Observational Study

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Does well-functioning hand constraint induce stress in forced-use therapy for children with unilateral cerebral palsy?

Eun Jae Ko, MD, PhD[a], In Young Sung, MD, PhD[b,*], Jin Sook Yuk, MPH, OT[c]

Abstract

This study investigated the stress induced by well-functioning hand constraint in forced-use therapy (FUT) for children with unilateral spastic cerebral palsy (CP).

Seventeen children with unilateral spastic CP (mean age 5.8 years) received FUT: 4-week unaffected upper limb immobilization with a short-arm Scotchcast and were encouraged to incorporate it to their daily routines and plays. They were evaluated at pretreatment, immediate post-treatment, and 6 months post-treatment. The Korea-Child Behavior Checklist (K-CBCL) was used to assess the stress degree; box and block test (BBT), Erhardt Developmental Prehension Assessment (EDPA), Quality of Upper Extremity Skill Test (QUEST), and Pediatric Motor Activity Log (PMAL), upper limb function; and Pediatric Evaluation of Disability Inventory (PEDI), daily living activities.

In the preschoolers, most scores of K-CBCL tended to increase after FUT; however, there was no significant change in all scale findings after FUT. In the school-aged children, most scores of K-CBCL tended to decrease after FUT; however, there was no significant change in all scale findings after FUT. The findings of the BBT, QUEST, PMAL how often and well subscales significantly improved post-treatment ($P < .05$).

The 4-week FUT with well-functioning hand constraint significantly improved the UL function and did not induce emotional and behavioral problems in children with unilateral spastic CP.

Abbreviations: ADL = activities of daily living, BBT = box and block test, cCIMT = classic constraint-induced movement therapy, CIMT = constraint-induced movement therapy, CP = cerebral palsy, EDPA = Erhardt Developmental Prehension Assessment, FDR = false discovery rate, FUT = forced use therapy, GMFCS = Gross Motor Function Classification System, K-CBCL = Korea-Child Behavior Checklist, mCIMT = modified constraint-induced movement therapy, OT = Occupational therapy, PEDI = Pediatric Evaluation of Disability Inventory, PMAL = Pediatric Motor Activity Log, QUEST = Quality of Upper Extremity Skill Test, UL = Upper limb.

Keywords: cerebral palsy, children, emotions, psychological, rehabilitation, stress

1. Introduction

Cerebral palsy (CP) is a non-progressive motor impairment syndrome caused by developing brain problems; affected patients have motor and balance function disabilities, asymmetry, and activities of daily living (ADL) performance limitations.[1] Many therapies can improve upper limb (UL) function in children with unilateral spastic CP: constraint-induced movement therapy (CIMT), hand-arm intensive bimanual training, neurodevelopmental treatment, and intramuscular botulinum toxin A injection, augmenting occupational therapy (OT).[2]

CIMT stimulates the affected arm by restraining the unaffected UL; it has many models. Classic CIMT (cCIMT) involves putting a full-arm cast on the unaffected UL for at least 2 weeks, with intensive training of the more-affected UL for at least 3 hours each day.[3] Modified CIMT (mCIMT) differs from cCIMT in the unaffected UL restraining type (e.g., sling, cast, mitt, and glove), structured training type, program duration (hours per day) and length (number of weeks), and training location, context, and provider (home/camp, individual/group, and therapist/parent).[4] Hybrid CIMT involves mCIMT followed by bimanual task-specific training.[5,6] Forced use therapy (FUT) restrains the unaffected UL and encourages the use of the affected UL in ADL performance at home without additional specific structured training programs.[7] Many studies have shown the effect of CIMT; CIMT modestly or strongly improves the movement quality and efficiency of the affected UL compared with usual care.[8]

However, the unaffected UL motion should be restricted in CIMT using casts, slings, mitts, or gloves. CIMT did not result in hand function decline of the unaffected UL, which is constrained therein;[7] however, there were some reported side effects,

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including skin redness, rash, or pinching[3] and increased irritability,[9,10] which led to CIMT withdrawal. Furthermore, we have been concerned that it may cause stress and psycho-emotional problems in children owing to restriction of the freely moving well-functioning UL. Nevertheless, no data have been reported on such to date. This study aimed to evaluate stress related to FUT in children with unilateral spastic CP by assessing their emotional and behavioral problems.

