



## Constraint-induced movement therapy in a patient with ataxia after cerebellar tumor resection

*Terapia por contensão induzida em paciente com ataxia após ressecção tumoral cerebelar*

*Terapia por contención inducida en paciente con ataxia tras la resección tumoral cerebelar*

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### Abstract

**Introduction:** Constraint-induced movement therapy (CIMT) is a very modern method used in rehabilitation to treat individuals with functional impairment of the upper limbs. **Objective:** To apply the CIMT method and evaluate its effects in a patient with ataxia after removal of a cerebellar tumor. **Method:** This is a longitudinal interventional study of the case report type. The participant complained of tremors and difficulty writing and typing with the right hand as a result of the cerebellar tumor's removal. She was subjected to CIMT daily in three-hour sessions for 10 days. The participant was evaluated with the Motor Activity Log (MAL) and the Wolf Motor Function Test (WMFT). **Results:** There was an improvement in the quantity, quality, speed, accuracy and strength with which the participant performed the tasks according to the MAL and WMFT scales. Improvement in writing was also identified and the report of better use of the member for daily tasks demonstrated the patient's satisfaction after the end of therapy. **Conclusion:** This case report

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demonstrated the benefits of the application of the CIMT method in a patient with ataxia as a consequence of a rare cerebellar tumor, improving task execution time, strength and functional improvement of the affected upper limb.

**Keywords:** Neurological Rehabilitation. Ataxy. Astrocytoma.

### Resumo

**Introdução:** A terapia por contensão induzida (TCI) é um método atual de reabilitação com intuito de tratar indivíduos com perdas funcionais nos membros superiores. **Objetivo:** Aplicar o método TCI e avaliar seus efeitos em uma paciente com ataxia após a remoção de tumor cerebelar. **Método:** Trata-se de um estudo longitudinal de caráter intervencionista do tipo relato de caso. A participante queixava-se de tremor e dificuldade de escrever e digitar com a mão direita em consequência da retirada de um tumor cerebelar. Foi tratada segundo método de TCI por três horas diárias durante 10 dias e avaliada pela Motor Activity Log (MAL) e Wolf Motor Activity Test (WMFT). **Resultados:** Houve melhora na quantidade, qualidade, velocidade, exatidão e força com que a participante executava as tarefas de acordo com as escalas MAL e WMFT. A melhora na escrita também foi identificada e o relato de melhor uso do membro para tarefas cotidianas demonstrou satisfação da paciente após o final da terapia. **Conclusão:** Esse relato de caso demonstrou os benefícios da aplicação do método TCI em uma paciente com ataxia como seqüela de um tumor cerebelar raro, o que proporcionou melhora no tempo de execução de tarefas força e ganho funcional no membro afetado.

**Palavras-chave:** Reabilitação Neurológica. Ataxia. Astrocitoma.

### Resumen

**Introducción:** La terapia por contención inducida (TCI) es un método actual de rehabilitación con el propósito de tratar individuos con pérdidas funcionales en los miembros superiores. **Objetivo:** Aplicar el método TCI y evaluar sus efectos en una paciente con ataxia después de la remoción del tumor cerebeloso. **Método:** Se trata de una investigación longitudinal de carácter intervencionista del tipo informe de caso. Una participante se quejaba de temblor y dificultad para escribir con la mano derecha, como consecuencia de la remoción de un tumor cerebeloso. Se la trató según el método de terapia inducida por tres horas diarias durante 10 días e, después de la evaluación del registro de actividades motoras Motor Activity Log (MAL) y Wolf Motor Activity Test (WMFT). **Resultados:** Se ha mejorado la cantidad, calidad, velocidad, exactitud y en la fuerza con que la participante desarrollaba sus tareas de acuerdo con las escalas MAL y WMFT. También se identificó una mejora en la escritura, además del relato de mejor uso del miembro para tareas de la vida cotidiana, lo que demuestra la satisfacción de la paciente después del fin de la terapia. **Conclusión:** Este relato de caso demuestra los beneficios de la aplicación del método TCI en una paciente con ataxia como secuela de un tumor cerebeloso raro, lo que proporcionó una mejora en el tiempo de ejecución de tareas, fuerza y proporciono provechos funcionales en el miembro afectado.

