

Research Article

A Pilot Study to Assess the Immediate Effect of Dynamic Carbon Ground Reaction Ankle Foot Orthoses on Balance in Individuals with Charcot-Marie-Tooth in a Clinical Setting

Burke K^{1,2}, Cornell K^{1,3}, Swartz Ellrodt A^{1,2}, Grant N^{1,2}, Paganoni S^{2,4,5} and Sadjadi R^{1,2*}

¹Charcot-Marie-Tooth (CMT) Center of Excellence, Massachusetts General Hospital, USA

²Department of Neurology, Massachusetts General Hospital, USA

³Cornell Orthotics and Prosthetics, USA

⁴Department of Physical Medicine and Rehabilitation, Spaulding Rehabilitation Hospital, USA

⁵VA Boston Healthcare System, USA

*Corresponding author: Sadjadi R, Department of Neurology, Massachusetts General Hospital, 165 Cambridge St, Suite 820, Boston, MA 02114, USA

Received: June 29, 2021; Accepted: July 27, 2021;

Published: August 03, 2021

Abstract

Charcot-Marie-Tooth (CMT) causes muscle weakness and atrophy generally in distal extremities, with or without sensory changes. These impairments contribute to impaired balance and gait and increase risk for falls and secondary injuries. Dynamic Carbon Ground Reaction Ankle Foot Orthoses (DCGR-AFOs) are one type of lower extremity orthosis that can be prescribed to help improve gait and balance in this patient population. To our knowledge, no studies have evaluated the immediate impact of DCGR-AFOs on gait and balance in this population. In this pilot study, 9 individuals with CMT and gait impairment were seen in clinical setting by a physical therapist and orthotist. Participants were asked to complete the modified Clinical Test of Sensory Interaction and Balance (mCTSIB) and tasks on the 4-Item Dynamic Gait Index (DGI) with and without bilateral DCGR-AFOs to assess static and dynamic balance. The average DGI scores were 6/12 without the DCGR-AFOs and 10/12 with the DCGR-AFOs. Improvements on the mCTSIB varied. The findings in this study suggest an immediate improvement in dynamic balance during ambulation with the use of DCGR-AFOs, as assessed by the 4-Item DGI. Data on static balance did not reach significance suggesting the need for future studies to further assess the effects of DCGR-AFOs on static standing balance, as well as the impact of training with physical therapists. This pilot study demonstrates that it is possible to demonstrate potential benefits of DCGR-AFOs with a gross fitting in a clinical setting, prior to referral to an orthotist for custom fitting.

Keywords: Charcot-Marie-Tooth; Inherited neuropathies; Orthotic devices; Ankle foot orthoses; Balance; Gait

Abbreviations

ABC: Activities-Specific Balance Confidence Scale; AFOs: Ankle Foot Orthoses; CGS: Change in Gait Speed; CMT: Charcot-Marie-Tooth; DCGR-AFOs: Dynamic Carbon Ground Reaction Ankle Foot Orthoses; DGI: Dynamic Gait Index; GHT: Gait with Horizontal Head Turns; GLS: Gait on Level Surface; GVT: Gait with Vertical Head Turns; mCTSIB: Modified Clinical Test of Sensory Interaction and Balance

Introduction

Hereditary sensory and motor polyneuropathies, or Charcot-Marie-Tooth (CMT) disease, refer to the most common type of hereditary neurologic disorders that cause primarily distal weakness, sensory loss, and foot deformity. As the disease progresses, the muscle weakness and sensory changes contribute to impaired balance and gait, putting individuals at increased risk for falls and secondary injuries. Different interventions are aimed toward optimizing balance and gait in these individuals. Physical therapy is routinely considered for patients with significant functional limitations. Physical therapists primarily focus on gait and balance training, core stabilization, functional exercises, lower extremity resistance training, as well as

stretching to prevent contractures and deformity. They review the need for bracing and assistive devices to improve function. In addition to physical therapy, referral to an orthotist for custom bracing can be very beneficial in optimizing safety and function in individuals with gait impairments [1-5].