2. Materials and methods

2.1. Study design and participants

This study was approved by the Ethical Committee of Asan Medical Center (reference number: 2016–0045), and the trial has been registered at Clinical Research Information Service (ref number: KCT0003454). Written informed consent was obtained before data collection.

Children who visited the outpatient clinic of the Pediatric Rehabilitation Medicine Division at Asan Medical Center from September 2011 to July 2016 were assessed for inclusion herein according to the following criteria:

1. unilateral spastic CP diagnosed by a pediatric physiatrist;
2. no severe UL paralysis that could affect performance of ADLs or plays;
3. no cognitive dysfunction rendering them unable to cooperate during testing or therapy;
4. ability to walk independently;
5. stable medical conditions; and
6. written informed consent provided by caregivers.

2.2. Intervention

For FUT, the unaffected UL of the participants was immobilized with a short-arm Scotchcast[7] for 4 weeks. The Scotchcast was applied from below the elbow to the fingertips by a trained occupational therapist. It was changed every week to determine if any complication, including abrasion or pressure ulcer, occurred. During 4 weeks, the children also received the specific structured OT program for the affected UL for 30 minutes, 1 session a week, which was an unchanged and regularly scheduled rehabilitation program. Functional OT was provided with concrete therapeutic goal setting. Stretching exercise of the affected UL and tasks of reaching, grasping, holding, and manipulating an object; making hand gestures; and weight bearing on the arm were practiced. They also had training on ADLs and plays, which included eating, grooming, dressing, and using the toilet. After structured functional OT, the children were encouraged to use their affected UL during ADLs at home with a short-arm Scotchcast on the unaffected UL. The main caregivers were instructed to encourage the children to use their affected UL more frequently at home and during play routines.

2.3. Outcome measurements

The children were evaluated by experienced occupational therapists at pretreatment, immediate post-treatment, and 6 months post-treatment. These 2 fixed occupational therapists involved in the evaluations were not the same occupational therapists involved in the delivery of the intervention program.

The Korea-Child Behavior Checklist (K-CBCL)[11,12] was used to assess the stress degree by evaluating emotions and behaviors. The primary outcomes were the findings of the broad-band behavior scales for the total, internalizing, and externalizing problems; the secondary outcomes were those of 7 or 8 narrow-band syndrome scales. The K-CBCL is a widely used caregiver report form identifying behavioral problems in children. It has 2 different versions, depending on the childrens age. The preschool version (K-CBCL/1½-5) contains 100 behavioral problem questions (rated 0–3) for children aged 1.5–5 years. These items provide scores for 7 narrow-band syndrome scales (emotional reactiveness, anxious/depressed, somatic complaints, withdrawn, sleep problems, attention problems, and aggressive behavior) and 3 broad-band behavior scales (internalizing, externalizing, and total behavioral problems). The school-age version (K-CBCL/6–18) contains 119 behavioral problem questions (rated 0–3) for children aged 6 to 18 years. These items provide scores for 8 narrow-band syndrome scales (anxious/depressed, withdrawn/depressed, somatic complaints, social problems, thought problems, attention problems, rule-breaking behavior, and aggressive behavior) and 3 broad-band behavior scales (internalizing, externalizing, and total behavioral problems). Higher problem scores indicate higher behavioral disturbance levels. The T-score cut-off points for the broad-band behavior scales determine the degree of deviance from normality, categorizing childrens conditions as clinical, borderline, or non-clinical.[11] Non-clinical findings indicate those within the normal range. The children with clinical and borderline findings were grouped into 1 group and those with nonclinical findings into the other group.

