

Congenital clubfoot: a review

Congenital clubfoot is a relatively common lower limb deformity that is increasingly managed by non-operative methods of serial manipulation and immobilization. This review looks at aetiology, examination techniques and severity scoring, and provides an overview of operative and non-operative methods of treatment.

Clubfoot or congenital talipes equinovarus is a lower limb abnormality that presents at birth with a characteristic deformity of forefoot cavus and adduction, heel varus and equinus of one or both feet. The deformity can be broadly classified as simple or complex, with atypical variants associated with other conditions such as arthrogyriposis and myelomeningocele accounting for up to 7.7% of all cases (Siapkara and Duncan, 2007). Recently there has been an important trend towards non-operative management, with widespread use of the Ponseti method as well as other techniques of manipulation and immobilization. This review will cover aetiology, examination techniques, severity scoring, and provide an overview of operative and non-operative methods of treatment.

Aetiology

Congenital talipes equinovarus represents a wide spectrum of deformity, and the aetiology appears to be multifactorial, with both genetic and environmental factors implicated. Clubfoot has an incidence of 1 per 1000 live births in the UK, with a male to female ratio of 2–3:1 and is bilateral in up to 50% of cases. Several studies have addressed the genetic factors; population, familial and twin studies have all found evidence of a genetic component, but the mode of inheritance does not follow a distinct pattern. There is a wide ethnic variation, with an incidence of up to 7 per 1000 in the Polynesian population. Incidence is increased among first-degree relatives, and siblings have a 3% risk of clubfoot (Siapkara and Duncan, 2007).

There are several theories that growth disturbance in utero as a result of different factors, including chemical teratogens, radiation, hormones and other environmental factors, can lead to clubfoot (Cummings et al, 2002). However, although the fetal foot progresses through stages of equinus, hindfoot and forefoot adduction followed by supination, plantar-flexion and subsequent development towards adult morphology

(Böhm, 1929), medial displacement of the navicular does not occur at any point during normal development. The moulding theory, first proposed by Hippocrates, attributes formation of the deformity to external uterine compression and oligohydramnios, but this is not supported by the timing of clubfoot development during the first trimester, when there is ample intrauterine volume.

A number of macroscopic and microscopic soft tissue abnormalities have also been implicated, although there is some debate as to whether they represent primary or secondary changes. Increased collagen synthesis, myo-fibroblasts and differential regional growth disturbance have all been implicated. The retraction fibrosis theory is based on the finding of increased fibrous tissue and myoblasts in calf musculature, and may contribute to recurrence (Ippolito and Ponseti, 1980). Investigators have found smaller and shorter calf muscles, and an accessory soleus muscle on the affected limb and anomalous tendon insertions (Porter, 1996). Other studies have found small or absent anterior tibial and dorsalis pedis arteries on the affected side, and a high prevalence of absent dorsalis pedis pulse in the parents of affected children. An increased ratio of type I:II muscle fibres and increased percentage of connective tissue in affected limbs, and the association with arthrogyriposis, spina bifida and sacral agenesis, all suggest a neurogenic aetiology (Handelsman and Badalamente, 1981).

Pathoanatomy

The complex deformity in clubfoot can be separated into four major components: forefoot cavus and adduction, heel varus and equinus, summarized by the mnemonic CAVE (Cooke et al, 2008) (*Figure 1*).

Although the foot appears supinated, forefoot cavus is the result of plantar flexion of the first ray and pronation of the forefoot relative to the hindfoot. In unilateral clubfoot, the affected lower limb may be shorter with a thinner calf and shorter foot.

The hindfoot is supinated and in equinus; the talus is wedge shaped and medially rotated, with the displaced talar head palpable dorsolaterally in the sinus tarsi. The calcaneus is plantarflexed, medially displaced and vertical, so that it may not be palpable. The mid-foot is adducted and supinated, with cuneiforms and metatarsals that are adducted but normal. The navicular is wedge shaped, medially displaced and vertical, exposing the talar head in the sinus tarsi, with the

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cuboid less severely displaced medially over the calcaneal head. The forefoot appears supinated, but is often pronated relative to the hindfoot in more severe cases, with plantarflexion of the first ray causing the cavus deformity.

