the technology, we did not provide online sessions and the whole training program was carried out at home by children and their parents without supervision, nor were quantitative measurements regarding the level of attention recorded. Finally, a cost-effects analysis could be useful for describing all aspects of this new approach.

Conclusions

This study shows the effects of a 3-week AOT home training program in promoting the UL goal-directed skills in children with UCP by means of a customized ICT platform, called Tele-UPCAT. To our knowledge this is the first RCT study, which opens new frontiers for the potential use of ICT solutions for providing feasible and effective AOT home training in childhood.

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Conflicts of interest
The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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Authors' contributions
All the authors contributed to the conceptualization and Methodology of the study. Elena Beani, Valentina Menici, Elisa Sicola and Giuseppina Sgandurra did the data curation; Giuseppina Sgandurra carried out the formal analysis, funding acquisition, investigation, project administration, software, supervision and validation; Elena Beani and Giuseppina Sgandurra drafted the manuscript, and all the other authors reviewed and edited the final version. All authors read and approved the final version of the manuscript.

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