Modified Constraint-Induced Movement Therapy for a 12-Month-Old Child With Hemiplegia: A Case Report

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KEY WORDS
• cerebral palsy
• hemiplegia
• modified constraint-induced movement therapy
• pediatrics

OBJECTIVE. This case report describes the use of modified constraint-induced movement therapy (CIMT) to improve upper-limb function in a 12-month-old child with right hemiplegia. It also describes parent concerns about CIMT and documents the short- and long-term effects of modified CIMT.

METHOD. The participant was assessed 5 times over a 7.5-month period using the Peabody Developmental Motor Scales–2, Pediatric Motor Activity Log, Toddler Amount of Use Test, and Knox Parent Questionnaire. CIMT included a nonremovable cast worn on the unaffected arm and approximately 8 hr per week of occupational and physical therapy for 2 weeks.

RESULTS. Benefits of improved upper-limb function measured immediately after CIMT were sustained at 6 months’ follow-up. No adverse events related to cast use were reported.

DISCUSSION. The findings from this case report suggest that CIMT was a safe intervention associated with improving upper-limb function for this young child with hemiplegia.


Constraint-induced movement therapy (CIMT) has emerged over the past decade as a promising intervention for reducing impairment and improving functional use of the affected upper limb in children with hemiplegia secondary to congenital brain injury (Charles & Gordon, 2005). Two distinct aspects of traditional CIMT for adults with stroke or children with cerebral palsy exist, which are based on the work of Taub and colleagues (Taub & Uswatte, 2003): (1) restraint of the unaffected upper limb and (2) intensive intervention involving mass practice. Two theoretical mechanisms have been offered to explain the therapeutic changes demonstrated in CIMT research for adults with stroke. First, patients with hemiplegia may experience a phenomenon known as learned nonuse, in which they do not realize the full potential of motor recovery in their affected upper limb because they have become used to their limited movement capability. Thus, one way in which CIMT is believed to be therapeutic is by diminishing the learned nonuse experienced by some patients. The terms developmental disuse (Gordon, Charles, & Wolf, 2005) and developmental disregard (Deluca, Echols, Law, & Ramey, 2006) have been used by pediatric CIMT researchers to describe this phenomenon in children with hemiplegia. Second, stroke researchers believe that CIMT may be effective in improving motor recovery in patients with hemiplegia because of increased size and shifting of cortical area neural firing after CIMT (Wittenberg et al., 2003). These findings support the idea that cortical reorganization is possible in patients with chronic neurological conditions such as stroke and may explain the observed improvements in use of the affected upper limb. Although whether these mechanisms can explain improvement from CIMT in children is not known, a recent
pilot study demonstrated evidence suggesting that cortical reorganization may also be possible in some children with hemiplegia (Cope et al., 2007).

Most studies on pediatric CIMT have experimented with its two key features by varying the type of restraint used and the frequency and intensity of intervention. Many of these studies are considered “modified” CIMT because they involve intervention intensity less than the traditional 4 to 6 hr per day (Taub, Ramey, DeLuca, & Echols, 2004). Three randomized controlled trials (Charles, Wolf, Schneider, & Gordon, 2006; Deluca et al., 2006; Taub et al., 2004) and one nonrandomized controlled trial (Eliasson, Krumlinde-Sundholm, Shaw, & Wang, 2005) have investigated the effectiveness of CIMT with children diagnosed with hemiplegia secondary to cerebral palsy. Moreover, nine studies with single-group or case report designs have investigated CIMT (Charles, Lavinder, & Gordon, 2001; DeLuca, Echols, Ramey, & Taub, 2003; Eliasson, Bonnier, & Krumlinde-Sundholm, 2003; Glover, Mateer, Yoell, & Speed, 2002; Karman, Maryles, Baker, Simpser, & Berger-Gross, 2003; Naylor & Bower, 2005; Pierce, Daly, Gallagher, Gershkoff, & Schaumburg, 2002; Sterr, Freivogel, & Voss, 2002; Yasukawa, 1990). Collectively, these studies have provided evidence that CIMT or modified versions of CIMT can be effective for improving upper-limb function in some children with hemiplegia (see Charles & Gordon, 2005, for a review).

