

Letters to the editor

'Clinical experience of constraint induced movement therapy in adolescents with hemiplegic cerebral palsy—a day camp model'

SIR—Children and adolescents with hemiplegic cerebral palsy (CP) have difficulty using their affected hand for skilled activities. It has been widely considered that various components contribute to decreased hand function, such as spasticity, weakness of the muscles, dysfunction of sensorimotor abilities, and the existence of mirror movements. Perception and cognition are also highly related to hand function but less investigated.¹ Thus the ability to use the hemiplegic hand is multifactorial. Little is known about the effect of therapeutic treatment of hand function in children and adolescents with hemiplegic CP, and there exist few intervention studies and ideas describing such training.²

Constraint induced movement therapy (CI therapy) is a recently developed treatment approach for adults after stroke. The neurological injury usually leads to depression in motor and perceptual function that is considerably greater than will eventually be the case after spontaneous recovery of the insult. It has been hypothesized that this is due to a 'learned non-use' phenomenon involving the actual use of the hemiplegic hand.³ One reason why individuals do not use their affected limb relates to the fact that there has been repeated evidence that using the hand has resulted in failure. To overcome learned non-use and improve function, it has been suggested that the well-functioning extremity needs to be restrained. A number of studies using CI therapy have been carried out, most of which have involved restraining the well-functioning upper extremity for a period of approximately two weeks, for 90% of the waking hours, while intensively training the affected arm. Results have demonstrated improvement that has remained stable months after intervention has ended.³ Only a few case studies and one randomized study of children with hemiplegic CP have examined this concept, all indicating a positive treatment outcome.^{4,5,6} These studies have not specifically examined groups of adolescents with congenital lesions resulting in hemiplegic CP, or whether they could gain increased hand function by using CI therapy. The hypothesis of learned non-use may not be of the same significance for children and adolescents with early brain lesion as it is for stroke patients, but the idea of restraining the dominant hand may well induce increased performance for these children and adolescents.

In more recent years therapists have been guided by a therapeutic approach based on motor learning theories. This suggests that motor behaviour emerges through multiple systems interacting within unique task and environmental contexts.⁷ To overcome motor dysfunction, repeated practice and experimentation with varied strategies and tasks are needed to find the optimal solutions to motor problems and to develop performance skills. It has been argued that through such repetition an individual's ability to perform tasks develops over time.⁸ Although the concept has been received favourably, it has rarely been utilized for intervention studies.

Our interest, therefore, was in exploring the effectiveness of treatment based on principles of motor learning and CI

therapy, by using an adapted model of CI therapy for a group of nine participants with hemiplegic CP aged 13 to 18 years (Appendix I). The participants were enrolled at a day camp for 7 hours a day, 5 days a week, over a two week summer period. During these 7 hours the participants wore a glove-like splint on the well-functioning hand which prevented them from being able to grasp. The camp programme, based on principles of motor learning, consisted of encouraging daily recreational activities which took place in a favourable and motivating setting. The activities included frisbee golf, basketball, boules, baseball, volleyball, canoeing, water games, and various fine motor skills games, including board, dice, and card games. The participants were also engaged in the preparation of meals as well as washing up and all food intake was performed with the hemiplegic hand. In addition, manipulative exercises were performed every morning. Activities were chosen based on the assumption that through activity performance, important aspects of hand function, like speed, precision, and manipulation, were exercised. Secondly, activities were chosen on the basis of the idea that skills and task performance improve in relation to practice.⁸ This latter statement was especially utilized for practising frisbee golf and in an in-hand manipulation shift task. These two activities were offered on a regular basis in conjunction with the hypothesis: 'you learn what you practice.'⁸ All activities at the camp were performed in a group which enabled the participants to support and collaborate with each other when they encountered difficulties. Similarly, they had each other to turn to for moral support when needed. Assessments were conducted before and after the day camp and at 5 month's follow-up (Appendix I).

After the day camp hand skills had improved in almost all measurements (Table I). Dexterity improved, demonstrated by decreased time to move objects of different size and characteristics (Jebsen Taylor Hand Function Test). The median decreased by approximately 20 seconds, which indicates that the participants learned to grasp and release objects more easily. Improved results of subtest 5 in the Bruininks-Oseretsky Test demonstrated increased coordination and precise movement in time. Likewise, both tasks measuring in-hand manipulatory skills improved, which demonstrated that the participants were more capable of using differentiated finger movements than before intervention. Motor skills used in common daily activities measured by Assessment of Motor and Process Skills demonstrated small but significant improvements. The grip strength, however, did not improve. To a great extent the improvements were sustained at 5 month's follow-up (Table I). The number of throws when playing frisbee golf were noted on the first and last day of the day camp (practised on 7 occasions). Interestingly, the participants made great progress in the game, a completely novel task when performed with the hemiplegic hand. This indicates that participants can learn what they practice, even with a hemiplegic hand.

