

# Proprioceptive neuromuscular facilitation in HTLV-I-associated myelopathy/tropical spastic paraparesis

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## ABSTRACT

**Introduction:** Human T cell lymphotropic virus type I-associated myelopathy/tropical spastic paraparesis (HAM/TSP) can impact the independence and motricity of patients. The aims of this study were to estimate the effects of physiotherapy on the functionality of patients with HAM/TSP during the stable phase of the disease using proprioceptive neuromuscular facilitation (PNF) and to compare two methods of treatment delivery. **Methods:** Fourteen patients with human T cell lymphotropic virus type I (HTLV-I) were randomly allocated into two groups. In group I (seven patients), PNF was applied by the therapist, facilitating the functional activities of rolling, sitting and standing, walking and climbing and descending stairs. In group II (seven patients), PNF was self-administered using an elastic tube, and the same activities were facilitated. Experiments were conducted for 1h twice per week for 12 weeks. Low-back pain, a modified Ashworth scale, the functional independence measure (FIM) and the timed up and go test (TUG) were assessed before and after the interventions. **Results:** In the within-group evaluation, low-back pain was significantly reduced in both groups, the FIM improved in group II, and the results of the TUG improved in group I. In the inter-group analysis, only the tone was lower in group II than in group I. **Conclusions:** Both PNF protocols were effective in treating patients with HAM/TSP.

**Keywords:** HTLV-I. Proprioceptive neuromuscular facilitation. Exercise movement techniques. Exercise therapy. Activities of daily living. Myelopathy.

## INTRODUCTION

Human T lymphotropic virus type I (HTLV-I) was first reported in Brazil in 1989 by Kitagawa et al.<sup>1</sup>. This virus has been estimated to be carried by 20 million individuals worldwide<sup>2</sup>. HTLV-I-associated myelopathy/tropical spastic paraparesis (HAM/TSP) is the most frequent neurological condition that has been linked to HTLV-I. It consists of a slowly progressive spastic paraparesis that is associated with neurogenic bladder disturbances and less conspicuous sensory signs. Lower limb weakness, steadily progressing to an abnormal, spastic gait, is the first symptom in approximately 60% of HAM/TSP patients<sup>2</sup>.

Franzoi et al.<sup>3</sup> found gait and sphincter control disturbances to be the most prevalent causes of disability in HAM/TSP patients. Weakness, old age, low-back pain, long disease duration, asymmetric onset of symptoms and spasticity substantially interfere with the ability to walk. Rehabilitation

plays a key role in restoring quality of life by developing the functional ability of each patient<sup>4</sup>.

The proprioceptive neuromuscular facilitation (PNF) technique was used as a therapeutic tool in this study. This method consists of a facilitation process through which therapists guide the patients' movements with adequate manual contraction and appropriate resistance, thus enabling and accelerating neuromuscular mechanical reactions<sup>5</sup>.

To test the hypothesis that functionality in HAM/TSP patients would improve with physiotherapy techniques, a treatment program was undertaken, using PNF associated with two different types of facilitation. In the first type, the therapist facilitated the patterns of functional movement, while in the second type, elastic tubing was used under the therapist's supervision to promote such patterns.

## METHODS

The initial sample included 30 outpatients who were consecutively selected from our neurological outpatient unit. All of the patients were randomly divided into two groups — group I (PNF performed with a therapist) and group II (PNF performed with the aid of an elastic tube) — by drawing lots using the fixed allocation randomization method. Allocation to the control and intervention groups was undertaken using a ratio of 1:1. This study used a convenience sample to

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apply the physical therapy protocols because it was difficult to group patients with HAM/TSP due to the following reasons: 1) patients with motor impairment found it difficult to travel to Physical therapy services two times per week for 20 sessions; 2) many of the patients were constrained because they presented with urinary incontinence; and 3) many did not have a free public transport pass, although they had the legal right to one. The studied sample consisted of 14 patients (7 women and 7 men) because sixteen patients were excluded for various reasons (one patient was in a car accident, 1 was diagnosed with an abdominal aortal aneurysm, 8 could not be transported by anyone, and 6 decided to continue treatment near their homes). All of the patients used medication for spasticity control before and during the study.