**Palabras clave:** Rehabilitación Neurológica. Ataxia. Astrocitoma.

### Introduction

Constraint-induced movement therapy (CIMT) is a motor and neurobehavioral approach used in rehabilitation to improve the upper limb's

function in patients with asymmetric use [1, 2]. The CIMT is based on the neuroplasticity theory and cortical reorganization. Studies have shown the effectiveness of this therapeutic intervention due to improvements in the functional performance of

the impaired upper limb in children with cerebral cancer, cerebral palsy or spinal cord injury. In adults, this effectiveness was proven in the treatment of the upper limbs of stroke victims and patients with general dysfunctions of the central nervous system [1, 3-9].

The current CIMT protocol requires total adherence on the part of patients and time availability to finish the treatment protocol, which is comprised by three stages. The first stage, called “shaping and task practice”, involves the repetitive training of tasks, three hours per day during two consecutive weeks. “Shaping” is a repetitive task that composes or simulates part of a functional activity, while “task practice” consists of a complete functional activity. In the second stage, the healthy upper limb is restricted by a glove for 90% of the waking hours. The third stage, called “transfer package”, involves the adherence-enhancing behavioral methods and transfers the gains obtained to the real world [3, 10].

The “transfer package” is essential for this technique and was developed to insert the CIMT in the patient’s daily activities by monitoring the impaired upper limb’s use during the therapy’s period. The third stage is comprised by (1) maintenance of a diary and/or detailed descriptions of the impaired upper limb’s use in activities of daily living, and (2) resolution of problems by introducing assistive and/or movement modification devices to improve the impaired upper limb’s use [3, 11].

Cerebellar ataxia is characterized by a group of disorders related to changes in gait and coordination resulted from dysfunction of the cerebellum and associated systems due to genetic inheritance or acquired causes, which leads to considerable disability. Among the causes of cerebellar ataxia are surgical lesions for ablation of tumors such as pilocytic astrocytoma [12, 13].

There are still few strategies for the neurological rehabilitation of cerebellar ataxia. Most strategies involve the intensive training of tasks focused on coordination, balance, posture, gait and activities of daily living, including conditioning programs, range of motion exercises and muscle strengthening [14, 15]. In addition to the traditional approach, non-invasive brain stimulation techniques and stochastic whole-body vibration therapy are also reported in the literature as effective [16, 17].

This study aims to apply for the first time the CIMT method in a patient with ataxia after resection of

cerebellar pilocytic astrocytoma and to evaluate its post-therapy functional performance.

## Methods

This is a longitudinal interventional study of the case report type, carried out at the School of Physical Therapy and Occupational Therapy (FFTO/UFPA). The participant was female, 42 years old and right-handed, with an undergraduate degree and employed as a laboratory technician.

The onset of the symptoms was reported in August 2016, with demand for emergency medical care, including periorbital edema, nausea, headache and hypertension. After her condition had been stabilized, a neurological evaluation was carried out, indicating normotonia (Ashworth 0) and normoreflexia (deep and superficial). In addition, muscle strength (MRC4+/5), range of motion (ROM), coordination and balance were preserved. Gait and appendicular ataxia, dysarthria, dysmetria, tremor and nystagmus were not identified. The superficial and deep sensitivity of all limbs was preserved except for the right upper limb, which showed paresthesia. Intense bouts of headache and nausea with no apparent cause were also recurrent. Nuclear magnetic resonance imaging revealed a solid tumor measuring 2.5 cm in the right cerebellar hemisphere. Given the family history of the patient, whose mother died due to a brain tumor, and the clinical presentation, the surgical procedure was indicated. The surgery was performed in November 2016 with total removal of the tumor, which was taken for histopathological analysis and characterized as grade 1 cerebellar pilocytic astrocytoma, with a high rate of glial cell proliferation.