By restraining the dominant hand, the participants were forced to use their hemiplegic hand more frequently and in a different manner than they would usually have done. The increased performance indicated that this intervention was indeed successful for participants with fairly mild hand dysfunction. Interestingly, this is at an age where the hand dysfunction may be considered to be in a persistent condition and it is, therefore, common practice to diminish frequency of treatment.⁹ Given this, it was surprising that such a short

intervention period resulted in sustained improvement of several measured parameters. The overall findings in the current study matched the results of earlier CI therapy research of stroke patients³ and case reports of children with hemiplegic CP.^{4,5,6}

The intervention programme used for this study was somewhat modified from the contemporary use of CI therapy. The participants were allowed to use their dominant hand as a supporting hand, but with no possibility for grasping which was prevented by the glove-like splint. The time using the splint was reduced compared with contemporary CI therapy, with the participants only wearing the restraining splint during the day camp. We were unsure if it would be possible to persuade them to wear it in their home settings as well; after the camp ended our opinion was that it might have been possible as the splint was not as frustrating to the children as had been anticipated. We did not use shaping as an operant-movement conditioning approach, where objectives are approached in small steps of progressively increasing difficulty, in the strict application of CI therapy.² However, the activities during the day camp were chosen to challenge the participants' abilities. The framework for intervention was based on principles of motor learning, principles that to our understanding, seem to be somewhat broader than shaping.

One of the questions we asked was whether manipulative tasks could be learned through practice in participants with hemiplegic CP. Manipulative tasks involve independent finger movements and in-hand manipulation, requiring extensive motor control. This is known to be deficient in all children with hemiplegia. With this in mind, two specific tasks, the shift

task and the rotation task, were assessed before and after treatment (Appendix I). The shift task was systematically and repeatedly practised during the day camp (10 to 15 minutes each day). Results revealed that the participants had improved in both the shift task and the rotation task after the day camp. But at follow-up, the results were only maintained for the trained shift task and not for the rotation task. The result suggests that task-specific training (at the shift task) was superior to general training and demonstrates the possibility of improving in-hand manipulation, which is not commonly reported.

There are few studies investigating conventional treatment of hand function in children with hemiplegic CP and the available studies show weak results.² Improvement of hand function is commonly related to the effect of hand surgery and, to some extent, botulinum toxin. Therefore, improvements relating to CI therapy are interesting. Although the results are promising, only a small group of individuals was enrolled at the camp. All but one were considered to have mild impairments. Repeated baseline measurements were not attained as this started as a clinical project. The participants' own experiences of the treatment outcomes were not specifically evaluated. However, at the post-assessment session, they informally described their own experiences and it was apparent that they were pleased with the changes that occurred. Comments were made such as: 'It means a lot, I feel more secure' and 'The hand function is better, which is an important feeling, I now know I can use it [that hand]'. We should note that several factors have contributed to these results, such as training in carefully selected activities, the effect of a group intervention, and the participants' own motivation to undergo training. We believe, however, that restraint of the dominant hand may be seen as a potentially important agent for improving hand function.

Table I: Results of intervention

	Measurements	Before intervent.	After intervent.	5-month follow-up
Upper limb coordination	Bruininks-Oseretsky Test, subtest 5 (points)	13 (11–17)	16 ^a (12–19)	16 ^b (12–19)
Dexterity	Jebsen-Taylor Hand Function Test (s)	72.5 (45.5–112)	49.3 ^a (36–108)	50 ^a (32–96.5)
Strength	Gripping, sustained strength (N)	130 (57–241)	128 (63–251)	135 (47–264)
Motor skills	AMPS (logits)	1.9 (1.2–2.3)	2.0 ^b (1.4–3)	1.9 (1.2–2.3)
Manipulation	Rotation task (points)	1 (0–3)	2 (0–4) ^b	1 (0–4)
Manipulation	Shift task (points)	3 (0–7)	6 (0–8) ^b	4 (0–8) ^b
Task performance	Frisbee golf ^c (throws)	20 (14–35)	14 (12–18) ^a	

Median (min – max) are reported for different assessments. Wilcoxon matched-pair test was employed to measure difference between scores before intervention and after intervention, as well as difference in scores before intervention and at follow-up. ^a $p < 0.01$; ^b $p < 0.05$. ^cResult after a total of 7 practice occasions. At 5 month's follow-up assessment of frisbee golf could not be performed, because of winter weather. Intervent, intervention. N, Newton.

