Effect of Modified Constraint Induced Movement Therapy and Hand Arm Bimanual Intensive Training on Upper Extremity Skills and Functional Performance in Children with Spastic Hemiplegic Cerebral Palsy

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ABSTRACT

Purpose: Spastic hemiplegia results into impairments of upper extremity that leads to activity limitations and affects quality of life. Current rehabilitation focuses on task oriented functional therapy or motor learning approaches. mCIMT and HABIT are training methods focused on the same principles.

Method: 26 participants (age 2-10 years) were matched in regards of age and side of affection to both groups, where subjects in mCIMT group wore a restrictive sling for 6 hours a day and performed repetitive massed practice of unimanual activities with sling: 2 hours per day for 10 days. Participants in group HABIT performed intensive bimanual activities for 6 hours a day for 10 days. Outcome measures QUEST and COPM were assessed in both the groups immediately after the intervention and 1 month post intervention.

Results: mCIMT and HABIT groups showed significant improvement from the pretest to immediate posttest in the QUEST and COPM (P < .0001) (15.26 -15.21 for QUEST, 12.64 - 13.94 for COPM performances and 14.24 - 14.43 for COPM satisfaction for group A-B respectively) which were maintained after 1 month of intervention. Moreover there was no significant difference between both the groups

Conclusion: Thus authors can conclude that, mCIMT and HABIT can be used equally to increase upper extremity skills and occupational performance in children with spastic hemiplegic cerebral palsy.

Keywords: Cerebral Palsy, mCIMT, HABIT, Hand Function, Task oriented approach

INTRODUCTION

Cerebral Palsy (CP) is described as a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. [1] There are other neurological symptoms likes dystonia, ataxia, athetosis and particularly spasticity secondary to brain pathology in children with cerebral palsy including other secondary disturbances like issues with sensation, perception, behavior, communication and cognition. [2] In developed countries, the prevalence data vary from 1.5-2.5/1000live births. [3,4] The prevalence of cerebral palsy is much higher in developing countries. [5]
Hemiplegic Cerebral Palsy accounts for 35% out of all types of cerebral palsy, more precisely referred as individual having unilateral impairments so mentioned as unilateral cerebral palsy. Out of all types of cerebral palsy, spastic are the most common type’s, accounts for 80% - as the most predominant tone. [6] Children with spastic hemiplegic cerebral palsy are presented with loss of upper motor neuron excitation which leads to poor selective motor control and weakness, as an impairment resulting into difficulty in performing coordinated task and affect activities of daily living. [7-8] Upper limb involvement is more pronounced as compared to lower limb, distal affection more than proximal in hemiplegic cerebral palsy. [9]

Children with unilateral cerebral palsy can be further qualitatively classified as per the International Classification of Functioning, Disability and Health-Children and Youth (ICF-CY). [10] They are classified on the basis of presence of changes in brain structure and function resulting in impairments of spasticity, muscle length, sensation, and weakness at the impairment level. Limitations in activity performance are common in self-care activities, [11] which leads to decreased activity and participation and affects quality of life. [12] In hemiplegic CP, hand use is affected in various perspective which hinders person- task- environment dynamic interplay in terms of spasticity, decreased selective motor control, grasp- lift synergy, postural control during uni-manual activities, affected feed forward mechanism for reaching and grasping affects both uni-manual and bimanual performance and capacity. [13-15] As upperlimb dysfunction leads to difficulty with day to day bimanual activities that hamper functional independence and participation in home, school and community, thus rehabilitation addressing UL dysfunction becomes need of an hour. [16] A number of UL rehabilitation approaches have been reported in children with unilateral CP. [16] Various traditional approaches works on impairments, on biomechanical, structural and neuro developmental frame of reference. [17] there has been growing evidence in literature showing positive effect of constraint induced movement therapy (CIMT) and bimanual therapy (BIT) for patients with hemiplegia. [17]

Classic CIMT consist of practicing unimanual activities for 6 hours per day and applying constraint for 90% of waking hours. mCIMT protocols similarly involve restraint of the non-affected limb with modification as name suggests in the type of restriction applied (e.g., glove, cast, sling) and are accompanied by a unimanual repetitive practice, differing in part from the classic CIMT in terms of the model of therapy (short-term intensity, long-term distribution model) and dose intervention. [16] The location, context and provider of training (house/camp, individual/group, therapist/parent) are also differentiated in relation to the classic CIMT. [17,18] Modified CIMT was adopted to make classic CIMT more child friendly, easy to use, to increase compliance and decrease the frustration. [16] HABIT includes: high intensity of treatment (6 hours per day for 2 – 3 weeks); the use of behavioral shaping theory [19, 20], and does not rely on physical assistance or handling but rather uses environmental adaptation for grading of activities of the child.