Ankle Foot Orthoses (AFOs) are commonly prescribed to compensate for ankle weakness and improve gait safety. Choosing appropriate AFOs relies on thorough evaluation of muscle strength, muscle length, sensory integrity, postural control, and gait mechanics by a physical therapist and orthotist. AFOs have been shown to improve walking ability in individuals with CMT [1-3] by improving postural control, gait mechanics and ambulation. When a person with CMT has anterior tibialis weakness and resultant foot drop during swing phase of gait, increased hip flexion during swing phase is a compensatory strategy that may be used to allow foot clearance and prevent tripping. This increases the work of ambulation and may contribute to secondary impairments, such as hip or low back pain. AFOs correct for the foot drop, thereby decreasing the excessive hip flexion during swing phase and reducing risk for tripping [2,3]. The improvement in hip flexion and tripping was seen with use of both the posterior leaf spring AFOs as well as the anterior AFOs [3].

Furthermore, anterior AFOs with an anterior elastic strap secured underneath shoelaces and providing resistance to ankle plantarflexion have been found to improve walking economy by reducing the energy cost of walking over time and distance [4].

Clinically, there have been reports that Dynamic Carbon Ground Reaction Ankle Foot Orthoses (DCGR-AFOs) [6] with anterior support have been effective in improving proper gait mechanics, by restoring predictable and consistent step length and foot placement, which in turn may decrease risk for falls and improve overall efficiency with gait. DCGR-AFOs assist with correcting foot drop during swing phase of gait for individuals with anterior tibialis weakness, but also assist stance phase by controlling the forward motion of the tibia for individuals with plantarflexion weakness and assist with push off at terminal stance, which can improve balance, stride length, and gait speed [7]. Furthermore, one study suggested that many individuals are resistant to meeting with an orthotist for an evaluation for bracing, and compliance with the use of AFOs in individuals with CMT can be low [8]. In this pilot study, the immediate effects of the DCGR-AFOs on static and dynamic balance were assessed in individuals with CMT *via* a gross fitting in the clinical setting. The hypothesis was that it is possible to demonstrate immediate improvements in static and dynamic balance with a trial of DCGR-AFOs in clinic, which may convince individuals to meet with an orthotist for a custom fitting of AFOs.

Methods

Study design

This study was a single-subject experimental design in which each participant served as their own control, performing all balance and gait outcomes with and without the use of DCGR-AFOs.

Participants

This pilot study looked at a targeted group of individuals with a diagnosis of CMT who were seen in the Massachusetts General Hospital CMT Center of Excellence between June 2018 and December 2018. To be eligible, they had to be between the ages of 18 and 90 years old, with a clinical diagnosis of CMT and self-reported ability to ambulate household distances with or without an assistive device. They also had to be seen on a day in which the clinic physical therapist and orthotist were available, and there was enough space for testing to complete without interruption to clinic. Eligible participants were seen by the physical therapist and orthotist in clinic and were determined to be appropriate for the trial of dynamic carbon ground reaction AFOs. This determination was made by the physical therapist and orthotist based on the individual's history and clinical presentation. Individuals reporting balance impairments, tripping during ambulation, or unsteady gait, who also demonstrated sensory ataxia and ankle weakness, were deemed appropriate to trial the AFOs. Participants were excluded if they had significant foot deformities that would require more significant customization, or if they denied any balance or gait impairments. Individuals were also excluded if they had evidence of other musculoskeletal or neurological deficits unrelated to CMT that may impact balance or gait or interfere with study participation. Eligible participants were informed of the study and study staff consented interested participants. The study was approved by the Partners Human Research Committee.



Figure 1: Dynamic carbon ground reaction ankle foot orthotic.

Intervention: Ground reaction AFOs

For this study, all participants trialed bilateral DCGR-AFOs with anterior support (Figure 1). The DCGR-AFOs have an anatomically shaped anterior tibia shell that stabilizes the tibia and a short strut that extends around the instep and offers medial and lateral support at the ankle. The strut connects the footplate, which goes in the shoe beneath the plantar aspect of the foot, to the anterior support, thereby resisting ankle plantarflexion during swing phase of gait. The carbon fiber material is flexible and does allow for tibial progression and ankle dorsiflexion during stance phase, with some resistance from the brace, thereby assisting with stability during stance phase [6]. The improved tibial progression can contribute to improved push off at terminal stance, improved balance, stride length, and gait velocity [7].