The posterior and medial soft tissue structures are thickened and contracted, including joint capsules, ligaments and muscles. The medial structures are shortened, including the deltoid and spring ligaments and the tibialis posterior tendon, which contribute to the adduction deformity. Contracted posterior ligaments and Achilles tendon with tight calf musculature contribute to the equinus deformity, and the peroneal muscles and tibialis posterior often have thickened tendon sheaths. Contracture of the plantar fascia contributes to the cavus deformity. As the tibialis anterior tendon becomes medialized it acts as a deforming force, increasing the supination and adduction deformities.

Examination

The child should be assessed soon after birth by an orthopaedic surgeon or a physiotherapist with training in clubfoot management. It is necessary to use a standardized, systematic approach including a 'head-to-toe' examination to exclude any associated abnormalities (e.g. spina bifida, arthrogryposis). Differential diagnoses include flexible positional talipes, congenital vertical talus and severe metatarsus adductus. In older children, note whether the heel is bearing any weight on standing, whether the foot is plantigrade, and look for abnormal distribution of plantar callosities.

The foot should be examined with the knee in extension and at 90° of flexion, to separate the contribution to the equinus deformity of gastro-soleus complex contracture from ankle joint stiffness (Cummings et al, 2002). The knee is held in 90° of flexion to provide a reference point for assessment of rotational deformity of the foot and internal tibial torsion. If the calcaneus is not palpable, the heel will feel small, soft and empty. The displaced talar head and neck are uncovered and palpable dorsolaterally in the sinus tarsi, and the medial malleolus may be difficult to palpate, and often lies in contact with the navicular. Midfoot stiffness can be assessed by attempting to abduct the forefoot onto the displaced talar head. Note the presence of a medial crease or curved lateral border of the foot (Figure 2), which relate to the severity of the deformity and prognosis (Dyer and Davis, 2006).

Severity scores

Clinical scoring systems have been designed to quantify the severity of the clubfoot, using a range of parameters. The Pirani (Pirani et al, 1999) and Diméglio scores (Diméglio et al, 1995) are the most commonly used in clinical practice, and both have excellent rates of inter-observer reliability and reproducibility (Flynn et al, 1998).

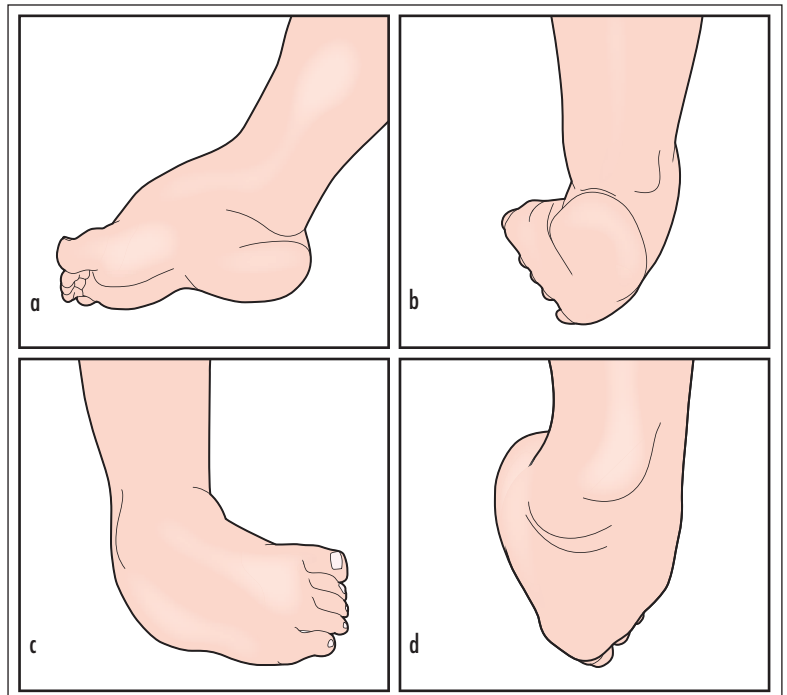


Figure 1. a. Cavus. b. Adductus. c. Varus. d. Equinus.

The Pirani severity score (Dyer and Davis, 2006) uses six clinical signs – three midfoot and three hindfoot score – giving a score out of six (Table 1). The parameters can be recorded at intervals during treatment with the Ponseti method to assess progress and as a prognostic indicator to determine if an Achilles tenotomy will be required.