Individually, each of these interventions might yield improvement in upper extremity function. There has been no conclusion about superiority of one of the approach over another. [21] While principles of these approaches guide the practice, still application of CIMT and HABIT is highly variable in terms of setting(Clinic- home, group- individual, Implementer caregiver- therapist), type of constraint( Mitt, sling, splint, cast).Perhaps, the most proficient variable is dosage, Prominent ranges in dosage variability found in the literature included: durations from 5 days to 70 days, intensities from 1 hour a day to 6 hours a day, and total number of hours from 12 to 210 hours. [17]
Moreover, it was hypothesized that, gains in unimanual practice/capacity might transfer to gain in bimanual performance. Evidence in literature shows that same intensity of CIMT compared to BIT was found to be equally effective in improving hand function. Moreover, results from CIMT would be secondary to the intensity of practice rather than the constraint as postulated by Gordon. So authors of the present study wanted to compare the effect of child friendly form of CIMT with HABIT, with high intensity-short duration dosage, with combination of 2 settings i.e. 1 hour-therapist guided therapy, 5 hours-caregiver guided therapy) on the quality of upper extremity use and on the functional individualized goal set by the family in the Indian population.

MATERIALS AND METHODS

Non blinded Quasi Experimental study was carried out in a convenient sample at OPD of Pediatric physical therapy department of General hospital & private pediatric set-ups. Sample size calculation was done using pilot study with SD of 7.29 at 5% level of significance with 80% power of study. The estimated sample size was 13 for each group. Children with confirmed diagnosis of spastic hemiplegic cerebral palsy, age-2 years to 10 years with active movements of shoulder, elbow, wrist of affected limb such that child is able to reach forward, able to actively perform reach, grasp, release activities with moderate level of muscle tone (1-2 Modified Ashworth Scale) were included in the study, whereas child diagnosed as quadriplegic or spastic diplegic cerebral palsy with severe muscle spasticity or contracture (MAS >3) or previous orthopedic surgery or serial casting or Botulinum toxin A injection in upper limb within 6 months prior to study entry were excluded.

All patients coming from various OPD of general hospital were referred to pediatric neurologist for confirm clinical diagnosis of hemiplegic CP. Subjects those who were following in inclusion criteria were recruited for the study. Nature and purpose of the study was explained to the parents of the patients prior to the study. Informed written consent was taken from the parents or legal guardians prior to the study. Oral consent was taken from the patient/subject who was verbal and having cognition to understand. Subjects were matched into 2 groups according to their age and side affected: 13 in each group: Group A: modified Constraint Induced Movement Therapy (mCIMT) Group B: Hand Arm Bimanual Intensive Training (HABIT). Baseline assessment of outcome measure was taken one day before commencing the intervention in either groups. Patients were treated in either of the group for 10 days: 5 days a week for 2 weeks. Individual sessions of 45-60 minutes were conducted by physiotherapist for three times/week for 6 weeks (i.e. from baseline assessment to the time when last assessment of outcome measure was taken, after 4 weeks of no experimental intervention period) in outpatient pediatric treatment room.

Group A: modified Constraint Induced Movement Therapy (MCIMT)

The modified constraint-induced movement therapy (mCIMT) protocol worked on 2 main principles of CIMT as explained by Taub et al that is constrain the extremity and repeated structured mass practice to the affected upper limb (2 hours of uni-manual activities). Child wore a restrictive glove on unaffected upper extremity for 6 hours a day and have performed repetitive massed practice of uni-manual activities with glove on for 2 hours per day for 10 days i.e. 5 days a week for 2 weeks, out of which 1 hour of uni-manual activities with glove on was done at department in presence of the therapist and rest one hour was done at the home. 6 hours of glove could be split into different sessions of minimum of 30 minutes of each session. Intention of the home program was to involve the families and
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engage an intensive period of practice to facilitate use of affected hand at home and community. Activities like catching, throwing of balls, stacking blocks, turning cards, opening zip, putting beans in jar, playing with Velcro, peg board and opening door focusing on grasp, release and hold were practiced as mass practice repetitively.