The DCGR-AFOs were not custom fit for each participant. In situations where the DCGR-AFOs did not fit in the participant's shoes, they were asked to use shoes provided by the orthotist, even if they did not match the participant's exact shoe size. There were no custom foot orthoses provided, which would have optimized the foot position in the shoe and on the AFO. This was a gross assessment of the impact of the DCGR-AFOs on balance and gait in the clinical setting where space and time constraints did not allow for individualized fitting of each brace. Participants who agreed they might benefit from AFOs were referred for a separate appointment with an orthotist for custom fitting, and to physical therapy for gait training with their custom AFOs. Outcomes for this study were not repeated with the custom AFOs as all outcomes were completed in a single visit prior to the participant receiving custom AFOs.

Outcome measures

The outcome measures chosen for this study were chosen by the clinic physical therapists as measures that would be practical to perform in a clinical setting without adding significant time to the clinical visit. They capture information on balance confidence, dynamic balance during ambulation, and static balance on different surfaces and with eyes open and closed.

Activities-specific Balance Confidence Scale (ABC): The ABC is a self-reported questionnaire that involves individuals rating their confidence for performing sixteen activities without falling or losing balance, on a 0-100% confidence scale [9-12]. This questionnaire was used to assess balance confidence and fall risk in our participants prior to them completing any of the gait or balance tasks. In this

questionnaire, 0% indicates they believe they cannot perform the activity without falling, whereas 100% indicates they have full confidence they can perform the activity without falling. The sum of all ratings is divided by 16 to give an average percent confidence for the entire questionnaire ranging from 0% to 100%. A score less than 67% indicates increased risk for falls and can accurately identify people who fall 84% of the time [9-12].

Four Item Dynamic Gait Index (DGI): Tasks from the 4-Item DGI were used in this study. The 4-item DGI is a reliable tool used to assess gait and balance [13]. The tasks from this outcome measure were chosen to detect changes in dynamic gait activities. The four tasks include:

- Ambulation on level surface
- Ambulation with changes in gait speed
- Ambulation with horizontal head turns
- Ambulation with vertical head turns.

Each item is scored 0-3 with a total maximum score of 12 points, where a 3 on each task indicates normal gait pattern. A higher total score indicates higher function. For each item, use of an assistive device or walking aid during ambulation, limits the individual to a score of 2 or lower, for a maximum score of 8 points. For the purposes of this study, use of an AFO was not considered an assistive device or walking aid in order to minimize risk of a ceiling effect. Any other assistive device (cane, walker, crutches, etc.) was still considered assistive devices and appropriately accounted for in the scoring of each item. If an individual demonstrated normal gait with no evidence of imbalance, they received a 3 on the task being tested. A score of less than 10/12 on the 4-item DGI indicates an increased risk for falls. Participants were asked to complete this test with and without the DCGR-AFOs [13]. The physical therapist scored each item and a stopwatch was used for each task that required measurements in gait speed.

Modified Clinical Test of Sensory Interaction and Balance (mCTSIB): The mCTSIB assesses an individual's balance under the following conditions:

- Feet together, eyes open, firm surface, arms across chest
- Feet together, eyes closed, firm surface, arms across chest
- Feet together, eyes open, foam surface, arms across chest
- Feet together, eyes closed, foam surface, arms across chest

Participants were asked to hold each condition for up to 30 seconds. If they were not able to hold it for 30 seconds, the time they were able to hold the position was recorded. Timing was stopped if the participant's arms moved from the original position, if their feet moved, or if they opened their eyes in conditions 2 and 4. They completed each condition 1-2 times. They were rated on a scale of 1-4 for each item on the amount of sway they demonstrated from minimal sway (1) to loss of balance/fall (4) [14,15]. Participants were asked to complete each portion of this test with and without the DCGR-AFOs. It has been demonstrated that there is no difference between scores on the mCTSIB with shoes on versus off [16]. Therefore, all participants were tested with shoes on for the mCTSIB.

