

Effects Of Constraint-Induced Movement Therapy As A Rehabilitation Strategy For The Affected Upper Limb Of Children With Hemiplegic Cerebral Palsy: Systematic Review Of The Literature

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ABSTRACT

Introduction: Hand function impairment is the main disability among children with hemiplegic cerebral palsy. They start to perform most motor tasks exclusively with their unaffected upper limb, thereby causing a phenomenon described as developmental disregard. To minimize this phenomenon, constraint-induced movement therapy (CIMT) is emerging as a rehabilitation strategy for improving the functional use of the affected upper limb. The aim of this study was to conduct a systematic review of the literature on the effects of CIMT among children with hemiparetic cerebral palsy.

Method: In this systematic review of the literature a search of Cocharane, APTA, Pub Medline, Science Direct data base was done to analyze the effects of CIMT on the functional performance of the affected upper limb among children with hemiparesis. Selection criteria: All randomized controlled trials, controlled clinical trials, case studies and pilot studies on CIMT, modified CIMT and Forced were selected. The review discusses how CIMT in adults is different from paediatric population, the different types of CIMT used in children, key features and variations in protocols currently in use with children, the different methods of constraint used in paediatric population, outcome measures used and the efficacy of CIMT. Results: There are 13 studies fulfilled the inclusion criteria, reported substantial gains in functional use of the hemiplegic upper limb following CIMT with children. However, there was considerable variation between the studies regarding of type of constraint used, intensity and duration of training, and outcome measures. Modified CIMT with reduced duration constraining and training is feasible for children. Conclusion: Although there is huge variation in protocols for administering CIMT it can be reasonably effective mode of training of upper limb in hemiplegic cerebral palsy.

Key words: Constraint-induced movement therapy, hemiplegia.

INTRODUCTION

Cerebral palsy (CP) is defined as a group of permanent disorders of movement and posture development that are attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain. It is characterized by the inability to control motor functions and potentially has a negative effect on such children's overall development by affecting the ability to explore, speak, learn and become independent^{1,2,3}. Spastic hemiplegia is one of the types of CP and accounts for more than one third of new cases. It causes spasticity and motor-functional disability contralateral to the brain damage^{2,4,5}.

Gordon,¹⁰ Charles¹⁴ and Wolf stated that impaired hand function is a major disability among children with hemiplegic cerebral palsy (HCP). Because of this, such children usually have considerable difficulty in using the affected upper limb to perform tasks, including two-handed activities, thereby under-mining their independence and quality of life.

Considering the principles of developmental disregard, an intervention strategy called Constraint-induced movement therapy (CIMT) has been developed. CIMT therapy is the first rehabilitative approach which takes into account not only remediation of motor dysfunction but also the problem of learned non use deriving from functional limitation⁶ Concept of learned non use was first described by Taub after his research on monkeys in which the somatic sensations were surgically abolished from upper limb. These animals had motor deficit because of this sensory deprivation, but the strength was preserved and were able to perform movements under visual control. However, deafferented monkeys did not use their insensate limb. Taub found that after a period of restraint of the unaffected limb, the monkeys began to use their affected limb in an effective and permanent way. Taub hypothesized that Function suppression which is typical of the deafferented monkeys may be due to learned non use. He proposed that reversal of functional suppression could be attained by restraining the unaffected limb. This constitutes the core of CIMT therapy.

DEVELOPMENTAL DISREGARD

Developmental disregard occurs as a result of the conditioned suppression of movement, secondary to unsuccessful efforts to make voluntary movements during the acute phase of a neurological injury. Such individuals initially

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try to use the affected limb but, because of physiological reactions in the nervous system during the acute phase of the injury, there are no observable voluntary movements⁶. Successive failure leads these individuals to learn to use the unaffected limb when performing tasks, thereby leading to learned nonuse which is called as developmental disregard in paediatrics. This nonuse is believed to be responsible for a gradual reduction in the cortical representation of the affected limb, which would prevent its use during the chronic phase of a neurological injury.

Impaired arm & hand function

Repeated failed attempts to use arm

Discouragement leads to avoidance

Developmental disregard

Flow chart: Developmental Disregard. (Taub, Stephan Page, Levine p.) Children with hemiplegia face unique challenges in learning to use their affected arm. Their early attempts to use their affected arm take a lot of effort and often fail to use hand, while their unaffected arm constantly succeeds in activities of hand.

