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A long-term observation on effects of modified constraint-induced movement therapy combined with bimanual training in children with hemiplegia in "The Pirate Group" program – case report

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Abstract

Study aim: The aim of the research was to present a unique intervention for restoring limb function in cases of hemiplegia based on a modified constraint-induced movement therapy (mCIMT) combined with bimanual training (BIT) carried out as part of "The Pirate Group" program, and long-term observation of its effects.
Material and methods: The study included five boys aged three to five years, diagnosed with cerebral palsy (CP) with a form of hemiplegia (unilateral cerebral palsy). The effectiveness of the therapy and its long-term effect were analysed based on the Small Kids-AHA test conducted before the therapy, after two weeks of therapy, and six months after the end of therapy.
Results: Some differences were noted between the global scores of Small Kids-AHA test obtained before and after two weeks of therapy. No differences were found between the global scores of Small Kids-AHA test obtained after two weeks of therapy and six months after the end of therapy.
Conclusion: Modified constraint-induced movement therapy combined with bimanual training carried out as part of "The Pirate Group" program is an effective therapeutic intervention, and its effects may persist for six months after the end of therapy.

Key words: BIT – Cerebral palsy – Hand therapy – Hemiplegia – mCIMT

Introduction

The most common cause of motor disability in early childhood is cerebral palsy (CP) [18, 36]. Hemiplegia is the most common form of CP among full-term children and the second most common (after diplegia) in preterm children [10, 45, 46]. It is characterized by paresis on one side of the body, opposite to the location of the brain lesion [10]. Children with hemiplegia caused by primary and secondary neuromuscular disorders have problems with performing basic activities, such as reaching, grasping, holding or releasing objects [7, 16, 34]. As most everyday activities, such as tying shoes, peeling a banana, dressing a doll or using scissors, require coordinated work by both hands, these limitations have a negative impact on functioning and participation in social life [4, 8, 23].

Paediatric occupational therapy seeks to increasing children's independence and participation in daily life roles [3, 35]. Occupation therapy for children focuses on the development of activities such as play, self-care, and fine motor task e.g. writing or drawing [47]. To achieve this goal, the functional status of the upper limb must be improved. Hand therapy is a specialty area of practice for occupational therapists working in the pediatric setting [13]. A number of therapeutic strategies have been developed to improve upper limb function. Studies indicate that the most frequently-used hand therapy methods are modified constraint-induced movement therapy (mCIMT) and bimanual training (BIT); both have been found to be effective, and the two have been found to bring very good results when used in combination [2, 5, 6, 11, 14, 15, 20, 36, 42, 44] . However, there are many discrepancies regarding the duration of immobilization of the more efficient upper limb and the total duration of therapy, and no guidelines exist regarding the optimal or minimum duration of effective therapy. Although reducing the duration of intensive therapy brings many benefits, including making it less burdensome for the child and the family [32] and reducing the cost of therapy. The length of therapy needed to achieve

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long-lasting effects is unclear. As such there is a need to verify the long-term effects of the analysed interventions and conduct long-term studies (follow-up).

As children with hemiplegia present a heterogeneous clinical picture, not to mention considerable differences in their response to upper limb rehabilitation, it is not easy to put theoretical models into practice. Furthermore, physiotherapy and occupational therapy constitute an integral part of the life of children with disabilities, and as such, therapy should be effective, interesting and targeted at the individual needs of the child. Therefore, creating an optimal therapy program for these children is a challenge. With this in mind, the aim of this study is to present a unique approach based on a combination of modified constraint-induced movement therapy with bimanual training, carried out as part of “The Pirate Group” program. It also presents the results of a long-term observation of its effects.

Material and methods

The study examined the outcomes of a unique therapeutic approach called “The Pirate Group” (Table 1). The choice of study design, i.e. a descriptive case report with non-probabilistic sampling, is in accordance with an established conceptual model describing case study research in occupational therapy practice [33].

The study received the permission of the Senate Committee on Research Ethics, the Józef Piłsudski Academy of Physical Education, Warsaw (consent no. SKE01-09/2021) and was conducted by following all principles described in the Declaration of Helsinki. Written consent for participation was expressed by the legal guardians of the children..

