

Prescription of foot and ankle orthoses for children with Charcot–Marie–Tooth disease: a review of the evidence

Grant Scheffers¹, Claire Hiller¹, Kathryn Refshauge¹, Joshua Burns^{1,2}

¹Faculty of Health Sciences, The University of Sydney, Australia, ²Institute for Neuroscience and Muscle Research, The Children’s Hospital at Westmead, Australia

Background: Charcot–Marie–Tooth disease (CMT) is the most common inherited peripheral neuropathy and is associated with debilitating lower limb impairments and activity limitations. These impairments and activity limitations are potentially amendable to the prescription of orthoses, yet there is no universal, literature-based consensus to inform the decision making process of whether or not orthoses are indicated for a particular child with CMT, and if so, what type of device.

Objectives: The aims of this paper were to: (1) review the lower limb impairments and activity limitations of children with CMT; (2) review the indications of commonly prescribed foot and ankle orthoses; and (3) formulate a clinical algorithm for the optimal prescription of foot and ankle orthoses for children with CMT.

Major findings: We conducted a comprehensive search of the major databases and reference lists of relevant articles and books. In general, in-shoe orthoses are indicated for children with CMT and pes cavus and foot pain and/or mild balance impairments, whilst ankle-foot orthoses are indicated for children with CMT and pes cavus, foot drop, global foot and ankle muscle weakness and/or ankle equinus, and moderate-severe balance impairments and/or difficulty walking.

Conclusions: A clinical algorithm is proposed to guide the prescription of foot and ankle orthoses for children with CMT. Further research is required to determine the efficacy of different foot and ankle orthoses, and the predictive ability of the proposed clinical algorithm, to improve the lower limb impairments and activity limitations of children with CMT.

Keywords: Adolescent, Ankle, Charcot–Marie–Tooth disease, Child, Foot, Orthotic devices

Introduction

Foot and ankle orthoses are prescribed to support, correct, and/or compensate for a wide variety of lower limb impairments, with an additional aim of improving the performance of everyday activities.¹ Charcot–Marie–Tooth disease (CMT) is the most common inherited peripheral neuropathy² and is associated with debilitating lower limb impairments and activity limitations, the onset and severity of which are variable among affected children.^{3–8} These impairments and activity limitations are potentially amendable to the prescription of orthoses, yet this hypothesis has not been adequately tested through high quality, randomized controlled trials. Currently, the prescription of foot and ankle orthoses for children with CMT is often based solely on clinical judgement; there is no universal, literature-based consensus to inform the decision making process of whether or not orthoses are indicated

for a particular child with CMT, and if so, what type of device. Therefore, the aims of this review were: firstly to review the lower limb impairments and activity limitations of children with CMT; secondly to review the characteristics, functions, and indications of commonly prescribed foot and ankle orthoses; and thirdly to formulate a clinical algorithm for the optimal prescription of foot and ankle orthoses for children with CMT. We conducted a comprehensive search of the Cochrane Neuromuscular Disease Group Trials Register, MEDLINE, EMBASE, CINAHL, AMED, reference lists of articles, and books.

Charcot–Marie–Tooth Disease

CMT is the collective name given to a group of inherited peripheral neuropathies. With a prevalence rate of 8–41 per 100 000 people, CMT constitutes the most common type of inherited peripheral neuropathy.² The name *Charcot–Marie–Tooth disease* refers to the three clinicians who first described the condition in 1886,^{9,10} although it is also known as ‘Hereditary Motor and Sensory Neuropathy’.^{11,12}

Correspondence to: Joshua Burns, Institute for Neuroscience and Muscle Research, The Children’s Hospital at Westmead, Locked Bag 4001, Westmead, New South Wales, 2145, Australia. Email: joshua.burns@health.nsw.gov.au

Historically, CMT has been difficult to diagnose due to the marked variation in phenotype seen among patients. The introduction of nerve conduction studies in the late 1950s led to a greater understanding of the pathophysiology behind CMT. In 1968, Dyck and Lambert proposed two main types of CMT based on electrophysiological findings: those characterized by demyelination and severely reduced nerve conduction velocities (e.g. CMT type 1) and those characterized by axonal degeneration and normal or only slightly reduced nerve conduction velocities (e.g. CMT type 2).^{11,12} Demyelination has previously been estimated to cause 76% of all cases of CMT, with the remaining 24% of cases caused by axonal degeneration.¹³ Further classification of CMT is based on the defective gene and mode of inheritance. CMT type 1A (CMT1A) is the most common sub-type, accounting for 67% of all demyelinating types of CMT.¹⁴ However, there is only a weak correlation between genotype and phenotype.¹⁵ CMT therefore remains principally a clinical diagnosis established by signs and symptoms, family history, and nerve conduction studies.

Lower Limb Impairments of Children with CMT

Impairments are defined as 'problems in body function and structure'.¹⁶ The lower limb impairments of children with CMT are a consequence of the progressive, length-dependent peripheral denervation that typifies CMT. The longest nerves are principally affected, resulting in primary impairments of symmetrical muscle atrophy, weakness, and reduced sensation of the distal limbs (feet and hands).³⁻⁸ Secondary impairments include high-arched (pes cavus) foot deformities, poor balance, ankle equinus, foot pain, and foot/leg cramps.

The following sections review the primary and secondary lower limb impairments of children with CMT. The main findings are summarized in Table 1. Much of the earlier research into the lower limb impairments of children with CMT was of low methodological quality due to the use of biased sampling methods, small sample sizes, and clinical tests with poor or untested validity, reliability, and/or sensitivity. However, these studies still provide an insight into the disease process and clinical features of CMT and have therefore been included in the review below.

Primary lower limb impairments of children with CMT

Atrophy and weakness of foot and ankle muscles

Muscle atrophy and weakness in children with CMT typically begins with the foot intrinsics, later progressing to include selected muscles in the anterior and lateral compartments of the leg (e.g. peroneus brevis and tibialis anterior).¹⁷⁻¹⁹ The respective antagonist

muscles (e.g. peroneus longus, tibialis posterior, and triceps surae) are relatively spared until later in the disease process. This unique pattern of selective muscle involvement was originally proposed after clinical observations of muscle atrophy in affected children and adults.^{8,19} Gallardo *et al.*⁴ have since used magnetic resonance imaging (MRI) to demonstrate atrophy-related changes in the foot intrinsics of patients with CMT1A, including three children. Interestingly, such changes were not evident in the extrinsic leg muscles of these children (e.g. peroneus brevis and tibialis anterior).