In the immediate postoperative period, the patient developed nausea and ataxia of the right hemisphere, especially of the extremities, and could not stand due to severe gait ataxia as well as dysarthria. In the 3 months after the postoperative period, the patient showed normotonia (Ashworth 0), normoreflexia (deep and superficial), muscle strength (MRC4+/5) and normal joint range of motion (ROM). Superficial and deep sensitivities were both preserved for all limbs. There were no further complaints of headache, nausea, dysarthria, or ataxic gait. Eye movement disorders were not identified. Coordination and balance remained compromised after surgery due to dysmetria and tremor of the upper right limb, making

some tasks requiring manual dexterity and accuracy harder, including writing and typing, pointed out as the patient's primary complaint. A slight right ankle tremor was also identified, but not involving movement or walking limitations.

### Instruments and procedures

In our research, evaluations were made with specific instruments for the CIMT method, all validated for the Brazilian Portuguese language. Before the instruments' application, the participant was submitted to the Mini Mental State Examination (MMSE) to assess her cognitive function, and the score obtained was 26. As inclusion criterion for the CIMT, it was necessary to have a score higher than the cut-off point according to education level in the MMSE [18].

Frequency and quality of daily use of the most affected upper limb were both assessed with the Motor Activity Log (MAL), and motor ability was evaluated with the Wolf Motor Function Test (WMFT). In addition to the scales, writing ability was also evaluated before and after the CIMT by having the patient sign her name.

Balance and gait were previously evaluated according to the Tinetti scale, in which the patient's score was 28, indicating low risk of falls and not compromising the exclusive exercises for the upper limb in the CIMT.

In the initial contact, the application of the technique and the study were explained, and the patient agreed to be submitted to the therapy and research protocol by signing a consent form, in which she was informed about all the risks, benefits and procedures.

The patient was evaluated in three moments. In the first one, general evaluation and screening for the CIMT were performed. In the second one, before the intervention, and in the third one, after the intervention, evaluations were performed with the MAL, WMFT and signature records.

The MAL is a structured interview that assesses how much and how the individual uses the most affected upper limb outside the therapeutic environment. The participant was asked standard questions about the amount of use of her arms (QT – Amount Scale or AS) and the quality of their movements (QL – How Well Scale or HW) during the indicated functional activities [19].

The scales are printed and put before the patient so that the score can be assigned, which could vary from 0 to 5, considering that: 0 = no use of the affected upper limb to perform a given task; 1 = very rare use; 2 = rare use; 3 = use for half the time the task was performed; 4 = regular use (often); and 5 = balanced use of the upper limb (normal). The total score was calculated from the arithmetic mean of these values [20, 21]. These scales were presented to the participant and doubts were clarified in advance so that she could choose the appropriate score according to the observed patterns.

The WMFT is a test developed to evaluate the benefits of CIMT in individuals with stroke sequelae [22, 23]. It aims to assess the motor skills of patients with moderate and severe motor impairment of the upper limbs.

The protocol contains 17 tasks, performed in order of difficulty (gross to fine motor skill), progressing from the proximal to the distal joints (shoulder to finger). The WMFT is divided into three parts: (1) time (the speed with which the task can be completed), (2) functional ability (the quality of movement when the task is completed) and (3) strength (ability to lift weight). All tasks are filmed and timed.

The final score is obtained from the mean time of the task's completion. The maximum time allowed for each task is 120 seconds. In our study, the test was filmed and later analyzed by two evaluators, who assigned scores for each task performed by the participant.

### Intervention

Before each intervention day, the MAL items were applied every other day, with 15 items on one day and 15 items on the other. The participant was instructed to use the most affected member on days when there was no intervention (weekend) and report the task in a diary, including difficulties and their overcoming.

The intervention consisted of training the affected extremity with 10 shaping tasks chosen based on the patient's needs, 5 having been performed per day. The chosen shaping tasks were consistent with their methodological concept, including tasks that can be evaluated by complexity and according to parameters such as counting time or number of repetitions within a preset time [21].

Shaping is a training method based on the principles of behavioral training, where the motor

goal is achieved in small steps [19]. Several studies indicate that the tasks are flexible in relation to the increase of the level of difficulty and complexity; their components are modified throughout the protocol according to the established progression parameters and the difficulty of each task varies according to the patient's improvement [20, 21, 24, 25]. For this, it is important to define how the task will be analyzed, establishing its parameter of knowledge of the results and which movements should be focused on [26].