Intervention – The Pirate Group

“The Pirate Group“ program has been carried out by qualified physiotherapists at the OLINEK Intensive Therapy Center in Warsaw since March 2016. Classes took place in the form of two-week programs running during weekdays, i.e. for 10 days, with each class lasting two hours a day: total therapy time is 20 hours. Every day, an hour and a half was devoted to mCIMT therapy, i.e. a total of 15 hours, and the final half an hour of each class was devoted to BIT therapy, i.e. a total of five hours.

Modified constraint-induced movement therapy therapy was performed by tying the more efficient hand (the entire upper limb – arm, forearm, hand) with a bandage. The classes were held in the form of play, in a specially arranged room, in groups of four children. A physiotherapist was assigned to each child. During therapy, the children performed tasks related to forming a crew (dressing up in pirate costumes, painting beards and mustaches, preparing provisions for the journey), repairing a ship (removing old nails, patching holes and painting sails), sailing (scrubbing the ship, hoisting the sails to the mast, folding the gangway, looking out for a deserted island through a telescope) or preparing for battle (looking for weapons, collecting

Table 1. Advantages and limitations of “The Pirate Group” therapy

	Advantages	Limitations
“The Pirate Group”	– group therapy – motivation, commitment, imitation, participation, cultivation of social skills, strengthening factor [2, 9, 30]	– limited availability (one center in Poland)
	– arranged environment – motivation, fun, reduces frustration and discomfort during upper limb immobilization [2, 39]	– no scientific reports regarding the long-term effect of therapy
	– a new, different, unknown way of therapy – greater commitment, adventure, willingness to participate in therapy [17, 39]	
	– protocol (using BIT# therapy every day) allowing the participants to implement new skills under the supervision of a therapist every day [39, 40]	
	– positive approach of the parent, commitment, good cooperation with the therapist [2, 39]	
	– fun – “One-armed pirate” – motivation, children are more willing to have their hand immobilized without frustration [2, 29, 39]	
	– satisfactory effects of therapy using a relatively short total therapy time (20 hours) [38, 39]	
	– the effectiveness of the therapy is scientifically confirmed [38]	

#BIT – bimanual training.

ammunition, improving sword-wielding techniques). In order to ensure the best quality of activities performed, depending on the situation, the therapists used wrist orthoses with a thumb hold and kinesio taping. During their two weeks in "The Pirate Group", the children did not participate in any other forms of therapy.

Participants

The study included five boys aged three to five years, diagnosed with cerebral palsy (CP) with a form of hemiplegia. All participated in "The Pirate Group", a two-week intensive therapy program, at the OLINEK Intensive Therapy Center in Warsaw. The characteristics of the subjects are presented in Table 2.

The inclusion criteria for the study were as follows: diagnosis of CP – hemiplegia, age between two to five years, GMFCS assessment at level I or II, MACS/mini-MACS assessment at levels I–III and CFCS assessment at level I–III, intellectual functioning enabling the child to cooperate and follow the therapist's instructions, consent given to participate in the study by the parent or legal guardian. The exclusion criteria comprised the following: lack of physiological well-being (tired, sleepy, sick child), occurrence of uncontrolled epileptic seizures, intolerance of activities, intolerance of limiting the activity of the more efficient upper limb, two or more absences from classes, withdrawal of consent from a parent or legal guardian.

Procedure

Among the participants, the activity of the affected upper limb was assessed at three points: (I) pre-intervention (baseline), i.e. before the start of therapy in "The Pirate Group", (II) post-intervention, i.e. after two weeks of therapy, and (III) follow-up, i.e. six months after the end of therapy. The children did not participate in any form of intensive rehabilitation program during the six-month period from the end of the program until the final assessment; however, they attended standard physiotherapy (one or two times a week) and used the services of other specialists as needed, e.g. a speech-language therapist or psychologist.

Before starting the therapy, a general interview was conducted with the parent. In addition, the child's affected upper limb was evaluated; this included assessments of the type of deformation of the wrist and fingers according to Zancolli classification, the type of thumb deformation according to House classification, types of grip and independence in self-care activities. The level of functioning was also evaluated based on the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS) and the Communication Skills Classification System (CFCS). Based on this information, the therapist completed the Hand Therapy Assessment (clinical document).