The Diméglio score (Diméglio et al, 1995) can also be used to classify clubfoot deformities and monitor the response to treatment. A scale of 1–20 is derived from the reducibility of four different parameters scored out of four using a goniometer, with four additional points (Table 2). The deformity can be assigned a grade I–IV with increasing severity. A score of 1–5 equates to grade I, 6–10 with grade II, 11–15 with grade III, and 16–20 with grade IV. The four parameters recorded are equinus, varus, derotation of calcaneo-forefoot block, and adduc-

Figure 2. Bilateral clubfoot deformity with medial crease and curved lateral border.



tion of the forefoot relative to the hindfoot. One point is added for each of the following findings: a marked posterior crease, a marked mediotarsal crease, plantar retraction or cavus, and the condition of the muscles, especially tibialis anterior.

Imaging

Antenatal ultrasound scanning can be used to diagnose clubfoot in utero; one study found a positive predictive value of 83% and a false positive rate of 17% (Bar-On

et al, 2005). Other studies have found a higher false-positive rate, especially for unilateral clubfoot (up to 29%), and higher rates of associated anomalies with bilateral clubfoot. Ultrasound can detect clubfoot as early as 12 weeks, but the false-positive rate may be raised during this period as a result of delayed skeletal maturation, and scanning is more accurate after 18–20 weeks of gestation. The degree of deformity cannot be reliably predicted antenatally, with one study finding 26% of antenatally diagnosed cases of clubfoot did not require any intervention (Tillett et al, 2000), although it may help parental preparation and early referral to a specialist unit.

Imaging is not routinely used in the clinical treatment of clubfoot, although radiographs are sometimes used to assist preoperative planning. Studies using three-dimensional computed tomography have demonstrated the inaccuracies of radiographical measurements, as the involved bones are mostly cartilaginous at birth with small ossification centres, but this is not used clinically. Serial magnetic resonance imaging scans have been used to accurately assess the response to different techniques of non-operative management (Pirani et al, 2001), but this is not indicated in clinical practice.

Management

The aims of treatment are to give the patient a flexible, painless, plantigrade foot with as near normal function as possible. Management can be non-operative or operative; the approach to managing virgin clubfoot in the infant or young child may differ greatly from managing complex, recurrent or neglected clubfoot. Regardless of which method of treatment is used, the deformities are never corrected completely, and the affected calf will often remain thinner and the foot shorter. However, even persistent medial navicular displacement is compatible with a normal-looking, pain free, functional plantigrade foot (Ponseti, 1992).

Non-operative

Non-operative management is based on serial manipulation and immobilization. The different techniques are all based on the principles of plastic deformation and stress-relaxation. Overcorrection or incorrect technique can result in a number of iatrogenic complications, e.g. rocker-bottom deformity.

Ponseti

Ignatio Ponseti first published his method of serial manipulations and castings in 1963 (Ponseti and Smoley, 1963), but it did not gain widespread popularity until a long-term follow-up study was published in 1995 (Cooper and Dietz, 1995). The study found that 78% of patients treated with the method had good or excellent long-term function, compared with 85% in an age-matched normal population without the deformity.

Table 1. The Pirani score

Midfoot score	Curved lateral border	Normal	0
		Medial distal deviation	0.5
		Severe proximal deviation	1
	Medial crease	No medial crease	0
		Moderate medial crease	0.5
		Deep medial crease	1
	Talar head coverage	Reduced talo-navicular joint	0
		Subluxed reducible talo-navicular joint	0.5
		Irreducible talo-navicular joint	1
Hindfoot score	Posterior crease	No crease	0
		Moderate crease	0.5
		Deep crease	1
	Rigid equinus	Normal dorsiflexion	0
		Plantigrade with knee extended	0.5
		Fixed equinus	1
	Empty heel	Hard heel	0
		Moderately soft heel	0.5
		Soft heel (calcaneum not palpable)	1

From Pirani et al (1999)

Table 2. The Diméglio score

Characteristics	Points			
	Equinus	Varus	Calcaneo-forefoot block derotation	Adduction
90° to 45°	1	1	1	1
90° to 20°	2	2	2	2
90° to 0°	3	3	3	3
90° to -20°	4	4	4	4
Other parameters				
Posterior crease	1			
Medial crease	1			
Cavus	1			
Poor muscle condition	1			
Score/20				

From Diméglio et al (1995)