At the end of each session, the patient was challenged to perform a functional task, which was

also evaluated in a simpler way and with fewer processes for more complex situations. At times, the participant was assisted by the therapist to perform the steps which she was unable to complete by herself, and verbal feedback was provided with each improvement in the tasks' performance. In this method, this activity is called Task Practice. It is a structured technique, which involves functional activities that are carried out integrally, in a period from 15 to 30 minutes [19]. In this case, the activities of setting the table, playing checkers or dominoes and painting another person's nails or doing their make-up were involved (Table 1).

**Table 1** – Description and progression parameters of Shaping tasks and Task practice

| Shaping                                 | Description (30 s/attempt)  | Progression parameters  |
|---|---|---|
| 1. Turning pieces                       | Turning as many domino pieces as possible on a table  | Number of pieces turned, size and weight of the pieces  |
| 2. Placing pieces in a box              | Checkers pieces were placed in a row on a table and the participant had to put them in a box  | Number and size of pieces, height and positioning of the box and distance between the box and the participant |
| 3. Stacking pieces                      | Stacking three domino pieces  | Number of stacks and type of pieces   |
| 4. Transferring pieces                  | Transferring circles in a string from one end to the other  | Number of circles, height of string and size of circles   |
| 5. Writing                              | Writing a letter or a word on a sheet of paper  | Number of letters, words or phrases   |
| 6. Transferring grains                  | Transferring grains from one container to another with a spoon  | Number of transfers, distance between containers and type of spoon  |
| 7. Transferring water                   | Transferring glasses with water from a bowl to the sink   | Number of transfers, distance between bowl and sink, height and type of cup                                   |
| 8. Transferring balls of dough          | Transferring balls of play dough from one plate to another with a fork  | Number of transfers, distance and height between the plates and the fork's position                           |
| 9. Typing                               | Typing a numerical sequence on a calculator   | Number of digits and size of the calculator's keys  |
| 10. Hanging clothes                     | Hanging clothes with as many preachers as possible  | Number of preachers, distance between them and the participant and type of preacher                           |
| Task practice                           | Description   |   |
| 1. Setting the table                    | The participant should set the table, arrange the dishes on the table and cut and serve food, always using the most affected member                     |   |
| 2. Playing a game of checkers/ dominoes | The participant should play a match of checkers or dominoes, making it necessary for her to handle small pieces with varied weights                     |   |
| 3. Painting someone's nails             | The participant should paint the nails of another person trying to have maximum coordination of the movement of the more affected upper limb            |   |
| 4. Doing someone's make-up              | The participant should do another person's make-up, with the utmost dexterity and precision to perform the task (applying eyeliner, shadow and mascara) |   |

## Results

After the intervention with the CIMT method, there was improvement in the quantity, quality, speed, accuracy and strength with which the participant performed the tasks. The participant

started to use the right upper limb more and with better quality of movement, including for previously unrealized activities, such as answering the telephone, brushing teeth, putting makeup on, picking up a cup by the handle and eating food with her hands.

Table 2 shows the time spent in each WMFT task before and after the CIMT. In no task the patient exceeded the 120 s needed for failing. There was a reduction in the execution time of several tasks, which led to a 2.14 s reduction in relation to the total average time the patient took to complete the WMFT tasks.

There was improvement in the MAL scores in both the quantity (QT) and quality (QL) of the most affected upper limb's movement, reflecting an increase in the mean of the total score from 2.83 to 4.3 points and from 2.63 to 4.65 points, respectively, values that are closer to normal (Table 3).

**Table 2** – Time of execution of the WMFT tasks before and after CIMT

| TASK                        | PRE-CIMT | POST-CIMT |
|-----------------------------|----------|-----------|
| Forearm on the table        | 0.97 s   | 1.03 s    |
| Forearm on the box          | 1.12 s   | 0.91 s    |
| Elbow extension             | 1.08 s   | 0.84 s    |
| Elbow extension with weight | 1.91 s   | 1.31s     |
| Hand on the table           | 0.88 s   | 0.59 s    |
| Hand on the box             | 1.13 s   | 0.97 s    |
| Reaching and withdrawing    | 1.00 s   | 0.81 s    |
| Lifting a can of soda       | 2.94 s   | 2.41 s    |
| Picking up a pencil         | 2.57 s   | 2.39 s    |
| Picking up a paper clip     | 4.67 s   | 5.93 s    |
| Stacking pieces             | 42.97 s  | 18.69 s   |
| Turning cards               | 13.12 s  | 10.06 s   |
| Turning a key               | 4.66 s   | 3.73 s    |
| Folding a towel             | 8.66 s   | 5.28 s    |
| Lifting a basket            | 3.07 s   | 3.75 s    |
| MEAN                        | 6.05 s   | 3.91 s    |