Many of the children participating in pediatric CIMT research to date have been school age. Those studies that have included children ages 0 to 18 months have provided evidence to suggest that even young children can benefit from CIMT (Deluca et al., 2006; Eliasson et al., 2005; Naylor & Bower, 2005; Taub et al., 2004). However, data for young children in these studies are not presented individually or in age subgroups, making it difficult to determine how young children respond to CIMT. Two case studies specifically examined the effects of CIMT or modified CIMT for young children ages 0 to 18 months (Deluca et al., 2003; Yasukawa, 1990). In both studies, a 15-month-old participant demonstrated positive outcome in functional upper-limb use from the combined restraint and intervention. The evidence from these studies suggests that young children with hemiplegia (0–18 months) can also experience improved upper-limb function after participating in CIMT or modified CIMT. However, more research is needed to verify this finding.

The application of CIMT to pediatric populations has prompted recent concerns that children may not be developmentally ready for the intense massed practice and restraint used in CIMT protocols (Hart, 2005). It is thought that the intensive nature of CIMT may cause stress and fatigue and present an excessive burden on families in terms of time and cost. It has also been suggested that restraint of the unaffected arm poses a safety risk, may cause unnecessary frustration and stress by forcing the child to use the more affected arm, and may negatively affect the unaffected arm and hand because of disuse during periods of development (Stanger & Oresic, 2003). Finally, concerns about pediatric CIMT have been raised by practicing therapists who suggest that 4 to 6 hr of treatment per day is not supported in current service delivery models and is unrealistic in terms of reimbursement from some payers (C. Cayo, personal communication, December 2005).

Concerns about pediatric CIMT are perhaps even more valid when considered for younger children (0–18 months) with hemiplegia. Although relatively few accounts of adverse events have been reported in pediatric CIMT literature that support these concerns (Crocker, MacKay-Lyons, & McDonald, 1997; Glover et al., 2002), further research is needed to determine whether the benefits of improved upper-limb function outweigh the potential risks. Our main aim in this investigation was to address this question by providing modified CIMT (mCIMT) to a 12-month-old child with hemiplegia. Our specific aims were to (1) identify parent concerns related to intensity of mCIMT for young children with hemiplegia and possible risks of restraining the unaffected upper limb, (2) describe the use of a mCIMT protocol in therapy management for a young child with hemiplegia, and (3) document the short- and long-term effects associated with mCIMT for a young child with hemiplegia.

Method

Design

We used a single-system design with one participant. The participant was evaluated at five different points over a period of 7.5 months: twice before mCIMT (2 weeks before and immediately before mCIMT) and 3 times after mCIMT (immediately after mCIMT, 2 weeks’ post-CIMT, and 6 months’ post-mCIMT). We used Pretest 1 and 2 to measure the degree of stability in outcome measures during 2 weeks of standard care. The two follow-up assessments determined whether changes measured immediately after mCIMT were sustained at 2 weeks and 6 months after mCIMT. Figure 1 shows the study design.

Description of Participant

At the start of this investigation, the participant was 12 months old and had been diagnosed with right hemiplegia with suspected cerebral palsy. She was adopted from a Central American country at 6 months of age and came to the United States in November 2004. No information was available regarding the birth mother’s pregnancy or delivery.
Magnetic resonance imaging was performed in December 2004 and revealed that the left frontal and temporal lobes were underdeveloped.

Before the start of this investigation, we noted that the participant had full passive range of motion in all right upper-extremity joints with a mild increase in muscle tone (greatest in the hand, as evidenced by nearly constant fistings). The participant would actively reach for toys with her right upper extremity only after verbal or physical prompting or gentle restraint; otherwise, the left side was used exclusively. Gross grasping with the affected right hand was attempted but not successfully. If both hands were free to engage in play, she most often used her left hand. If a task required use of bilateral upper extremities, she would use her right hand to assist her left. The participant had difficulty tracking an object from midline to the right and from her right side to midline. We noted that she stopped tracking the object (when moving from midline to the right) at about 45° and then redirected her gaze back to midline.

