Night splinting does not increase ankle range of motion in people with Charcot-Marie-Tooth disease: A randomised, cross-over trial

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**Question** What is the effect of wearing splints at night to stretch the plantarflexors on dorsiflexion range of motion (ROM) in people with Charcot-Marie-Tooth disease? **Design** Randomised, assessor-blinded, cross-over trial. **Participants** 14 people (1 dropout) aged 7 to 30 years with Charcot-Marie-Tooth disease Type 1A and with ≤ 15 degrees dorsiflexion range of motion (ROM). **Intervention** A splint holding the ankle in maximum dorsiflexion was worn nightly on one leg for 6 weeks followed by the opposite leg for the subsequent 6 weeks. **Outcome measures** The primary outcome was dorsiflexion ROM; secondary outcomes were inversion ROM, and dorsiflexion, evasion, and inversion strength, measured before and after splinting, and three months later. **Results** There was no significant difference between the experimental and the control intervention in terms of ROM or strength. Wearing the splint at night increased dorsiflexion ROM by 1 degree (95% CI –3 to 4; p = 0.72) and evasion ROM by 1 degree (95% CI –1 to 3; p = 0.28) compared to not wearing the splint. Wearing the splint increased dorsiflexion strength by 41 N (95% CI –53 to 135; p = 0.38), reduced evasion strength by 6 N (95% CI –112 to 101; p = 0.92) and reduced inversion strength by 8 N (95% CI –110 to 95; p = 0.88) compared to not wearing the splint. **Conclusion** Wearing night splints does not increase ankle ROM or strength in people with Charcot-Marie-Tooth disease Type 1A. [Refshauge K, Raymond J, Nicholson G, van den Dolder PA (2006) Night splinting does not increase ankle range of motion in people with Charcot-Marie-Tooth Disease: A randomised, cross-over trial. Australian Journal of Physiotherapy 52: 193–199]

Key words: Physical Therapy Modalities, Charcot-Marie-Tooth Disease, Peripheral Neuropathy, Stretching, Pes Cavus

**Introduction**
Charcot-Marie-Tooth disease is a demyelinating neuropathy involving peripheral nerves and causing gross slowing of motor and sensory nerve conduction velocity. The most common form of Charcot-Marie-Tooth disease is Type 1A, and the development of foot and ankle problems, particularly muscle weakness, in this group is common (Holmes and Hansen 1993). The typical pattern of muscular weakness begins in the intrinsic muscles of the feet, followed by weakness in peroneus brevis and longus, tibialis anterior, extensor digitorum longus and extensor hallucis longus (Sabir and Lyttle 1983). This pattern of weakness results in muscle imbalance: the dorsiflexors become weak while the plantarflexors remain relatively strong, ultimately leading to contracture of the Achilles tendon. Secondary to this muscle imbalance, people with Charcot-Marie-Tooth disease typically develop a pes cavus deformity, with inversion of the calcaneus, adduction of the forefoot, and clawing of the toes (Alexander and Johnson 1987, Roper and Tibrewal 1989).

Currently, management of foot deformities in Charcot-Marie-Tooth disease involves surgical intervention (Alexander and Johnson 1987, Fenton et al 1984). However, clinical results of the most common surgical procedure – triple arthrodesis – appear to deteriorate with time (Medhat and Krantz 1988). The prevention of Charcot-Marie-Tooth disease foot deformities, and consequently the avoidance of surgery, should therefore be a high priority for people with Charcot-Marie-Tooth disease. Despite this, there are no high-quality, well-controlled trials evaluating the effects of any conservative management strategies for people with Charcot-Marie-Tooth disease.

Clinicians aim to prevent the development of plantarflexion contracture in Charcot-Marie-Tooth disease using muscle stretching. This method of conservative management is typically employed among younger people, or in those whose symptoms are potentially reversible. One intervention used is night splinting, which could provide effective treatment for people with Charcot-Marie-Tooth disease in two ways. First, night splinting allows the ankle to be placed in a dorsiflexed position for a prolonged period, applying a sustained stretch to the ankle plantarflexor muscles. Second, the use of night splints represents a more patient-friendly mode of stretching than alternatives such as lower limb serial casting.