This consists of placing the unaffected upper limb in a cast and training of the contralateral limb. Evidence from using CIMT among adults who have suffered stroke suggests that it has beneficial results. These have been identified mainly through tests on the levels of upper-limb impairment and focal disability and through subjective measurements. Such tests have suggested that intensive behavioral training helps to transfer the gains achieved in the clinic to life outside the clinic. However, the use of this therapy among children with HCP is still controversial. Thus, the aim of this study was to conduct a systematic review of the literature on the effects of CIMT on children with hemiplegic cerebral palsy.

PHYSIOLOGICAL BASIS OF CIMT:

Two possible mechanisms for the observed effects are believed to be a) Overcoming the learned non-use of the more affected arm (i.e. increased use of the more affected arm) and b) Use dependent cortical reorganization.⁶ Cortical reorganization could be a possible explanation for the recovery. They also studied motor cortex mapping using transcranial magnetic stimulation before & after CIMT intervention and found that there is increase of motor output area size and MEP amplitudes, indicating enhanced neuronal excitability in the damaged hemisphere for the target muscles. Some author demonstrated changes in the activation of the motor cortex after CIM therapy using fMRI⁸. Functional magnetic resonance imaging showed bilateral sensorimotor activation after therapy⁹.

IMAGING FINDINGS IN PEDIATRIC CIMT

Numerous studies showed that the brain continuously reorganizes itself to adapt to environmental demands, and the size of the cortical representation of a body part in adults depends on the amount of use of that part. CIMT, with its emphasis on repetitive practice, was viewed as a possible model of the application of principles of neuroplasticity to rehabilitation. Nudo, in a series of studies demonstrated that repetitive practice with specific UE muscles in the adult squirrel monkey resulted in expansion of the motor cortex representing those muscles, and further, that training with the affected limb following motor cortex lesioning in monkeys resulted in cortical reorganization, so that the area surrounding the lesion, which is not normally involved in control of the hand, was recruited to participate in movement of that hand.^{8,9} In a direct evaluation of the ability of CIMT to produce cortical reorganization, used focal transcranial magnetic stimulation (TMS) to map the cortical motor output area of a hand muscle in 13 chronic stroke patients before and after CIMT. They found an approximate doubling of the size of the excitable motor cortex that could elicit movement in the more involved arm and hand after CIMT, in a pattern suggesting recruitment of cortex adjacent to lesioned areas. They described their results as “. . . the first demonstration in humans of long-term alteration in brain function associated with a therapy-induced improvement in the rehabilitation of movement after neurological injury.” Grotta et al. [2004] recently replicated these findings in a small pilot study with eight adults who had a stroke and were in the acute phase (2-weeks poststroke). They also found a strong correlation between the number of TMS activation points and functional test scores. They concluded that, if implemented within the first 2 weeks after stroke, CIMT “ is probably not harmful and it may accelerate recovery. TMS noninvasively demonstrates the biological effect of CIMT on brain reorganization.”⁸ used functional magnetic resonance imaging (fMRI) to investigate cortical reorganization after CIMT in two adults disabled from chronic stroke. In addition to their functional gains, both patients showed new cortical activity post-CIMT during a sequential finger-tapping task, including in areas bordering the infarcted region, in bilateral association motor cortices, and in ipsilateral primary motor cortices.

There are as yet few studies of possible neuroplastic cortical changes in children following CIMT, such as those described above with adults, but early reports are positive. report a case study with an 8- year-old boy with congenital right hemiplegic CP and left caudate infarction, who received a modified form of CIMT. The protocol included 3 weeks of continuous casting of the unaffected arm, in conjunction with conventional OT 1 hr per week. Clinical measures of motor function, fMRI, and magnetoencephalography were done before and after therapy and 6 months later. Both frequency of use and quality of movement of the hemiplegic hand were improved following removal of the cast, compared to

pretreatment testing. In addition, fMRI showed increased cortical activation in the sensorimotor cortex contralateral to the hemiparetic hand after the 3-week treatment interval. These changes were maintained at the 6-month follow-up. The authors conclude that, "This is the first report, to our knowledge, of cortical reorganization after modified constraint-induced movement therapy in a child with hemiplegic CP." Interestingly, the timing of the magnetoencephalography amplitude changes, which were coregistered with the fMRI data, supported the conclusion that increased contralateral cortical activation after CIMT was due to increased peripheral feedback to the sensorimotor cortex, rather than to increased activation of contralateral motor cortex. Since this feedback is necessary for perceptual awareness of movement, CIMT may contribute to increased hand use through the reduction/ resolution of inattention to contralesional space, or hemispatial neglect.