Group B: Hand Arm Bimanual Intensive Training (HABIT): Children practiced bimanual activities 6 hours per day for 10 weekdays (60 hours). 1 hour hand arm intensive bimanual program was done at department with the therapist and other 5 hours bimanual task was done at home. Care was taken that child uses both affected and unaffected hand in order to perform bimanual activity. Bimanual activities like stacking bricks, tying shoe laces, opening lock and key, buttoning shirt, eating food with spoon, opening bottle, throwing large ball, stacking cards were done.

Caregivers were asked to keep an exercise diary, to log the total period of practice done, restraint device worn per day and any issues arising from use of the glove. For subject in HABIT group care givers reported about how many hours’ bimanual activities was done at home. Along with it, caregivers in both the groups reported about the activities done at home. Outcome measures were taken at the end of intervention and after 4 weeks of completing the intervention.
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Outcome measures: The Quality of Upper Extremity Skills Test (QUEST), impairment based descriptive measure designed to evaluate quality of movement patterns and hand function in children with cerebral palsy. [25] The QUEST involves evaluation of 36 items of upper extremity function in four domains: dissociated movement, grasp, protective extension, and weight-bearing. Affected and non affected sides were tested in this study. The Canadian Occupational Performance Measure (COPM) [26] is a client-centered measure based on persons’ perception of their occupational performance in self-care abilities, productivity and leisure activities. Parental responses regarding perception of occupational performance were recorded in our study secondary to the age of the child. This adaptation has been supported and adopted by Cusick et al. [27] in his findings, where it was demonstrated as acceptable internal consistency reliability for performance (mean alpha = 0.73) and satisfaction (mean alpha = 0.82), content and construct validity and responsiveness using this approach. Both outcomes were reliable and valid measure for measuring quality and parents’ perception for performance in CP [25-26]

This study was approved by institutional ethics committee.

Statistical Analysis:
Statistical analysis was done using SPSS version 16 and Microsoft Excel 2007. Comparison between the mean values of different variables measured post treatment in the two groups was performed using wilcoxon and paired t test for QUEST and COPM respectively. Comparison between pre –post measures within the group was performed using wilcoxon test for all the variables in both the groups.

To analyse the carry over effect of the approach, data was collected after 4 weeks of post intervention. Comparison of the mean values of different variables were done between and within groups after 4 weeks.

Level of significance was kept at 5% with confidence interval (CI) at 95%.

RESULTS
Basic demographics characteristics of both the group

<p>| TABLE 1. Demographic characteristics of subjects in 2 groups: |</p>
<table>
<thead>
<tr>
<th>Groups</th>
<th>NO. of patients</th>
<th>Mean Age (years)</th>
<th>SD</th>
<th>MALE</th>
<th>FEMALE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Group A</td>
<td>13</td>
<td>4.64</td>
<td>2.4</td>
<td>9(69.2%)</td>
<td>4(30.76%)</td>
</tr>
<tr>
<td>Group B</td>
<td>13</td>
<td>4.73</td>
<td>2.2</td>
<td>10(76.9%)</td>
<td>3(23.07%)</td>
</tr>
</tbody>
</table>

All the subjects in both the groups were matched 1-1 with each other according to their age and side affected. There was no significant difference between both the groups in regards of age and side affected at baseline assessment with (p = 0.743 for age and p= 1.00 for side affected). The percentage of girls and boys in both the groups were 30.76 and 69.2% in group A whereas 23.07 and 76.9% in group B.

Overall children spend an average time in training of 39.1/50 hours in mCIMT group and 41.2/50 hours in HABIT group at home. So compliance for home treatment comes out to be 78.2% and 82.4% in mCIMT and HABIT group respectively.

