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Evaluation of effectiveness of modified pediatric Constraint-induced movement therapy on unilateral cerebral palsy by Remort Intervention

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ABSTRACT: Pediatric Constraint-Induced Movement Therapy was conducted for two weeks through online sessions to evaluate the effectiveness of remote therapy to avoid exhaustion of mothers from commuting their children with cerebral palsy (CP). Participants were six patients with unilateral CP, to whom a series of home practices were managed and performed by mothers on behalf of therapists, including cast restraint of the non-paralyzed upper extremity, motor activity log (MAL) administration, and other relevant home practices. Peabody Developmental Motor Scales (PDMS, the second edition), Pediatric Evaluation of Disability Inventory (PEDI), and Pediatric Motor Activity Log (PMAL) were employed as evaluation indexes to assess the effectiveness of pre- and post-intervention and in the follow-up evaluation one month after the intervention. As a result, all participants completed the program. Results indicated that the average duration of fiberglass cast fitting was 4.23 hours per day. Furthermore, PDMS, PEDI, and PMAL scores improved significantly during the intervention. In addition, intervention effects endured until the follow-up evaluation one month after the intervention, except for the PEDI Functional Skills (PEDI-FS) score. As previously mentioned, the study's findings confirm the effectiveness of Pediatric Constraint-Induced Movement Therapy (P-CIMT) when performed remotely.

KEYWORDS: Cerebral palsy; Occupational therapy; Spastic hemiplegia.

1. INTRODUCTION

Cerebral palsy (CP) is defined as a no progressive motor impairment caused by brain injury inflicted at a very early stage of human development [1]. Several types of CP are confirmed, among which is the prevalence of unilateral CP, which stands out in CP cases of term infants [2]. Children with unilateral CP exhibit various types of motor impairment. A study reported that they exhibit muscle weakness, contracture, and lack of selective kinetic control abilities, indicating dysfunctions more severe in the upper extremities than in other parts [3]. These dysfunctions inhibit sound motor development at the upper extremity, which probably would negatively impact children with unilateral CP in developing self-sustainability in terms of their activities of daily living (ADL) [4][5]. Most of the children with unilateral CP are barely capable of using the paralyzed upper extremity or hand. Thus, they will quickly learn to perform most activities by using only the non-paralyzed (or less-paralyzed) upper extremity or hand (developmental disuse) [6]. This tendency of developmental disuse is problematic because it possibly leads to secondary dysfunction, including soft tissue shortening and limitation in the range of joint motion. Taub et al. of the University of Alabama developed a therapeutic intervention applicable to children with unilateral CP, Pediatric Constraint-Induced Movement Therapy (P-CIMT), in 2004 based on the therapeutic intervention for adult patients with hemiplegia, Constraint-Induced Movement Therapy (CIMT) [7][8]. Several reviews have already been published regarding the effectiveness of P-CIMT [9] [10] [11] [12] [13] [14]. In this study, the following three specific P-CIMT protocols were employed: 1) Participants should be fitted with a fiberglass cast at the non-paralyzed upper extremity for two weeks (Constraint); 2) Therapists and mothers should attend face-to-face interview sessions daily to confirm that children use their paralyzed hands (MAL administration); and 3) Allocate daily home practice (HP). Numerous studies have reported depression or exhaustion in mothers of children with CP, demonstrating that daily commutation to and from care facilities

would increase mothers' stress. Consequently, it is not easy to complete the P-CIMT program since not many papers have been published internationally. In this study, monitoring the use of paralyzed hands was conducted remotely for a short duration to resolve the challenge. Moreover, the modified Pediatric Constraint-Induced Movement Therapy（m-P-CIMT) with protocols that aimed to reduce the mother's burden, where children with unilateral CP performed concentrated daily practices at home with their mothers without visiting clinics, was conducted to evaluate its effectiveness.

