

Contralateral force irradiation for the activation of tibialis anterior muscle in carriers of Charcot-Marie-Tooth disease: effect of PNF intervention program

Irradiação contralateral de força para a ativação do músculo tibial anterior em portadores da doença de Charcot-Marie-Tooth: efeitos de um programa de intervenção por FNP

Paula C. Meningroni¹, Carolina S. Nakada¹, Luciana Hata¹, Amanda C. Fuzaro¹, Wilson Marques Júnior², João E. Araujo¹

Abstract

Objective: To evaluate the response of the tibialis anterior (TA) muscle following a five-week protocol with contralateral irradiation force through Proprioceptive Neuromuscular Facilitation (PNF) diagonals in patients with demyelinating polyneuropathy associated with Charcot-Marie-Tooth disease type 1A (CMT-1A). **Methods:** The study included 12 patients of both sexes. They were treated twice-weekly for 5 weeks. At each session, they performed the following diagonal patterns: chopping, extension-adduction with internal rotation (EAIR) and flexion-abduction with internal rotation (FAIR). The diagonals were repeated four times, in both upper and lower limbs, with each repetition lasting six seconds on average. During execution, the response of the TA muscle was recorded by a surface electromyograph disregarding the initial and final two seconds of each diagonal. The mean RMS values of the four repetitions were normalized in percentage. The initial and final data were analyzed through the *t* test for paired samples with significant *p*-values <0.05. **Results:** The contralateral force irradiation with the chopping diagonal to the left and to the right increased the percentage RMS values of the TA muscle in the last session when compared with the values of the first session (*t*=-3.94 and *t*=-3.87, respectively). Similarly, the EAIR diagonal increased the percentage RMS values of the TA muscle in the last session when compared with the values of the first session (*t*=-3.3 and *t*=-4.58, respectively). The only diagonal that did not produce higher values of contralateral force irradiation in the TA muscle, left and right, was the FAIR (*t*=-2.31 and *t*=-1.55). **Conclusion:** These results may justify the use of a treatment program with PNF diagonals in patients with CMT-1A who have difficulty activating the TA muscle.

Key words: Charcot-Marie-Tooth Disease; proprioceptive neuromuscular facilitation technique; peripheral demyelination; electromyographic evaluation.

Resumo

Objetivo: Avaliar a resposta do músculo tibial anterior (TA) após um protocolo de cinco semanas com irradiação contralateral de força através de diagonais de facilitação neuromuscular proprioceptiva (FNP) em pacientes com polineuropatia desmielinizante associada à doença de Charcot-Marie-Tooth do tipo 1A (CMT-1A). **Métodos:** Participaram deste estudo 12 pacientes, de ambos os sexos. Eles foram tratados em uma frequência de duas vezes por semana, durante cinco semanas. Em cada sessão, foram utilizadas as diagonais de Chopping, extensão-adução com rotação interna (EARI) e flexão-abdução com rotação interna (FARI). As diagonais foram repetidas quatro vezes, em ambos os membros superiores e inferiores; cada diagonal tinha duração média de 6 segundos. Durante as execuções, a resposta muscular do TA foi registrada por um eletromiógrafo de superfície, desprezando-se os 2 segundos iniciais e finais de cada diagonal. A média dos valores de Root Mean Square (RMS) das quatro repetições foi normalizada em porcentagem. Os dados iniciais e finais foram submetidos ao teste em *t* para amostras pareadas com valores de *p* significativos <0,05. **Resultados:** A irradiação de força contralateral, através da diagonal de Chopping à direita e à esquerda, aumentou os valores de RMS em porcentagem do músculo TA na última sessão quando comparados com os valores da primeira sessão (*t*=-3,94) e (*t*=-3,87), respectivamente. Na mesma direção, a diagonal EARI aumentou os valores de RMS em porcentagem do músculo TA na última sessão quando comparados com os valores da primeira sessão (*t*=-3,3) e (*t*=-4,58), respectivamente. A única diagonal que não produziu valores maiores de irradiação de força contralateral nos músculos TA direito e esquerdo foi a diagonal FARI (*t*=-2,31) e (*t*=-1,55). **Conclusão:** Esses resultados podem justificar a utilização de um programa de tratamento através de diagonais de FNP em portadores de CMT-1A que possuam dificuldades na ativação do músculo TA.