MECHANISMS FOR EFFECTIVENESS OF CIMT: CURRENT FORMULATION

As these and other imaging studies have demonstrated, learned nonuse, in addition to preventing the individual from attempting to regain use of the affected UE after a period of recovery, also leads to a contraction of the cortical motor and sensory representation of the affected limb. The conditions created by CIMT, particularly intensive, massed, repetitive practice, are precisely those shown to be effective in inducing cortical plasticity and adaptive reorganization after injury. Explanations for the efficacy of CIMT have thus been modified to incorporate evidence of use-dependent cortical reorganization after CIMT. CIMT produces a permanent increase in hemiparetic arm use through two linked, but independent mechanisms. First, by providing a situation in which the patient now receives positive reinforcement for use of the weaker arm, while at the same time experiencing negative consequences for efforts to use the constrained stronger arm, the nonuse of the more impaired arm learned in the acute and early subacute periods is counter conditioned. At the same time, increased use of the more impaired arm through sustained, repetitive practice, induces cortical

reorganization; specifically expansion of the cortical region controlling the arm to areas adjacent to the lesion and to homologous areas of ipsilateral cortex. Once a "critical mass" of intensity and duration of therapy is reached, the impaired limb has "recaptured" sufficient cortical space to sustain its use. Given the presumed heightened plasticity of the developing brain, the next logical step was to extend CIMT to the pediatric population, as first proposed by Taub.

METHOD

A literature search with no limits on data or language was conducted in the databases Cochrane, APTA, Pub Medline, Science Direct data base was done. Selection criteria: All randomized controlled trials, controlled clinical trials, and case studies and pilot studies comparing CIMT, modified CIMT and Forced were selected. The review discusses how CIMT in adults is different from paediatric population, the different types of CIMT used in children, key features and variations in protocols currently in use with children, the different methods of constraint used in paediatric population, outcome measures used and the efficacy of CIMT. The key words used were constraint-induced therapy, cerebral palsy, hemiplegia, modified constraint induced movement therapy (mCIMT) and Forced use. The articles identified by the search strategy were evaluated in terms of the following inclusion criteria: (1) study design: randomized controlled trial; (2) population composed of children affected by cerebral palsy that caused hemiparesis; (3) All three types of Constraint Therapy, 4) Children 1-18 years old with hemiplegia and 5) all outcome measures. Selection criteria: 1) Randomized controlled trials 2) Controlled clinical trials 3) Case studies & 4) Pilot studies. In the initial search 41 articles. After abstract screen: 18 articles. After full-text screen. 13 articles included in systematic review.

DATA ANALYSIS

Because of the insufficient data presented in some studies and the variety of different instruments used for measurements, no statistical analysis (meta-analysis) was performed. Thus, a descriptive analysis of the results is presented below.

STUDY	Study Design	Constraint given	Outcome measure	Result
Jeanne R Charles 2006 10	RCT . N 5 22; Ages 4–8 yr Hemiplegic CP (Control: 11 CIMT: 11)	Sling 6 hr/day (breaks from sling, not to exceed 30 min daily) and 10–12	Jebsen–Taylor Test of Hand Function, Bruininks–Oseretsky Test of Motor Proficiency, Caregiver Functional Use Survey, Grasp strength, Two-point discrimination and Muscle tone.	CIMT group had increased speed. increased dexterity increased functional use CIMT greater than control group No changes in measures of level of grip strength, Sensation and tone. Maintenance of gains 6 months post intervention

Table contd.