Data analysis

The primary outcomes of this study were the total score on the 4-Item DGI and performance on the mCTSIB. Participants' scores with the DCGR-AFOs were compared to those without AFOs. A secondary outcome was how many participants agreed to be referred for an evaluation by an orthotist for custom bracing. The ABC was used to assess balance confidence and fall risk in our participants.

Results

Participants

A total of 11 individuals agreed and were consented to participate in this study. After meeting with the physical therapist and orthotist, one individual was deemed not appropriate for the AFOs being used in this study, as she required significant customization for foot and ankle orthoses given foot and ankle deformities. A second individual was consented, but not able to come at a time when the orthotist was in clinic and was therefore excluded from the study. A total of nine eligible individuals participated in this study. All nine participants completed all outcome measures during their clinical visit.

Participants included 6 women and 3 men, all Caucasian, with a mean age of 52 (range 28-72). Six participants had CMT1A, one with CMT4C, and two with unknown gene status. All nine participants presented with ankle dorsiflexion (manual muscle testing <5/5) and/or plantarflexion weakness (unable to rise on toes) noted on exam, and all participants presented with at least mild sensory impairments in their lower legs and feet, with two participants presenting with a positive Romberg and another four with negative Romberg demonstrated with increased sway. Romberg testing was not reported in the other four participants.

Balance confidence

The average percent confidence on the ABC scale was 67.5% with a range from 48.1% to 85.6% balance confidence. Four participants had scores less than the cut-off of 67% to predict increased risk for falls.

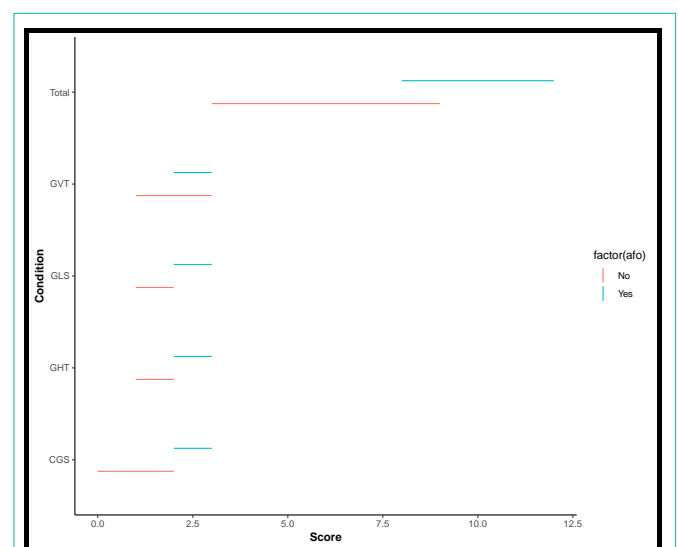


Figure 2: Results for 4-Item DGI - Mean Scores.

GVT: Gait with Vertical Head Turns; GLS: Gait on Level Surface; GHT: Gait with Horizontal Head Turns; CGS: Change in Gait Speed.

Table 1: Results for 4-Item DGI - Mean Scores.

	No AFOs	with AFOs	Improvement with AFOs	Significance
Gait level surface	1.6	2.7	1.1	
Change in gait speed	1.3	2.3	1	
Gait with horizontal head turns	1.6	2.3	0.8	
Gait with vertical head turns	1.8	2.7	0.9	
Total Score	6.2	10	3.8	p<0.001

Table 2: Results for mCTSIB.