To assess UL function, the box and block test (BBT),[13] Erhardt Developmental Prehension Assessment (EDPA),[14] Quality of Upper Extremity Skill Test (QUEST),[15] and Pediatric Motor Activity Log (PMAL)[16] were employed; to assess ADL performance, the Pediatric Evaluation of Disability Inventory (PEDI) was used.[17] The BBT[13] was developed to assess gross manual dexterity by measuring the number of blocks shifted from 1 side to the other. The total score of the affected hand was calculated in this study. The EDPA[14] measures the functional age of the hand by observing prehensile behavior patterns during reaching, grasping, manipulating, and releasing of dowels, cubes, and pellets in children aged between 1 month and 6 years. The EDPA for the affected hand was employed in this study. The QUEST[15] is a criterion-referenced test designed to assess movement patterns, which form the basis of UL function of children aged between 18 months and 8 years. It measures performance in 4 domains: dissociated movement, grasp, weight bearing, and protective extension. It has good test-retest and inter- and intra-observer reliabilities.[18] The total score in the QUEST was used herein. The PMAL is a parent-report measure of the use of the affected UL in everyday activities in children with hemiplegic CP aged from 7 months to 8 years; it involves 22 activities and was developed as an outcome measurement tool for evaluating CIMT effectiveness in children with CP.[16] The "how often" scale measures the amount of use and the "how well" scale, the movement quality in the affected UL. The PEDI[17] measures the functional capacity and performance of children aged between 6 months and 7.5 years in 3 subscales:

1. self-care;
2. mobility; and
3. social function.

It has very good reliability and responsiveness.[19,20] Additionally, data on age, sex, Gross Motor Function Classification System (GMFCS) level,[21] and hemiplegic side (right or left) were collected.

2.4. Data analysis

Data were analyzed using SPSS for Windows version 18.0 (SPSS Inc., Chicago, IL, USA), and means and standard deviations were obtained with the threshold for statistical significance set at $P < .05$. The Wilcoxon signed rank test was used to compare the continuous variables and McNemar test to compare the categorical variables at pre-treatment, immediate post-treatment, and 6 months post-treatment. Additionally, false discovery rate (FDR)-controlling procedure (Benjamini-Hochberg procedure[22]) was used to control Type I errors, because conducting multiple comparisons could increase Type I errors. FDR-controlling procedure has greater power than other family-wise type I error controlling procedures (such as the Bonferroni correction).[23] FDR-adjusted *P* value was suggested.

3. Results

3.1. Baseline characteristics

Seventeen children were enrolled, and there was no follow-up loss. Their baseline characteristics are shown in Table 1. Their mean (SD) age was 5.8 (2.9) years, and there were 6 boys and 11 girls. They consisted of 12 children with right hemiplegic side and 5 with left hemiplegic side. All children had GMFCS level I.

3.2. Stress after forced-use therapy

The emotional and behavioral problems measured using the K-CBCL before and after FUT are shown in Tables 2 and 3. In K-CBCL/1½-5, most raw scores tended to increase after FUT; however, there was no significant change in the findings of all broad-band behavior and narrow-band syndrome scales after FUT (Table 2). In K-CBCL/6–18, externalizing behavioral problem and aggressive behavior significantly decreased after FUT when Wilcoxon signed rank test was used ($P < .05$). However, they did not show significant difference when FDR-controlling procedure was used. Furthermore, the remaining raw K-CBCL scores tended to decrease after FUT; however, they were nonsignificant (Table 2).

In the T-score analysis, the number of children with non-clinical total behavioral problems increased from 11 to 13 after FUT, and that with non-clinical externalizing behavioral problems increased from 10 to 13 after FUT (Table 3). However, these changes were not significantly different.

Table 1

Baseline characteristics of the children with cerebral palsy.

	Values
Number	17
Age (years)	5.8 (2.9)
Sex (Male: Female)	6: 11
GMFCS level (I: II: III: IV: V)	17: 0: 0: 0: 0
Hemiplegic side (Right: Left)	12: 5

Values are presented as mean (standard deviation) or numbers. GMFCS = Gross Motor Function Classification System.

Table 2

Changes in the emotional and behavioral problems of the preschoolers and school-aged children after forced-use therapy.

