

CASE STUDY

Effect of modified constraint-induced movement therapy on lower extremity hemiplegia due to a higher-motor area lesion

KENJI NUMATA¹, TAKASHI MURAYAMA², JUN TAKASUGI², & MASARU OGA¹

¹Department of Physical Therapy, Ibaraki Prefectural University of Health Science, Ibaraki, Japan and ²Graduate School of Medicine, Department of Integrative Neurophysiology, Chiba University, Chiba, Japan

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Abstract

Background: Modified CIT (mCIT) was performed in a stroke patient with a lesion in the left medial frontal region including the supplementary motor area (SMA), which is part of the higher-motor area, and whose primary symptom was motor ignition difficulty observed at the start of voluntary movement of the right leg.

Case study: mCIT was performed from awakening to bedtime (not including bath time) with his non-affected limb fixed with a knee splint while he was in the hospital ward. Two days (the total time he wore the appliance was 19.5 hours) after the intervention was introduced, voluntary movement of the right leg occurred, and functional improvement was observed.

Conclusions: These findings are speculated to be related to the facts that the unilateral SMA strongly contributes to movement of the ipsilateral limb and that the plasticity of the SMA, which is a higher-motor area, is greater than that of primary areas. It is probable that different regions of the brain have different plasticity, resulting in differences in the process of functional recovery and the level of recovery.

Keywords: Rehabilitation, constraint-induced movement therapy, motor ignition difficulty, supplementary motor area

Introduction

Wolf et al. [1] performed so-called constraint-induced movement therapy (CIT), a new type of rehabilitation therapy in which paralysed upper limb of chronic patients with hemiplegia due to stroke was exercised intensively while the movement of their non-paralysed upper limb was constrained and reported that the therapy is effective for functional improvement of patients with mild-to-moderately severe motor upper extremity paralysis. In addition, Levy et al. [2] reported that, as a result of comparison between activated areas of the brain before and after CIT intervention using functional magnetic resonance imaging, cortex including the primary motor area (M1) around the lesion, in which no activation had been observed before intervention, the contralateral M1 to the affected limb and

bilateral supplementary motor areas (SMA) were activated in the functional recovery process of the paralysed upper limb. It has been reported that such functional recovery of paralysed limbs by CIT is based on the plasticity of the brain as reflected in phenomena including reorganization of the neural circuits of the damaged brain [2–5].

During CIT intervention, patients experience a great deal of mental and physical distress due to restricted movement of the non-paralysed arm. Currently, therefore, modified CIT in which the tasks and period of intervention are modified (mCIT) to decrease such distress experienced by patients is commonly performed [6–9]. Meanwhile, earlier studies on the efficacy of CIT mainly dealt with stroke patients with relatively mild spastic paralysis of the upper limb. As there have been few reports on the effectiveness of CIT on the lower

limbs [10], validation of this approach requires more research. One of the purposes of this study was to examine the efficacy of CIT specifically for lower limb treatment, although a variety of symptoms are observed in patients with post-stroke dysfunction. This study performed mCIT in a post-stroke patient who had motor ignition difficulty of the lower limb, which was associated with lesions in the medial frontal region including SMA (i.e. the higher motor area) and resulting impaired movement such as walking impairment and investigated its effect on functional improvement.

Case report

The subject was recruited from a community-based, free-standing inpatient rehabilitation unit in an acute care hospital. The subject of this study was a patient with hemiplegia due to stroke who fully understood the purpose and content of this study and gave written informed consent. The patient was a 72-year-old, right-handed man. He had no significant past medical history. He was enrolled in this study on day 14 after cerebral infarction.

Although weakness of his right upper and lower limbs and difficulty in walking were observed at the time of onset, no disturbance of consciousness was observed and physical therapy was started on the day of the consultation admission. He received bedside physical therapy in the acute ward for the first 2 weeks. He then continued this therapy in the hospital facilities for physical therapy after being transferred to the rehabilitation ward. He was discharged from hospital on day 78 after the stroke.

MRI findings

Fluid attenuated inversion recovery (FLAIR) imaging on the day of onset showed a high density area in the medial frontal area of the left cerebral hemisphere from the anterior cingulate gyrus (Figure 1(a)) to the sub-cortical supplementary motor area (Figure 1(b)). In addition, multiple lacunar infarcts were observed in the left and right sub-cortical regions.