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STUDY	Study Design	Constraint given	Outcome measure	Result
Edward Taub, 200411	RCT. Casted N 5 18; Ages 7 months–8 yr; Hemiplegic CP (Control:9CIMT: 9)	Long arm cast; 24 hr/day and 21 days 6 hr/day; 21 consecutive days;	Emerging Behaviors Scale (EBS), the Pediatric Motor Activity Log (PMAL), and blinded ratings of videotaped sessions of the Toddler Arm Use Test (TAUT).	spontaneous use, : scores on lab tests of motor function of CIMT group Maintained gains at 3 and 6 months follow-up testing.
John K. Willis 200212	RCT: FORCED USE THERAPY Age Group:1-8 years Hemiparetic CP (Control:13 CIMT:12)	Plaster cast ;1month Regular OT & PT Was going on	Peabody Developmental Motor Scales (PDMS)	The 12 treatment (casted) children improved 12.6 PDMS points after 1 month of casting; the 13 control children improved 2.5 points. Improved PDMS scores persisted 6 months later when 7 treatment children returned. Similar results were obtained in the crossover when 10 control children received casts
Jeanne R Charles 200713	CCT 8 children; mean age 8y 7mo [SD 2y 6mo]; range 5–11y),	children wore a sling on their non-involved upper extremity for 6 hours per day during 10 out of 12 consecutive days	Jebsen–Taylor Test of Hand Function, the Speed and Dexterity subtest (no. 8) of the Bruininks–Oseretsky Test of Motor Proficiency	The second intervention resulted in further improvement on these measures. Results indicate that intensive practice associated with CIMT may be retained long term, and that continued improvements may occur after a second intervention dose.
Andrew M. Gordon, 200614	CCT; 20 children of age 4 to 13 years	Wore a sling for 6 hours per day for 10 of 12 consecutive days	Jebsen–Taylor Test of Hand Function, Bruininks–Oseretsky Test of Motor Proficiency, Caregiver Functional Use Survey, Grasp strength, Two-point discrimination and Muscle tone.	Significant improvements in involved hand- movement efficiency and environmental functional limitations, which were retained through the 6-month posttest. However, there were no differences in efficacy between younger and older children. CIMT is not age dependent.
N Kuhnke 200815	CCT Nine patients (five males, four females; mean age 16y [SD 6y 5mo], range 11–30y)	6 hrs per day for 12 consecutive days	Wolf motor function test (WMFT).	Significant improvement in the quality of upper extremity movements in both groups. Only in patients with preserved crossed projections, however, was this amelioration accompanied by a significant gain in speed, whereas patients with ipsilateral projections tended to show speed reduction.
Ann-Christin Eliasson 200516	CCT modified N 5 41; Ages 18 months–4 yr; Hemiplegic CP (Control:20 CIMT:21)	Fabric glove with thermoplastic splint; 1–2 hr/day and 2 months	Assisting Hand Assessment (AHA)	CIMT group demonstrated increased with gross UE test tasks. Maintained gains 6 months post

Table contd.

STUDY	Study Desgin	Constraint given	Outcome measure	Result
Trenna L. Sutcliffe, 200717		modified constraint-induced movement therapy for 3 weeks.	Clinical, functional magnetic resonance imaging and magnetoencephalography measurements, Pediatric Motor Activity Log	Magnetoencephalography showed increased cortical activation in the ipsilateral motor field and contralateral movement evoked field after therapy. Cortical reorganization was maintained at the 6-month follow-up. This is the first study to demonstrate cortical reorganization after any version of constraint-induced movement therapy in a child with hemiplegia.
Julie D Ries 200618	CS 28-month-old girl	6hrs per day for 10 days	Tardieu and Tardieu, AROM, strength, and motor control	significant improvement in 2-handed function in young children who were treated with an adapted and much less intensive model of PCIT than the Taub et al19model that we used with our patient.
	CS	30 weeks using a modified CIMT protocol consisting of four one-hour sessions per week	Action Research Arm Test	modified CIMT may be useful in the treatment of upper extremity dysfunction caused by a childhood brain injury
Rebekah Miller 200519	CS child was 15 months of age	full-arm, bivalved cast 3 weeks of intensive intervention (6 hours a day)	Peabody Developmental Motor Scales ²⁶ (PDMS), (2) the Denver Developmental Screening Tool ²⁷ (DDST), (3) the Pediatric Motor Activity Log (PMAL), and (4) the Toddler Arm Use Test (TAUT).	Child developed new behaviors throughout both interventions. During intervention 1, the child developed independent reach, grasp, release, weight bearing (positioned prone on elbows) of both UEs, gestures, self-feeding, sitting, and increased interactive play using both UEs.
Stephanie C DeLuca, 200320	mCIMT: N=6; Age=6-15yrs	(custom-made thermoplastic resting splint held in place with straps) 2 hours a day, 5 days a week for 4 weeks.	Quality of Upper Extremity Skills Test.	Children and parents were positive about mCIT. The use of mCIT is a promising intervention for children with chronic acquired hemiparesis.
Anne Gordon [2007];²¹ CE Naylor [2005]²²	Mcimt: N=9 Age:2.5-5yrs.	gentle restraint, with an adult holding the child's unaffected hand during the activities	Quality of Upper Extremity Skills Test	modification of constraintinduced movement therapy may be an effective way of treating young children with hemiplegia.