Palavras-chave: doença de Charcot-Marie-Tooth; técnica de facilitação neuromuscular proprioceptiva; desmielinização periférica; avaliação eletromiográfica.

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¹ Laboratory of Bioengineering, Neuropsychological and Motor Behavior, Department of Biomechanics, Medicine and Locomotor Rehabilitation, Faculty of Medicine of Ribeirão Preto, Universidade de São Paulo (FMRP/USP), Ribeirão Preto (RP), Brazil

² Department of Neuroscience and Behavioral Sciences, FMRP/USP

Correspondence to: João Eduardo de Araujo, Laboratório de Bioengenharia, Neuropsicobiologia e Comportamento Motor, Departamento de Biomecânica, Medicina e Reabilitação do Aparelho Locomotor, FMRP/USP, Av. dos Bandeirantes, 3.900, CEP 14049-900, Ribeirão Preto (SP), Brazil, e-mail: arauoje@fmrp.usp.br

Introduction

Charcot-Marie-Tooth disease (CMT) is the most prevalent hereditary peripheral neuropathy, affecting one in every 2500 people in the world^{1,2}. CMT has variations, with 70% of all cases being type 1A (CMT-1A). The disease is an autosomal dominant demyelinating polyneuropathy associated with a DNA duplication of chromosome 17³⁻⁶. CMT-1A produces typical functional changes, such as reduction in the conduction speed of the peripheral nerve⁷⁻¹⁰, hypo- or areflexia, sensitivity loss, distal muscle atrophy and subsequent progressive functional disability, which accompanies the evolution of the disease¹¹⁻¹³. The lower limbs (LL) of patients with CMT are the most affected, and one of the characteristic changes is foot drop, which is a result of a muscular atrophy of variable severity. Foot drop in CMT-1A is primarily generated by a weakness of the tibialis anterior (TA), but with significant contribution of weakness of the extensor digitorum longus and extensor hallucis longus^{14,15}. Despite the importance of dorsiflexion during normal gait and despite the fact that foot drop is a common characteristic of CMT, no proposals were found in the literature for the treatment of this type of pathology¹⁴.

Muscle biopsies of patients with CMT-1A submitted to a 12-week strengthening protocol showed that, with an adequate protocol, there will be muscular adaptation to strength training¹². Aitkens et al.¹⁶ and Kilmer et al.¹⁷ demonstrated that low- and moderate-intensity strengthening protocols (with weights on the fist and ankle, 12 weeks of treatment with three weeks in submaximal regime) in patients with neuromuscular diseases, including CMT-1A, improve upper and lower limb muscle strength¹⁶. In contrast, an intensive strengthening protocol with progressive training frequency, volume and, especially, intensity (12 weeks, one set of ten repetitions progressing to five sets, and three times progressing to four times a week) increases the risk of training-induced injury in patients with neuromuscular disease¹⁷. Accordingly, as demonstrated by Lindeman et al.¹⁸, it seems that the low to moderate-intensity strengthening protocols (three times a week with weights adapted to the strength of each patient) are safe for patients with CMT-1A. However, Chetlin et al.¹², in a classic 12-week moderate-intensity strengthening protocol, observed that the female patients with CMT-1A reached 80% of the normal strength in six out of eight exercises; but, in this study, no positive modifications were found in the dorsiflexion muscles.

Several physical therapy methods have been used for LL motor rehabilitation and strengthening. Proprioceptive Neuromuscular Facilitation (PNF) can be mentioned among the most commonly used methods. PNF aims to improve neuromuscular performance by stimulating muscle and joint proprioceptors with muscle force irradiation techniques¹⁹.