The inclusion criteria were as follows: meeting the World Health Organization diagnostic guidelines for HAM/TSP<sup>6</sup>; disease duration of at least one year<sup>7</sup>; classified between 1 and 4 on the modified Ashworth scale<sup>8</sup>; and a score of 7, 6 or 5 on the functional independence measure scale (patients with independent gait with crutches or independent gait under supervision)<sup>9</sup>. The exclusion criteria were as follows: concomitant neurological disease identified by the attending physician; coinfection with HIV; diagnosis of diabetes, alcoholism or orthopedic disease; patients who had undergone physical therapy within six months prior to the protocol; and patients in wheelchairs.

The protocol included assessments by the same physical therapist at three different times. Sixty days after the initial assessment, a similar second examination was performed. Therapeutic interventions were performed immediately after visit two. After twenty sessions, the end-points were checked. All of the assessments were made by one of the authors, and both groups were treated by two different physical therapists, who were thoroughly trained to offer standardized treatment to both groups.

The protocol included a careful case history and examination. The low-back pain of the patients was assessed using a yes/no question. Spasticity was estimated using the modified Ashworth scale<sup>8</sup>. Hip and knee segments (flexion and extension) were assessed, and both sides were compared. When measuring tone, scores 0 and 1 were considered as such; score 1+ was recorded

as 2, score 2 corresponded to 3, and score 3 corresponded to score 4. The functional independence measure (FIM)<sup>9</sup> was estimated using the daily living activity scale. Cognitive and communicative items were excluded because they were beyond the scope of this study. Therefore, the minimum total score was 13, and the maximum score was 91. Balance and mobility were assessed using the timed up and go test (TUG)<sup>10</sup>.

Group I underwent treatment with PNF to facilitate rolling, sit-to-stand movements and walking and stair climbing, and group II was treated with PNF using a 2-meter-long blue elastic tube<sup>11</sup> to facilitate the same functions. The patterns that were used are shown in **Table 1**. Five repetitions, twice each, were performed, and two specific PNF techniques were used in both groups: rhythmic initiation and a combination of isotonic<sup>5</sup>. In group II, the rhythmic initiation was applied by the therapist in a two-stage progression, involving passive and active phases. In the active progression, the patient was alone, and the resistive movement was applied by an elastic tube.

Data analysis (using Sigma Stat software, version 3.11, Systat Software Inc. San Jose. California) was performed at two levels. The first step evaluated whether the disease had progressed from the initial stage to the stage 60 days later within each group (McNemar's test was used for nominal data related to low-back pain, Wilcoxon's test was used for tone, and a paired t test was used for FIM and TUG) and 60 versus (vs.) 60 days between the groups (the Chi-square test was used for nominal data unrelated to low-back pain, the Mann-Whitney test was used for tone, and Student's t test was used for the FIM and TUG) to ascertain whether the groups were homogeneous. In the second step, the number of times of testing, the 60 days and post intra-groups were analyzed (McNemar's test was used for low-back pain, Wilcoxon's test was used for tone, and a paired t test was used for the FIM and TUG). To analyze the time of testing, the post vs. post inter-groups were used (the Mann-Whitney test was used for low-back pain and tone, and Student's t test was used for the FIM and TUG). All of the tests were performed considering a significance level of  $\alpha=0.05$ .

TABLE 1 - The patterns used in the activities that were performed by the two groups.