0-2 weeks:
Comparison between groups:

<table>
<thead>
<tr>
<th>Table 2: Between group analysis for 0-2 weeks</th>
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<tbody>
<tr>
<td>Outcomes</td>
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<tr>
<td>---------</td>
</tr>
<tr>
<td>QUEST</td>
</tr>
<tr>
<td></td>
</tr>
<tr>
<td>COPM P</td>
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<td></td>
</tr>
<tr>
<td>COPM S</td>
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<td></td>
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</tbody>
</table>

There was no significant difference between the groups in regards of QUEST, COPM P and COPM S with Z: -.314 and p = 0.753 for QUEST and p = 0.667, 0.958 for COPM respectively.
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Comparison within the groups:
There was a significant difference in regards of QUEST, COPM performance and COPM satisfaction in both the groups with p<0.001 for all the variables.

To analyze difference in QUEST and COPM scores after 2weeks of intervention in both the groups, Wilcoxon signed rank test was used which showed significant difference in all outcome measures within the groups considering pre-post test data with p value-0.001 for both groups.

<table>
<thead>
<tr>
<th>TABLE 3</th>
<th>With-in group analysis for 0-2 weeks</th>
</tr>
</thead>
<tbody>
<tr>
<td>GROUPS (QUEST)</td>
<td>PRE TREATMENT</td>
</tr>
<tr>
<td>GROUP – A</td>
<td>MEAN SD</td>
</tr>
<tr>
<td>73.11</td>
<td>1.93</td>
</tr>
<tr>
<td>GROUP B</td>
<td>70.83</td>
</tr>
<tr>
<td>GROUPS (COPM P)</td>
<td>PRE TREATMENT</td>
</tr>
<tr>
<td>GROUP – A</td>
<td>MEAN SD</td>
</tr>
<tr>
<td>15.79</td>
<td>1.24</td>
</tr>
<tr>
<td>GROUP B</td>
<td>17.4</td>
</tr>
<tr>
<td>GROUPS (COPM S)</td>
<td>PRE TREATMENT</td>
</tr>
<tr>
<td>GROUP – A</td>
<td>MEAN SD</td>
</tr>
<tr>
<td>15.24</td>
<td>1.44</td>
</tr>
<tr>
<td>GROUP B</td>
<td>16.4</td>
</tr>
</tbody>
</table>

For 0-6 weeks (after 1 month of intervention):
Analysis for 0-6 week was done between 15 subjects as there were 07 subjects in group A and 08 subjects in group B .Reasons for drop outs are expressed in the flow chart.

Comparison between groups:
There was no significant difference between the groups in regards of QUEST, COPM P and COPM S with Z: -0.338 and p=0.735 for QUEST and p=0.309, 0.265 for COPM Performance and satisfaction respectively.

<table>
<thead>
<tr>
<th>Table: 4: Between group analysis for 0-6 weeks</th>
</tr>
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<tbody>
<tr>
<td>Outcomes</td>
</tr>
<tr>
<td>QUEST</td>
</tr>
<tr>
<td>Group B</td>
</tr>
<tr>
<td>COPM P</td>
</tr>
<tr>
<td>Group B</td>
</tr>
<tr>
<td>COPM S</td>
</tr>
<tr>
<td>Group B</td>
</tr>
</tbody>
</table>

Comparison within the groups:
There was a significant difference in regards of QUEST, COPM Performance and COPM satisfaction in both the groups with p<0.018 ad 0.012 for all the variables for both the groups.

As there was no significant difference between groups, so effect size was calculated.

Effect size was moderate to strong for the effect within group with d- 8.64, 9.08, 7.57 for group A for all three variables whereas, it was 7.61, 6.52 and 6.53 for group B.