While the theory behind stretching may be biologically plausible, the evidence supporting the use of stretch in humans is inconclusive. In a systematic review of stretching trials, Harvey et al found an 8 degree (95% CI 6 to 9) change in joint range of motion (ROM) in healthy, able-bodied participants in a meta-analysis of four moderate-quality trials. However, recent evidence suggests that improvement in ROM following a stretching program in this type of...
The cohort may be due to increased tolerance of stretch rather than actual changes in muscle extensibility (Follp et al 2006). Among high-quality (PEDro score > 6), randomised, controlled trials in clinical populations with either restricted range of motion or contractions, small treatment effects have been found (eg, Ben et al 2005), whereas others have found no effect with stretching (Moseley et al 2005; Harvey et al 2000; Harvey et al 2003). Ada et al (2005), in a single-blind randomised controlled trial (PEDro score 8), found that a 4-week stretching program reduced the development of some contractures in stroke patients. Therefore, evidence for the use of stretching to improve ROM remains equivocal.

One potential reason for the lack of effectiveness is insufficient duration of stretch. Typically, up to 30 min of stretch was applied in these studies. An increased duration could be applied by using night splints. Night splints were found to be effective in boys with Duchenne muscular dystrophy (Hyde et al 2000), although the high drop-out rate renders caution in interpreting the results of this trial. Serial casts were used successfully to treat ankle contracture in patients with traumatic brain injury (Moseley 1997). These data provide preliminary evidence to suggest that prolonged stretch, such as that provided by night splints, may be more effective than shorter duration stretches.

Despite its feasibility and widespread clinical use, the effectiveness of night splints on dorsiflexion ROM in Charcot-Marie-Tooth disease is unknown. The primary aim of the present study, therefore, was to examine the effect of long duration stretches applied using night splints, on dorsiflexion ROM in Charcot-Marie-Tooth disease. We hypothesised that if the foot was maintained in a dorsiflexed position overnight for several weeks, the strong ankle plantarflexors would be maintained in a lengthened position and there would be some increase in the length of the posterior calf and peroneus longus muscles. A secondary aim of this study was to examine changes in eversion ROM and ankle strength.

Method

Design We used a randomised, single-blind, cross-over design with repeated measures taken at regular time intervals to determine the effect of stretching (Figure 1). We elected to use a cross-over design because Charcot-Marie-Tooth disease Type 1A is not rapidly progressive and the pool of patients is quite small. At the initial assessment, the treating physiotherapist randomly selected the leg to be splinted first by tossing a coin after baseline measurements were completed. After six weeks, the splint was changed to the opposite leg. At 12 weeks, the splint was removed and participants discontinued splinting on either leg. Throughout the entire protocol, participants were requested to avoid performing additional stretches or exercises that deviated from their normal routine. Participants attended a physiotherapy department on four occasions: at Week 0 (baseline), Week 6 (end of the intervention for the first leg), Week 12 (end of intervention for the second leg) and at Week 26 (final follow-up). The same assessor, who was blinded to group allocation, made all measurements for each participant. Blinding was ensured by performing the random allocation after the assessor had left the physiotherapy department and by regularly instructing participants that they were not to discuss the intervention with the assessor on any of the measurement occasions. The order of assessments was randomised for each visit. The study was approved by the local human research ethics committee and participants provided informed consent prior to randomisation and data collection.

Participants Participants were recruited from among patients who attended the Molecular Medicine Laboratory at Concord Repatriation General Hospital. To be included, participants required genetic tests confirming that their Charcot-Marie-Tooth disease was Type 1A. Participants also required restricted range of passive dorsiflexion in both ankles (ie, ≤ 15 degrees dorsiflexion from plantargrade). This range is less than both the American Academy of Orthopaedic Surgeons normal range for dorsiflexion of 20 degrees (Greene and Heckman 1994) and the average range for dorsiflexion of 18 degrees (SD 7) in a sample of 300 males and females aged 15–34 years (Moseley et al 2001). Participants were excluded if they had previously had surgery to either foot, had sustained a recent ankle sprain or fracture of either leg, or had undergone any physiotherapy intervention or stretching program within the last six months. People older than 30 years were not recruited for this study as it was expected that older participants would not be as responsive to the intervention.