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Appendix I: Participants and assessment measures

Participants

Eight participants with mild hemiplegic CP and one with moderate hemiplegic CP. All participants attended mainstream schools. Participants were motivated to participate by applying themselves

Design

Assessments took place before intervention, after intervention, and at 5 month's follow-up

Assessments

1. Subtest 5 of Bruininks-Oseretsky Test of Motor Proficiency,¹⁰ modified so that unimanual items were executed with hemiplegic hand (maximum score, 21)
2. Jebsen Hand Function Test,¹¹ evaluated by time (s) it takes to accomplish tasks with hemiplegic hand
3. Sustained grip strength during 10 seconds was measured using electronic measuring device Grippit¹² (AB Detektor, Gothenburg, Sweden)
4. Assessment of Motor and Process Skills:¹³ a standardized observation measuring individuals' occupational performance in daily life. Tasks were scored in accordance with manual. Only motor skill item was reported in this report
5. Two tasks investigating in-hand manipulation using Exner's¹⁴ classification, rotation task (turning small pegs 180° using a pegboard), and shift task (moving a pencil linearly after it has been grasped with finger pads, from point to top of pencil). Through use of video analysis, rotation task was scored on a rating scale from 0 to 4 and shift task from 0 to 8
6. Frisbee golf is a game in which the goal is to transverse a 350 foot long course, ending with a frisbee basket, in fewest number of throws. Amount of throws needed to get frisbee in frisbee basket was recorded

'Use of selective serotonin reuptake inhibitors in children with Pervasive Developmental Disorder: risk of treatment emergent mania'

SIR—We read with interest the recent paper by Delong et al.¹ in which he describes the treatment of 129 children with Pervasive Developmental Disorder (PDD) with fluoxetine. Such large studies are important and help to clarify the role of selective serotonin reuptake inhibitors (SSRIs) for young people with PDD. Delong et al. identified a subgroup with above average intelligence and a family history of bipolar disorder who responded particularly well on several behavioural measures. It was noted that five of the children developed bipolar disorder during follow-up, though it was not clear whether they were in the treatment responsive group.

A recognized side effect of treatment with SSRIs is precipitation of mania. In adults, the scale of the risk has been found to be between 3.7%² and 17%³ though there are no figures for children and adolescents. A family history of bipolar disorder is also a recognized risk factor for developing mania. In effect, Delong et al. are proposing treating a high risk group with a mania inducing agent.

Diagnosis of mania in children with PDD is notoriously difficult. In the large study by Wozniak et al.⁴ they found irritability and hostility (rather than elation) to be the most typical presenting symptoms. Delong and colleagues do not make any mention of this potential risk, although they note that a limiting factor for tolerance of fluoxetine was almost always behavioural activation and/or irritability or agitation. The possible link between fluoxetine use and the five children who developed mania is discounted as direct induction due to the time delay between treatment initiation and symptom development. However, more complex models of causation, with multiple factors interacting over time, are increasingly being recognized in psychiatry.

In summary, our conclusion is that greater vigilance is required when using SSRIs in this group of children who are already genetically vulnerable to developing mania, and to whom another known risk factor for mania is being introduced. Because mania may not be recognized in this group due to its atypical presentation, quantitative as well as qualitative changes in behaviour need to be looked for and assessed.

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'DeLong replies'

SIR—We appreciate the letter from Drs Bates and Wilson. We are in complete agreement regarding the possibility of precipitating mania by using selective serotonin reuptake inhibitors (SSRIs) in patients with bipolar diathesis. We also think this is an important side-effect limiting SSRI treatment in some autistic children. This is one of several factors emphasizing the similarities between autism and major affective disorder/bipolar disorder.

Any difficulties caused by such escalation during treatment with SSRIs can be minimized by discussing at the outset the possibility of increased activation or agitation resulting from SSRI treatment, so the medication can be stopped promptly if such side-effects occur. In some cases, the SSRI is so beneficial that it is thought worth while to add a mood-stabilizer to the SSRI to permit the continued use of the latter.

However, we would suggest that one cannot be sure that every instance of hyperactivity induced by SSRIs represents the precipitation of mania. As one of the early proponents of the concept of juvenile bipolar disorder in the US,¹ I agree with the difficulty of diagnosing mania in children.

The five children who developed bipolar disorder during follow-up had all been SSRI responders, treated for more than 3 years.

We agree that great vigilance is required when using SSRIs in the autistic spectrum group of children. We try to discuss all aspects of the problem with families before instituting such trials. The marked beneficial effect in some children, sustained over years, in our judgment, makes the effort worthwhile.

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Erratum

'Folic acid and prevention of birth defects'

Van Dyke et al.

DMCN Vol 44: 426–29

The primary author of the above mentioned annotation wishes to point out the following corrections to the article:

p 427, para 5, line 6 should read: 'Increased folic acid intake could potentially reduce the prevalence of some types of congenital heart disease.'³⁴

p 428, para 3, line 4 should read '... heart disease. Individual differences in specific enzymes involved with folic acid metabolism and associated amino acid pathways may potentially be risk factors for NTD and possibly other abnormalities. Polymorphisms of specific enzymes, particularly MTHFR, have been linked to specific metabolic effects.'⁵⁹

References:

- p 426, para 5, line 9: delete ref. 7.
p 427, Table I: delete refs. 37 and 51; add ref. 56
p 427, para 1, line 9: add ref. 41.
p 427, para 8, line 9: change ref. 24 to 50
p 427, para 4, line 11: add ref. 10

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