Table 2. Characteristics of five boys diagnosed with hemiplegia

No.	Fetal age (weeks)	Birth weight (grams)	Age at examination (years)	Affected body side	GMFCS*	MACS/mini-MACS^	CFCS~	Zancolli#	House$	Comorbid disorders
1	39	3200	5	R	1	2	2	2a	1	Visual impairment
2	40	2700	5	R	2	2	3	2	3	Epilepsy, speech impairment, behavioral disorders
3	38	3470	3	R	1	2	1	1	1	Epilepsy
4	38	3650	5	R	1	2	1	2	1	Visual impairment
5	40	4200	5	L	2	2	3	1	3	Visual impairment, intellectual disability, hydrocephalus

*GMFCS – Gross Motor Function Classification System; ^MACS/mini-MACS – Manual Ability Classification System; ~CFCS – Communication Skills Classification System; #Zancolli – types of wrist and finger deformations according to the Zancolli's classification; $House – type of thumb deformity according to House's classification.

In order to establish individual therapy goals for the tested children, and to verify the effectiveness of the therapy and its long-term effects, each child also received the Small Kids-AHA test at three time points, *viz.* before therapy, after two weeks and again at six months after the end of therapy. The Small Kids-AHA test was administrated by a certified physiotherapist who was not involved in the therapy.

Instruments

The following data were collected from medical records (Hand Therapy Assessment, used at the OLINEK Intensive Therapy Center): fetal age, birth weight, date of birth, affected side of the body, assessment in the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS) and Communication Skills Classification System (CFCS), type of wrist and finger deformities according to Zancolli’s classification, type of thumb deformities according to House’s classification, comorbid disorders.

The Small Kids-AHA test (a version of the Kids-AHA test for younger children) is intended for children aged from 18 months to five years, with unilateral movement disorders (brachial plexus paresis, hemiplegia). It has been repeatedly tested for validity and reliability [21, 23, 26, 28]. The Small Kids-AHA is a non-invasive research tool carried out as a play activity, so children take part willingly, spontaneously and without coercion [26, 37]. All guidelines for the proper conduct of the test are described in detail in the Kids-AHA test instructions [27]. Briefly, the activity of the upper limb used as the assisting hand (i.e. less efficient hand) is assessed during activities performed with both hands during a 15–20 minute session. The child plays with a validated set of toys, forcing bimanual work, and the entire play session is recorded. Based on the recording, the examiner analyzes the degree of cooperation of the upper limbs during play and makes an assessment. The test comprises 20 tasks, each of which is scored from 1 to 4 points, where 4 indicates “effective”, 3 – “somewhat effective”, 2 – “ineffective”, 1 – “does not do“; the total score ranges from 20 to 80 points, with a higher score indicating a higher ability level. An improvement of at least four points in the summary assessment indicates a clinically relevant improvement in the activity of the upper limb [22, 26].

Results

Figure 1 presents the total AHA score (global result of the Small Kids-AHA test) at pre-intervention (baseline), post-intervention (after two weeks of therapy) and at follow-up (six months after the end of therapy). Differences were noted between the pre-intervention and post-intervention results. All participants demonstrated an increase in total AHA score by more than four points, i.e. a clinically relevant change (Participant 1 improved by seven points, Participant 2 by five points, Participant 3 by nine points, Participant 4 by five points, Participant 5 by 13 points). Most participants showed an improvement at follow-up compared to baseline.

No clinically relevant differences in total AHA score were noted between post-intervention and follow-up. The outcomes increased by one point in two participants (Participant 1 and Participant 3), decreased by two points in one participant (Participant 4) and decreased by three points in the other two participants (Participant 2 and Participant 5).