<table>
<thead>
<tr>
<th>Table: 5: Effect size between groups:</th>
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<tbody>
<tr>
<td>Outcome</td>
</tr>
<tr>
<td>QUEST</td>
</tr>
<tr>
<td>COPM P</td>
</tr>
<tr>
<td>COPM S</td>
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</tbody>
</table>

<table>
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<tr>
<th>Table 6: Effect Size within groups</th>
</tr>
</thead>
<tbody>
<tr>
<td>Outcome</td>
</tr>
<tr>
<td>QUEST</td>
</tr>
<tr>
<td>COPM P</td>
</tr>
<tr>
<td>COPM S</td>
</tr>
</tbody>
</table>

Comparison within the groups:
There was a significant difference in regards of QUEST, COPM Performance and COPM satisfaction in both the groups, Wilcoxon signed rank test was used which showed significant difference in all outcome measures within the groups considering pre-post test data with p value-0.001 for both groups.
DISCUSSION

Results showed statistically significant difference in QUEST and COPM for both group A & B after 2 weeks of intervention with (p = 0.001). Children with hemiplegic cerebral palsy as also known as unilateral cerebral palsy learn to use only unaffected hand for daily tasks which leads to behavioral suppression of use of affected hand and is termed as developmental disregard. [28] Thus therapy must create experience that should change the behavioral suppression of developmental disregard and must reward the use of affected limb in simpler tasks like stabilizing an object. CIMT is considered as one of the method of achieving this experience. [28] Various modified and distributed form of CIMT has been proposed. One of them which were used in this study was to constraint hand for 6 hours per day and 2 hours of unimanual activities for 2 weeks. This modified way was used to make the protocol more child friendly. [16]

There is insufficient evidence to support the use of a specific type of constraint over another. [21] Constraint used in this study can be considered as key point to make it child-friendly as it was a gentle restraint in order to reduce frustration, discouragement in using constraint and reduces fail.

Results from present study are consistent with other studies in showing a significant improvement in upper limb function after mCIM therapy in children with unilateral impairments. [29] Morris and
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Taub has proposed two mechanisms to be responsible for increased use of the more affected extremities, overcoming learned non-use and inducing use-dependent cortical re-organization \[30-31\] which basically works on motor learning principle to strengthen new motor pattern to perform an activity more efficiently.

HABIT was implemented on the basis of the experience with CIMT for children with unilateral impairments, \[31\] mainly to focus on an area that leads to activity limitation and participation restriction. HABIT uses principles of specificity of training in form of repetitive practice and plasticity. Neurophysiology literature specifically motor behavior states that practicing active bilateral movements may result in a facilitation effect from the non paretic arm to the paretic arm. Thus both arms work as coordinated unit in the brain. \[32\] In his research, Gordon asserts HABIT as name suggest is 6 hour intensive training of bimanual activities that helps in shaping of motor cortex, motor planning, coordination and in turn improves quality of use of U/E. \[19\]

Moreover results shows there is no significant difference in effect of MCIMT and HABIT on quality of upper extremity skills with (p=0.753). Both the intervention works on motor learning principle with shaping of motor task with intensive practice which might leads to plasticity. Similarly as a child engages in intense practice using their affected extremity it is thought that the neural connections in their brain change. The altered neural connection can create new pathways (sprouting) or change pre-existing pathways (unmasking) in order to enhance functional movement (occupational performance) (Pendleton & Schultz-Krohn, 2006), that is what occurs with intensive therapy of HABIT as supported by Nichole Hayes. \[33\] Our result is even supported by study done by Sakzewski where it was postulated that both the therapies found to be equally effective in improving hand functions in children with congenital hemiplegia. \[34\]

Results showed significant improvement within both groups A and B in COPM for performance and satisfaction with (p = 0.001). Traditional management for children with hemiplegic cerebral palsy works mainly at impairment level to reduce tone, increase range of motion and to increase strength of the limb for function. \[35\] However MCIMT and HABIT both of this technique works on activity limitation and participation restriction and not on impairment level. \[36\] COPM for performance and satisfaction is goal based subject’s perception about their performance and satisfaction related to their activity. Training focusing on specific goal that are meaningful to child and can be transferred to the daily functioning leads to significant change in parental perception of their child performance regardless to type and intensity of training.