2. METHODS

The study was conducted under the approval of the Research Ethics Committee of the Tokyo Metropolitan Government Bureau of Public Health, with informed consent obtained from participants and their guardians. Participants were recruited via social networking services. Adaptive criteria were set as follows: Children aged 6–8 years who were diagnosed with unilateral CP; those who were categorized in levels II or III in the Manual Ability Classification System (MACS) (Eliasson et al., 2006) scores; those who exhibited evident paralysis in the paralyzed upper extremity; and the family's willingness to participate in the intervention to become capable of controlling the paralyzed arms. Exclusion criteria were set as follows: Those with medical problems unassociated with CP; severe muscle tone (Modified Ashworth score > 3.5); those who underwent botulinum toxin therapy in the upper extremity within the past six months or with scheduled treatment within the study period; and those with balance problems precluding fiberglass cast fitting. Six participants deemed eligible for the study received a detailed explanation regarding the m-P-CIMT program from the first author of this paper and participated in the program after providing their consent in writing for program participation. This study was conducted following the ethical principles of the Declaration of Helsinki, and with the approval of the Ethics Committee of the Syonan university of medical science (Approval number 24008), with informed consent obtained from participants and their guardians.

Evaluation

All participants were instructed to visit our rehabilitation center one month before, one day before, on the program's final day, and one month after the program for evaluation. The second author of this paper is a physical therapist who completed a pediatric CIMT Training course at the University of Alabama and evaluated the participants' overall bodily function and usage frequency and movement quality of their paralyzed upper extremity and hand. Two subscales in the Peabody Developmental Motor Scales, 2nd edition (PDMS) [18] were employed for physical function evaluation, which was the Grasping (PDMS-G; 26-item, total scores of 52 points) used for grasping motion ability assessment and the Visual-Motor Integration (PDMS-V; 72-item, total scores of 144 points) used for visual-motor integration ability evaluation, respectively. These two subscales consist of 98 items, all of which are evaluated by a three-point scale as follows: 0 points: Incapable of or unwilling to move; 1 point: Capable of moving but not at a standard level of movement; and 2 points: Capable of movement at a standard level of movement. The evaluation using the Pediatric Evaluation of Disability Inventory（PEDI) was also conducted to assess ALD function assessment of children with CP [19]. PEDI is a scale that determines the level of independence in ADLs. The Functional Skills Scales (FS) were designed to sample meaningful sub-tasks of a set of complex functional activities. The Caregiver Assistance Scale (CA) is a measure of the extent of help the caregiver provides in typical daily situations. Part I evaluates the functional abilities of the child in the areas of self-care (73 items), mobility (59 items), and social function (65 items), with a score of 1: when the child performs the assessed item and of 0: when the child cannot perform it. Part II is related to the need for help provided by the caregiver for the performance of 20 items in the same areas evaluated in the first part, self-care, mobility, and social function, with the following scores: 5–Independent, 4–Supervision, 3–Minimum assistance, 2–Moderate assistance, 1–Maximum assistance and 0–Total assistance. In addition, a series of structured interviews were conducted to confirm how often participants used the paralyzed upper extremity (How Often scale: HO), as well as how well they performed these tasks (How Well scale: HW) in terms of 22 items of daily activities conducted outside of the clinic, by using the Pediatric Motor Activity Log（PMAL) [20]. A six-point scale ranging from 0 to 5 was used to give a score to each item. The higher the HO scores, the more often the participant used a paralyzed hand, and higher HW scores led to better movement quality. The total score of each scale was calculated using the item's mean scores.

Intervention

All six participants were elementary school students, and the intervention was conducted for 15 consecutive days during spring break. Program protocols employed in this study included 1) Participants should be fitted with a fiberglass cast at the non-paralyzed upper extremity for two weeks (Constraint); 2) Mothers should confirm daily that the children use their paralyzed hands (MAL administration), and 3) Allocate daily home practice (HP). Two fiberglass casts were prepared and used alternately to keep them clean by washing the ones not in use. Participants' favorite Japanese animation hero characters were illustrated on these casts so that the mothers could