Activities group I/group II

- 1 - Rolling supine to lateral (patterns of bilateral lower extremity flexion, with knee flexion, for lower trunk flexion-right)
- 2 - Transition from sitting to standing (patterns of bilateral, asymmetrical upper extremity flexion with neck extension-left)
- 3 - Standing on one leg/Therapist in front of the patient\*
- 4 - Weight shift forward and backward/Therapist in front of the patient\*
- 5 - Forward gait/Therapist in front of the patient\*
- 6 - Walking backward/Therapist behind the patient\*
- 7 - Walking sideways/Therapist at the patient's side\*
- 8 - Going up stairs/Therapist behind the patient\*
- 9 - Going down stairs/Therapist in front of the patient\*

\*The facilitation of numbers 3-9 was performed using parallel bars.

**Ethical considerations**

The study was approved by the local ethics committee, and all of the subjects signed an informed consent form (Protocol #134/02).

**RESULTS**

In group I, the mean age at onset was 47.7±14.8 years old, whereas, in group II, it was 50.4±12.4 years old. The average disease duration in group I was 5.9±5.5 years, and that in group II was 6.1±4.2. Groups I and II were similar at baseline, as no significant differences in the variables of age at assessment or duration of disease were detected (p=0.35 and 0.46, respectively).

Seventy-two percent of the subjects reported difficulty in walking, 21% reported difficulty getting on the bus, and 7% reported falls. Ninety-three percent reported lower limb weakness, and 7% reported low-back pain as their first symptom. Low-back pain and sphincter disturbances were frequent symptoms. The groups were similar with regard to walking aids: without support (group I - 57%, group II - 43%), help with a crutch (group I - 29%, group II - 28%) and help with two crutches (group I - 14%, group II - 29%).

When comparing data within and between the initial and 60 days groups, the values for low-back pain, right lower limb tone, the MIF and the TUG test were not statistically significant (Table 2).

A significant reduction in the number of individuals with low-back pain was detected when comparing the 60 days and post-treatment time points in group I (p=0.015), and the same findings occurred in group II (p=0.004, Figure 1).

The statistical analysis in groups I and II showed no significant differences in the tone of the right leg after either treatment (p=0.1 and p=0.07, respectively). However, between the groups, a statistically significant reduction in group II was found (p=0.01). In the left lower limb, groups I and II showed

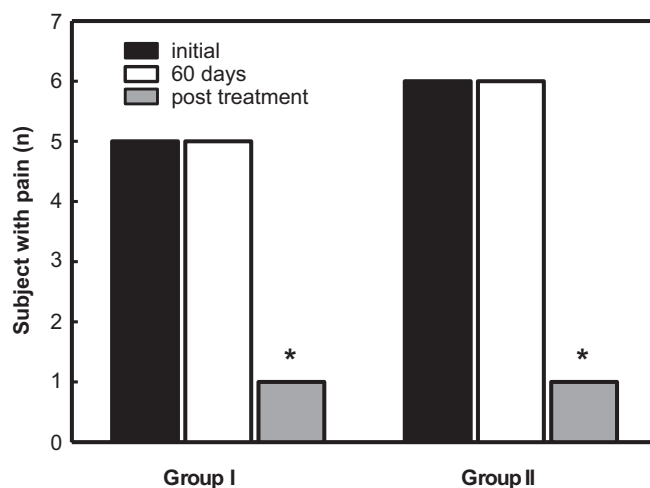


FIGURE 1 - Numbers of patients in groups I and II who had pain. Group I: performed with a proprioceptive neuromuscular facilitation (PNF) therapist. Group II: PNF performed with the aid of an elastic tube. \*statistically significant difference between post-treatment and initial treatment and after 60 days in groups I and II.

no significant differences after either treatment (p=0.15 and 0.84, respectively).

The comparison between groups showed no statistically significant differences (p=0.51).

The FIM parameter at 60 days was not different post-treatment for group I (p=0.17); however, this difference was statistically significant in group II (p=0.01). Inter-group analysis showed no significant differences (Figure 2).