Manual muscle testing was also previously used to assess selective muscle involvement in patients with CMT.^{8,19} Dynamometry has since been shown to be a more valid, reliable, and sensitive measure of muscle strength than manual muscle testing.²⁰⁻²² Burns *et al.*³ used hand-held dynamometry to assess the foot and ankle inversion, eversion, plantarflexion, and dorsiflexion strength of 81 children with CMT1A. When compared to age-equivalent norms, this sample of affected children was shown to have foot and ankle muscle weakness. Furthermore, the mean inversion-to-eversion strength ratio was 1.2 (SD, 0.3; range, 0.5-2.2) and the mean plantarflexion-to-dorsiflexion strength ratio was 3.3 (SD, 0.7; range, 2.0-5.1). This contrasts to normal inversion-to-eversion and plantarflexion-to-dorsiflexion strength ratios for children of 1.0 and 2.6, respectively.²⁰ It can be surmised that whilst the degree to which individual extrinsic foot muscles in children with CMT1A become weak remains unclear, it seems each of the muscular compartments of the legs do weaken, in particular the evertors in the lateral compartment and the dorsiflexors in the anterior compartment.

Reduced sensation

Only vibration and proprioceptive sense have been assessed in children with CMT (four children, all with CMT type 1).⁸ In this study by Sabir and Lyttle, each child demonstrated normal sensation, although seven of the remaining eight adult patients included in the study demonstrated moderate-severe impairments. These findings suggest that vibration and proprioceptive sense are implicated at a later stage of the CMT disease process, yet a larger sample size of affected children is needed to confirm this hypothesis.

Secondary lower limb impairments of children with CMT

Pes cavus

Pes cavus is a multiplanar foot deformity characterized by an abnormally high medial longitudinal arch; it also commonly features a varus (inverted) hindfoot, a plantarflexed (downward) orientation of the first metatarsal/forefoot, an adducted (internally directed) forefoot, and claw toes (Fig. 1). The development of

Table 1 Studies of lower limb impairments and activity limitations in children with CMT

Study	Design and participants	Main findings	Limitations of study
Vanasse and Dubowitz (1981), ⁶ England	Longitudinal (but only baseline impairment data); 14 children with CMT type 1 aged 4 months–12 years.	Impairments: weakness of foot/ankle muscles (71%); atrophy of ankle muscles (62%); reduced sensation (29%); pes cavus (21%); ankle equinus (14%); frequent falls as initial manifestation (43%) Activity limitations: developmental delay in first year of life (self-reported, 36%); walking (self-reported abnormal gait at disease onset, 43%), running (self-reported inability to run at disease onset, 43%) Impairments: weakness of foot/ankle muscles (100%); reduced sensation, i.e. touch, pain, and cold (75%) vibration and position (0%); pes cavus (100%) Activity limitations: not ascertainable from data	Biased sampling method, small sample size; poor methodological quality of clinical tests; activity limitations not quantified
Sabir and Lyttle (1984), ⁸ Canada	Cross-sectional; 4 children with CMT type 1 aged 7, 8, 10, and 11 years.	Impairments: self-reported onset of pes cavus in first decade (45%); second decade (5%). <10 years: weakness of foot/ankle muscles (0%); reduced sensation (5%); pes cavus (22%). 10–20 years: weakness of foot/ankle muscles (50%); reduced sensation (69%); pes cavus (87%). Activity limitations: self-reported onset of clumsiness in walking and running/frequent tripping in first decade (45%); second decade (5%). Impairments: 0–4 years: weakness of ankle muscles (0%); atrophy of foot intrinsics (17%); atrophy of ankle muscles (0%); reduced sensation (0%); pes cavus (25%). 5–10 years: weakness of ankle muscles (10%); atrophy of foot intrinsics (50%); atrophy of ankle muscles (0%); reduced sensation (50%); pes cavus (50%). 11–19 years: weakness of ankle muscles (71%); atrophy of foot intrinsics (71%); pes cavus (100%). Activity limitations: walking (self-reported clumsy walking) 0–4 years (17%); 5–10 years (40%); 11–19 years (57%)	Biased sampling method, small sample size; poor methodological quality of clinical tests; ankle equinus not assessed; frequency of symptoms and activity limitations not ascertainable. Small sample size; poor methodological quality of clinical tests; ankle equinus not assessed; activity limitations not quantified.
Berciano et al. (1989), ⁷ Spain	Cross-sectional; children with CMT type 1 aged <10 years (n=18) and 10–20 years (n=16).	Impairments: self-reported onset of pes cavus in first decade (45%); second decade (5%). <10 years: weakness of foot/ankle muscles (0%); reduced sensation (5%); pes cavus (22%). 10–20 years: weakness of foot/ankle muscles (50%); reduced sensation (69%); pes cavus (87%). Activity limitations: self-reported onset of clumsiness in walking and running/frequent tripping in first decade (45%); second decade (5%). Impairments: 0–4 years: weakness of ankle muscles (0%); atrophy of foot intrinsics (17%); atrophy of ankle muscles (0%); reduced sensation (0%); pes cavus (25%). 5–10 years: weakness of ankle muscles (10%); atrophy of foot intrinsics (50%); atrophy of ankle muscles (0%); reduced sensation (50%); pes cavus (50%). 11–19 years: weakness of ankle muscles (71%); atrophy of foot intrinsics (71%); pes cavus (100%). Activity limitations: walking (self-reported clumsy walking) 0–4 years (17%); 5–10 years (40%); 11–19 years (57%)	Small sample size, limited to CMT1A; poor methodological quality of clinical tests; ankle equinus not assessed; activity limitations not quantified.
Garcia et al. (1998), ⁵ Spain	Longitudinal; 12 children with CMT1A aged 0–4 years (n=12), 5–10 years (n=10), and 11–19 years (n=7).	Impairments: weakness of foot/ankle muscles (0%); atrophy of foot intrinsics (100%); pathological MRI* findings in foot intrinsics (100%) and extrinsics (0%); reduced sensation (33%); pes cavus (67%); ankle equinus ≤10° (100%) Achilles tendon shortening (100%) Activity limitations: nil (Functional Disability Scale=normal, 100%) Impairments: decreased mean ankle DF† ROM‡, foot/ankle strength, balance, and power for all ages versus age-equivalent norms; decreased walking endurance for affected adolescents; ankle instability (72%); frequent trips (63%) and falls (47%); pes cavus (34.5%); foot drop (4%); foot pain (27%); foot/leg cramps (36%). Activity limitations: walking (trips/falls, endurance, temporospatial); jumping (long jump).	Biased sampling method, small sample size, limited to CMT1A; poor methodological quality of clinical tests (except MRI*); frequency of symptoms not ascertainable. Biased sampling method, sample limited to CMT1A; sensation not tested.
Gallardo et al. (2006), ⁴ Spain	Cross-sectional; 3 children with CMT1A aged 8, 16, and 17 years.	Impairments: weakness of foot/ankle muscles (0%); atrophy of foot intrinsics (100%); pathological MRI* findings in foot intrinsics (100%) and extrinsics (0%); reduced sensation (33%); pes cavus (67%); ankle equinus ≤10° (100%) Achilles tendon shortening (100%) Activity limitations: nil (Functional Disability Scale=normal, 100%) Impairments: decreased mean ankle DF† ROM‡, foot/ankle strength, balance, and power for all ages versus age-equivalent norms; decreased walking endurance for affected adolescents; ankle instability (72%); frequent trips (63%) and falls (47%); pes cavus (34.5%); foot drop (4%); foot pain (27%); foot/leg cramps (36%). Activity limitations: walking (trips/falls, endurance, temporospatial); jumping (long jump).	Biased sampling method, small sample size, limited to CMT1A; poor methodological quality of clinical tests (except MRI*); frequency of symptoms not ascertainable. Biased sampling method, sample limited to CMT1A; sensation not tested.
Burns et al. (2009), ³ Australia	Cross-sectional; 81 children with CMT1A aged 2–16 years (mean age 8.5, SD‡ 3.3 years).	Impairments: weakness of foot/ankle muscles (0%); atrophy of foot intrinsics (100%); pathological MRI* findings in foot intrinsics (100%) and extrinsics (0%); reduced sensation (33%); pes cavus (67%); ankle equinus ≤10° (100%) Achilles tendon shortening (100%) Activity limitations: nil (Functional Disability Scale=normal, 100%) Impairments: decreased mean ankle DF† ROM‡, foot/ankle strength, balance, and power for all ages versus age-equivalent norms; decreased walking endurance for affected adolescents; ankle instability (72%); frequent trips (63%) and falls (47%); pes cavus (34.5%); foot drop (4%); foot pain (27%); foot/leg cramps (36%). Activity limitations: walking (trips/falls, endurance, temporospatial); jumping (long jump).	Biased sampling method, small sample size, limited to CMT1A; poor methodological quality of clinical tests (except MRI*); frequency of symptoms not ascertainable. Biased sampling method, sample limited to CMT1A; sensation not tested.