The participant received occupational and physical therapy services from January 2005 to the start of this investigation in June 2005. Presenting concerns were hemiplegia in the right upper and lower extremity with fisting of the right hand and decreased use of the right arm and leg leading to delayed fine and gross motor skills. Frequency of services ranged from once a week to two times a week for 45- to 60-min sessions of both occupational and physical therapy (average of 4 hr per week). All treatment sessions took place in the family’s home with either parents or grandmother present. Family members were consistent with follow through of the therapists’ recommendations (HF and DB were the treating therapists).

Treatment sessions during the 6 months preceding start of mCIMT focused on increasing right upper- and lower-extremity awareness and spontaneous use. Therapy sessions also worked on visual tracking skills (to address right visual field neglect). A neoprene thumb splint was fashioned for her right hand to be worn during waking hours and assist with thumb opposition for grasping of toys. The participant tolerated the splint well and was noted to be more successful with grasping items in her right hand while wearing the splint.

**Procedures**

During the first 2-week period, the participant received standard care, which included two 60-min sessions per week of both occupational and physical therapy for a total of 4 hr per week. Following this period, the participant received mCIMT for 2 weeks, including more intensive occupational and physical therapy (8 hr per week) and a nonremovable cast on the unaffected upper limb. After the 2-week period of CIMT, the cast was removed, and the participant resumed a standard level of occupational and physical therapy. Follow-up assessments were taken at 2 weeks and 6 months after mCIMT.

**Modified CIMT.** The mCIMT sessions differed from the standard care sessions in two ways. First, the therapists doubled the amount of time they typically spent with the participant (from 4 hr per week to 8 hr per week). However, the goals and activities selected were the same as in standard care. Second, the physical therapist devoted more time to treatment of the affected upper limb during the 2 weeks of CIMT than during standard care. The occupational therapist and physical therapist collaborated on intervention activities to be consistent from session to session. Intervention focused on bilateral hand use and functional unilateral hand use; sensory stimulations for limb awareness; weight bearing in various positions; and trunk strengthening and promotion of transitional movements involving the upper extremities such as side-sitting, four-point stance, and pull-to-stand. Within each session, therapists altered the task frequently and used novel objects to maintain interest and motivation. Activities selected were age appropriate to the child.

**Restraint.** Both removable and nonremovable casts were considered. A nonremovable cast was determined to be the best method of restraint for this particular child on the basis of discussion with parents, therapists, and researchers (see Figure 2). The cast was made on the Friday before the start of mCIMT on the following Monday so that any...
modifications to ensure proper fit could be made; however, no changes were required. The cast was made of lightweight waterproof casting material (Coban self-adhesive elastic wrap; 3M, St. Paul, MN). The cast was a short-arm design (i.e., it started at the proximal aspect of the forearm and ended at the fingertips). The thumb was posted out from the hand. This design allowed for freedom of elbow movement, making dressing easier and yet limiting use of the unaffected upper extremity. The cast was removed at the end of the 2-week period of mCIMT.

**Outcome Measures**

The participant’s right upper-extremity function was measured using three assessments. The Peabody Developmental Motor Scales–2 (PDMS–2; Folio & Fewell, 2000) is a criterion referenced and normed measure of gross motor and fine motor function for children ages 0 to 83 months intended to show developmental level and detect change after therapy. Items from the Grasping and Visual–Motor Integration (VMI) subtests were used to compare upper-extremity function before and after CIMT; both affected and unaffected sides were tested and scored separately. We report raw scores here; a higher score indicates a better performance. We used this measure because it was commonly used within the agency employing the therapists and has been shown to be valid and reliable (Folio & Fewell, 2000). The Pediatric Motor Activity Log (PMAL; Taub et al., 2004) is a 22-item parental interview used to measure the child’s upper-extremity functioning in activities at home. Upper-limb use is scored on two ordinal scales, How Often and How Well. The How Often scale ranges from 0 (never uses the affected arm for task completion) to 5 (uses affected arm on almost all occasions for task completion); the How Well scale ranges from 0 (unable to use affected arm for task completion) to 5 (uses affected arm in a way that is normal for child’s age). Scoring for both scales involved averaging across the 22 items, with a higher score indicating a better performance. The parent was interviewed by one of the treating therapists. The Toddler Amount of Use Test (TAUT; Taub et al., 2004) is a tool intended to measure the degree of spontaneous use and quality of movement of the affected upper extremity during play activities. The TAUT is a 17-item assessment that is scored using the same How Well and How Much scales as in the PMAL; only the How Well scale was used here. For this investigation, the TAUT was videotaped and later scored by an occupational therapist not involved in this investigation. To control for expectation bias, the rater was unaware of the order of assessment dates. Scoring was the same as for the PMAL. Both the PMAL and TAUT were selected to be consistent with other investigations into pediatric CIMT and to facilitate comparison of treatment effects across studies. Neither the PMAL nor the TAUT have been subjected to reliability or validity studies. The neoprene thumb splint described earlier was not worn during assessments.