	(A) Pre-treatment	(B) Immediate Post-treatment	(C) 6 months post-treatment	*P* value (A-B)	*P* value (B-C)	*P* value (A-C)	FDR-adjusted p-value (A-B)	FDR-adjusted p-value (B-C)	FDR-adjusted p-value (A-C)
K-CBCL/1½-5 (n=9)									
Behavior scales									
Total behavioral problem	38.2 (19.2)	42.6 (23.6)	43.8 (28.2)	.104	.715	.553	.278	.794	.679
Internalizing behavioral problem	9.4 (7.1)	10.6 (7.9)	11.2 (8.7)	.167	.715	.611	.278	.794	.679
Externalizing behavioral problem	15.2 (7.8)	16.8 (9.9)	17.2 (11.1)	.136	.461	.446	.278	.794	.637
Syndrome scales									
Emotional reactiveness	2.9 (2.5)	3.4 (2.6)	5.5 (3.8)	.257	.267	.168	.317	.794	.475
Anxious/depressed	2.3 (1.2)	3.0 (2.3)	2.9 (2.1)	.285	.854	.257	.317	.854	.475
Somatic complaints	2.1 (3.3)	1.9 (3.1)	1.2 (2.0)	.157	.357	.276	.278	.794	.475
Withdrawn	2.1 (1.6)	2.2 (1.6)	2.7 (1.4)	.317	.285	.285	.317	.794	.475
Sleep problems	2.4 (1.8)	3.0 (2.4)	3.2 (2.9)	.285	.414	.285	.317	.794	.475
Attention problems	4.6 (2.4)	4.8 (2.4)	4.6 (2.5)	.157	.593	1.000	.278	.794	1.000
Aggressive behavior	10.7 (6.4)	12.0 (8.5)	12.7 (9.3)	.136	.715	.235	.278	.794	.475
K-CBCL/6–18 (n=8)									
Behavior scales									
Total behavioral problem	22.6 (13.4)	19.1 (9.4)	22.0 (17.5)	.058	.786	.499	.213	1.000	.784
Internalizing behavioral problem	5.4 (3.8)	4.3 (2.9)	3.8 (2.1)	.141	.713	.144	.247	1.000	.784
Externalizing behavioral problem	5.5 (4.0)	4.0 (2.8)	4.9 (5.7)	.026*	.891	.339	.213	1.000	.784
Syndrome scales									
Anxious/depressed	2.1 (1.6)	1.9 (1.6)	2.3 (2.9)	.317	.713	.713	.436	1.000	.871
Withdrawn/depressed	2.1 (1.8)	1.9 (1.6)	2.1 (2.4)	.414	1.000	1.000	.506	1.000	1.000
Somatic complaints	1.1 (2.0)	0.5 (1.1)	0.6 (1.1)	.102	.317	.180	.247	1.000	.784
Social problems	3.1 (2.6)	3.1 (2.9)	3.1 (2.4)	1.000	1.000	.914	1.000	1.000	1.000
Thought problems	0.6 (0.7)	0.8 (0.9)	1.0 (1.8)	.564	.655	.705	.620	1.000	.871
Attention problems	5.0 (3.7)	4.8 (3.5)	4.9 (3.6)	.157	.713	.492	.247	1.000	.784
Rule-breaking behavior	1.4 (1.5)	1.1 (1.4)	1.1 (1.7)	.157	1.000	.317	.247	1.000	.784
Aggressive behavior	4.1 (3.3)	2.9 (2.2)	3.8 (4.3)	.041*	1.000	.496	.213	1.000	.784

Values are presented as mean (standard deviation). K-CBCL1½-5=preschool version of the Korea-Child Behavior Checklist, K-CBCL/6–18=school-age version of the Korea-Child Behavior Checklist,.
* $p < .05$ by the Wilcoxon signed rank test.
FDR = false discovery rate.

Table 3

Number of children assessed to have non-clinical, borderline, and clinical emotional and behavioral problems after forced use therapy (n=17).