encourage children to put them on by talking about hero characters, such as, "Let's put on this cool weapon, just like your hero!" Mothers were also instructed to allow their children to put off the cast if they did not want to wear it and record the time they did not wear casts. Regarding item 2, PMAL administration, mothers were instructed to record only the HW scale in PMAL so the daily score increase would boost the mother's motivation to encourage children to use paralyzed hands. Regarding item 3 concerning HPs, mothers and participants were told to perform several ADL-related tasks and various types of toy play selected from the Bank of Shaping, which was developed at the University of Alabama, at home, having fun without pressure. The first author participated in daily online meetings with the mothers of six participants using an internet meeting system provided by Zoom Video Communications, Inc. to confirm 1) Cast wearing duration, 2) HW scale score in MAL, and 3) HP performance status, and answered questions or requests for consultation from mothers. Participants wore fiberglass casts from the first to the 13th day. On the 14th and 15th days, they practiced with their mothers to use both hands at home without wearing fiberglass casts. All six participants and their mothers visited the medical center together one month before the intervention program to receive an explanation of the m-P-CIMT program outlines and precautions from the first author.

Data Analysis

A matched pair test of Wilcoxon was employed for the statistical analysis of data obtained a day before and on the program's final day to clarify changes in scales. In addition, it was used to compare results obtained immediately after the program completion and those obtained in the follow-up evaluation to confirm if the intervention effects would be sustained for a month. In addition, Cohen's formula was employed to determine effect sizes, which were set as follows: 0.8 and above scores were deemed "large effect," 0.5 and above were deemed "intermediate effect," and 0.2 and above were deemed "small effect," respectively [21].

Table 1 clinical characteristics at baseline (n = 6).

Child	Affected Side	Gender	Age, Yr:Mo	MACS
1	Right	Male	6.8	Ⅱ
2	Right	Female	6.4	II
3	Right	Male	7.2	III
4	Left	Male	8.6	III
5	Left	Female	8.7	Ⅱ
6	Left	Female	4:2	II

3. RESULTS

All six participants (three boys and three girls, mean age 7.54 years old, standard deviation=1.05) completed the m-P-CIMT program, including all relevant protocol practices and evaluation sessions. No one withdrew from the program. Table 1 shows the participants' baseline data. The average duration in which participants wore the fiberglass cast during the awakening time was 4.23 hours (5.1 hours was the longest duration, and 3.1 hours was the shortest).

Table 2 indicates the score fluctuation in PDMS, PEDI, and PMAL obtained at pre-, post-, and follow-up of the intervention. PDMS-G to evaluate the skill activities of paralyzed upper extremities showed significant improvements during the intervention, exhibiting p=0.01 in comparison between pre-and post-intervention, along with the score of 0.84 in Cohen's d at p=0.01. In contrast, no significant difference was observed between post-intervention and one-month follow-up evaluation. The PDMS-V results showed a score of 0.84 in Cohen's d at p=0.01 in comparison between pre-and post-intervention, demonstrating a significant improvement during the intervention. In contrast, no significant difference was observed between post-intervention and one-month follow-up evaluation. Regarding the PEDI self-care items, the participant's functional skills (FS) exhibited intermediate improvement, indicating a score of 0.39 in Cohen's d at p=0.01. However, a significant difference was observed between post-intervention and one-month follow-up evaluation, showing that the effect did not endure. The caregiver assistance scale (CAS) score exhibited a substantial improvement in comparison between pre-and post-intervention, showing a score of 1.66 in Cohen's d at p=0.01. In contrast, no significant improvement was observed between post-and one month after the intervention. In addition, the HO scale in PMAL showed an improvement with a score of 1.79 in Cohen's d at p=0.02 in comparison between pre-and post-intervention. No significant difference was confirmed between post- and one month after the intervention. The HW scale showed a significant improvement with a score of 1.66 in Cohen's d at p=0.02 in comparison between pre and post-intervention. At the same time, the comparison between post- and one month after the intervention did not indicate a significant difference.