Neurological/neuropsychological findings

The muscle tone of his right upper and lower limbs (paralysed limbs) on day 14 after the stroke showed moderate hypotonia and slightly increased deep tendon reflexes. Superficial sensation was slightly decreased; deep sensation was normal. Trömner and Babinski responses were present; toe grasping response was absent. Grasp response and instinctive grasp response [11] were present in the right arm. Brunnstrom motor recovery stages [12] (BR stages)

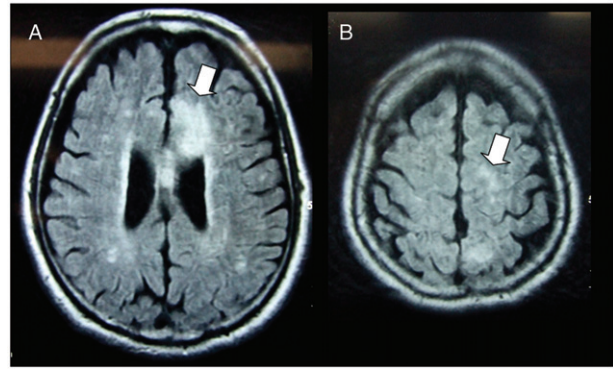


Figure 1. Flair imaging on day of onset: (a) anterior cingulate gyrus; (b) lesion in the sub-cortical region of the supplementary motor area (arrowheads).

were used to determine the severity of hemiparesis. BR stages indicating the severity of hemiparesis are as follows: Stage 1, flaccid: no apparent muscle tone. Stage 2, spastic stage: presence of muscle spasticity or associated movement. Stage 3, synergy stage: presence of stereotyped of motor synergy. Stage 4, movement deviating from the basic, synergies: begin to break stereotyped of motor synergy. Stage 5, relative independence of the basic synergies: selective movement of different joint is adequate. Stage 6: normal or near normal. According to the BR stage, the level of paralysis of both upper limbs and fingers was 6. Assessment of the legs according to BR stage criteria was not possible because there was no voluntary movement in the hip, knee or ankle joint. His consciousness was clear; although neuropsychological findings showed slight verbal perseveration, good communication was maintained and the Folstein mini-mental state test score was 25/30 [13]. Agnosia, apraxia and aphasia were not observed. No limitation of the range of motion was observed in any joint.

Function of lower limbs

In this patient, no voluntary movement of the hip, knee and ankle joint of the right leg (paralysed limb) was observed in the lying, sitting or standing position or during transitional movements between the positions. When in the standing position, he was unable to bear weight on the right leg (paralysed limb). However, although he had difficulty in initiating the first step, once he started walking, movement of the right leg occurred and he was able to walk in a straight line without a walking aid device and both the stance phase and the swing phase of his right and left legs were symmetrical, indicating a normal walking pattern. When he started walking, the first step was initiated by the left leg (non-paralysed limb), which was followed by the swing of

the right leg (paralysed limb). When he tried to turn to the right from walking straight, steps of his right leg stopped, interfering with the continuity of walking. He had no difficulty turning to the left. Despite the absence of any obvious motor paralysis, motor ignition difficulty was apparent in the lower limbs at the start of walking and the patient had difficulty initiating walking. Once started, however, it became possible to keep walking. This is a phenomenon called 'gait apraxia' [14, 15]. In addition, other symptoms, such as cessation of walking, are observed when patients such as the present case change direction to either the right or the left. Therefore, the present case could be considered to have right unilateral gait apraxia.

Course of physical therapy until mCIT intervention

On the day before mCIT intervention, neurological/neuropsychological tests were conducted and voluntary movement and movement of the right leg during walking were examined; the results of those tests were consistent with the test results obtained on day 14 post-stroke. On the day of onset, the patient was started on physical therapy, which was administered for 6 days a week (40 minutes a day). Physical therapy consisted of conventional physical therapy procedures such as weight bearing on the right leg in the standing position, balance training, walking exercise and range of motion exercise and Bobath-based treatment [16]. After the start of physical therapy, he started walking exercise using parallel bars on day 3 post-stroke, walking exercise on crutches on day 16 post-stroke and walking exercise without a stick under supervision on day 21 post-stroke. In all types of walking, no movement of the right leg was observed at the start of walking and he was at risk of falling due to tripping. Bobath-based treatment was performed mainly to cause voluntary movement of the right leg, but no effect was observed. He was able to walk around the hospital ward on his own using a walking frame, which is associated with a low risk of falling. He was also able to perform self-care such as eating, grooming, changing his clothes, bathing and toileting.

Methods of intervention

mCIT intervention was introduced on day 68 after the start of physical therapy. The mCIT intervention period had been set so as to start 5 days earlier. When the patient himself or three physical therapists confirmed any change in right lower limb function, however, the intervention was to be terminated at that point. A knee splint (Nippon Eizai Co., Ltd.) was applied to the left knee, i.e. the



Figure 2. Knee splint (Nippon Eizai Co., Ltd.) used for modified constraint-induced therapy (mCIT). The appliance was placed on the left leg (non-paralysed limb) of the patient (the photograph shows the appliance placed on a healthy person).