Analysis of the TUG test, performed at the 60 days vs. post times in group I, showed a statistically significant difference (p=0.047), but this difference was not observed in group II (p=0.10). In the inter-group analysis, no significant differences were observed (p=0.45). The results are displayed in Figure 3.

TABLE 2 - Data concerning the initial stage of statistical analysis.

Variables	Initial versus 60 days		60 days versus 60 days
	Group I=7	Group II=7	Group I versus Group II
Low-back pain (number of patients)	5/7 versus 5/7 p=1.0	6/7 versus 6/7 p=1.0	5/7 versus 6/7 P=0.25
Right lower limb tone	3 (1-4) versus 3 (1-4) p=1.00	2 (1-4) versus 2 (1-4) p=1.00	3 (1-4) versus 2 (1-4) p=0.52
Left lower limb tone	3 (1-4) versus 3 (0.7-9) p=1.00	2 (1-4) versus 2 (1-4) p=1.00	3 (1-4) versus 2 (1-4) p=0.88
Functional independence measure	81.7±1.32 versus 81.3±1.3 p=0.2	80.14 ±1.37 versus 81.14±1.33 p=0.5	81.3±1.3 versus 81.14±1.33 p=0.94
Timed up and go test	24.1±3.7 versus 23.5±3.9 p=0.54	27.3±7.8 versus 25.9±7.9 p=0.48	23.5±3.9 versus 25.9±7.9 p=0.80

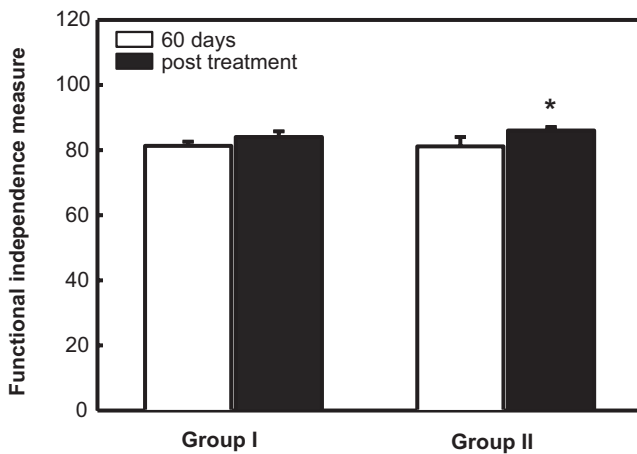


FIGURE 2 - Evaluation of the functional independence measure (mean and standard deviation). Group I: performed with a proprioceptive neuromuscular facilitation (PNF) therapist. Group II: PNF performed with the aid of an elastic tube. \*statistically significant difference between post-treatment and after 60 days for groups I and II.

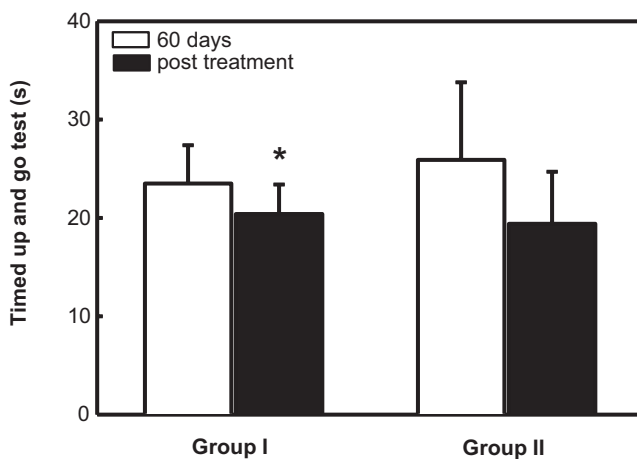


FIGURE 3 - Evaluation of the timed up and go test (mean and standard deviation). Group I: performed with a proprioceptive neuromuscular facilitation (PNF) therapist. Group II: PNF performed with the aid of an elastic tube. \*statistically significant difference between post-treatment and 60 days for groups I and II.