	(A) Pre-treatment		(B) Immediate Post-treatment		(C) 6 months post-treatment							
K-CBCL	Non-clinical	Clinical and borderline	Non-clinical	Clinical and borderline	Non-clinical	Clinical and borderline	*P* value (A-B)	*P* value (B-C)	*P* value (A-C)	FDR-adjusted p-value (A-B)	FDR-adjusted p-value (B-C)	FDR-adjusted p-value (A-C)
Behavior scales												
Total behavioral problem	11 (64.7)	6 (35.3)	13 (76.5)	4 (23.5)	13 (76.5)	4 (23.5)	.500	1.000	.500	.750	1.000	.938
Internalizing behavioral problem	14 (82.4)	3 (17.6)	14 (82.4)	3 (17.6)	14 (82.4)	3 (17.6)	1.000	1.000	1.000	1.000	1.000	1.000
Externalizing behavioral problem	10 (58.8)	7 (41.2)	13 (76.5)	4 (23.5)	12 (70.6)	5 (29.4)	.250	1.000	.625	.750	1.000	.938

Values are presented as numbers (%).
FDR = false discovery rate, K-CBCL = Korea-Child Behavior Checklist.

3.3. Forced-use therapy effects on upper limb function

After 4 weeks of FUT, the findings of the BBT, EDPA cube subscale, QUEST, PMAL how often and well subscales, and PEDI self-care function significantly improved when Wilcoxon signed rank test was used ($P < .05$, Table 4). However, only BBT, QUEST, PMAL how often and well subscales improved significantly when FDR-controlling procedure was used ($P < .05$). These improvements were sustained in most measurements at 6 months post-treatment. Particularly, the self-care, mobility, and social functions in the PEDI consistently improved even after the intervention ($P < .05$) either using Wilcoxon signed rank test or FDR-controlling procedure.

4. Discussion

This is the first study to discuss FUT-related stress in children with unilateral spastic CP. The 4-week FUT did not significantly increase the emotional and behavioral problems measured using the K-CBCL. In the preschoolers, most scores tended to increase after FUT; however, there was no significant change in all scale findings after FUT. In the school-aged children, the raw K-CBCL scores tended to decrease after FUT. Among them, externalizing behavioral problem and aggressive behavior significantly decreased after FUT when Wilcoxon signed rank test was used ($P < .05$), but did not show significant difference when FDR-controlling procedure was used. Since conducting multiple comparisons could increase Type I errors, the results by FDR-controlling procedure are more reliable than ones by the Wilcoxon signed rank test. In other words, there is no significant differences in the raw K-CBCL scores after FUT in the school-aged children. The tendency of better results regarding emotional and behavioral problems in the school-aged children than in the preschoolers herein are probably because the former can understand better the aim of the FUT and discomfort of restricting the unaffected UL than the latter. Further, the former might have more motivation to participate actively in their home and play routines, leading to better UL function and ADL performance. These results may have increased the self-esteem of the school-aged children, leading to mental stability. In the T-score analysis, the number of children with nonclinical total and externalizing behavioral problems moderately increased; however, these changes were not significantly different. As FUT did not increase emotional and behavioral problems, it could be concluded that FUT does not induce stress in children with unilateral spastic CP.

Table 4

Changes in hand function after forced use therapy (n=17).

	(A) Pre-treatment	(B) Immediate Post-treatment	(C) 6 months post-treatment	*P* value (A-B)	*P* value (B-C)	FDR-adjusted p-value (A-B)	FDR-adjusted *P* value (B-C)
BBT	12.9 (7.2)	16.4 (8.5)	17.4 (9.8)	.002*	.202	.005†	.474
EDPA (Dowel)	6.9 (1.7)	7.2 (1.7)	7.1 (1.7)	.102	.317	.113	.528
EDPA (Cube)	6.9 (1.7)	7.2 (1.7)	7.3 (1.7)	.025*	1.000	.050	1.000
EDPA (Pellet)	8.1 (1.9)	8.4 (1.9)	8.5 (1.9)	.059	1.000	.079	1.000
QUEST	78.4 (12.5)	79.6 (12.9)	75.6 (23.3)	.001*	.441	.005†	.474
PMAL (How often)	28.9 (19.6)	43.5 (27.9)	37.9 (23.4)	.001*	.569	.005†	.813
PMAL (How well)	33.5 (26.9)	46.1 (30.3)	45.1 (32.1)	.001*	.887	.005†	1.000
PEDI (Self-care function)	56.4 (14.1)	57.6 (13.3)	61.6 (11.1)	.041*	.001*	.068	.005†
PEDI (Mobility function)	52.6 (6.5)	52.8 (6.3)	55.2 (4.5)	.180	.003*	.180	.010†
PEDI (Social function)	51.6 (12.4)	51.9 (12.2)	56.3 (9.0)	.063	.001*	.079	.005†