Note: *magnetic resonance imaging; †standard deviation; ‡dorsiflexion; §range of motion.



Figure 1 Pes cavus, illustrating an abnormally high medial longitudinal arch, a varus (inverted) hindfoot, a plantarflexed (downward) orientation of the first metatarsal/forefoot, an adducted (internally directed) forefoot, and claw toes (author's own photograph).

pes cavus is a cardinal manifestation of CMT and is thought to be related to selective atrophy and weakness of the foot intrinsics, peroneus brevis, and tibialis anterior muscles.^{8,19}

Pes cavus is a common but variable feature amongst children with CMT.^{3–8} The wide variation in frequency of pes cavus amongst children with CMT (21–100%) might reflect the heterogeneity which is inherent with the use of small sample sizes. Alternatively, or in addition, the wide variation in frequency of pes cavus might be related to the subjectivity of simply observing the presence/absence of pes cavus. A more valid and reliable way to quantify the severity of pes cavus is through the use of the Foot Posture Index (FPI), which is a standardized, six-item observational assessment tool.²³ The only study assessing children with CMT which used the FPI was conducted by Burns *et al.*,³ whereby pes cavus was reported in 28 of 81 (34.5%) children with CMT1A. Furthermore, weakness and imbalance of the extrinsic foot muscles were significantly related to the development of pes cavus. In the same study, pes cavus was shown to be more frequent amongst adolescents (63%) than young children. A longitudinal study of children with CMT1A by Garcia *et al.* also noted an increased frequency of pes cavus with age from 3 of 12 (25%) children aged 0–4 years to 5 of 10 (50%) children aged 5–10 years and 7 of 7 (100%) children aged 11–19 years,⁵ again demonstrating the progressive nature of CMT.

Poor balance

Balance is the functional outcome of a complex interaction between the sensorimotor systems of the body. A study by Burns *et al.*³ reported balance to be impaired in children of all ages with CMT1A, with 58 of 81 (72%) children also complaining of ankle instability during walking. The authors assessed

balance with the Bruininks–Oseretsky Test of Motor Proficiency (2nd Ed), a valid and reliable measure of motor ability in children,²⁴ although results for static balance were not separated from those for dynamic balance.

Poor balance in children with CMT could be secondary to a number of associated impairments, including pes cavus, foot and ankle muscle weakness, and reduced sensation. Pes cavus has been shown to shift the area of pressure under the foot laterally during walking,²⁵ which could contribute towards the development of inversion instability in children with CMT.^{1,26} Foot and ankle muscle weakness is likely to undermine the ability to make postural adjustments, especially during walking.^{27,28} Furthermore, reduced sensation has been shown to create a greater dependence on vision to make postural adjustments.²⁹

Ankle equinus

A limitation of ankle dorsiflexion range of motion, or ankle equinus, has been reported in children with CMT and is thought to result from contracture of the plantarflexors and/or intrinsic muscles of the foot, with subsequent shortening of the Achilles tendon.^{3,4} Burns *et al.*³ reported the average passive dorsiflexion range of motion to decrease from 27° in childhood to 23° in adolescence among 81 children with CMT1A. The authors used a weight-bearing lunge test with the knee flexed and measured the angle of ankle dorsiflexion with a digital inclinometer. Reference values for dorsiflexion range of motion using this method in healthy children are generally greater than 30°. In another study by Gallardo *et al.*,⁴ passive dorsiflexion range of motion was reported to be only 10° in each of the three children studied with CMT1A. The authors measured the angle of ankle dorsiflexion in non-weightbearing using a goniometer, yet this measurement tool is reported to have questionable validity and reliability.³¹ Irrespective of the method, it is clear that dorsiflexion range of motion is limited to some extent in most children with CMT.