## DISCUSSION

Studies on the contributions of different PNF techniques in HAM/TSP are not available. Specific programs for sensory-motor functional rehabilitation constitute well-proven, effective approaches for neurological deficits that are secondary to HTLV-I<sup>12</sup>. Lannes et al.<sup>13</sup> proposed scales to support physiotherapeutic assessments and kinesiotherapy strategies that aimed at functional rehabilitation and gait improvement in HAM/TSP patients. Some of these scales were used in the present study.

The motor disability of patients with HAM/TSP and low-back pain significantly interferes with daily activities and

can lead to functional limitations<sup>14</sup>. Initial weakness, mostly proximal, and lower limb spasticity occur, followed by low-back pain, sphincter disturbances and balance impairment. These symptoms limit activities of daily living, such as transfers and walking up or downstairs<sup>2-4,15</sup>. Drug approaches have been limited to symptomatic treatment, while physical therapy can attenuate the effects of motor myelopathy<sup>12,16,17</sup>.

HAM/TSP progression occurs markedly during the first years of the disease<sup>18</sup>. However, disease-induced dysfunctions can improve, which allows for functional benefits in patients, as was demonstrated in this study because the subjects in both treatment groups showed no disease progression. To the best of our knowledge, this study was the first to control the stability of the disease before physiotherapy.

Regarding first symptoms, our study was consistent with that of Araujo et al.<sup>2,19,20</sup>, who found weakness of the lower limbs as the main symptom (93%), followed by lumbar pain (7%). The main functional activities that were affected in our two groups, i.e., walking (72%), followed by climbing on the bus (21%) and falls (7%), were similar to those found in the literature.

Among the variables that were chosen for analysis, pain was the most constant. Pain frequently leads to changes in activities of daily living and professional restrictions in HAM/TSP subjects. Tavares et al.<sup>14</sup> found that 75.5% of patients among 90 HAM/TSP patients exhibited low-back pain that significantly impacted daily activities.

Among the rehabilitation techniques, PNF was chosen because it aims to enhance the performance of the neuromuscular system by stimulating muscle and joint proprioceptors using basic processes, such as muscular strength radiation<sup>5</sup>. The stimulation of strong muscle groups facilitates muscular activation<sup>21</sup>, which has resulted in functional recovery and improvements in gait patterns in patients with neurological disorders<sup>13,22</sup>. However, despite being commonly used in clinical practice, PNF has lacked evidence of scientific effectiveness in HAM/TSP.

In this study, PNF provided a significant improvement in low-back pain, activities of daily living (FIM), mobility and balance (TUG) in both groups. Because the treatment was applied differently in each group, a comparative analysis was performed to detect the putative supremacy of either treatment form. The results showed no significant differences, suggesting that the application of PNF with an elastic band and therapist supervision or PNF with direct facilitation were equally effective.

In this study, PNF reduced low-back pain. Kofotolis Kellis<sup>21</sup> also found a reduction in low-back pain, leading to the reduction in the functional limitations of patients after applying PNF. The reduction in low-back pain is important because of its positive impact on quality of life, social integration and the work activities of the patients<sup>23,24</sup>.

Spasticity is a significant limiting factor in the day-to-day lives of these patients. The modified Ashworth scale has been widely used as a tool to quantify spasticity<sup>8</sup>, although it has only rarely been used in HAM/TSP<sup>3</sup>. Ideally, hypertonia assessment should be performed during self-motion and passive mobilization because the muscle activation patterns

and spasticity increases differ between these two situations<sup>15</sup>. In our study, spasticity was assessed during passive mobilization using the modified Ashworth scale. Although no significant differences were observed in the intra-group analysis, a trend toward right-leg tone reduction was detected in group II and was responsible for the significant differences that were found when the two groups were compared. The favoring of the right lower limb might be related to right-side dominance. Although both sides are affected, patients tend to prioritize voluntary movement on the dominant side. The tone reduction in these patients was correlated with the physical therapy because no anti-spastic medications were modified.