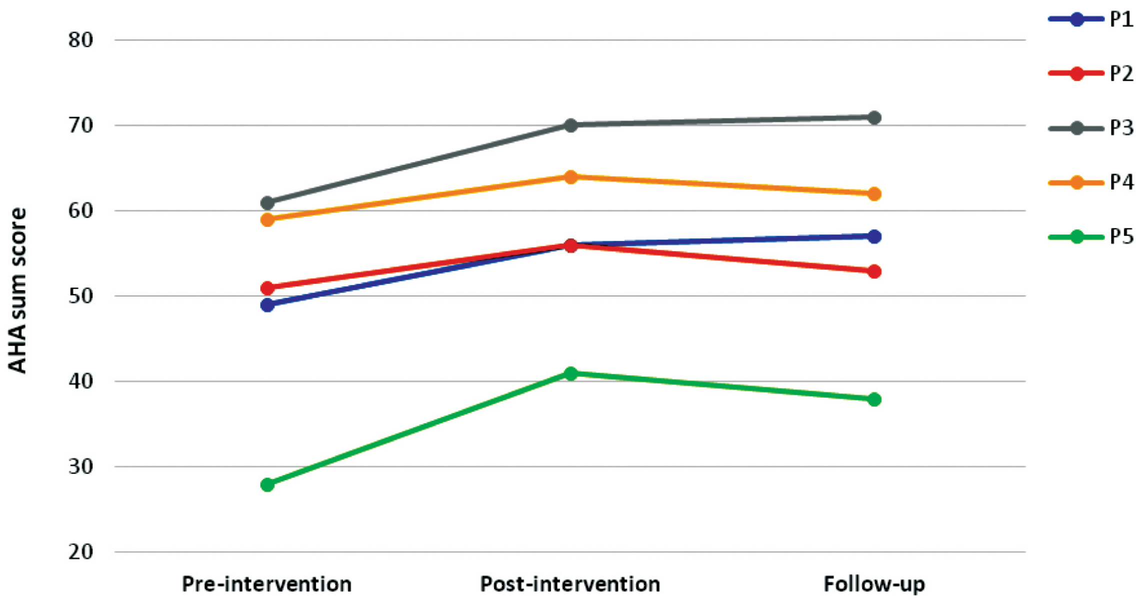

Figure 1. Results of the Small Kids-AHA test (sum score) at different stages of the study according to participant

Note: P1 – participant 1; P2 – participant 2; P3 – participant 3; P4 – participant 4; P5 – participant 5.

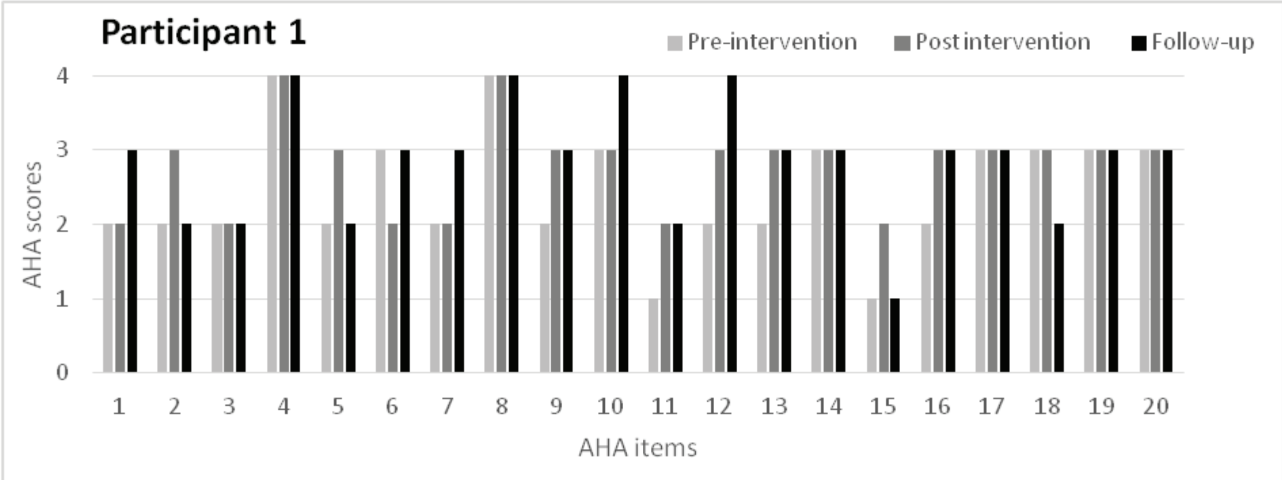

Figure 2. The Small Kids-AHA test item scores (1–20) for Participant 1 before intervention (pre), after two weeks of intervention (post) and at six months after the intervention (follow-up)

Note: 1 – initiates use; 2 – amount of use; 3 – chooses assisting hand when closer to objects; 4 – stabilizes by weight or support; 5 – reaches; 6 – moves upper arm; 7 – moves forearm; 8 – holds; 9 – grasps; 10 – stabilizes by grasp; 11 – varies type of grasp; 12 – releases; 13 – moves fingers; 14 – grip force regulation; 15 – manipulates; 16 – readjusts grasp; 17 – coordinates; 18 – orients objects; 19 – proceeds; 20 – flow in bimanual task performance.