Intervention Pre-formed splints hinged at the ankle were fitted to the participants. The splint was adjusted into dorsiflexion by the treating physiotherapist until participants felt a stretch in their calf muscles which could be tolerated during sleeping. Participants were encouraged to increase the amount of dorsiflexion if they felt that the stretch was insufficient. The splint also maintained the ankle in neutral inversion/eversion. Participants were instructed to wear the splint for the whole night, and only to remove it to walk short distances (eg, to the toilet) and then replace it. Participants, or their parents, were asked to record the number of hours that they wore the splint each night using a simple custom-designed, compliance diary.

Outcome measures Passive dorsiflexion ROM assessed using the Lidcombe Template according to the protocol described by Moseley and Adams (1991) was the primary outcome. Participants were positioned in supine, with a small roll placed under the knee to provide a small amount of knee flexion so that dorsiflexion ROM could be measured without limitation from the gastrocnemius muscle. A standardised force (69 N for participants ≤ 12 years old and 118 N for participants > 12 years old) was applied to the metatarsal heads in a line parallel to the long axis of the tibia. The amount of force being applied was monitored using a spring balance. Although the force applied was standardised, the differences in foot length meant there was variation between participants in terms of the torque applied, which ranged from 5.5 to 8.1 Nm for participants ≤ 12 years old and 10.3 to 16.8 Nm for participants > 12 years old. The landmarks used for measurement of dorsiflexion angle were the head of fibula, lateral malleolus, inferior border of the calcaneus, and head of the fifth metatarsal. A photograph was taken from a standardised distance, and the dorsiflexion angle calculated from joining the landmarks. This method is highly reliable (ICC ≥ 0.97) (Moseley and Adams 1991).

Passive eversion ROM was measured with subjects positioned in prone, using the method described by Norkin and White (1985). The calcaneus was moved laterally into eversion to the end of range and a goniometer was used to measure the eversion angle. The distal end of the goniometer was aligned with the midline of the calcaneus and the proximal end with the midline of the lower leg. The midpoint between the two

Table 1. Mean (SD) score, mean (SD) difference within interventions, and mean (95% CI ) difference between interventions for all outcomes for the experimental intervention (n = 26 legs) and the control intervention (n = 26 legs) for Periods 1 and 2.

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Score</th>
<th>Difference within interventions</th>
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<td>658 (194)</td>
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<td>859 (266)</td>
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<td>Inversion force (N)</td>
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<td>1076 (360)</td>
<td>1126 (345)</td>
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</tbody>
</table>

Exp = Experimental, Con = Control; * = A at Week 0 + B at Week 6, ** = A at Week 6 + B at Week 0; † = A at Week 6 + B at Week 12, †† = A at Week 12 + B at Week 6

Figure 1. Design and flow of participants through the trial.
malleoli was aligned with the axis of the goniometer.

Isometric dorsiflexion, eversion and inversion strength was measured using a hand-held, digital dynamometer. For dorsiflexion, the transducer was placed over the metatarsals, for eversion over the fifth metatarsal and for inversion over the first metatarsal. Participants performed three maximal voluntary contractions of each of the three muscle groups and the results were averaged to account for possible fatigue in weakened muscles.

Data analysis  Power calculations conducted *a priori* indicated that 13 participants and therefore 26 test legs would provide sufficient power (> 80%) to detect a difference of 5 degrees in dorsiflexion ROM, assuming a standard deviation of 6 degrees. We selected 5 degrees change in dorsiflexion ROM *a priori* as the minimum clinically-relevant difference. This is consistent with others (eg, Ben et al 2005, Moseley et al 2005).

Within group differences (ie, post-test minus pre-test scores) were calculated for dorsiflexion and inversion ROM, and dorsiflexion, eversion and inversion strength for all experimental legs (n = 26) and control legs (n = 26) during Periods 1 and 2. A paired t-test was used to compare night splinting versus control on each of these change scores. To check there was no order effect of the night splinting, the same analysis was conducted on the experimental legs (n = 26) and control legs (n = 26) during Period 1 only. Due to the cross-over design, the data collected at six months could not be classified as resulting from either the experimental or control intervention. Therefore, within group differences were calculated for each outcome measure from Week 0 to Week 26. In order to present independent data, the 13 legs that were splinted first were chosen arbitrarily to be examined.