	No AFOs		With AFOs		Differences (Time AFOs - no AFOs; Sway no AFOs - AFOs)	
	Min	Max	Min	Max	Min	Max
Condition 1 Time Held	20	30	30	30	10	0
Condition 1 Sway Score	1	2	1	1	0	1
Condition 2 Time Held	1.9	30	8.9	30	7	0
Condition 2 Sway Score	1.5	4	1	4	0.5	0
Condition 3 Time Held	17.6	30	15	30	-2.6	0
Condition 3 Sway Score	1	3.5	1	4	0	-0.5
Condition 4 Time Held	1	30	1.4	30	0.4	0
Condition 4 Sway Score	2	4	3	4	-1	0

Dynamic balance during ambulation

Results for the 4-item DGI are presented in Table 1 and Figure 2. For the 4-item DGI, the average score without the AFOs on was 6/12, with range of 3/12 to 9/12. The average score with the use of the DCGR-AFOs was 10/12, with a range of 8/12 to 12/12. All of the participants improved at least one point on the 4-item DGI. Average improvement was 3.7 points with a range of 1-point improvement to 6-point improvements. The average percent improvement in this test was 76.9% with a range of 12.50% to 166.7%.

For task number 1, Gait on a Level Surface (GLS), one participant did not demonstrate an improvement, six participants improved by one point, and two improved by two points. For task number 2, gait with Changes in Gait Speed (CGS), two participants did not demonstrate improvement with AFOs, five improved by one point and two improved by two points. For task number 3, Gait with Horizontal Head Turns (GHT), two participants did not demonstrate an improvement, while the other seven improved by one point. Finally, for task number 4, Gait with Vertical Head Turns (GVT), three participants did not demonstrate an improvement in AFOs, four improved by one point, and two improved by two points. For the group, there was a significant improvement on each of the items as well as the total score with use of the DCGR-AFOs compared to without the AFOs.

Static balance

Results for the mCTSIB are presented in Table 2. For condition one of the mCTSIB, flat surface with eyes open, one participant was able to increase time held, while the other eight participants were able to hold for the full 30 seconds with and without the use of DCGR-AFOs. Sway scores improved in three participants and were unchanged at minimal sway with and without DCGR-AFOs. The average time held was 28.9 seconds without AFOs and 30 seconds with the DCGR-AFOs.

For condition two of the mCTSIB, flat surface with eyes closed, three participants demonstrated improved time held, while the other six were able to hold for the full 30 seconds with and without the use of DCGR-AFOs. Sway scores improved in seven participants and were unchanged in two participants, one with mild sway and the other with loss of balance both with and without the use of the DCGR-AFOs. The average time held was 24.0 seconds without AFOs and 27.0 seconds with DCGR-AFOs.

For condition, three of the mCTSIB, foam surface with eyes open, two participants improved time held with use of DCGR-AFOs, one demonstrated a decline in time held and the other six participants were able to hold for the full 30 seconds with and without the use of DCGR-AFOs. Sway scores improved in three participants declined in one participant and were unchanged in five participants with four demonstrating minimal sway and one demonstrating mild sway with and without the use of DCGR-AFOs. The average time held was 28.2 seconds without AFOs, and 28.3 seconds with DCGR-AFOs.

For condition four on the mCTSIB, foam surface with eyes closed, four participants demonstrated improved time held with use of DCGR-AFOs, three demonstrated a decreased time held, and two demonstrated no change. Sway scores improved in one participant, declined in one participant and remained unchanged in the other seven participants with all seven demonstrating loss of balance in this condition with and without the use of DCGR-AFOs. The average time held was 7.7 seconds without AFOs on and 10.2 seconds with DCGR-AFOs.

All participants demonstrated improvement in at least one of the conditions on the mCTSIB with use of the DCGR-AFOs. Any decline in performance, seen in 5/9 individuals, was observed when the individual was asked to stand on the foam. There were no significant differences in performance on any of the four conditions studied.

Referral to orthotist

All 9 participants (100%) agreed to being referred to an orthotist for custom fitting of AFOs and referral to physical therapy for gait training with bilateral AFOs.

Discussion

The findings in this study suggest a potential benefit of performing a gross assessment of standing static and dynamic balance with and without the use of AFOs in a clinical setting. In this pilot study, there was an immediate improvement in dynamic balance during ambulation with the use of DCGR-AFOs, with significant improvements on the 4-Item DGI. Standing static balance was improved in at least one of the conditions on the mCTSIB with use of DCGR-AFOs, but overall there were no significant differences with or without the DCGR-AFOs. With each of the participants also agreeing to be referred to an orthotist for custom fitting of AFOs, this study also suggests that a trial of the bilateral AFOs may help create buy-in, although we did not assess how many were in agreement prior to the AFO trial.