Previous techniques advocated pronation of the forefoot (Kite, 1972), which actually increased the cavus deformity. The Ponseti method differs in that the cavus deformity is corrected first by supinating the forefoot to bring it into correct alignment with the hindfoot. This is followed by simultaneous correction of the varus, adduction, and equinus of the hindfoot by gentle manipulation with counter pressure applied over the head of the talus (Figure 3). Treatment with this method can be performed successfully with 4–10 casts changed every 5–7 days by an orthopaedic surgeon or physiotherapist, and progress can be monitored with serial Pirani scores. The toe-to-groin casts are applied with the knee flexed to 90° and the final cast is applied with the foot in an overcorrected position of 70° abduction and 20° ankle dorsiflexion (Figure 4). Ponseti has authored a free handbook that explains his method (Staheli, 2009).

The corrected position is maintained in an abduction brace with the ankles dorsiflexed in 60° of external rotation. The brace should only be removed for up to 1 hour a day for the first 3 months and is worn only at night for a further 2–4 years. The correct continued use of a brace is important to prevent recurrence of clubfoot, and parent education and compliance are essential for successful treatment. Non-compliance with the brace is implicated in a large number of cases of failed conservative treatment or recurrence (Dobbs et al, 2004).

Depending on the severity of the initial deformity and the response to treatment, the castings may be supplemented by a percutaneous tendo Achilles tenotomy to correct residual equinus (in 70–90% of patients) and less frequently a tibialis anterior transfer (required in 15–30%). Tendo Achilles tenotomy can be performed on infants under local anaesthetic in an outpatient clinic setting with no need for tendon lengthening or grafting, and there are no reported cases of failure of tendon healing. Tibialis anterior tendon transfer is indicated if there is persistent excessive supination, and involves transfer of its insertion to the lateral cuneiform. Most cases of recurrence or relapse can be successfully treated with a further course of serial manipulation and immobilization, although resistant cases may require surgical treatment.