Values are presented as mean (standard deviation).
* $P < .05$ by the Wilcoxon signed rank test.
† $P < .05$ by FDR-controlling procedure (Benjamini-Hochberg procedure).
BBT = box and block test, EDPA = Erhardt Developmental Prehension Assessment, FDR = false discovery rate, PEDI = Pediatric Evaluation of Disability Inventory, PMAL = Pediatric Motor Activity Log, QUEST = Quality of Upper Extremity Skills Test.

CIMT was developed by Edward Taub in 1993, and the protocol originated from basic research on monkeys.[24,25] CIMT was developed to reverse the "learned non-use," in which initial motor depression following insult results in negative experience of using the affected limb, but positive experience of using the unaffected limb. It was introduced as a promising new therapy for adults with hemiparesis consequent to stroke in the mid-1990s.[26]

Studies discussing CIMT effectiveness in children started from 2004 with various models. In the study by Taub et al,[3] cCIMT was conducted in children with hemiparesis associated with CP (aged 7–96 months). cCIMT involved putting a full-arm cast on the unaffected UL for 21 consecutive days and intensive training of the more-affected arm for 6 hours each day; it yielded major and sustained improvements in motor function. mCIMT was introduced to make it more child-friendly;[4] mCIMT in children with CP (aged 4–8 years) for 6 hours per day for 10 out of 12 consecutive days improved the movement efficiency and dexterity of the involved UL, but not strength, sensibility, or muscle tone.[4] There were also studies on hybrid CIMT. Six-week mCIMT (three 3-hour sessions per week), followed by 2-week bimanual task-specific training, in children with unilateral spastic CP (aged 2.5–8 years) improved the spontaneous use of the affected limb.[6] In another study, 2-week mCIMT (restricting the unaffected arm for 10 hours per day and training of the affected arm for 3 hours per day), followed by 1-week bimanual functional training, in children with CP (mean age, 5 years and 6 months) was effective in promoting daily living functioning. A recent meta-analysis[8] demonstrated that CIMT modestly or strongly improved the movement quality and efficiency of the affected UL compared with usual care, but showed weak treatment effects for most outcomes compared with equal bimanual OT doses. CIMT effects may then be induced by intensive treatments of 3–6 hours per day rather than the constraint itself.

Although CIMT showed good treatment effects in previous studies and was recommended as an intensive training program, it is very difficult to perform in real hospital settings. Therefore, FUT, which does not require additional daily intensive training program at the hospital, but encourages patients to use the affected UL in performing ADLs with the cast on, can be a practical and cost beneficial method in many different clinical circumstances. In a previous study,[7] children with hemiplegic CP (mean age, 33.2 months) received 6-week FUT, in which unaffected limb was restricted with a short-arm Scotchcast and regularly scheduled rehabilitation program was provided. It showed that 6-week FUT improved the affected hand function. Forced use of the affected UL is then important, and home program and daily activity participation is more important than treatment duration. In the study by Willis et al,[9] 12 children with hemiparesis (aged 1–8 years) wore a plaster cast on the unimpaired arm for 1 month and continued their routine visits for OT and physical therapy. After 1 month, the Peabody Developmental Motor Scale scores significantly improved compared with those of the control group, and these improvements persisted for 6 months. However, in the study by Eugster-Buesch et al,[27] 2-week home-based FUT did not significantly improve the UL function in children with hemiplegic CP (mean age, 9.8 years). Herein, FUT consisted of unaffected hand constraint using a forearm Soft/Scotchcast for 6 hours per day for 2 weeks; however, only 45% maintained constraint for 6 hours per day, and 55% of the main caregivers stated that the FUT program was exhausting. The lack of compliance and short treatment duration may be related to the unsatisfactory result.