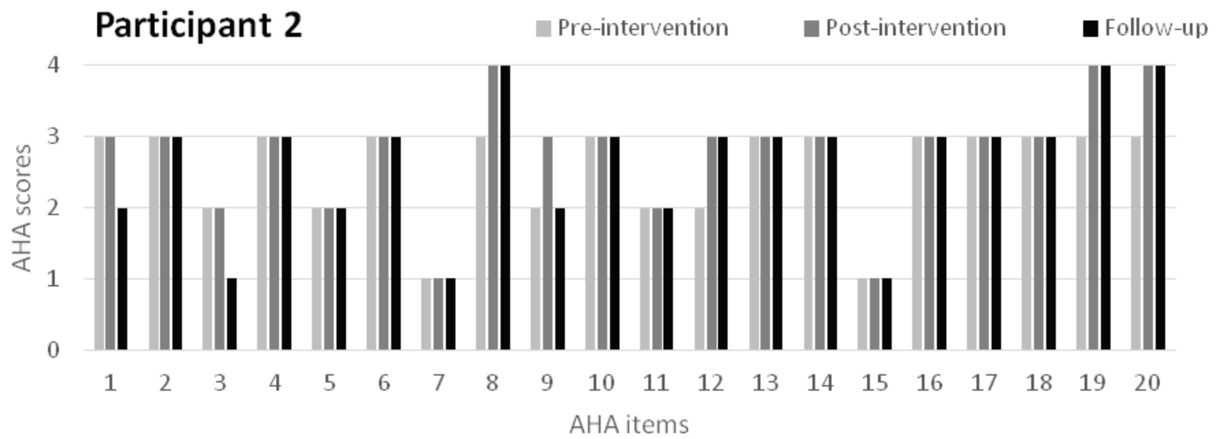

Figure 3. The Small Kids-AHA test item scores (1–20) for Participant 2 before intervention (pre), after two weeks of intervention (post) and at six months after the intervention (follow-up)

Note: 1 – initiates use; 2 – amount of use; 3 – chooses assisting hand when closer to objects; 4 – stabilizes by weight or support; 5 – reaches; 6 – moves upper arm; 7 – moves forearm; 8 – holds; 9 – grasps; 10 – stabilizes by grasp; 11 – varies type of grasp; 12 – releases; 13 – moves fingers; 14 – grip force regulation; 15 – manipulates; 16 – readjusts grasp; 17 – coordinates; 18 – orients objects; 19 – proceeds; 20 – flow in bimanual task performance

Participant 1 presented improvements and sustained effect for four items (9, 11, 13, 16) of the Small Kids-AHA; no effect was observed for seven items (3, 4, 8, 14, 17, 19, 20) (Figure 2). The score fell from end of intervention to follow-up in four items (2, 5, 15, 18), while an improvement in follow-up was noted for five items (1, 6, 7, 10, 12).

Participant 2 presented improvements and sustained effects for four items (8, 12, 19, 20) of the Small Kids-AHA; no effect was observed for thirteen items (2, 4, 5, 6, 7, 10, 11, 13, 14, 15, 16, 17, 18) (Figure 3). The score fell from the end of the intervention to follow-up in three items (1, 3, 9), and no improvement in follow-up was noted for any items.

Participant 3 presented improvements and sustained effects for seven items (1, 3, 6, 9, 10, 12, 14) of the Small Kids-AHA; no effect was observed for ten items (2, 4, 8, 11, 13, 15, 16, 17, 19, 20) (Figure 4). The score fell from end of intervention to follow-up in one item (5), but rose in two items (7, 18).

Participant 4 presented improvements and sustained effects for four items (10, 11, 15, 16) of the Small Kids-AHA, no effect was observed for fourteen items (1, 2, 3, 4, 5, 7, 8, 9, 12, 13, 17, 18, 19, 20) (Figure 5). The score fell from end of intervention to follow-up in two items (6, 14), and no increase was noted in any items.