According to the irradiation principle, the stimulation of strong and preserved muscle groups produces activation of the injured and weak muscles, facilitating muscle activation²⁰.

However, in spite of the undocumented clinical evidence, the scientific evidence that suggests the effectiveness of this type of treatment is limited regarding strengthening and improvement of LL muscle activation post-demyelinating polyneuropathy, associated or not to CMT-1A disease²¹. Thus, the present study aims to evaluate the response of the TA muscle following a five-week protocol with contralateral force irradiation through PNF diagonals in patients with demyelinating polyneuropathy associated with CMT-1A disease.

Methods

Participants

Twelve patients with Charcot-Marie-Tooth disease took part in this study, including seven men and five women. All of them displayed functional changes in the LL. The participants' age ranged from 22 to 64 years. The subjects were recruited at the Neurogenetics Clinic of the Department of Neurology, Psychiatry and Medical Psychology of Hospital das Clínicas da Faculdade de Medicina de Ribeirão Preto da Universidade de São Paulo (HCFMRP/USP). The pathology diagnosis was carried out by the family history, an electrophysiological test and/or a genetic test. The inclusion criteria were: good cognition, ability to walk with and without aid. The exclusion criteria were: heart arrhythmia, high blood pressure, cardiovascular problems, serious respiratory problems and inability to attend the sessions. The use of medication for the CMT treatment was allowed. The patients who met the inclusion criteria signed the informed consent form to take part in this study.

The project was approved by the Human Research Ethics Committee of HCFMRP/USP, protocol 10354/2007, and registered at the National Database of Ethics and Human Research (SISNEP) under CAAE - 0434.0.004.000-07, as being ethically and methodologically adequate, according to resolution 196/96 of the National Health Council (CNS).

Experimental design

The participants were assembled in a single group and treated with PNF diagonals. The objective was to produce contralateral force irradiation on the tibialis anterior muscle (TA). The participants were assessed during the first session and reassessed during the last session.

Stimulation protocol

The participants were treated with PNF diagonals twice a week for five weeks. The physical therapists who applied the PNF treatment were certified by the International Proprioceptive Neuromuscular Facilitation Association (IPNFA). Each specific diagonal was always applied by the same physical therapist. The consultation and evaluation took place at the Rehabilitation Center (RC) of HCFMRP/USP. The PNF diagonals were chosen according to the potential and functionality of the segments of each patient. The diagonals were: chopping (patient sitting without support, with upper limbs in flexion of shoulder and extension of elbow), extension-adduction with internal rotation (EARI) and flexion-abduction with internal rotation (FAIR; patient in supine position). All the diagonals were carried out to the right and left. The chopping diagonals use the upper limb strength in the diagonal movement to stimulate trunk and lower limb contraction. In comparison, the EARI and FAIR patterns use the strength of one of the lower limbs to activate the lower limb contralateral muscles. During the performance of the diagonals, the patients were asked to push the arms down toward the left/right knee (chopping), push the foot down and out (EARI) and pull the foot up and lift the leg out (FAIR). The maximum tolerable resistance of each patient was used, which enabled the execution of the movement diagonally throughout its range of motion. Each diagonal was repeated four times with a mean duration of six seconds. The rest interval between the diagonals was determined individually for each patient. Each session lasted 40 minutes.

Electromyography analysis

Surface electromyography (SEMG) analyses were carried out through electrodes on the motor points of the TA muscle to quantify the muscle activation response just before the first and last sessions of the five-week protocol with PNF. SEMG was recorded using a four-channel electromyography and bio-feedback system (EMG System do Brasil, São José dos Campos) with disposable bipolar electrodes at a fixed distance. For the collection, the area where the electrodes would be placed was shaved with a disposable razor, exfoliated with a nail file to remove dead cells, and cleaned with alcohol 70%²²⁻²⁴. The electrodes were then placed at the motor point of the TA muscle. All the skin preparation and electrode placement procedures followed the recommendations of the European SENIAM project (Surface Electromyography for the Non-Invasive Assessment of Muscles)²⁵. The EMG signal was collected during the execution of the PNF diagonals. Because each diagonal had a mean duration of 6 seconds, the initial and final two seconds

of each collection were discarded. The software DATAQ Instruments Hardware Manager was used for data acquisition.