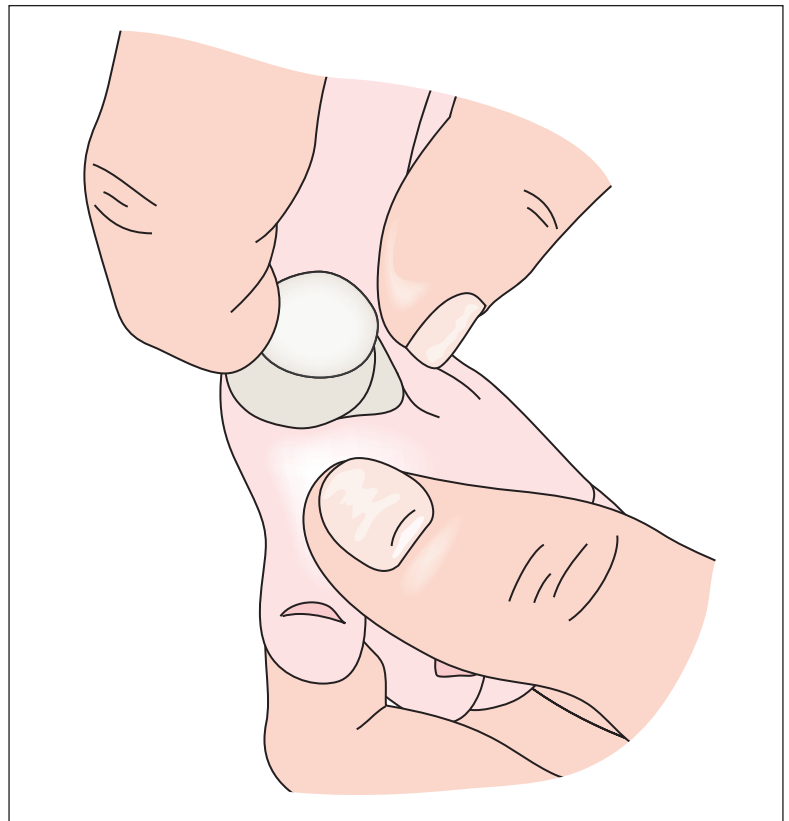


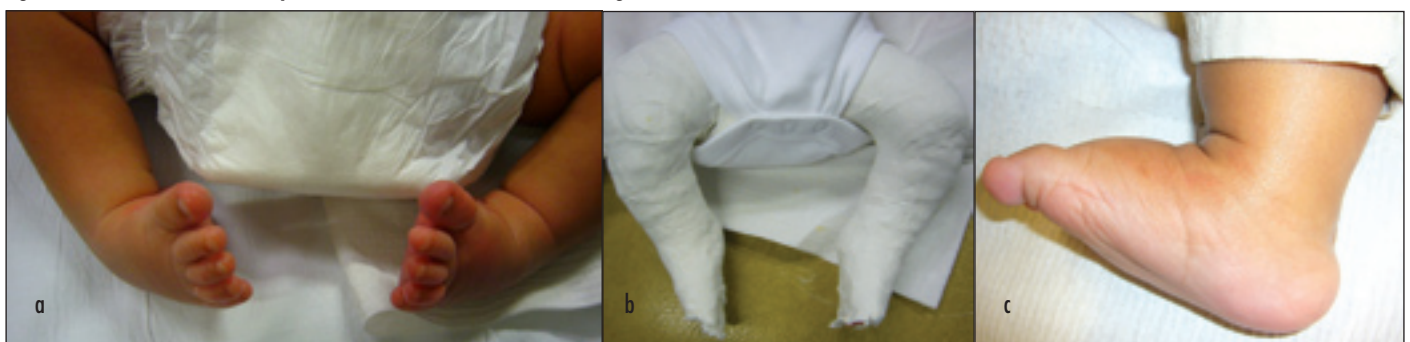
Figure 3. Correction of varus, adduction and equinus with gentle pressure over the uncovered head of the talus.

French

The French technique described by Bensahel et al (1990) is based on an intensive daily regimen of soft tissue massage, manipulation and taping by a specialist physiotherapist over the course of several months. Some authors have advocated the additional daily use of a continuous passive motion machine to maintain the degree of correction.

The gradual sequential correction of the deformities can lead to an excellent outcome, although the recurrence rate and need for secondary surgery is currently higher than with the Ponseti method. It also takes longer, and requires greater resources in terms of physiotherapy sessions as well as a high degree of parent understanding and compliance (Bensahel et al, 1990).

Figure 4. a. Bilateral clubfoot at presentation. b. Bilateral Ponseti casting. c. Position after final cast.



Operative

The incidence of surgery for the primary treatment of clubfoot has decreased dramatically as non-operative manipulation techniques have grown in popularity. The approach to surgery can be the 'one size fits all' principle described by Turco (1979) or the 'à la carte' approach of Bensahel, in which the minimum surgery is performed by tailoring the operation to the individual, reducing the risk of overcorrection. Surgery is mainly indicated after failure of non-operative management, delayed presentation, or in cases of recurrent or complex clubfoot associated with other syndromes. The relatively common 'limited surgery' procedures of Achilles tendon release and tibialis anterior tendon transfer are often not considered as surgical management, but rather as augments to non-operative methods.

Primary surgery (soft tissue release)

Soft tissue procedures include posteromedial, plantar and lateral release of contractures. An extensive posteromedial release is performed for residual hindfoot equinus and varus, and involves ankle and subtalar joint capsulotomy, medial ligament complex release, tibialis posterior lengthening and flexor tendon release. Plantar release is performed to correct cavus deformity, and an additional lateral release of talo-navicular and calcaneo-cuboid joint capsules may also be required. A 25-year follow-up study of patients who underwent soft tissue correction found that only 50% had good or fair results with no excellent results and 87% required further surgery. They also found a high incidence of persistent pain, weakness and residual deformity (Köse et al, 1999).

Revision surgery

Revision surgery is indicated for recurrence after soft tissue correction in approximately 25% of patients (Cummings, 2002). In patients under 2 years of age, a repeat soft tissue release may be attempted before resorting to extensive bony osteotomies, which are more commonly performed after the age of 4 years, in conjunction with further soft tissue release.

Forefoot adduction and supination are the most common persistent deformities. Numerous procedures have been described for the correction of residual adduction deformity, including a lateral calcaneocuboid closing-wedge resection and fusion in children over 4 years. Medial cuneiform opening-wedge osteotomy can be performed in children over 8 years of age with additional dorsal wedge osteotomies of middle and lateral cuneiforms to allow concomitant correction of cavus and rotational deformities (Dobbs et al, 2006). An opening osteotomy has the advantage of preserving foot length, but can only achieve a limited degree of correction as the lateral column is left intact. Supination and adduction deformities can be addressed by a combined medial cuneiform opening-wedge osteotomy and cuboid closing-wedge osteotomy.