**Table 3** – Results of the MAL before and after CIMT

| ACTIVITY                       | MAL QT   |           | MAL QL   |           |
|--------------------------------|----------|-----------|----------|-----------|
|                                | PRE-CIMT | POST-CIMT | PRE-CIMT | POST-CIMT |
| Flipping the light switch      | 3        | 5         | 3        | 5         |
| Opening a drawer               | 0.5      | 4.5       | 3        | 4.5       |
| Removing an item from a drawer | 0        | 4         | 0        | 4.5       |
| Picking up a phone             | 0        | 4         | 0        | 3.5       |

(To be continued)

**Table 3** – Results of the MAL before and after CIMT

| ACTIVITY                            | MAL QT   |           | MAL QL   |           |
|-------------------------------------|----------|-----------|----------|-----------|
|                                     | PRE-CIMT | POST-CIMT | PRE-CIMT | POST-CIMT |
| Cleaning a kitchen conter           | 3        | 5         | 4        | 4.5       |
| Getting out of a car                | 4        | 5         | 4        | 5         |
| Opening a refrigerator              | 5        | 5         | 4        | 5         |
| Opening a door with the knob        | 3        | 5         | 3        | 4.5       |
| Using a remote control              | 1        | 4         | 1        | 3.5       |
| Washing hands                       | 5        | 5         | 4        | 5         |
| Opening and closing taps            | 5        | 5         | 4        | 5         |
| Drying hands                        | 5        | 5         | 4        | 5         |
| Putting on socks                    | 5        | 4         | 4        | 4         |
| Taking soff socks                   | 5        | 4.5       | 4        | 4.5       |
| Putting on shoes                    | 5        | 4.5       | 4        | 4         |
| Taking off shoes                    | 5        | 4.5       | 4        | 4         |
| Getting up from an armchair         | 5        | 5         | 4        | 5         |
| Pulling a chair to sit on it        | 5        | 5         | 4        | 5         |
| Pulling a chair closer to the table | 5        | 5         | 4        | 5         |
| Picking up a glass or bottle        | 1        | 3         | 1        | 3.5       |
| Brushing teeth                      | 0        | 3         | 0        | 3         |
| Applying makeup, lotion or cream    | 0        | 3         | 0        | 3         |
| Picking up a key to lock the door   | 4        | 5         | 3        | 4.5       |
| Writing on paper                    | 1        | 2.5       | 1        | 3         |
| Carrying an object                  | 4        | 5         | 3        | 4.5       |
| Using a fork or spoon               | 3        | 4         | 3        | 4         |
| Brushing hair                       | 2        | 4         | 3        | 4         |
| Grabbing a cup by the handle        | 0        | 4         | 0        | 2.5       |
| Buttoning up a shirt                | 0,5      | 2.5       | 3        | 2         |
| Eating snacks with hands            | 0        | 4         | 0        | 4         |
| MEAN                                | 2.83     | 4.3       | 2.63     | 4.65      |

There was considerable improvement in the motor characteristics used in the patient's writing, before and after the CIMT, as shown in Figure 1. In signature "A", there was less control of tremor, generating circular movements with low amplitude that overlapped in the tracing of the letter. Better control over tremor could be noticed in signature "B", the features of which were firmer and without overlapping. As this was one of the patient's main complaints, the method applied contemplated her expectation.



**Figure 1** – Signature of the patient before and after the CIMT.

The evaluation of muscle strength consisted of two parameters of evaluation of the WMFT itself. One item consisted in evaluating the amount of load attached to the wrist with which the patient could lift her arm and position her hand on a box without compensating with the rest of her body, while the other consisted in the evaluation of grip force using a manual dynamometer. In both tests, increase in muscle strength was identified, according to Table 4. This finding implies an improvement in the ability to load and transfer objects with the affected arm.