The Knox Parent Questionnaire (KPQ; Knox & Evans, 2002) was used to assess parental perception of changes in their child’s performance over the course of the study. This questionnaire allowed for rating of change in 16 different skills related to self-care, mobility, communication, and play. Parents were asked to respond (yes, no, or don’t know) to the question “In the past week, have your child’s abilities changed in any way?” The items parents said “yes” to were also rated in terms of significance on a 5-point Likert scale (0 = very negative change, 5 = very positive change). The KPQ was filled out by the parent at three points, once after each week of CIMT and at the 2-week follow-up.

**Results**

**Tolerance for Cast Use and Increased Intensity of Treatment**

Parent and therapist reports suggested that the participant tolerated the cast well from the beginning. There were no incidents associated with cast use, such as compromised

**Figure 2. Nonremovable cast.**
safety or skin breakdown. At the end of the 2-week period of mCIMT, the parents were pleased with the observable changes in their child’s arm and hand function, and except for some sleep disruption secondary to not being able to suck the thumb on the unaffected side, the parents had positive feedback about the restraint aspect of mCIMT. In fact, after the cast was removed, the parents asked for another cast cut in such a way (bivalved) so that they could remove it and reapply it. In terms of the increased treatment intensity, therapists noticed a temporary decrease in upper-limb movements around the 4th day; they interpreted this as muscle soreness secondary to increased use. Parents and therapists generally felt the participant tolerated the increased intensity well. Therapists were able to decide to divide the 2-hr session into separate 1-hr sessions within a day if it was determined that the 2-hr continuous time would not be tolerated. Some sessions were separated with a break for lunch, to accommodate a nap, or for travel time from community experiences (e.g., playgroup at Gymboree) to the family’s home.

Immediate Effects Associated With mCIMT

Table 1 presents scores for the PDMS–2, PMAL, and TAUT at all testing times. Scores for the VMI subtest of the PDMS–2 were stable across pretest measures but increased immediately after mCIMT (change score = +18 points). Items on which the participant improved included unilateral right hand use for inserting shapes into a formboard, removing and placing pellets and cubes, and scribbling with a crayon. Scores from the PDMS–2 showed a gradual trend of improved performance for items in the Grasping subtest but no large increases immediately after mCIMT. On the PMAL How Often scale, the participant’s performance was stable over the pretesting period but improved immediately after mCIMT (change score = +2.04 points). On the PMAL How Well scale, the participant was also relatively stable across the two pretest sessions and then improved immediately after mCIMT (change score = +1.5 points). Scores for the TAUT How Well scale showed little change across pretesting but a modest improvement immediately after mCIMT (change score = +0.6 points).

Follow-Up Effects Associated With mCIMT

At 2-week follow-up, scores for both subtests of the PDMS–2 improved slightly. At 6 month follow-up, scores on the Grasping subtest improved (change = +9.0 points compared with posttest), and scores for the VMI subtest increased (change = +16 points compared with posttest). Scores on the How Often and How Well scales of the PMAL changed slightly at 2-week follow-up (change = −0.8 and +0.3 points, respectively) but improved at the 6-month follow-up (change = +1.05 and +0.9 points, respectively). Scores for the TAUT at 2-week and 6-month follow-ups changed little compared with post-mCIMT scores.