Post-hoc analysis was conducted to determine whether baseline dorsiflexion ROM affected the outcome, ie, whether treatment was more effective in those who were most impaired. The difference between the change in dorsiflexion ROM due to night splinting and the change in dorsiflexion ROM due to the control was regressed against baseline dorsiflexion ROM.

Data are presented as mean (SD) and paired difference (95% confidence intervals). The level of significance was set at *p* = 0.05.

### Results

**Flow of participants through the trial**  Fourteen participants (8 females, 6 males) with Charcot-Marie-Tooth disease (with typical bilateral involvement), aged 15 years old (SD 8, range 7–30), volunteered to participate in the study. No participant used walking aids although one participant used a cane because of impaired vision, and six participants used orthotics for arch support. The flow of participants through the trial is shown in Figure 1. One participant dropped out of the study within 3 weeks of commencing the intervention, citing that the splint was too uncomfortable.

**Compliance with intervention**  Participants generally complied well with wearing the splints, wearing them for an average of 7 hours per night (SD 2, range 4–9). The splints were worn for an average of 37 of a possible 42 nights (SD 8, range 18–42), ie, 88%. The approved protocol was followed for the duration of the study.

**Effect of intervention**  The baseline dorsiflexion ROM at baseline was 8º (SD 6) from plantargrade (Table 1), which is approximately 10–12º less than the reported norm. The splints were worn for an average of 37 of a possible 42 nights (SD 8, range 18–42), ie, 88%. The approved protocol was followed for the duration of the study.

![Table 2. Mean (SD) score, mean (SD) difference within interventions, and mean (95% CI) difference between interventions for all outcomes for the experimental intervention (n = 13 legs) and the control intervention (n = 13 legs) for Period 1 only.](image)

<table>
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<td>Inversion force (N)</td>
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<td>1115 (370)</td>
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Exp = Experimental, Con = Control; * = A at Week 0, ** = B at Week 0; † = A at Week 6, †† = B at Week 6
intervention. By the end of 26 weeks, dorsiflexion ROM had increased 3º (95% CI –2 to 8). Furthermore, there was no relationship between magnitude of effect of night splinting (difference between the change in dorsiflexion ROM due to night splinting and the change in dorsiflexion ROM due to the control) and the magnitude of initial restriction in dorsiflexion ROM ($r^2 = 0.02$, $p = 0.53$, Figure 2).

The average eversion ROM at baseline was 5º (SD 3) from neutral (Table 1). Wearing the splint at night had no significant effect on eversion ROM (paired difference 1º, 95% CI –1 to 3, $p = 0.28$; Table 1). There was also no effect found when the legs from Period 1 only were analysed (paired difference 0º, 95% CI –2 to 2, $p = 1.00$, Table 2). By the end of 26 weeks, eversion ROM had increased 1º (95% CI –2 to 4).

The average dorsiflexion strength at baseline was 648 N (SD 194) for the experimental legs and 658 N (SD 194) for the control legs (Table 1). Wearing the splint at night had no significant effect on dorsiflexion strength (paired difference –41 N, 95% CI –53 to 135, $p = 0.38$, Table 1). There was also no effect found when the legs from Period 1 only were analysed (paired difference 19 N, 95% CI –24 to 62, $p = 0.35$, Table 2). By the end of 26 weeks, dorsiflexion strength had increased 6 N (95% CI –54 to 66).

The average eversion strength at baseline was 862 N (SD 291) for the experimental legs and 859 N (SD 266) for the control legs (Table 1). Wearing the splint at night had no significant effect on eversion strength (paired difference –6 N, 95% CI –112 to 101, $p = 0.92$, Table 1). There was also no effect found when the legs from Period 1 only were analysed (paired difference –5 N, 95% CI –73 to 63, $p = 0.87$, Table 2). By the end of 26 weeks, eversion strength had increased 26 N (95% CI –90 to 141).