Foot pain

Foot pain is associated with the development of pes cavus and increased loading under the forefoot during gait,^{32,33} as demonstrated by adults with various types of CMT.²⁶ A study by Burns *et al.*³ reported 22 of 81 (27%) children with CMT1A complained of foot pain, and foot pain was more common in older children. A long-term implication of prolonged foot pain may be muscle inhibition and disuse atrophy.³⁴

Foot/leg cramps

Cramps are sudden muscle spasms which can temporarily impair function and provoke pain.³⁵

The mechanism of cramps is related to disturbances of the central and peripheral nervous system and skeletal muscle.^{36,37} According to a study by Burns *et al.*³, foot/leg cramps are a relatively common complaint of children with CMT, affecting 29 of 81 (36%) children with CMT1A, and are more prevalent in older children. A separate study by Burns *et al.*³⁸ found the presence of foot/leg cramps was the strongest independent predictor of poor quality of life amongst a sample of 70 children with CMT1A.

Lower Limb Activity Limitations of Children with CMT

Activity limitations are defined as ‘difficulties an individual may have in performing activities’.¹⁶ The following section reviews the lower limb activity limitations of children with CMT. The main findings are summarized in Table 1. Activity limitations currently identified in children with CMT are difficulty walking^{3,5-7} and jumping.³

Difficulty walking

Difficulty walking is a common complaint of children with CMT. Approximately 50% of children with CMT type 1 report a clumsy gait,⁶ generally beginning within the first decade of life,⁷ with 51 of 81 (63%) of children with CMT1A reporting frequent trips and 38 of 81 (47%) reporting frequent falls.³ Children with CMT1A have also been shown to walk more slowly than age-equivalent norms, with a reduced step length and wider base of support.³

Foot drop is one factor that can cause difficulty walking for patients with CMT. Foot drop is where dorsiflexion weakness and/or ankle equinus causes the foot to fall into excessive plantarflexion during swing phase of walking.³⁴ Foot drop obstructs normal advancement of the lower limb and requires compensatory recruitment of more proximal muscles to avoid toe drag. Foot drop also disrupts the shock absorption and forward progression of momentum that normally occur with heel strike of stance phase because the foot lands with the foot flat or with toe contact instead.³⁴ Dorsiflexion of the ankle during swing phase and heel strike during stance phase is normally achieved in healthy children by 2 years of age.^{39,40}

Foot drop, when defined as absence of heel strike, was said to be a relatively uncommon feature amongst children with CMT1A (3/81; 4%) by Burns *et al.*³ Yet foot drop has been shown to be a universal sign amongst adults with CMT (64/64; 100%).⁴¹ In the latter study by Vinci and Perelli of Italy, foot drop was defined as the inability to actively dorsiflex the foot against gravity past neutral (mild), or in severe cases of foot drop, -10° or less of active dorsiflexion range of motion. Whilst a standardized method of assessing foot drop is needed, it appears

likely that foot drop is also much more prevalent in adults with CMT than children due to the progressive nature of the disease.

Children and adults with various types of CMT are thought to compensate for foot drop by increasing hip and knee flexion during swing phase of walking (i.e. ‘steppage’ gait).⁴² Whilst this compensatory strategy maintains walking ability, the resulting gait is slow, difficult, and tiring. It is thought that the steppage gait adopted by patients with CMT and foot drop causes premature fatigue of the hip flexor muscles and limits walking endurance.⁴³ Burns *et al.*³ used the 6-minute walk test to demonstrate a reduced endurance capacity of children with CMT1A when compared to age-equivalent norms, but only in adolescent participants. It might be that the temporospatial gait parameters (e.g. walking speed and step length) are beginning to mature by the time of adolescence in the healthy population,⁴⁴ resulting in improved walking efficiency and endurance. In contrast, the progressive development of dorsiflexion weakness and pes cavus in adolescent patients with CMT is associated with abnormal temporospatial gait parameters.³

Difficulty jumping

Jumping is an important part of a child’s play. In a study by Burns *et al.*³, children with CMT1A have been shown to have a reduced jumping ability, as measured by a standing long jump, when compared to healthy children. Jumping is thought to require an explosive recruitment of bi-articular muscles such as the gastrocnemius,⁴⁵ so poor jumping ability in children with CMT might reflect ankle plantarflexor weakness. However, only 3/81 (4%) of the children with CMT1A demonstrated difficulty tip toe walking, which is considered to be a gross indication of plantarflexor weakness.³ Similarly, none of the three children in the study by Gallardo *et al.*⁴ were said to have difficulty tip toe walking. These results suggest that jumping ability in children with CMT might also be limited by other factors such as poor balance and/or ankle equinus.

Orthoses

Orthoses are prescribed for some children with CMT as an important component of their overall treatment. The purpose of prescribing orthoses has traditionally been defined as:

‘Devices applied direct and externally to the patient’s body with the object of supporting, correcting or compensating for an anatomical deformity or weakness, however caused. It may be with the additional object of assisting, allowing or restricting movement of the body.’¹

Orthoses for patients with foot and ankle impairments include in-shoe orthoses and ankle-foot

Table 2 Characteristics, function, and indications of in-shoe orthoses

Orthoses	Superstructure	Trim lines	Mechanism	Function	Indications
Foot orthoses ⁴⁶⁻⁴⁹ (Fig. 2A)	N/A	N/A	Resist* movement of hindfoot (frontal plane); cushion foot deformity	Correct varus/valgus hindfoot; compensate pes cavus/planus by redistributing plantar pressure	Varus/valgus hindfoot and poor balance; pes cavus/planus and foot pain
Heel orthoses ^{48,49} (Fig. 2B)	N/A	Inferior to malleoli	Resist* movement of subtalar joint (frontal plane)	Correct calcaneovarus/valgus; compensate mediolateral foot instability	Calcaneovarus/valgus foot and poor balance
UCBL† orthoses ^{46,48,49} (Fig. 2C)	N/A	Inferior to malleoli	Resist* movement of hindfoot (frontal plane) and forefoot (transverse plane)	Correct pes cavus/planus; compensate mediolateral foot instability	Pes cavus/planus and poor balance

Note: *decreases the range, velocity, or force of a motion⁵³; †University of California Biomechanics Laboratory.

orthoses (AFOs). The characteristics, functions, and indications of various types of in-shoe orthoses and AFOs are summarized in Tables 2 and 3, respectively.