Spasticity can be explained by myelopathy, which is an inflammatory process of chronic and progressive demyelization at lower levels of the spinal cord. The inflammation mostly involves the cortico-spinal tracts and causes motor impairments in the lower limbs<sup>15</sup>. Carr and Shepherd<sup>25</sup> reported that repeated strengthening exercises could reduce spasticity and improve muscle neural control and the maintenance of its extensibility.

The quadriceps and plantar flexors, as well as spasticity, are essential to maintaining function in HAM/TSP<sup>15,16</sup>. In this study, PNF directly facilitated functional tasks, such as rolling, the transition from sitting to standing, gait phases and going up/downstairs. These tasks require hip, knee and ankle mobility, which are facilitated by spasticity reduction.

The FIM scale<sup>26</sup> was chosen in the present study because it allows for functional assessments at different times. This instrument is able to measure the degree of aid that patients require to perform motor and cognitive tasks. Because no significant differences were observed between the groups at baseline, our results demonstrate that the intervention improved independence and reinforced the need for physical therapy in these patients.

In accordance with the work of Araújo et al.<sup>7,18</sup> and Smadja<sup>19</sup>, the patients had independence to walk, regardless of the 6-year disease time span, suggesting no relationship between the degree of disability and the disease duration. This finding differs from the study of Cruickshank et al.<sup>20</sup>, who found progressive worsening of gait over seven years that led to patients requiring wheelchairs. Although our patients had relative motion independence at treatment onset, the score improvements indicated the effectiveness of the intervention.

Fecal/urinary incontinence significantly impacts the quality of life of patients with spinal lesions<sup>27</sup>. Urologic manifestations are present in 90% of patients with HAM/TSP<sup>27</sup>. Group I showed a lower percentage (1.1%) of bladder control, and an improvement in sphincter function scores was noticed in group II, making these patients more independent compared with group I. After PNF, the independence scores increased in group I, although not significantly, with 56% showing complete independence. In group II, the improvement was statistically significant, with 68% of the patients achieving the maximum MIF score. This finding suggests that parameters such as gait and up-/downstairs walking improved to a greater extent.

The TUG test is an assessment tool that has been widely used to analyze balance and mobility<sup>28</sup>. In our study, the patients' conditions did not evolve negatively, which demonstrated that

the intervention prevented progression to severe loss of balance and mobility.

In group I, the patients were considered stable according to Steffan et al.<sup>29</sup> during the initial testing, and after 60 days, there were three semi-independent patients (42.8%) and four dependent patients (57.1%). Post-treatment, four patients were classified as semi-independent (57.1%), and three patients were classified as dependent (42.8%). In group II, at initial testing and after 60 days, one patient was considered independent (14.3%), three were semi-independent (42.8%), and three were dependent (42.8%), whereas after treatment, five were semi-independent (71.4%), and only one was dependent (14.3%). According to Podsiadlo Richardson<sup>30</sup>, when using this test, scores greater than 20 seconds indicate significant deficits in physical mobility and risk of falls. The score for group I after 60 days was 23.5 seconds, and it was 20.4 seconds post-treatment. In group II, the scores were 25.9 and 19.4 seconds, respectively. These data illustrate baseline walking difficulty and treatment effectiveness in balance improvement.

The relatively small sample size and the lack of adductor muscle tone evaluation, as well as the analysis of spasticity degree only by passive mobilization, were among the weaknesses of the present study. However, these factors did not impact our data.

This study showed that both physical therapy protocols, which are based on PNF applied by the therapist or with the aid of an elastic tube, were effective in the treatment of patients with HAM/TSP. If the number of professionals is insufficient to meet patient needs, the elastic tube technique can be used under the supervision of a therapist, without treatment quality concerns.

## CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

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