Moreover, major part of intervention (50 hours for each group) was implemented as home program to make it more feasible for parents, increase parental participation and provide natural environment for intensive practice. Compliance was found to be 78.2% and 82.4% for mCIMT and HABIT group respectively at home. Home based therapy for CIMT and Bimanual OT were investigated with effective results as quoted in a meta-analysis. \[16\] Compliance achieved in bimanual training was 85% as compared to 70 % of CIMT which suggest home practice sounds to be easier for bimanual group and supports the finding present in this study. \[16, 37\]

Post intervention carry over effect of mCIMT and Habit were investigated which showed the significant difference for both the group after 1month of intervention. There was drop out of total 11 subjects secondary to distance of center, school got started, patient got shifted or due to family issues. These changes seen after constraint therapy can be attributed to change in efficiency, ease and facilitation of use in the environment. Along with that a study showed that Functional magnetic resonance imaging showed bilateral sensory motor
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Activation before and after therapy and a shift in the laterality index from ipsilateral to contralateral hemisphere after therapy. mCIMT is creating a window of opportunity where children learn to use affected limb more efficiently in everyday activities which is supported by study, where effects were maintained for 1 month and 6 month after intervention respectively. [39-40]

HABIT is a form of functional training mainly targeted on coordination of the two hands using structured task practice embedded in bimanual play and functional activities with key concept of repetitive intensive practice. Studies done in adult stroke shows significant cortical reorganization after bimanual training. [41] Reorganization of corticocerebellar circuits with inclusion of contralesional motor cortex and ipsilesional cerebellum may be a key mechanism of bimanual therapy. [42] Bleyenheuft states that improvements in upper extremity function are believed to be associated with neuroplastic changes, presenting differential corticospinal developmental reorganization (ipsilateral and contralateral). Moreover, intensive practice can increase in activation and size of motor area controlling the affected hand as quantified by imaging techniques. [43]

As HABIT focuses primarily on functional activities which help to improve bimanual coordination thus significant improvement is seen in quality of U/E skills as well as activities of daily living which would have led to carry over change in perception of child’s performance. These results are supported by a study that showed the significant increase in functional use of affected upper extremity remained same after 1 month of intervention. [44]

Theories of motor learning and neural plasticity are the key principle for both: unimanual as well as bimanual approach, where mCIMT approach works on use dependent reorganization by intensive training of impaired hand, while other uses bimanual use along with functional activities for relearning of motor control in impaired hand [36] which justifies results in our study. Moreover, a systematic review was done to compare constraint induced movement therapy and bimanual training in children with unilateral cerebral palsy, which shows that both interventions produce similar improvements in unimanual and bimanual capacities and functional performance. [45] An another study with approach of bimanual training with and without constraint on hand functions shows that both interventions were similarly effective for improving use of affected hand in bimanual tasks shows similar results as shown in our study. [46]

As there was no significant difference between the effect of two approaches so, effect size was calculated which came out to be large effect size with group mCIMT having larger effect as compared to HABIT group. This was supported by Sakzewski [16] in a meta analysis that postulated that, studies comparing intensive unimanual therapy (CIMT, mCIMT) or hybrid therapy with standard care of a lesser dose have shown modest to strong treatment effects across most outcomes. [47-48] whereas, trials comparing intensive unimanual therapy (eg, mCIMT) with an equivalent dose of bimanual training have reported weak to modest treatment effects on most outcomes [49-51]

There were few limitations in our study such as the restraint of the unaffected arm for mCIMT and intensive training for HABIT were accurately controlled during the practice in therapist’s presence where as it was difficult to control at home during intervention in both groups. Moreover none of the tests measured the bimanual coordination or unimanual / bimanual ability individually. Proper accurate methods of restraining and applying intensive training at home for both mCIMT and HABIT should be incorporated. Future implications to be incorporated are to assess effect of unimanual capacity over bimanual performance should be measured after intervention. Effect of mCIMT followed by
HABIT should be assessed on quality and bimanual coordination for activities of daily living.

**CONCLUSION**

This study concluded that modified Constraint Induced Movement Therapy (MCIMT) and Hand Arm Bimanual Intensive Training (HABIT) both are effective in improving quality of upper extremity skills & occupational performance - satisfaction in spastic hemiplegic CP. Moreover, carry over effect of both the intervention was maintained even after completion of treatment protocol as assessed after 4 weeks of intervention. Thus, both mCIMT and HABIT can be used interchangeably in improving upper extremity skills & occupational performance - satisfaction in spastic hemiplegic cerebral palsy.

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