In-shoe orthoses

There are a number of different types of in-shoe orthoses described in the literature. The most commonly referred to in-shoe orthoses are custom-made foot orthoses, heel cup orthoses, and University of California Biomechanics Laboratory (UCBL) orthoses (Fig. 2).⁴⁶⁻⁴⁹ Custom foot orthoses can be made from a range of different materials, such as rigid polypropylene plastic, semi-rigid cork, or soft foam.⁴⁸⁻⁵⁰

Custom foot orthoses have been shown to be more effective than sham orthoses in reducing and redistributing abnormal ground reaction forces during walking in participants with bilateral pes cavus of any etiology, including 16 adults with CMT.⁵¹ Normalizing ground reaction forces also correlated to improved function, quality of life, and reduced foot pain. It seems logical that custom in-shoe foot orthoses would have a similar effect for children with CMT and pes cavus.

Heel cup orthoses are made of plastic which is specifically moulded to grasp the medial, posterior, and lateral aspects of the calcaneus.^{49,50} In this way, heel cup orthoses are designed to correct a varus or valgus hindfoot deformity during weight-bearing. UCBL orthoses are similar to heel cup orthoses in that they grasp the calcaneus, but they usually extend just proximal to the metatarsal heads to provide additional abduction/adduction control of the forefoot.^{47,49,50}

Ankle-foot orthoses

Ankle-foot orthoses are orthotic devices that incorporate some portion of the foot and leg and are indicated where more control is necessary than can be provided by in-shoe orthoses. There are a number of different types of AFOs described in the literature. The most commonly referred to plastic AFOs are

supramalleolar AFOs, posterior leaf spring AFOs, hinged AFOs, hinged AFOs with plantarflexor stops, spiral AFOs, hemispiral AFOs, solid AFOs, and floor reaction AFOs (Fig. 3).^{46,47,49,50,52,53} Carbon-fibre AFOs are a more recent development but were not included in this review due to the scarcity of evidence regarding their use.

AFOs feature a foot plate, a foot control, an ankle control, and a superstructure.^{46,52} The foot plate acts as a shoe insert to support the foot. The foot control and ankle control are arbitrary terms that can be described in terms of the amount and direction of movement that is permitted at the foot and ankle (i.e. free, held, stopped, resisted, or assisted motion).^{46,49,54} The superstructure refers to the metal uprights or plastic shell of the AFO, although traditional metal superstructures are rarely used anymore; they have largely been replaced by the development of heat-moulded plastic superstructures since the late 1960s and early 1970s.⁵⁵ The superstructures of AFOs terminate at some point below the knee; they act to provide leverage for the orthoses and influence how much foot and ankle movement is permitted. For example, supramalleolar AFOs have superstructures which terminate just proximal to the malleoli, thus providing moderate leverage and moderate control. By comparison, AFOs with full length calf coverage superstructures (e.g. posterior leaf spring AFOs, hinged AFOs, hinged AFOs with plantarflexor stops, solid AFOs, and floor reaction AFOs) provide superior leverage and superior control. Spiral and hemispiral AFOs are unique in that they have superstructures which spiral from the foot plate and around the leg, providing moderate, multidirectional leverage and moderate, multidirectional control.

The borders, or trim lines, of plastic superstructures also influence how much foot and ankle movement is permitted.^{46,49,56} For example, the trim lines of posterior leaf spring AFOs lie posterior to the malleoli, thus permitting relatively normal foot and ankle movement. Spiral and hemispiral AFOs also

Table 3 Characteristics, function, and indications of ankle-foot orthoses (AFOs)

Orthoses	Superstructure	Trim lines	Mechanism	Function	Indications
Supramalleolar AFOs* ^{46,47,49,50} (Fig. 3A)	Proximal to malleoli	Anterior to malleoli	Hold† hindfoot (frontal plane) and forefoot (transverse plane)	Correct pes cavus/plenus; compensate mediolateral foot instability	Pes cavus/plenus and poor balance (not corrected by UCBL‡ orthoses)
Posterior leaf spring AFOs* ^{47,49,50,52,53} (Fig. 3B)	Full length calf coverage	Posterior to malleoli	Assist‡ ankle DFI and resist‡ ankle PF** during swing phase	Compensate foot drop	Foot drop (due to ankle DFI weakness and/or ankle equinus $\geq 5^\circ$) and poor walking
Hinged AFOs* ^{46,47,49,50,52} (Fig. 3C)	Full length calf coverage	Anterior to malleoli	Hold† hindfoot (frontal plane) and forefoot (transverse plane)	Correct pes cavus/plenus; compensate mediolateral foot instability	Pes cavus/plenus and poor balance (not corrected by supramalleolar AFOs*)
Hinged AFOs* with PF** stops ^{46,47,49,50,52,53}	Full length calf coverage	Anterior to malleoli	Hold† hindfoot (frontal plane) and forefoot (transverse plane) and stop†† PF**	Correct pes cavus/plenus; compensate mediolateral foot instability; compensate foot drop	Pes cavus/plenus, poor balance, foot drop (due to ankle DFI weakness and/or ankle equinus $\geq 5^\circ$), and poor walking
Spiral AFOs* ^{46,50,52}	Spirals 360° from medial foot plate to medial tibial condyle	Posterior to malleoli	Resist‡ movement of ankle (sagittal plane), hindfoot (frontal plane), and forefoot (transverse plane), especially pronation, and assist‡ DFI during swing phase	Correct pes planus; compensate mediolateral foot and anterior/posterior ankle instability; compensate foot drop	Global foot weakness and poor balance and/or walking (with/without pes planus and/or foot drop)
Hemispiral AFOs* ⁵⁰	Spirals 180° from lateral foot plate to medial tibial condyle	Posterior to malleoli	Resist‡ movement of ankle (sagittal plane), hindfoot (frontal plane), and forefoot (transverse plane), especially supination, and assist‡ DFI during swing phase	Correct pes cavus; compensate mediolateral foot and anterior/posterior ankle instability; compensate foot drop	Global foot weakness and poor balance and/or walking (with/without pes cavus and/or foot drop)
Solid AFOs* ^{46,47,49,52,53} (Fig. 3D)	Full length calf coverage	Anterior to malleoli	Hold† ankle (sagittal plane), hindfoot (frontal plane), and forefoot (transverse plane)	Correct/support pes cavus/plenus; compensate mediolateral foot and anterior/posterior ankle instability; compensate foot drop; support ankle equinus ($\geq 0^\circ$); compensate PF†† spasticity	Global foot weakness and poor balance and/or walking (not corrected by spiral/hemispiral AFOs*, with/without pes cavus/plenus and/or foot drop); and/or ankle equinus ($\geq 0^\circ$, not corrected by hinged AFOs* with/without PF** stops); PF†† spasticity
Floor reaction AFOs* ^{46,47,49,50,53} (Fig. 3E)	Full length calf coverage and anterior band at proximal third of the tibia	Anterior to malleoli	Hold† ankle (sagittal plane), hindfoot (frontal plane), and forefoot (transverse plane) and assist‡ knee extension during stance phase	Correct pes cavus/plenus; compensate mediolateral foot instability; compensate weak ankle PF**, weak knee extension, knee flexor contracture and/or over-lengthened Achilles tendon	Crouch gait pattern (excessive hip flexion, knee flexion, and ankle dorsiflexion during stance phase, with/without pes cavus/plenus and/or poor balance)