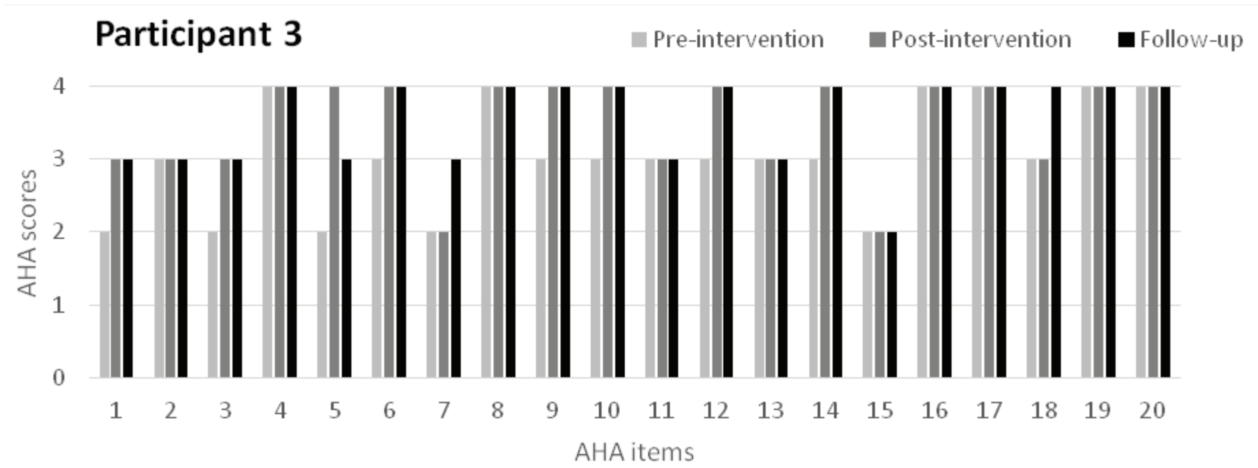

Figure 4. The Small Kids-AHA test item scores (1–20) for Participant 3 before intervention (pre), after two weeks of intervention (post) and at six months after the intervention (follow-up)

Note: 1 – initiates use; 2 – amount of use; 3 – chooses assisting hand when closer to objects; 4 – stabilizes by weight or support; 5 – reaches; 6 – moves upper arm; 7 – moves forearm; 8 – holds; 9 – grasps; 10 – stabilizes by grasp; 11 – varies type of grasp; 12 – releases; 13 – moves fingers; 14 – grip force regulation; 15 – manipulates; 16 – readjusts grasp; 17 – coordinates; 18 – orients objects; 19 – proceeds; 20 – flow in bimanual task performance.

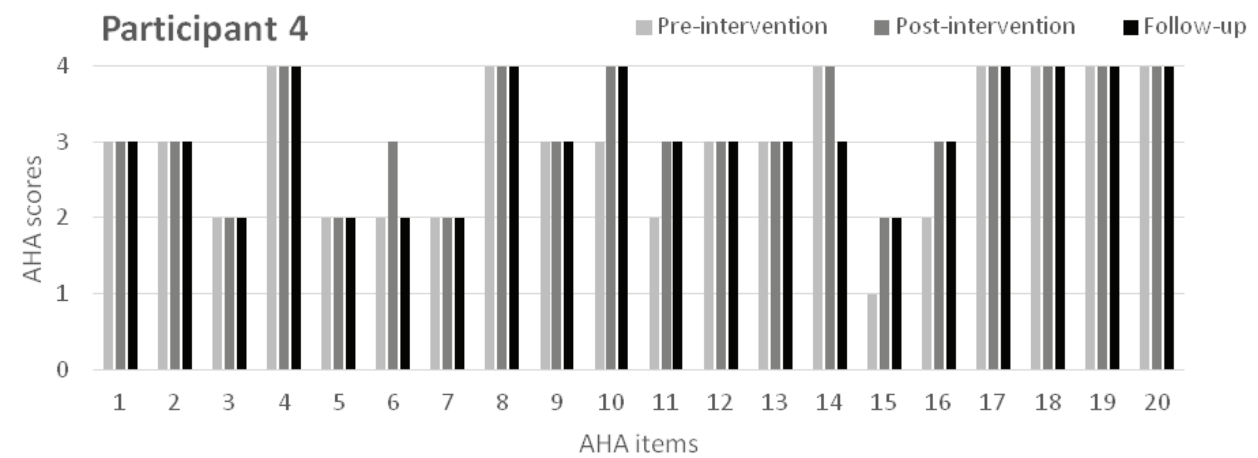

Figure 5. The Small Kids-AHA test item scores (1–20) for Participant 4 before intervention (pre), after two weeks of intervention (post) and at six months after the intervention (follow-up)