The participants in this study were individuals with CMT, and resultant distal lower extremity weakness, balance and gait impairments who, as demonstrated by the ABC questionnaire, had decreased activity balance confidence. Four of the nine (44%) participants reported ABC scores that fall below the cut-off score for being at increased risk for falls, and all participants reported some amount of decreased confidence on the questionnaire. This highlights the need for physical therapy interventions and consideration for lower extremity bracing in this population to improve balance confidence, and decrease risk for falls.

On the 4-Item DGI, all participants were below the cut-off of 10/12 without AFOs on, indicating increased fall risk, consistent with their ABC scores. With DCGR-AFOs, only 3/9 (33%) remained below the fall risk cut-off. This data should be taken with caution, as the DCGR-AFOs were not considered assistive devices for scoring on the DGI to allow for an equal comparison of gait with AFOs to gait without AFOs. The improvement in scores does represent improvement in gait activities with use of the DCGR-AFOs. DCGR-AFOs were found to have an immediate impact on balance and gait. All participants demonstrated improvements in ambulation tasks, with eight demonstrating at least 33% improvement, and three participants demonstrating greater than 100% improvement in their scores with the use of the DCGR-AFO braces. Pereira et al. [17] completed a similar study looking at the immediate effects of using AFOs on kinematics of gait and balance reactions in individuals with CMT. This study differs from that one in that they used a movement analysis system and infrared cameras to evaluate gait kinematics and balance reactions, which is not always practical for the clinical setting where both time and space are limited. They found that while using AFOs on the DGI, their participants performed 8.4% better when compared to not using orthoses. This study was completed to explore whether these immediate effects could be observed in a clinical setting with outcome measures that are commonly used by physical therapists to assess fall risk, postural control and gait. Similar to Pereira et al. the findings in this study did demonstrate an immediate improvement in balance and gait measures with the use of ground reaction AFOs. The improvement of 76.9% on the 4-item DGI in this

study is significantly greater than their findings, which may be due to the low scores the participants had at baseline without AFOs on, and the scoring exception to allow for maximum scores with use of the AFOs if gait was normalized with use of the bilateral DCGR-AFOs. These findings are also consistent with previous findings that people with CMT who wore AFOs regularly were more severely affected than those who were not wearing AFOs. [18] This improvement suggests that the walking tasks on the 4-Item DGI can be used as a clinical assessment to determine the potential benefits of AFOs for individuals with CMT and assist in the recommendation for a referral to an orthotist for custom fitting of such braces. However, given the adjustment to the scoring system, descriptive gait analysis and other outcomes measures should be considered for use in the clinic as well.

For static balance, there were no significant changes noted across the study population. Many of the individuals were able to hold the conditions of the mCTSIB for the required 30seconds without loss of balance, suggesting a possible ceiling effect. Five of the nine participants (56%) demonstrated decline in performance on either condition 3 or 4 when they were asked to stand on foam with the use of DCGR-AFOs. The ground reaction force with respect to the ankle and lower extremity joints is expected to be more unstable on the foam. Increased training with ground reaction braces would be expected for standing or walking on surfaces such as foam. This would be important to consider when individuals are ambulating on uneven or soft surfaces, such as sand or grass, and may warrant more focused gait and balance training with a physical therapist, if an individual were to be fit with such braces. All but one of the participants demonstrated improvement with eyes closed standing on a firm surface while wearing the DCGR-AFOs, most in that their sway scores improved from mild to minimal sway. This suggests an almost immediate learning effect to utilize the anterior support and ground reaction to find ground and stabilize oneself when visual cues are absent. Four of the nine participants (44%) were able to demonstrate improvement on foam with eyes closed with the DCGR-AFOs, suggesting that they may be able to utilize at least minimal ground reaction to find ground and improve stability without visual cues even on the uneven surface. An alternative explanation may be improved confidence with the DCGR-AFOs, which was not assessed in this study.