Note: *ankle-foot orthoses; †elimination of all motion in prescribed plane,⁵³ ‡University of California Biomechanics Laboratory; §increases the range, velocity, or force of a motion,⁵³ ¶dorsiflexion; ††decreases the range, velocity or force of a motion;⁵³ **plantarflexion; ††inclusion of a static unit to deter an undesired motion in one direction,⁵³ ‡‡plantarflexor.

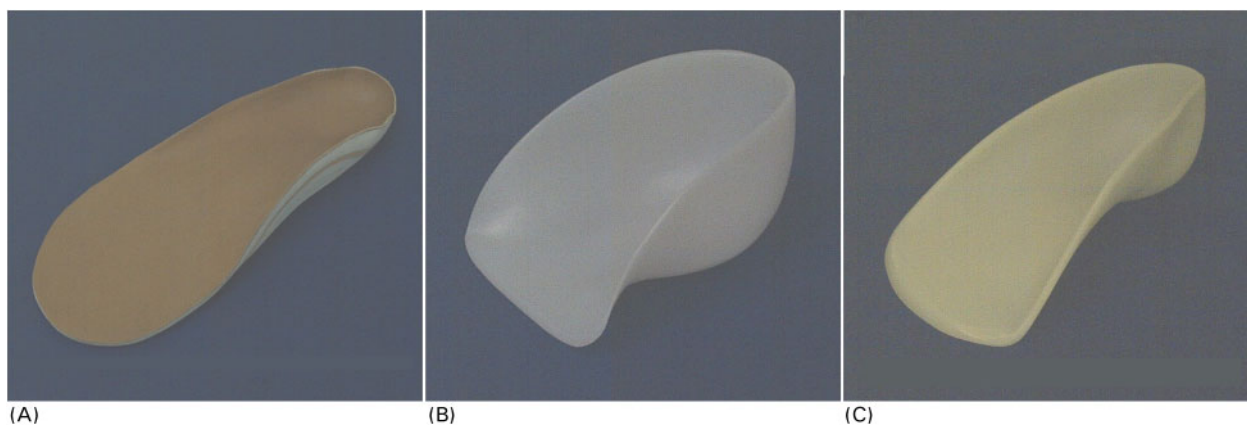


Figure 2 (A) Semi-rigid foot orthosis (courtesy of The Children’s Hospital at Westmead, Orthotics Department). (B) Heel cup orthosis (courtesy of The Children’s Hospital at Westmead, Orthotics Department). (C) University of California Biomechanics Laboratory (UCBL) orthosis (courtesy of The Children’s Hospital at Westmead, Orthotics Department).

have trim lines posterior to the malleoli, but the orientation of the superstructures provide moderate, multidirectional resistance to foot and ankle movement. Superstructures with trim lines anterior to the malleoli (e.g. supramalleolar AFOs, hinged AFOs, hinged AFOs

with plantarflexor stops, solid AFOs, and floor reaction AFOs) incorporate the ankle and forefoot, acting to hold the foot in the frontal and transverse anatomical planes.

Other qualities of AFOs which influence how much foot and ankle movement is permitted are the type