4. DISCUSSIONS

In this study, six children with unilateral CP participated in the m-P-CIMT program at home through online sessions to evaluate the effect of a remote intervention aimed at avoiding stress due to commutation to and from medical facilities. All participants successfully completed the program with protocols in line with the original protocols. A study in the business field reported that remote work would increase workers' productivity and enhance their work-life balance [22]. A support style introduced in this study to implement programs at home should be a promising option that can be widely applicable to the CIMT practices aimed at habituating subjects to use their paralyzed upper extremities in ADLs. The average duration of wearing the fiberglass cast during the awakening time was 4.23 hours, suggesting that the cast-wearing duration was relatively more extended in this study than in previous studies, one of which reported that the participants did not wear casts at home [23]. Illustrations of Japanese animation hero characters were printed on these casts. Several studies examined the intervention effect of hero modeling. They reported that introducing the topic of hero characters to encourage children to conduct a specific action was an effective method for altering children's behavior [24][25].

Table 2.
PRMS measurement results(n=6)

Outcome Measure		Pre	Post	*p*	Cohen's d Pre-Post	1-Mo Follow-Up
PDMS	Grasping	38.70 (1.72)	40.25(2.13)	0.01	0.61	43.29 (5.91)
	Visual	87.65(10.61)	101.17(13.98)	0.01	0.84	106.43 (20.93)
PEDI	FS	49.42(2.98)	52.70(3.16)	0.01	0.39	54.89 (7.18)
	CA	51.08 2.22)	58.85(2.61)	0.01	1.66	60.66 (8.85)
PMAL	HO	0.56(0.30)	2.10(0.33)	0.02	1.79	1.81(0.35)
	HW	2.15(0.45)	2.55(0.27)	0.01	0.98	2.42(0.31)

Note: Values indicated in pre-, post-, and 1-Mo Follow-up are average scores (standard deviations). Grasping tasks were evaluated using the Peabody Developmental Motor Scales (PDMS, 2nd edition) and its sub-scale, PDMS-G. PDMS-V is another PDMS sub-scale integrating evaluation of visual and physical movements. A subscale of PEDI called PEDI-FS is used to assess functional skills. PEDI-CAS is another sub-scale of PEDI to evaluate CAS.

Thus, the hero character on the fiberglass cast most likely contributed to the success of the present study. PDMS, PEDI, and PMAL results exhibited significant improvements during the intervention. In addition, all scales except for the PEDI-FS indicated the duration of intervention effects up to the one-month follow-up of the intervention, suggesting a possibility that the attempts to use the paralyzed upper extremities successfully led to the habituation because the practice was not conducted at medical facilities but at home, where the practice could easily be associated with ADLs. Moreover, Page et al. reported that therapists often struggle to organize practice tasks that participants feel attractive [26]. In this study, participants' mothers, who know their children's preferences in plays and daily activities better than anyone else, assisted them in using paralyzed upper extremities instead of therapists. Mothers' assistance at home would likely achieve the same degree of benefits as task practices with therapist assistance.

Study Limitations

The study did not have a control group, and the sample size was too small to provide a sufficient level of data to test the efficacy of this study. Further comparative investigation should be mandatory with an increased sample size and a control group without the knowledge of CIMT intervention.

5. CONCLUSIONS

The modified-pediatric-CIMT was conducted on six participants with unilateral CP capable of voluntary movement of paralyzed upper extremities. The m-P-CIMT was implemented remotely at home through online sessions with therapists. The intervention effects were evaluated using PDMS, PEDI, and PMAL as evaluation indexes. Protocols included wearing the fiberglass casts on non-paralyzed upper extremities of the participants, MAL administration by mothers, and implementation of home practices with mothers. All participants completed the program by finishing all protocols and evaluation sessions. Scores of PDMS, PEDI, and PMAL exhibited significant improvements during the intervention, and the effect endured up to the one-month follow-up after the intervention, except for the PEDI-FS. The results suggest that the remotely implemented P-CIMT can achieve the same effects as the P-CIMT conducted at medical facilities with one-on-one sessions with therapists. The authors

declare no conflicts of interest associated with this manuscript. The authors have no conflicts of interest directly relevant to this article's content.

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