Note: 1 – initiates use; 2 – amount of use; 3 – chooses assisting hand when closer to objects; 4 – stabilizes by weight or support; 5 – reaches; 6 – moves upper arm; 7 – moves forearm; 8 – holds; 9 – grasps; 10 – stabilizes by grasp; 11 – varies type of grasp; 12 – releases; 13 – moves fingers; 14 – grip force regulation; 15 – manipulates; 16 – readjusts grasp; 17 – coordinates; 18 – orients objects; 19 – proceeds; 20 – flow in bimanual task performance.

Participant 5 presented improvements and sustained effects for six items (1, 8, 10, 14, 17, 19) of the Small Kids-AHA; no effect was observed for seven items (3, 4, 6, 13, 15, 18, 20) (Figure 6). The score fell from end of intervention to follow-up in five items (2, 5, 7, 9, 11), but rose in two items (12, 16).

Discussion

Studies have confirmed that the combination of modified constraint-induced movement therapy with bimanual training (mCIMT-BIT) is an effective form of therapy [2, 5, 6, 12, 24, 42]. Recent research by Pawlak [38] found that mCIMT-BIT therapy conducted as part of “The Pirate Group” program is an effective way of improving the spontaneous activity of the upper limb involved in bimanual tasks among children with hemiplegia. These results are consistent with those of foreign studies [2, 12, 42]. However, what distinguishes the approach taken by “The Pirate Group” program from previous schemes is that a significant improvement was noted in the activity of the upper limb of the examined children after only a relatively short total length of therapy (20 hours over two weeks).

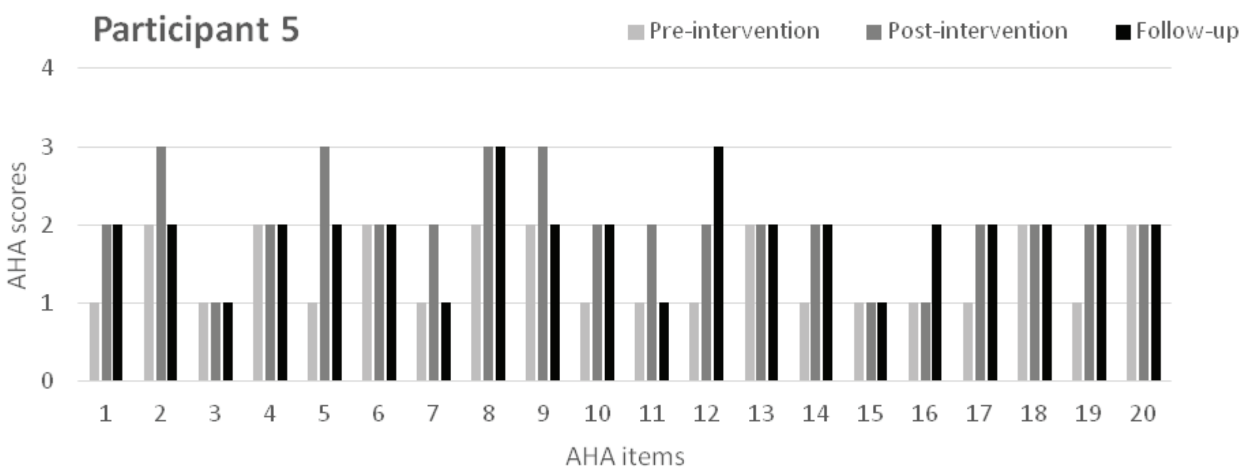

Figure 6. The Small Kids-AHA test item scores (1–20) for Participant 5 before intervention (pre), after two weeks of intervention (post) and six months after the intervention (follow-up)

Note: 1 – initiates use; 2 – amount of use; 3 – chooses assisting hand when closer to objects; 4 – stabilizes by weight or support; 5 – reaches; 6 – moves upper arm; 7 – moves forearm; 8 – holds; 9 – grasps; 10 – stabilizes by grasp; 11 – varies type of grasp; 12 – releases; 13 – moves fingers; 14 – grip force regulation; 15 – manipulates; 16 – readjusts grasp; 17 – coordinates; 18 – orients objects; 19 – proceeds; 20 – flow in bimanual task performance.