non-paralysed limb of the patient (Figure 2). Before starting mCIT intervention, a safety evaluation was conducted by three physical therapists while the patient wore the knee splint in the hospital ward. The therapists determined that there was no risk of falling although, with the knee splint, he suffered a slight loss of balance when standing up from a chair or bed and during transfers. When walking (with a walking frame), a reduction in speed due to shortened step length was observed and it took the patient slightly longer to turn around in a narrow space. However, there were no safety problems. Thus, it was determined that the patient's daily performance during the mCIT intervention period could be carried out without monitoring. He was supposed to wear the appliance from 6:30 (awakening time) to 19:30; he did not have to wear it during bath time. He was only instructed to maintain the same lifestyle as before mCIT intervention and no additional movements or tasks were imposed. During the intervention, he wore the appliance and underwent the same physical therapy programme as before, except for range of motion exercise. However, when fatigue or pain beyond physical and/or mental limits resulted from wearing the appliance, he was allowed to remove it temporarily.

Assessment before and after intervention

In addition to BR stage and neurological tests before and after the mCIT intervention, the patient's performance was video-taped and evaluated. In the physical therapy facility, single joint movements of the hip, knee and ankle joints of the right lower limb in sitting, recumbent and standing positions were recorded. Co-ordinated movements of these multiple joints (voluntariness, speed of movements and the range of motion), transitional movements from the recumbent to the standing position, walking (walking straight, turning to the right or the left) and others were also recorded. Furthermore, in the ward, routine daily activities (movements on the bed, transfer to a toilet or other places, transfer by walking, etc.) were also video-taped. Three physical therapists evaluated voluntariness and automatism of the right limb and the appropriateness of movements and then compared the results before vs after the intervention.

Results

On the evening of day 2 of mCIT, the patient reported movement of his right leg; three physiotherapists confirmed voluntary movement and terminated the intervention. Based on the patient's record, the total time he wore the appliance was 19.5 hours (day 1 of mCIT: 11 hours, day 2 of mCIT: 8.5 hours) (Table I). The patient reported physical and mental distress due to application of the appliance on day 1 of mCIT; he paid more attention to the affected limb and gradually became aware of functional improvement from the morning of day 2 of mCIT (Table I).

Among functional changes in his right leg observed after the intervention, voluntary control of hip, knee and ankle joint movements, both separately and jointly, became possible. The scope of these joint movements covered the full range of motion in recumbent, sitting and standing positions. On the other hand, a slight delay in initiating a motion and a minor reduction in speed were recognized as compared to the left (non-paralysed) limb. However, since these motions and actions were observed to be at the same functional level as those of the non-paralysed leg, a classification of BR stage 6 was made. In addition, autonomous movement of his right leg, associated with a normal movement pattern, occurred during transitional movement between those positions. Weight bearing on the right leg (paralysed limb) in the standing position became possible, difficulty in swinging his right lower limb forward at the start of walking resolved and voluntary walking became possible. In addition, halting of walking of the right leg, which was observed when turning to the right while walking, resolved and continuous walking become possible. In the ward, appropriate voluntary and/or autonomous right leg movements appeared in all of the patient's motions with both a normal movement pattern and stability. Since he was able to walk anywhere without a walking aid device, it was determined that he was at the self-walking level. In general, all of his motions were very close to the normal levels and no differences from healthy individuals were recognized.

No changes were observed in the results of neurological tests such as deep tendon reflexes, superficial sensation, deep sensation and pathological reflexes. Grasp response and instinctive grasp

Table I. Patient's record during modified constraint-induced therapy (mCIT) intervention (actual descriptions given by the patient). He reported physical and mental distress due to the splint applied to the knee on day 1; he paid more attention to the right leg and was beginning to be aware of the occurrence of movement on day 2.

Time		Activities and movements/self-observation
<i>Day 1</i>		
6:30 (waking up) to 13:00	○	<ul style="list-style-type: none"> • Skin regions where the splint was applied felt very hot. • I went out of the hospital with a visitor. • I had difficulty standing and sitting. • I was unable to stand up without bearing weight on my right leg.
13:00 to 15:00	×	
15:00 to 19:30 (going to bed)	○	
<i>Day 2</i>		
7:00 (waking up) to 14:00	○	<ul style="list-style-type: none"> • I was constantly very conscious of my right foot. • I was able to walk with the tip of my right foot pointing forward. • Bathing: when I got into the bathtub, I was able to put my right leg in first without difficulty. • It was extremely troublesome and a heavy burden to wear the appliance on the right foot. • I can tolerate it for 2 more days at most; I think I will tolerate it longer if I realize the actual effect after the 2-day period.
14:00 to 14:30	×	
14:30 to 16:00 (removal)	○	
The total time I wore the appliance on day 2 was 19.5 hours		

○: I used the splint, ×: I did not use the splint.

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response of the right arm were positive, with no improvement. Improved function was maintained until discharge from the hospital, on day 8 after the completion of mCIT (day 78 post-stroke).