Statistical analyses

Because the diagonals were repeated four times in each segment, the mean of the root mean square (RMS) values for each diagonal was considered (RMS execution 1+ RMS execution 2+ RMS execution 3+ RMS execution 4 / 4 = RMS of diagonal). The data of the RMS values obtained in the first session were normalized and converted to percentage. Accordingly, the mean RMS of each diagonal at the beginning of the treatment corresponds to 100% of TA muscle activation. The RMS values obtained in the last session were normalized to percentage, considering the values obtained in the first assessment. After normalization, data from the first and last sessions were tested using the *t* test for paired samples.

Results

Data analysis showed an increase in percentage RMS values in the last session of treatment compared to the first. The contralateral force irradiation, through the chopping diagonal to the right and to the left, increased the RMS values in percentage of TA muscle in the last session when compared to the values of the first session ($t=-3.94$ and $t=-3.87$, respectively; $p<0.05$; Figure 1). Similarly, the EARI diagonal increased the percentage RMS values of the TA muscle in the last session compared to the first session ($t=-3.3$ and $t=-4.58$, respectively; $p<0.05$; Figure 2). The only diagonal that did not produce higher values of contralateral force irradiation in the right and left TA muscles was the FAIR diagonal ($t=-2.31$ and $t=-1.55$, respectively; $p>0.05$; Figure 3).

Discussion

It is well documented that in CMT-1A there is a significant reduction in nerve conduction velocity through the peripheral nerves^{7,8} and that this modification is more pronounced in the LL of the patients¹⁴. This neural modification leads to distal muscle atrophy and inability to walk^{12,13}. In the present study, all patients showed a higher RMS activation in the TA muscle in the final assessment at the end of the PNF protocol. To obtain this result, we used an indirect treatment based on the concept of muscle force irradiation which occurs ipsilateral and contralateral to the performed diagonal^{19,26}. In 1979, Markos²⁷ showed the feasibility of this treatment modality by increasing the EMG activity and the degree of hip flexion in normal individuals. In contrast, the present data refer only to the contralateral force irradiation to the performed diagonal.

The choice of this modality of PNF treatment was based on the evidence that low- and moderate-intensity training increases muscle strength in patients with CMT-1A¹⁶, and that high-intensity protocols can cause muscle injuries in these patients¹⁷. By using the strongest muscles furthest to the target of the force irradiation, muscle injury was prevented in the participants in the present study. During the protocol, upper and lower limb diagonals were used. In these segments, the diagonal patterns were selected according to the greatest ability and potential for strength generation of the participants.

Muscle activation data, obtained through SEMG, are dependent on the biophysical characteristics of the muscles and motor neurons^{28,29}. Although the muscle trophism was not measured before and after the protocol, it is believed that the ten PNF sessions, which activated TA through muscle force irradiation, did not produce a significant increase in muscle trophism, which could justify such results. As demonstrated by Gabriel, Kamen and Frost³⁰ in 2006, the increase in EMG amplitude without hypertrophy is evidence of change in neural conduction to the muscle. In that sense, it is believed that the greater RMS activation of TA, through muscle force irradiation phenomena, may be due to an improvement in peripheral nerve conduction³¹. However, this hypothesis needs further confirmation, because such a phenomenon in this pathology has yet to be described.

Among the patterns used in this study, only the contralateral FAIR pattern was not able to increase the RMS activation values of the TA. A possible explanation for the low contralateral muscle force irradiation is that, in this diagonal, the facilitated pattern would be plantar flexion and not dorsiflexion. As for the other diagonals used in the upper and lower limbs, they were suitable for the production of contralateral dorsiflexion to the resisted movement. However, future investigations are needed in other groups of patients with CMT-1A to identify a biomechanical characteristic that might interfere in the mechanisms of muscle force irradiation, justifying the poorer performance in the force irradiation in this diagonal pattern.