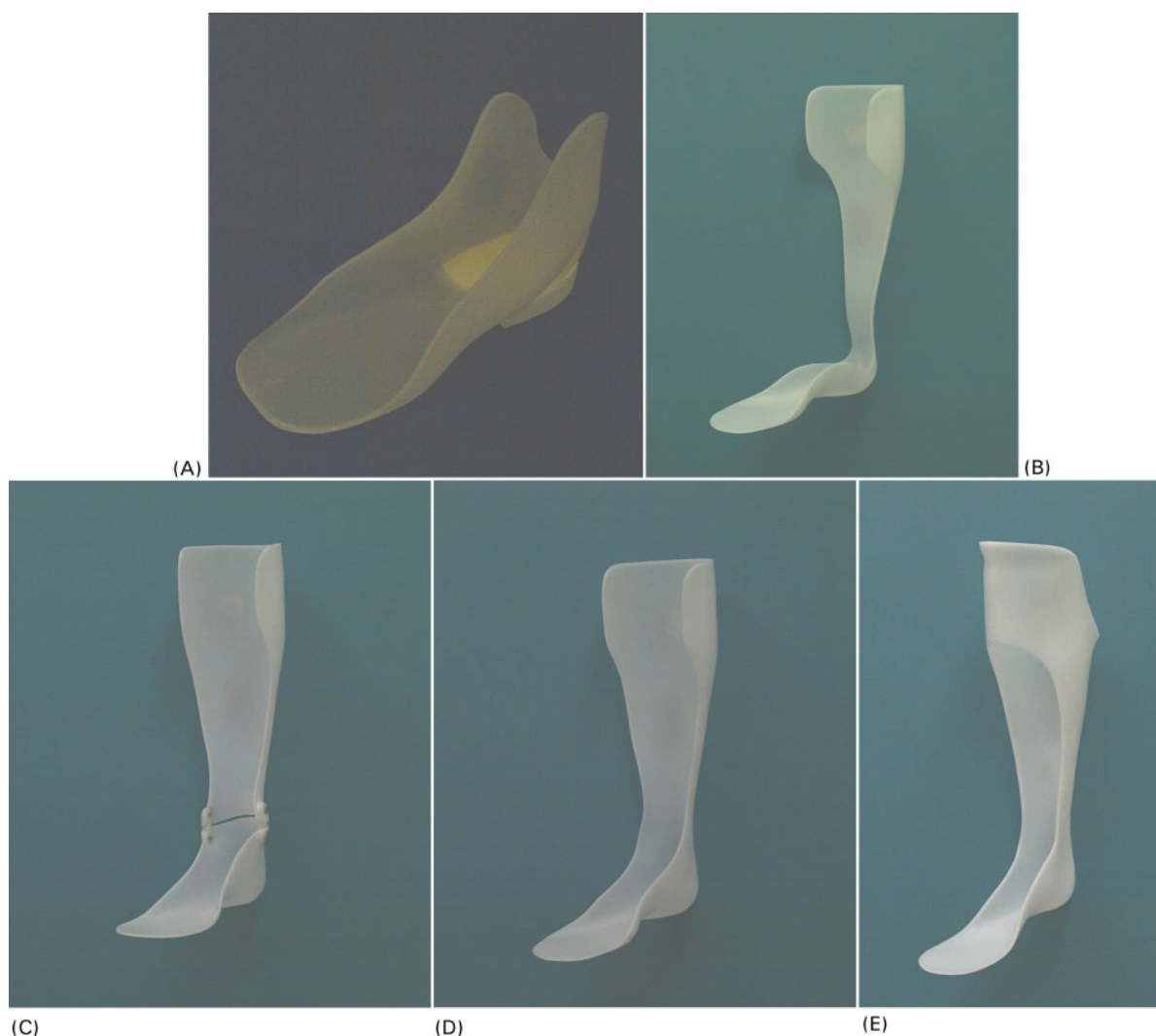


Figure 3 (A) Supramalleolar orthosis (courtesy of The Children’s Hospital at Westmead, Orthotics Department). (B) Posterior leaf spring AFO (courtesy of The Children’s Hospital at Westmead, Orthotics Department). (C) Hinged AFO (courtesy of The Children’s Hospital at Westmead, Orthotics Department). (D) Solid AFO (courtesy of The Children’s Hospital at Westmead, Orthotics Department). (E) Floor reaction AFO (courtesy of The Children’s Hospital at Westmead, Orthotics Department).

and rigidity of the plastic.⁵⁰ Most AFOs are made from polyethylene, which is relatively flexible. Polypropylene is a stiffer plastic and is used where greater strength is required.⁵⁰

It is also important to note that a proper-fitting shoe is essential for efficacious lower limb orthotic prescription.^{52,57} Many parts of a shoe can be modified but is beyond the scope of this review.

Indications for Foot and Ankle Orthoses in Children with CMT

A clinical algorithm for the optimal prescription of foot and ankle orthoses for children with CMT is presented in Table 4. The algorithm is based on the preceding information detailing the lower limb impairments and activity limitations of children with CMT and the characteristics, functions, and indications of commonly prescribed foot and ankle orthoses. The algorithm was formulated by grouping together various combinations of lower limb impairments and activity limitations to reflect the diverse range of clinical presentations seen in children with CMT and arranging them in a hierarchical order of increasing severity. It follows that the corresponding orthoses offer an increasingly greater amount of control.

The most fundamental level of orthoses prescription for children with CMT is custom foot orthoses for foot pain associated with pes cavus. However, if the pes cavus is also causing poor balance, orthoses capable of correcting the deformity and compensating for mediolateral foot instability are required. UCBL orthoses, supramalleolar orthoses, and hinged AFOs are all designed to provide an increasingly greater level of foot and ankle control.^{46,47,49,50,52} The difference between the amount of control they each provide is attributable to the leverage and trim lines of the superstructure. UCBL orthoses technically do not have a superstructure; they terminate below the malleoli and so provide negligible mediolateral foot

control. The superstructures of supramalleolar AFOs terminate just proximal to the malleoli and the trim lines lie anterior to the malleoli, thus providing moderate mediolateral foot control. The superstructures of hinged AFOs encompass the full length of the calf, terminating just distal to the knee crease, and the trim lines also lie anterior to the malleoli, thus providing superior mediolateral foot control. It is likely that custom foot orthoses could also be incorporated into the foot plate of supramalleolar orthoses and hinged AFOs (as well as other AFOs) to reduce foot pain where indicated, although this practice is not reported in the literature for children with CMT.

UCBL orthoses, supramalleolar orthoses, and hinged AFOs are all designed to permit normal, unrestricted foot and ankle kinematics in the sagittal anatomical plane during walking. However, sagittal plane control is required if foot drop is present. If foot drop is present in isolation, posterior leaf spring AFOs are indicated.^{47,49,50,52,53} Posterior leaf spring AFOs are inherently flexible because the trim lines taper at the ankle control and lie posterior to the malleoli. This design allows posterior leaf spring AFOs to bend under the load of body weight during stance phase of walking, thus allowing relatively normal foot and ankle kinematics in the sagittal plane. During swing phase, posterior leaf spring AFOs are designed to spring back at the ankle control to assist dorsiflexion and resist plantarflexion to compensate for foot drop. Posterior leaf spring orthoses should only be prescribed if foot drop is causing reduced walking ability (e.g. frequent trips/falls or abnormal temporospatial gait parameters).