To confirm whether this length was sufficient to yield long-term effects, the present study also followed the participants over a six-month follow-up period. Our findings confirm that the mCIMT-BIT therapy carried out as part of "The Pirate Group" program is an effective therapeutic intervention. Also, the follow-up results taken six months after the end of the therapy are promising.

Play has a key role in pediatric occupational therapy, and particularly in mCIMT therapy. It is an integral part of childhood, and one that significantly affects physical, mental, emotional and social development. Through play, children learn about the world and develop their interests [3, 47]. However, during regular therapy, verbal or "physical" cues are constantly repeated to encourage children to complete the task correctly. This external stimulation is frustrating for both the child and the therapist and often results in failure or incomplete completion of the task. Also, generally speaking, mCIMT-BIT therapy is an intensive, long-term intervention that requires focus and involvement from the child. It also requires the more efficient hand to be restricted, which may cause discomfort, anxiety, rebellion and frustration [17, 32, 31].

In contrast, while playing, thanks to certain adaptations, children forget about their limitations and become fully involved in the activity [2, 47]. "The Pirate Group" program was designed in such a way that the entire therapy was embedded in the convention of playing, not only with the therapist, but also with other children; such approaches have been found to increase the effectiveness of the therapy [1, 19, 41]. The arrangement of the environment, i.e. a pirate ship, pirate costumes (for children and therapists), the decoration of the room (an old trunk, balloons, ropes, plush parrots, monkeys), props (a pirate flag, a map, mugs, toys imitating swords, guns) were not without significance. When passing through the door into the room, the children set off on a thrilling journey, forgetting about the difficulties associated with everyday rehabilitation. As noted by other researchers, the arranged environment has a positive impact on commitment and motivation, and thus on the results of therapy [2, 19, 43].

Another important factor in favor of "The Pirate Group" approach is the fact that the children work in a group with their peers. Playing in pairs requires cooperation, introduces an element of healthy competition, but also gives a sense of support, security and happiness, which motivates children to participate in therapy [1, 2, 9, 17, 25, 30]. Furthermore, as the children work in a small group with a physiotherapist assigned to each participant, the therapists are able to adjust the level of difficulty of each task to the skills of a specific child, irrespective of their personal teamwork ability and level of functioning. Working in "The Pirate Group" provides children with many challenges and difficult problems to solve, but the tasks are presented in such a way that the children have fun, willingly repeat activities and are absorbed in playing with their peers.

Furthermore, our own observations, and those of other authors, indicate that parents are also more involved in play-based therapy [2, 17]. Showing parents that mCIMT-BIT therapy can be fun inspires them to use it at home. It has also been found that play therapy can also alleviate parental stress [31].

It is important to emphasize the uniqueness of the protocol used in "The Pirate Group" program: unlike previous approaches, in which a block of mCIMT therapy lasting a few days or weeks is followed by BIT training, the present approach consists of a short block of mCIMT therapy followed by one of BIT therapy every day. This is the only example of such an approach to mCIMT-BIT therapy to be published to date.

Although evidence-based therapies are based on scientific reports, the feasibility of therapies is often limited by reality and diversity of patient's cases. The challenge for therapists is to conduct therapy that is effective and targeted at the individual needs of the child, while also being interesting and motivating for the child to participate. In this regard, the described combination of mCIMT with BIT carried out as part of "The Pirate Group" program deserves special attention. Such an approach, grounded in the convention of play, with its arranged environment, group work, unique protocol, short therapy time and relatively short immobilization of the more efficient upper limb, makes the therapy friendly to both the children and their families. However, it should be borne in mind that the data was acquired from a small group of participants, thus have should be treated as a pilot study, and further studies are needed. Nevertheless, these results confirm the authors' belief in the uniqueness of "The Pirate Group" program.

Conclusions

"The Pirate Group" program was found to achieve very good results and with ongoing effects after therapy. We therefore encourage all physioterapists and occupational therapists in rehabilitation centers to create similar new thematic programs embedded in the convention of fun. However, it should be remembered that the selected intervention should not only provide a challenge and strive to achieve goals, but it should also give a sense of success, provide motivation and be family friendly.

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