The average inversion strength at baseline was 1074 N (SD 391) for the experimental legs and 1076 N (SD 360) for the control legs (Table 1). Wearing the splint at night had no significant effect on inversion strength (paired difference –8 N, 95% CI –110 to 95, $p = 0.88$, Table 1). There was also no effect found when the legs from Period 1 only were analysed (paired difference 2 N, 95% CI –130 to 135, $p = 0.97$, Table 2). By the end of 26 weeks, inversion strength had increased 108 N (95% CI –63 to 279).

**Discussion**

We found from this randomised cross-over trial that wearing night splints for six weeks had no effect on ankle dorsiflexion ROM, or eversion ROM or strength of ankle muscles. This lack of difference in dorsiflexion ROM was not due to lack of power because the study was adequately powered to find a difference of five degrees in dorsiflexion ROM, the primary outcome. Furthermore, although there was not perfect compliance with wearing the splint, participants complied better than we had anticipated, wearing the splint on most nights, and for the majority of each night. In other words, in the majority of cases, a prolonged dorsiflexion stretch was undertaken on most nights. The findings of this study represent a novel contribution to the evidence concerning treatment strategies for patients with Charcot-Marie-Tooth disease.

Stretching studies in other clinical populations have also...
failed to find a treatment effect. In randomised, controlled trials conducted on spinal cord-injured participants, four weeks of stretching, 30 minutes a day for five days per week yielded no treatment effect on either hamstring (Harvey et al 2003) or plantarflexor extensibility (Harvey et al. 2000). In the same patient population (Ben et al. 2005), but with a longer duration of treatment (12 weeks) only a small treatment effect was found (mean 4°, 95% CI 2 to 6). Moseley and colleagues (2005) also failed to find a treatment effect for reducing contracture following ankle fracture.

The lack of effect of splinting on dorsiflexion ROM may be because the intensity of the stretch to the plantarflexors was inadequate. While compliance for wearing the splint was higher than we had anticipated, possibly because in the younger sample there was parental enthusiasm for trying the intervention, the stretch actually delivered by the splint may have been insufficient to effect a length change in the plantarflexor muscles. Participants reported that it was difficult to maintain the heel fully in the splint during the night. In such cases, only a very mild stretch may have been applied, or in the worst case, sustained plantarflexion may have been encouraged during the night, rather than maintaining dorsiflexion.

Although we did not quantify the torque applied with the night splints, thus preventing any comparison with other stretching studies that have done so, it is interesting to note that studies which have applied relatively large stretch torques, possibly more than in our study, also failed to find clinically-relevant treatment effects. For example, Ben and colleagues (2005) used one-legged standing on a 15 degree wedge in their participants with spinal cord injury for 30 minutes, three times a week for 12 weeks, but found only a small treatment effect (mean 4°, 95% CI 2 to 6). Therefore, in our study, even though we used a longer duration of stretch, it could be argued that a change was unlikely as a result of our relatively conservative approach. Nevertheless, the results are interesting because they show that a technique in common clinical use is not effecting change to the targeted impairment. It may be that a combination of both high intensity and prolonged duration stretch is required to gain a clinically-worthwhile effect.

We received unsolicited feedback from many of the participants that, despite initial reluctance to wear the splints, they intended to continue wearing the splints after completion of the study. These participants reported that, during the period of wearing the splints, they felt better; they perceived that their ankle was less likely to roll over and sprain, they felt more stable while walking, and it was easier to run. Considering these anecdotal reports, it may be unwise to reject the wearing of splints at this preliminary stage. A possible option for future studies may be to use a questionnaire to measure ankle instability, or to measure function using an individualised assessment, eg, the Patient Specific Functional Scale (Stratford et al 1995) where the participants are asked to nominate specific activities that they have difficulty performing, and then rate their ability to perform these activities on a 0 to 10 scale. Such a scale is individualised and is responsive to change in functional status (Pengel et al. 2004).

In conclusion, our study is the first to investigate conservative management of loss of ROM in Charcot-Marie-Tooth disease. Other conservative techniques require investigation, eg, prolonged standing on a wedge, orthotic use, and strengthening exercises. Attention should also be given to the outcome measures to ensure that they are both patient-relevant and sensitive to change. They could perhaps include a measure of quality of gait (such as stride length or stability of foot placement) and of ankle instability.

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