Individuals with CMT demonstrate significant variability of the biomechanical impairments impacting their gait. Often the abnormal gait patterns are explained by foot drop during swing phase and plantar flexor failure, with decreased push-off at terminal stance [19]. As a result, these individuals ambulate with a compensatory steppage gait, in which they demonstrate exaggerated hip flexion during swing phase of gait to clear their foot, or more of an unpredictable gait pattern with slowed gait speed, increased base of support, decreased step length and variable foot placement. With these compensatory patterns, there is an increased work of ambulation and high-energy consumption, as well as potential for secondary injuries higher up the kinematic chain [17]. The benefits of the DCGR-AFOs with anterior support used in this study include supporting the foot during swing phase to compensate for the foot drop, and eliminating the need for the exaggerated hip flexion. They also likely improve the proprioceptive input in the lower legs, and improve tibial progression and ankle dorsiflexion during stance phase. The improved tibial

progression can contribute to improved push off at terminal stance, improved balance, stride length, gait velocity, and overall improved gait efficiency, although gait parameters were not measured in this pilot study. This is consistent with other findings demonstrating that both rigid and semi-rigid AFOs improve gait deviations, and that semi-rigid AFOs improve proprioceptive inputs in individuals with lower extremity neuropathy [20,21]. The DCGR-AFOs offer the benefits of both rigid and semirigid AFOs by supporting the ankle and improving proprioceptive input to improve gait mechanics.

This study did not include follow-up with participants to determine if they did receive custom AFOs. However, all participants agreed in clinic to be referred to an orthotist for custom fit AFOs and outpatient physical therapy for gait training if they receive the AFOs. Establishing this buy-in is important to optimize care for individuals with CMT. Future research is needed to determine both the compliance and long-term benefits of these braces for individuals with CMT.

Limitations and Future Directions

This pilot study has several limitations including a small sample size and the fact that the DCGR-AFOs were not custom fit to each individual. All individuals were asked to use the DCGR-AFOs in this study as time and space in the CMT clinic did not allow a thorough assessment to determine most appropriate AFOs for each individual. Individuals were also asked to wear footwear that fit the brace and did not necessarily match their shoe size, which was practical for a gross clinical assessment, but may have limited the benefits of the braces. Data was not collected on which participants were able to use their own footwear as opposed to a shoe to accommodate the brace. This may have provided insight into the varied data seen across participants in the mCTSIB test especially. We also did not complete a full assessment of ankle strength, which may have explained some of the differences across participants.

With this pilot study, it was demonstrated that a gross assessment of balance and gait with and without AFOs might help establish buy-in by demonstrating to the patient a potential for improved gait. Tasks from the 4-item DGI may be used to assess dynamic balance. While the mCTSIB was able to detect change in static balance for some of the participants, it did not reach significance. Future research should look at a larger sample size, as well as alternative outcome measures that may be better able to demonstrate potential benefits of the AFOs, especially given the need in this study to alter the scoring of the DGI to allow for equal comparison of gait with and without AFOs. Validated outcomes for comparing gait with and without AFOs, including the use of simple wearables, should be considered for future studies rather than the tasks in the 4-Item DGI, as this outcome is not validated with our modified scoring. Consideration for time and resources in the clinical setting should be made and outcomes that can be completed without disrupting clinic flow should be examined further in a larger population of individuals with CMT.

Future research is also needed to assess long-term benefits, patient satisfaction, and compliance, as well as to determine which types of tasks AFOs are most beneficial for individuals with CMT. It would be beneficial to compare different types of AFOs to help determine appropriate prescription based on clinical findings. Furthermore, it would be important to examine the benefits of gait training with a

physical therapist once fit with custom AFOs.

Conclusion

The findings in this study suggest that a gross assessment of balance and gait with and without AFOs in clinic may be helpful for individuals with CMT who are considering ankle bracing. In this pilot study, there was an immediate improvement in dynamic balance during ambulation with the use of DCGR-AFOs. Data on static balance did not reach significance suggesting the need for future studies to further assess the effects of AFOs on static standing balance, as well as the impact of training with physical therapists. The trial of bilateral DCGR-AFOs in clinic may assist with creating buy-in from the patient to then be referred to an orthotist for custom fitting and to physical therapy for focused gait training.