In this study, the 4-week FUT improved the UL function, as evaluated using the BBT, QUEST, PMAL how often and well subscales when using FDR-controlling procedure ($P < .05$), which is consistent with previous reports.[7,9] The self-care, mobility, and social function improvements in the PEDI at 6 months post-treatment were thought to be the result of natural development over time.

Previous studies on CIMT only focused on UL function improvements, and these improvements were related to longer constraint durations. Side effects may occur owing to affected UL constraint for long durations. There were some side effects of CIMT or FUT reported in previous studies, including mild skin redness, rash, or pinching.[3] Furthermore, by restricting the unaffected UL in CIMT or FUT, ADL performance difficulties could be induced in children with hemiplegic CP. This could lead to psycho-emotional burden and stress. In the study by Willis et al,[9] some children were withdrawn from the study and removed their casts because of irritability and/or complaints regarding wearing the cast. In the study by Crocker et al,[10] 1 child undergoing FUT appeared irritated, withdrew from play activities, and often removed and hid the splint from his main caregiver, leading to study withdrawal. However, no study has discussed such problems related to CIMT or FUT to date.

Mental health disorders are of significant public health interest, especially in patients with disability. Children with CP are susceptible to mental health disorders because of various physical risk factors and factors that affect social development, including mobility restriction, communication problem, and developmental comorbidity. Whitney, Warschausky, and Peterson[28] found that patients with CP aged 6 to 17 years had a high prevalence of mental health disorders (anxiety and behavioral/conduct problems) even after accounting for physical risk factors (physical activity, sleep duration, and pain); this concludes that physical factors do not fully account for a higher mental health disorder prevalence. Since mental health is as important as the physical health in these children, the findings of this study is meaningful.

5. Limitations

In interpreting the study results, several limitations must be considered. First, a small number of children from only 1 organization were included. Second, the relationship between FUT and stress was investigated in this study; therefore, stress related to cCIMT, mCIMT, and hybrid CIMT is still unknown. Third, although PEDI was used as an instrument that evaluates independence in daily living, it has a limitation in measuring daily activities in real life. Finally, the results cannot be generalized to all children with CP because all participants had GMFCS level I. Further testing with a larger cohort is needed.

6. Conclusions

This is the first study that has investigated the stress related to well-functioning hand constraint in FUT in children with unilateral spastic CP. Four weeks of FUT did not increase emotional and behavioral problems and consequently stress. As FUT significantly improved UL function and did not induce emotional and behavioral problems, the results herein have implications for clinicians working with children with CP for using FUT safely to improve UL function.