Knox Parent Questionnaire

Several important changes were reported on the KPQ after both the 1st and the 2nd weeks of mCIMT. Specifically, changes were noted in three aspects of function: (1) right upper-limb function, (2) gross motor skills, and (3) speech and communication. Use of the affected upper limb was reported by parents as being more frequent and spontaneous. Examples were seen in attempts to push the arm through a shirt sleeve, splashing more with the right arm during bathing, and attempting to hold the handle of a cup and drink. During play activities, parents noted increased ability to reach over head and grasp objects (e.g., placed on top of head) and increased strength to grasp and pull. Parents also noted much better ability to release objects from the right hand, as in placing objects into a container. Use of the index finger and thumb to pick up small objects for finger feeding improved during the 2-week period of mCIMT. Finally, parents noted many of these changes improved over the course of the 2 weeks of mCIMT (e.g., pushing the arm through the sleeve became smoother and stronger by the end of the 2-week period). Table 2 shows the number of items scored by parents on the KPQ as positively changed over the 2 weeks of mCIMT and at follow-up. The table also shows the number of items that were rated at 4 or 5 (positive or very positive changes).

In the area of gross motor skills, parents noted that during the 2-week period of mCIMT, their child became much better at pull-to-stand as she performed this task more frequently and at an increased variety of surfaces or supports. Also, quadruped creeping improved in terms of quantity and quality (weight bearing on right upper extremity changed from dorsum of the hand to the palm). The child also became more fluid at getting up into a sitting position, performing this task more quickly and with greater frequency.

Table 1. Outcome Measures Over Five Assessment Periods

<table>
<thead>
<tr>
<th>Outcome Measure</th>
<th>Pretest 1</th>
<th>Pretest 2</th>
<th>Posttest</th>
<th>2-Week Follow-Up</th>
<th>6-Month Follow-Up</th>
</tr>
</thead>
<tbody>
<tr>
<td>PDMS–2</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Grasping</td>
<td>28</td>
<td>30</td>
<td>33</td>
<td>37</td>
<td>39</td>
</tr>
<tr>
<td>VMI</td>
<td>40</td>
<td>44</td>
<td>62</td>
<td>63</td>
<td>78</td>
</tr>
<tr>
<td>PMAL</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>How Often</td>
<td>0.47</td>
<td>0.56</td>
<td>2.6</td>
<td>1.8</td>
<td>3.65</td>
</tr>
<tr>
<td>How Well</td>
<td>1.75</td>
<td>1.7</td>
<td>3.2</td>
<td>2.9</td>
<td>4.1</td>
</tr>
<tr>
<td>TAUT How Well</td>
<td>3.2</td>
<td>3.0</td>
<td>3.6</td>
<td>3.5</td>
<td>3.53</td>
</tr>
</tbody>
</table>

Note. PDMS–2 scores are for affected hand. PDMS–2 = Peabody Developmental Motor Scales–2; VMI = Visual–Motor Integration subtest; PMAL = Pediatric Motor Activity Log; TAUT = Toddler Amount of Use Test.
Parents rated the area of communication as the most significantly changed. Both receptive and expressive language increased during play interactions and general communication. At the start of the 2-week mCIMT period, the participant could say only 1 or 2 words; according to parent report, she verbally expressed approximately 20 words after mCIMT. She was also using signs more spontaneously and consistently.

Effect of Cast on Unaffected Upper Extremity

The PDMS–2 was also administered to the left (unaffected) upper extremity to determine whether continuous use of a short-arm cast would have adverse effects on grasping and visual–motor skills. Scores for both Grasping and VMI subtest items showed a continued improvement over time with no evidence of diminished performance during cast use (see Table 3).