Residual cavus may require tarsal or calcaneal osteotomies in children over 6 years of age, an Akron-type mid-tarsal osteotomy, which also allows varus-valgus correction, or even hindfoot-midfoot arthrodesis. Residual hindfoot varus or valgus can be addressed with an opening- or closing-wedge osteotomy.

Salvage surgery

The options for salvage surgery include triple arthrodesis, although the long-term outcome is variable, with one study finding 65% of patients had a poor result at follow up 13 years postoperatively, and the more limited talonavicular arthrodesis for patients with talonavicular osteoarthritis, dorsal subluxation and pain, which has satisfactory results 4 years postoperatively (Wei et al, 2000). Talectomy can be used to correct hindfoot deformity, with one study finding 75% of patients with a good or fair result at 20-year follow up, although two thirds required further surgery (Legaspi et al, 2001).

Ilizarov frames and Taylor spatial frames have been used in conjunction with various osteotomies and distraction osteogenesis techniques to correct residual deformities, although results are variable and there remains a high rate of recurrence and further surgery.

Late presentation (neglected clubfoot)

Delayed presentation is rare in the UK, but represents a significant problem worldwide, particularly in developing nations. Successful non-operative management is possible when commenced before 6 months of age, and one study from Brazil reports successful treatment with the Ponseti method in children up to 9 years of age, although the period of casting was longer (4 months) and one third required posterior release (Lourenço and Morcuende, 2007). Neglected clubfoot often warrants extensive surgical correction with a combination of soft tissue releases, wedge osteotomies, tarsal bone excisions and triple arthrodesis.

Discussion

The recent resurgence in the use of the Ponseti method has contributed to a huge reduction in the rate of corrective surgery for uncomplicated clubfoot (Morcuende et al, 2004). Parent education plays an important role in compliance with treatment and final outcome.

Clubfoot can cause significant deformity and have an enormously detrimental effect on quality of life if left untreated. It represents a great challenge in the developing world, where the incidence is higher and access to health education and treatment is limited. Although there are expanding programmes to identify children in developing countries for non-operative treatment soon after birth, there is still a large population who present late with severe deformities, which warrant extensive surgical correction.

Looking to the future, several refinements to the Ponseti method have been proposed, including the use of custom-fit dynamic orthoses instead of plaster of Paris,

which may improve compliance (Chen et al, 2007). Continuous passive motion has been used successfully, although compliance is a problem, and Botox injections have been used in conjunction with mobilization techniques (Alvarez et al, 2005).

Conclusions

Current evidence suggests that the best outcomes can be achieved with early treatment using the Ponseti or French methods of manipulation and immobilization. All additional procedures must be carefully planned, appropriately timed and tailored to the individual patient to keep surgical intervention to a minimum (Dobbs and Gurnett, 2009).

The increasing use of the Ponseti method worldwide has led to a greater understanding of the deformities involved, reduced the need for corrective surgery and provided a sustainable, cost-effective means of managing clubfoot in the developing world. **BJHM**

Figures 1 and 3 are redrawn from figures from www.global-help.org by kind permission of the editor, Lynn Staheli.

Conflict of interest: none.

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KEY POINTS

- Congenital clubfoot or congenital talipes equinovarus is a lower limb deformity that represents a wide spectrum of deformity with a prevalence of 1 in 1000 in the UK and a male:female ratio of 2–3:1.
- The aetiology of congenital talipes equinovarus is not fully understood, but it is thought to be multifactorial, and congenital talipes equinovarus is also associated with other conditions such as arthrogryposis and myelomeningocele.
- The four main characteristic components of the deformity are summarized by the mnemonic CAVE: forefoot Cavus, midfoot Adduction, hindfoot Varus, and hindfoot Equinus.
- The recent resurgence in the use of the Ponseti method has contributed to a huge reduction in the rate of corrective surgery for uncomplicated clubfoot, and parent education plays an important role in compliance with treatment and final outcome.
- Late presentation represents a huge challenge in the developing world, where access to health care and health education is limited, but progress is being made, for example in Malawi, with the use of dedicated non-physician specialists to perform the Ponseti treatment method in the community.