If foot drop is present in combination with pes cavus and causing difficulty walking and impaired balance, hinged AFOs with plantarflexion stops are indicated. These orthoses are designed to provide both sagittal and mediolateral plane control.^{46,47,49,50,52,53} The trim lines of the superstructures lie anterior to the

Table 4 Clinical algorithm for the optimal prescription of foot and ankle orthoses for children with CMT

Impairments and activity limitations	Orthoses
Pes cavus and foot pain	Foot orthoses
Pes cavus and poor balance	UCBL* orthoses
Pes cavus and poorer balance (not corrected by UCBL* orthoses)	Supramalleolar orthoses
Pes cavus and poorer balance (not corrected by supramalleolar AFOs†)	Hinged AFOs†
Foot drop and poor walking	Posterior leaf spring AFOs†
Foot drop, poor walking, pes cavus, and poor balance	Hinged AFOs† with PF‡ stops
Global weakness of foot/ankle muscles and poor walking and/or balance (with/without pes cavus and/or foot drop)	Hemispiral AFOs†
Global weakness of foot/ankle muscles and poorer walking and/or balance (not corrected by hemispiral AFOs†, with/without pes cavus and/or foot drop)	Solid AFOs†
Pes cavus and/or ankle equinus ($\geq 0^\circ$, not corrected by hinged AFOs† with/without PF‡ stops)	Solid AFOs†

Note: *University of California Biomechanics Laboratory; †ankle-foot orthoses; ‡plantarflexion.

malleoli, thus affording adequate mediolateral stability to correct the pes cavus and compensate for poor balance. Whilst the plantarflexion stops compensate for foot drop during swing phase of walking, they also create a greater knee flexion moment at heel strike during stance phase. This increased knee flexion moment must be tolerated by the knee extensor muscles. Hinged AFOs with plantarflexion stops should therefore only be prescribed if the patient demonstrates poor walking and balance in the presence of adequate knee extensor strength.

Where poor walking and/or poor balance are implicated in combination with global weakness of the foot and ankle muscles, multidirectional control is required. Hemispiral AFOs are designed to resist, but not eliminate, movement of the foot and ankle in all directions.⁵⁰ The superstructures of hemispiral AFOs begin at the lateral aspect of the foot plate and wrap 180° around the posterior calf, terminating near the medial tibial condyle. In this way, the orientation of hemispiral superstructures is well designed to correct pes cavus deformities. Also, the multidirectional control provided by hemispiral AFOs compensates for foot drop during swing phase of walking and foot and ankle instability during stance phase. However, resisting movement of the foot and ankle during stance phase will also disrupt normal foot and ankle kinematics and function.

If hemispiral AFOs are inadequate to compensate for the extent of global weakness of the foot and ankle muscles and poor walking and/or poor balance, solid AFOs are indicated. Solid AFOs are designed to hold the foot and ankle in all anatomical planes and reduce the degrees of freedom.^{46,47,49,52,53} However, the high degree of stability afforded by solid AFOs will further disrupt normal foot and ankle kinematics and function during walking, as well as during the performance of other everyday activities such as standing up and sitting down.

Alternatively, solid AFOs can be prescribed to support ankle equinus deformities, so long as the ankle still permits at least plantargrade alignment. Solid AFOs can also be prescribed to support pes cavus deformities which cannot be corrected by hinged AFOs (with or without plantarflexion stops). It is thought however that surgery is indicated if the varus hindfoot component of a pes cavus deformity cannot self-correct.⁵⁸ Similarly, if the interface pressures between the orthoses and the body are intolerable for the patient, serial casting or surgical management is required.^{1,18}

Discussion

This review of lower limb impairments and activity limitations of children with CMT and commonly

prescribed foot and ankle orthoses has enabled the formulation of a clinical algorithm for the optimal prescription of foot and ankle orthoses for children with CMT. The clinical algorithm is depicted in Table 4. In general, in-shoe orthoses are indicated for children with CMT and pes cavus and foot pain and/or mild balance impairments, whilst ankle-foot orthoses are indicated for children with CMT and pes cavus, foot drop, global foot and ankle muscle weakness and/or ankle equinus, and moderate–severe balance impairments and/or difficulty walking.

It follows that the optimal prescription of foot and ankle orthoses for children with CMT requires valid and reliable clinical testing of these factors. The study by Burns *et al.*³ provides good examples of such tests: pes cavus can be assessed using the FPI; muscle strength using dynamometry; balance using the Bruininks–Oseretsky Test of Motor Proficiency; and walking ability using an instrumented walkway (e.g. GAITRite). In this way, clinical findings can be compared to normative data to ascertain the degree of impairment. The patient's progress over time and response to any subsequent treatment interventions (e.g. orthoses) can then be monitored also.

It is important to work collaboratively with the patient and their family. The implications of the clinical findings, along with the proposed treatment options, should be clearly explained to the patient and their family. If a particular type of orthoses is deemed to be the most appropriate therapeutic intervention, it is important to gain informed consent and improve patient compliance by discussing the possible benefits and disadvantages of the orthoses. A study by Vinci and Gargiulo⁵⁹ of Italy demonstrated poor compliance amongst adults with CMT who were prescribed orthoses. The authors surmised that adults with CMT are not likely to wear orthoses if they cause adverse physical and psychological consequences which outweigh their potential benefits. Orthoses have also been shown to have a negative psychological impact on children who wear them for spinal deformities.^{60,61} Together, these findings reiterate the importance of determining a patient's suitability for orthoses on an individual basis. Furthermore, it is essential that the correct type of orthoses is prescribed so that adequate support, correction, and/or compensation is provided and any interference to normal lower limb kinematics is minimized. Orthotists can also enhance patient compliance by improving the cosmesis of orthoses for children with colourful patterns and designs.

Finally, whilst the clinical algorithm presented in Table 4 aids clinical reasoning, it is not evidence-based. Further research is still required to determine the efficacy of different foot and ankle orthoses, and the predictive ability of the proposed clinical

algorithm, to improve the lower limb impairments and activity limitations of children with CMT. Ideally, the evidence-based research framework pertaining to the prescription of orthoses for children with cerebral palsy^{62,63} should to be replicated for children with CMT. For this, high quality randomized controlled trials and subsequent systematic reviews are essential.

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