**Table 4** – Evaluation of muscle strength before and after CIMT

| TEST                               | PRE-CIMT    | POST-CIMT   |
|------------------------------------|-------------|-------------|
| Functional strength (arm on box)   | 2.5 kg      | 4.5 kg      |
| Palmar grip strength (dynamometer) | 63.0 pounds | 68.6 pounds |

## Discussion

There are still few studies with effective strategies for rehabilitation of patients with cerebellar ataxia. [16] This study shows for the first time the application of CIMT in a patient with cerebellar ataxia as sequela of pilocytic astrocytoma resection. The effects of the application of the CIMT for three hours over two weeks demonstrate functional improvement in several aspects such as quality and quantity of movement,

palmar grip strength and decrease of the execution time of the WMFT tasks. Although other alternatives may be used to treat these disorders, some of them do not follow published protocols or require expensive equipment and complex management [16, 17]. On the other hand, CIMT, in addition to its low cost, already has consistent evidence of its effectiveness in several clinical conditions [1-10].

The MAL demonstrated differences both quantitatively and qualitatively in relation to the affected upper limb. Of the 30 MAL tasks, eight that were previously classified as "almost normal" were considered "normal", demonstrating greater functionality of the patient's limb. In the MAL item named "taking clothes out of a drawer", it was noted that classification went from "never done" to "normal". In addition, a relevant item added in the evaluation before and after application of the CIMT, the patient's signature, was also assessed, demonstrating an improvement in coordination during writing.

Doussoulin et al. [3] applied modified CIMT both in its collective and individual modalities for three hours during ten days, to promote the functional independence of patients with upper limb weakness caused by a stroke. The study concluded that both modalities generated functional independence. In a meta-analysis [27] where the effect of bilateral and unilateral upper limb training was assessed, evaluating the function and activities of daily life after a stroke, it was shown that CIMT is more effective than bilateral training for increasing the upper limb's functional capacity.

In the study by Ji & Lee [28], virtual reality was associated with CIMT for the rehabilitation of patients with stroke sequelae, being effective for recovering the function of these patients' upper limbs, offering yet another very modern feature that can be used concomitantly with the TCI. Batool et al. [29] showed that the CIMT's application promoted a statistically significant improvement in motor function and self-care of the hemiplegic upper limb when compared to other therapies in patients aged 35-60 years old.

Many studies have shown the benefits of applying CIMT in patients with cerebral palsy to improve the affected limb's function, but it is also significantly efficient when compared to other rehabilitation methods [6, 30-32]. The study by Sparrow et al. [5] performed with children with brain tumor sequelae, showed beneficial results both qualitatively and quantitatively in the movement of the trained upper

limb; in addition, the gains were maintained in the follow-up evaluations for three months.

CIMT promotes cortical plasticity and reorganization, and several studies in humans demonstrate that this neural reorganization can be facilitated in rehabilitation with repetitive training, practice of specific tasks, sensory training, and practice of mental activity [33]. In the case of cerebellar lesions, it is known that the cerebellum has a high degree of plasticity, even in adult life, which occurs due to changes in its architecture and to the new connections established in a process of synaptic reorganization [34]. However, our results show that the 10-day CIMT performed three months after the postoperative period was able to promote significant functional improvement.

The present study also had limitations, such as the intervention having been conducted in only one patient and without a control group. This fact can be explained because the occurrence of pilocytic astrocytoma is rare in adults: the incidence rate in this population is 4.8 per million inhabitants per year [12, 35]. To minimize this limitation, all evaluated parameters were compared before and after 10 days of CIMT.

The current CIMT protocol applied to functional limitation caused by cerebellar ataxia resulted from surgical ablation is potentially effective, but more studies with a larger number of patients and with other types of ataxia that may benefit from this therapy are needed to provide the desired functional improvement.

## Conclusion

This case report revealed that the application of the CIMT method in a patient with ataxia caused by the resection of a rare tumor in the cerebellum was efficient and improved task execution time, strength, writing and functional improvement of the affected upper limb.

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