Discussion

Difficulty in initiating voluntary movement of the patient's right leg and gait disturbance were not improved immediately before mCIT (day 68 after the start of physical therapy). However, although he complained of physical and mental distress due to the intervention with knee immobilization on day 1 after starting mCIT, he paid more attention to the affected limb and was beginning to be aware of the occurrence of movement on day 2 of mCIT intervention. The functional improvement resulting from the short-term intervention was inconsistent with the temporal functional changes after the onset of stroke and was considered to be due to the mCIT intervention.

No neurological sign or paralysis indicating impairment of the corticospinal tract was observed in his upper and lower limbs from the onset. In an experiment with monkeys, SMA cells were activated when initiating motions intentionally or carrying out multiple motions sequentially [17, 18]. Based on this result, these cells have been considered to be related to higher-order aspects of motor control. Damage of the medial surface of the frontal lobe including the SMA causes no or only slight motor paralysis, but is associated with signs and symptoms of the upper limbs such as alien hand signs [19, 20] and an instinctive grasp response [11], which occurs irrespective of the patient's will, as in this patient. The main symptom in this patient was the absence of voluntary movement of the ankle, knee and hip joint, which interfered with transitional movements. At the start of walking, difficulty in initiating the first step of the right leg was observed. However, although normal gait was observed once walking straight was initiated, walking stopped when he tried to turn to the right. Gait apraxia occurs in patients whose bilateral SMAs are impaired [14, 15]. Although he was able to walk in a straight line once walking was started, motor ignition difficulty of the right leg was observed and walking stopped when making a turn [15, 21]; therefore, this patient was considered to have right unilateral gait apraxia due to a lesion of the left SMA. It has been confirmed that the bilateral SMAs are activated before the start of walking [22] and during walking [23, 24] in healthy persons. Goldberg et al. [19] reported that a lower motor sub-routine is executed irrespective of the patient's

will when the SMA is damaged. Therefore, walking of patients with gait apraxia is considered to be executed by inferior centres responsible for locomotion [25, 26] such as the mesencephalic locomotor region and the ponto-bulbar locomotor region. Both the instinctive grasp response of the right arm and motor ignition difficulty of the right leg observed in this patient were due to damage of the SMA and were considered abnormalities of high-level voluntary motor control.

In this patient, functional improvement was achieved by intervention over a very short period (2 days); although this may have been because the damage of the SMA was located in the sub-cortical area, it may have been because of specific properties of the SMA as a higher motor area. It has been suggested that the unilateral SMA strongly contributes to movement of the ipsilateral limbs, because in simple movement of unilateral limbs, the SMA of the hemisphere opposite the limbs is activated, whereas the bilateral SMAs are activated in complicated movements [18]. In addition, it has been reported that the bilateral SMAs as well as primary motor and pre-motor areas and cerebellum in the hemisphere opposite the lesion are activated in the process of motor recovery from paralysis using CIT intervention [2, 3, 27]. From these findings, it was speculated that the SMA plays an important role in functional improvement and that in this patient the SMA of the contralateral side contributed to the functional improvement. In addition, Pons et al. [28] reported that the monkey's repair capacity after cortical damage is greater in higher-order areas than in primary areas. It has been reported that the transmission efficiency of synapses in the cerebral cortex is changed by a change in the stimulation frequency in a short period [29] and that neuronal activities in the primary motor area [30] and parietal area [31] are changed immediately after new motor learning. The motor recovery achieved in this patient in a short time may have been due to the facts that activities of synapses and neurons in the brain have high plasticity and that the medial surface of the frontal lobe including the SMA as a higher motor area has a high repair capacity.

This patient was discharged from hospital on day 8 after mCIT intervention; until then functional maintenance was confirmed. However, functional changes after discharge from hospital could not be followed because of ethical reasons associated with the study. Still, it is reported that function will improve further after CIT [32, 33]. That is probably because the improved functions are used more frequently in daily life.

The brain has various structures, which constitute complex and diverse nerve networks. It is probable that such different regions of the brain have different

functional activity and repair capacity, resulting in differences in the process of functional recovery and the level of recovery.

Watson et al. [34] reported that the s-shaped curve (plotting functional recovery against time, namely, after a gradual rise in the early onset stage, the curve rises sharply during the convalescent stage and then reaches a plateau) appears with recovery of specific motor tasks, as is implicitly acknowledged by a majority of clinicians, may not necessarily be the true or only a model of recovery, because there are some patients who achieve a fresh 'burst' of renewed progress at a much later stage. Recovery is possible, regardless of the s-shaped curve, when therapy is specifically tailored to the factors unique to each patient's impairment or recovery potential. The remarkable recovery observed in the present case is attributable to mCIT being ideally suited to the causes of the impairment. In the future, the restorative effect of CIT on different affected areas needs to be confirmed in individual cases.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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