DISCUSSION:

Out of all the 13 studies 7 studies used conventional CIMT, 5 studies used modified CIMT & 1 used FUT. Duration of constraining was varying from 24hrs to 1hr & training duration was from 6hrs to 30mins. Several different types of restraints were used in all the 13 studies, which are as follows: Holding a child's hand (Naylor 2005), Using a glove or mitt (Eliasson 2004), Forearm splints (Crocker 1997; Glover 2002), Slings (Charles 2001; Gordon 2005), Short arm casts (Willis 2002) and Long arm casts (Deluca 2003).

AGE DEPENDENCY AND EFFECTIVENESS OF CIMT

There is no specific age group selected. There is wide range in studies varying from 1 to 18yrs. Is CIMT age dependent? Gordon et al. (2006) studied age-dependence of the effectiveness of CIMT. Children divided into older group (9-13) and younger group (4-8). No difference in outcomes between older and younger. So author concluded that CIMT is not age dependent.

REPEATED COURSE OF CONSTRAINT INDUCED MOVEMENT THERAPY

A recent therapeutic intervention, constraint-induced movement therapy (CIMT), has been shown to improve movement efficiency and quality of movement in the involved hand of children with hemiplegic cerebral palsy (CP). In Jeanne R Charles (2007) study, investigate the long-term effects of CIMT and the effect of a second course on involved limb function using an ABABA design. Eight children with mild to moderate hemiplegic CP (six males, two females; mean age 8y 7mo [SD 2y 6mo]; range 5-11y), who had received a CI therapy intervention 12 months before this study, participated in a second intervention. In both interventions, the children wore a sling on their non-involved upper extremity for 6 hours per day during 10 out of 12 consecutive days and were engaged in play and functional activities that provided structured practice using the involved upper extremity. The results indicated initial improvements in movement efficiency, as measured by the Jebsen-Taylor Test of Hand

Function, the Speed and Dexterity subtest (no. 8) of the Bruininks-Oseretsky Test of Motor Proficiency, and caregivers' perceptions of amount of use and quality of movement of the involved limb, were retained 12 months after the first intervention. The second intervention resulted in further improvement on these measures. Results indicate that intensive practice associated with CIMT may be retained long term, and that continued improvements may occur after a second intervention dose.

COMPLICATIONS

No specific report of complications and/or adverse effects of treatment reported but few studies reported Withdrawal/Dropouts. In 13 studies 5 authors reported dropouts & 3 are reason provided. Three subjects rejected the

glove constraint (Eliasson et al, 2005), one withdrawal due to uncooperative child (Willis et al, 2002) and one withdrawal due to lack of subject's tolerance of the intervention (Charles et al, 2006)

IS THERE A SPECIFIC PROTOCOL?

There is a wide variety of protocol, frequency and duration of therapy and constraint

Lack of adequate rationale for the choice of protocol, so it remains unclear as to which parameters and what combination of constraint, therapy and duration is most effective in achieving the desired outcomes in paediatric population.

The evidence does not yet answer many questions

Specifically, the evidence does not yet answer many questions:

What type of constraint should the child wear?

How many hours should the constraint be worn daily?

What is the ideal duration of constraint therapy in order to support lasting functional gains?

Should therapeutic activities be completed while wearing the constraint and if so, should these activities be supported by a therapist, a caregiver, or some combination?

At what ages is constraint therapy most beneficial?

Are there ages or levels of functioning for which constraint therapy is not beneficial?

Is constraint therapy equally beneficial for children with various diagnoses or functional skill levels?

CONCLUSION

The use of constraint induced therapy in pediatrics appears promising but the level of evidence is still weak.

LIMITATIONS

Unable to compare across studies due to variety of variables, Small sample sizes and low level of evidence is also included in the systemic review as included some case series and pilot studies also.

FUTURE RESEARCH

Comparison of protocols, Structured practice alone compared to a constraint intervention as to know what works better in paediatric population. Long term outcomes greater than 12 months and RCT's with higher power studies.

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