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References

[1] Pellegrino L. Batshaw ML. Cerebral palsy. When Your Child Has a Disability: The Complete Sourcebook of Daily and Medical Care Baltimore, MD: Brookes Publishing; 2000;275–87.
[2] Sakzewski L, Ziviani J, Boyd R. Systematic review and meta-analysis of therapeutic management of upper-limb dysfunction in children with congenital hemiplegia. Pediatrics 2009;123:e1111–22.
[3] Taub E, Ramey SL, DeLuca S, et al. Efficacy of constraint-induced movement therapy for children with cerebral palsy with asymmetric motor impairment. Pediatrics 2004;113:305–12.
[4] Charles JR, Wolf SL, Schneider JA, et al. Efficacy of a child-friendly form of constraint-induced movement therapy in hemiplegic cerebral palsy: a randomized control trial. Dev Med Child Neurol 2006;48:635–42.
[5] de Brito Brandao M, Mancini MC, Vaz DV, et al. Adapted version of constraint-induced movement therapy promotes functioning in children with cerebral palsy: a randomized controlled trial. Clin Rehabil 2010;24:639–47.
[6] Aarts PB, Jongerius PH, Geerdink YA, et al. Effectiveness of modified constraint-induced movement therapy in children with unilateral spastic cerebral palsy: a randomized controlled trial. Neurorehabil Neural Repair 2010;24:509–18.
[7] Sung IY, Ryu JS, Pyun SB, et al. Efficacy of forced-use therapy in hemiplegic cerebral palsy. Arch Phys Med Rehabil 2005;86:2195–8.
[8] Sakzewski L, Ziviani J, Boyd RN. Efficacy of upper limb therapies for unilateral cerebral palsy: a meta-analysis. Pediatrics 2014;133:e175–204.
[9] Willis JK, Morello A, Davie A, et al. Forced use treatment of childhood hemiparesis. Pediatrics 2002;110(1 Pt 1):94–6.
[10] Crocker MD, MacKay-Lyons M, McDonnell E. Forced use of the upper extremity in cerebral palsy: a single-case design. Am J Occup Ther 1997;51:824–33.
[11] Oh KJ, Kim YA. Manual for the Korean version of ASEBA Preschool Forms & Profiles. Seoul: Huno Inc; 2013:15–28
[12] Oh KJ, Kim YA. Manual for the Korean version of ASEBA School-Age Forms & Profiles. Seoul: Huno Inc; 2011:13–24.
[13] Mathiowetz V, Volland G, Kashman N, et al. Adult norms for the Box and Block Test of manual dexterity. Am J Occup Ther 1985;39:386–91.
[14] Erhardt RP, Beatty PA, Hertsgaard DM. A developmental prehension assessment for handicapped children. Am J Occup Ther 1981;35:237–42.
[15] DeMatteo C, Law M, Russell DJ, et al. The reliability and validity of Quality of upper extremity skills test. Phys Occup Ther Pediatr 1993;13:1–18.
[16] Deluca SC, Echols K, Law CR, et al. Intensive pediatric constraint-induced therapy for children with cerebral palsy: randomized, controlled, crossover trial. J Child Neurol 2006;21:931–8.
[17] Haley SM, Coster WJ, Ludlow LH, et al. Pediatric evaluation of disability inventory (PEDI): Development, Standardization and Administration Manual. Boston, MA: New England Medical Center Hospitals. PEDI Research Group 1992.
[18] Haga N, van der Heijden-Maessen HC, van Hoorn JF, et al. Test-retest and inter- and intrareliability of the quality of the upper-extremity skills test in preschool-age children with cerebral palsy. Arch Phys Med Rehabil 2007;88:1686–9.
[19] Berg M, Jahnsen R, Froslie KF, et al. Reliability of the pediatric evaluation of disability inventory (PEDI). Phys Occup Ther Pediatr 2004;24:61–77.
[20] Vos-Vromans DC, Ketelaar M, Gorter JW. Responsiveness of evaluative measures for children with cerebral palsy: the gross motor function measure and the pediatric evaluation of disability inventory. Disabil Rehabil 2005;27:1245–52.
[21] Palisano R, Rosenbaum P, Walter S, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol 1997;39:214–23.
[22] Benjamini Y, Hochberg Y. Controlling the false discovery rate: a practical and powerful approach to multiple testing. J R Stat Soc Ser B (Methodological) 1995;57:289–300.
[23] Wikipedia. False discovery rate 2019 [cited 2019 Dec 17]. Available from: https://en.wikipedia.org/wiki/False_discovery_rate.
[24] Taub E. Ince LP. Somatosensory deafferentation research with monkeys: implications for rehabilitation medicine. Behavioral Psychology in Rehabilitation Medicine: Clinical Applications. Baltimore, Md: Williams & Wilkins; 1980;371–401.
[25] Taub E. Movement in nonhuman primates deprived of somatosensory feedback. Santa Barbara, CA: Journal Publishing Affiliates; 1977; 335–74.
[26] Morris DM, Crago JE, DeLuca SC, et al. Constraint-induced movement therapy for motor recovery after stroke. NeuroRehabilitation 1997;9: 29–43.
[27] Eugster-Buesch F, de Bruin ED, Boltshauser E, et al. Forced-use therapy for children with cerebral palsy in the community setting: a single-blinded randomized controlled pilot trial. J Pediatr Rehabil Med 2012;5:65–74.
[28] Whitney DG, Warschausky SA, Peterson MD. Mental health disorders and physical risk factors in children with cerebral palsy: a cross-sectional study. Dev Med Child Neurol 2019;61:579–85.