Thus, the present results justify the use of a treatment program through PNF diagonals in patients with CMT-1A who have difficulty in activating the TA muscle. Furthermore, the phenomenon of contralateral muscle force irradiation, through the patterns of PNF diagonals used in this study, is an effective therapeutic tool as described in the method's manuals.

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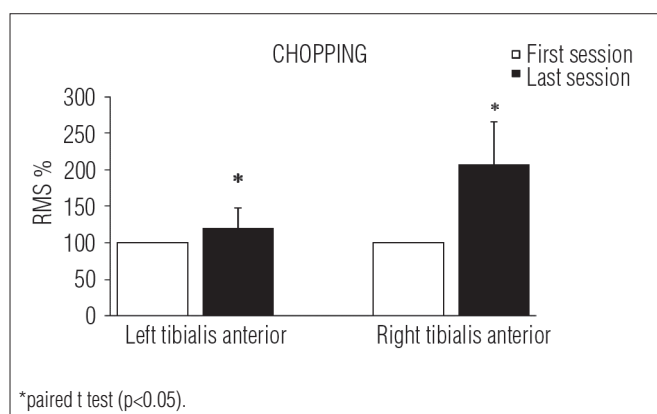


Figure 1. Graphic representation of the percentage RMS values in the left and right tibialis anterior muscle in the first and last session with the chopping PNF diagonal to the left and to the right. The columns represent the mean and the bars represent \pm SEM. Number of subjects = 12.

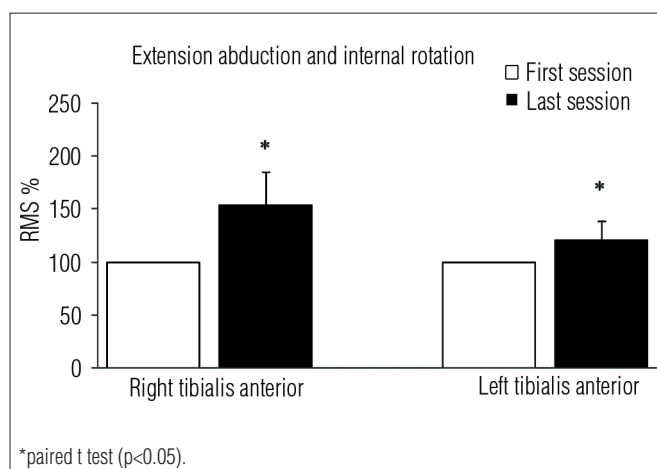


Figure 2. Percentage RMS values in the right and left tibialis anterior muscle in the first and last session with the extension adduction and internal rotation PNF diagonal. The columns represent the mean and the bars represent \pm SEM. Number of subjects = 12.

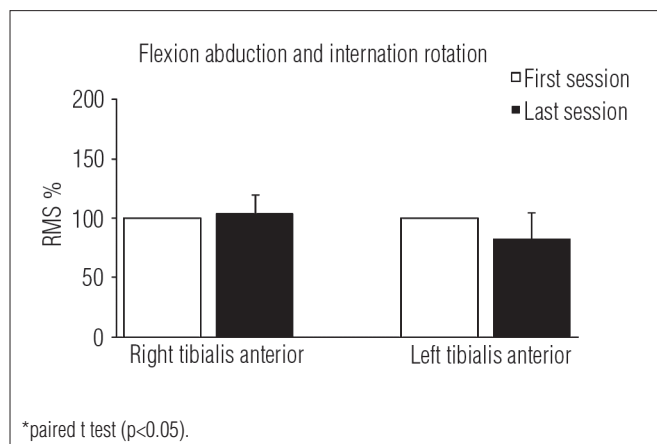


Figure 3. Percentage RMS values in the right and left tibialis anterior muscle in the first and last session with the flexion abduction and internal rotation PNF diagonal. The columns represent the mean and the bars represent \pm SEM. Number of subjects = 12.

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