Discussion

This investigation of a 12-month-old child with hemiplegia demonstrated a positive link between mCIMT and increased use of the affected upper limb with little evidence of adverse events. These findings suggest that for this child the benefits associated with mCIMT surpassed the risks. The results of improved upper-limb function reported here are consistent with other investigations into pediatric mCIMT specifically investigating young children. One difference between this investigation and previous studies was that the total amount of therapy (16 hr) was far less (90–126 hr in Deluca et al., 2006, and 28 hr in Glover et al., 2002). This difference is significant because it suggests that important clinical changes observed in a short period of time may be related to treatment intensities that are feasible for the therapists within the context of their service delivery model. This finding supports the use of mCIMT protocols with greater-than-standard amounts of therapy but at a level of intensity that is not excessive for the child, parent, or therapist. Another difference between this study and some previous CIMT investigations was the type of restraint used. We used a nonremovable cast with this child on the basis of likelihood of compliance with a less restrictive restraint, whereas other investigations into mCIMT for young children used splints or slings. It is difficult to separate the relative contribution of the restraint and the intensive rehabilitation because the design did not control for these two factors independently. It is likely that the cast played some role in the changes observed because it was worn continuously for 2 weeks, forcing the child to use the nonaffected upper limb. Also, the total amount of therapy was increased by 8 hr, an amount that may appear small but actually represented twice as much as was received before starting mCIMT. Two investigations have examined the effects of a restraint only and have reported beneficial effects (Sung et al., 2005; Willis, Morello, Davie, Rice, & Bennett, 2002). We found no studies that experimentally compared the effects of different restraint devices.

The size of the treatment effect measured for this participant was similar to those in studies that have used similar outcome measures. For example, Taub et al. (2004) reported change scores for the PMAL How Much and How Well scales ranging from 1.3 to 2.0, which is similar to that reported here (1.5). Also, the 15-month-old child in DeLuca et al. (2003) improved by 1.1 and 1.7 points, respectively, on the PMAL How Much and How Well scales. Whether the magnitude of changes reported here and elsewhere represent a “minimally clinically important difference” is not known. For example, a change of 1.5 points on the PMAL representative of meaningful and important arm use as perceived by the parent? Is a change of 0.5 enough? The parents in this study were not specifically asked about the changes made on the PMAL or TAUT; however, responses to the KPQ suggest that the parents felt the changes made in upper-limb movement were important to the child’s functional arm use at home.

The measure of visual–motor skill (PDMS–2) and functional upper-limb use (PMAL and KPQ) suggested impressive changes in function of the upper limb. However, improvements in grasping ability with the affected hand were not as significant as measured by the PDMS–2 Grasping subtest. Grasping performance using whole hand or finger grasp patterns is known to develop later than reaching or whole arm movements (Gordon, 1994), which may explain why improvements on the Grasping subtest were less impressive than for the VMI subtest. A longer period of intervention may be needed to affect hand function.
Our final aim in this project was to determine whether beneficial effects observed immediately after mCIMT would be sustained at 2-week and 6-month follow-ups. Results showed that three of five outcomes (PMAL How Well and How Often scales and TAUT) decreased slightly at 2 weeks but were improved relative to posttest data at 6 months. This finding suggests that gains measured at posttest were not only sustained but also improved 6 months after mCIMT.

These findings should be cautiously interpreted because of the following limitations. First, because the child was physician referred for participation in mCIMT, the findings may have been biased in favor of a positive outcome because this child was viewed as likely to benefit. Second, the use of measures without known psychometric properties (PMAL and TAUT) may lead to errors in data. The fact that all measures except the TAUT were consistent in showing positive results, however, suggests that the findings are believable. Third, the findings of sustained performance over the 6 months following mCIMT may have been influenced by continued standard care, occasional use of the cast, and maturation. The fact that substantial improvements after mCIMT were preceded by stability across the two baseline measures suggests that maturation is not a plausible explanation for changes observed immediately after mCIMT. Moreover, amount of parent involvement in home-based intervention was not quantified during this study and may also have accounted for positive changes observed. Finally, the findings have limited clinical generalizability because only one child participated.

Showing that a modified version of CIMT is positively associated with upper-limb function for a 12-month-old child in this study provides new evidence suggesting that mCIMT may be an effective form of intervention for younger children with hemiplegia. However, little is known about severity subgroups of children (or adults) with hemiplegic cerebral palsy who are most likely to benefit from mCIMT. Moreover, little is known about the optimal frequency, duration, and intensity of treatment; type of treatment; or the type of restraint device that is most effective. It appears that even a modest increase in therapy intensity and casting is associated with a positive effect. Future research with larger samples and longer follow-up periods is needed to address these issues for clinicians to make effective decisions about using mCIMT with young children. ▲

Acknowledgments
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References