Acknowledgements

We would like to thank all our patients who participated in this study. Additionally, we thank Dr. Anita Myers, the primary developer and copyright holder of the ABC scale, for granting us permission to use the ABC scale for this study.

References

- Corrado B, Ciardi G, Bargigli C. Rehabilitation management of the Charcot-Marie-Tooth Syndrome. A systematic review of the literature. *Medicine*. 2016; 95: e3278.
- Guillebaste B, Calmels P, Rougier PR. Assessment of appropriate ankle-foot orthoses models for patients with Charcot-Marie-Tooth disease. *American Journal of Physical Medicine and Rehabilitation*. 2011; 90: 619-627.
- Ramdharry GM, Day BL, Reilly MM, et al. Foot drop splints improve proximal as well as distal leg control during gait in Charcot-Marie-Tooth disease. *Muscle Nerve*. 2012; 46: 512-519.
- Menotti F, Laudani L, Damiani A, et al. An anterior ankle-foot orthosis improves walking economy in Charcot-Marie-Tooth type IA patients. *Prosthetics and Orthotics International*. 2014; 38: 387-392.
- Charcot-Marie-Tooth Association. A guide to physical and occupational therapy for CMT. 2019.
- Allard USA. Professional Instructions. 2019.
- Danielsson A and Sunnerhagen KS. Energy expenditure in stroke subjects walking with a carbon composite ankle foot orthosis. *Journal of Rehabilitation Medicine*. 2004; 36: 165-168.
- Vinci P and Gargiulo P. Poor compliance with ankle-foot-orthoses in Charcot-Marie-Tooth disease. *European Journal of Physical and Rehabilitation Medicine*. 2008; 44: 27-31.
- "Activities-Specific Balance Confidence Scale." Shirley Ryan Ability lab: Rehabilitation Measures. 2018.
- Powell LE and Myers AM. "The Activities-specific Balance Confidence (ABC) Scale". *The Journals of Gerontology: Series A*. 1995; 50: M28-M34.
- Myers AM, Powell LE, Maki BE, et al. "Psychological indicators of balance confidence: relationship to actual and perceived abilities". *J Gerontol A Biol Sci Med Sci*. 1996; 51: M37-M43.
- Myers AM, Fletcher PC, Myers AH, and Sherk W. "Discriminative and evaluative properties of the Activities-specific Balance Confidence (ABC) scale". *J Gerontol A Biol Sci Med Sci*. 1998; 53A: M287-M294.
- Marchetti GF and Whitney SL. "Construction and validation of the 4-item dynamic gait index". *Physical Therapy*. 2006; 86: 1651-1660.
- Modified Clinical Test of Sensory Organization and Balance. 2018.
- Cohen H, Blatchly CA, Gombash LL. A study of the clinical test of sensory interaction and balance. *Phys Ther*. 1993; 73: 346-351.

16. Whitney SL and Wrisley DM. The influence of footwear on timed balance scores of the modified clinical test of sensory interaction and balance. *Arch Phys Med Rehabil.* 2004; 85: 439-443.
17. Pereira RB, Felicio LR, de Sa Ferreira A, et al. Immediate effects of using ankle-foot orthoses in the kinematics of gait and balance reactions in Charcot-Marie-Tooth disease. *Fisioter Pesqui.* 2014; 21: 87-93.
18. Ramdharry GM, Pollard AJ, Marsden J, Reilly MM. Comparing gait performance of people with Charcot-Marie-Tooth disease who do and do not wear ankle foot orthoses. *Physiother. Res. Int.* 2012; 17: 191-199.
19. Don R, Serrao M, Vinci P, et al. Foot drop and plantar flexion failure determine different gait strategies in Charcot-Marie-Tooth disease. *Clinical Biomechanics.* 2007; 22: 905-916.
20. Rao N and Aruin AS. The effect of ankle-foot orthoses on balance impairment: single-case study. *J Prosthet Orthot.* 1999; 11: 15-19.
21. Aruin AS and Rao N. Ankle-foot orthoses: proprioceptive inputs and balance implications. *J Prosthet Orthot.* 2010; 22: 34-37.