What is new in idiopathic clubfoot?

Ryan M. O'Shea¹ · Coleen S. Sabatini²

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Abstract Idiopathic clubfoot has a tremendous worldwide prevalence. If left untreated, the deformity has severely disabling effects on mobility and quality of life. Given its prevalence and significance, numerous studies are published on this condition every year. In this article, we attempt to highlight important themes and findings of studies published on idiopathic clubfoot over the past 3 years.

Keywords Clubfoot · Talipes equinovarus · Ponseti method · Pediatric foot · Congenital foot deformity · Pediatric orthopedics

Idiopathic clubfoot is one of the most common pediatric foot deformities, affecting approximately 1–4 per 1000 live births. The deformity is characterized by cavus of the midfoot with forefoot adductus and hindfoot equinovarus. If left untreated, the deformity will persist into adulthood and severely impair gait and adversely affect the lives of those born with this condition. Given the large prevalence of disease worldwide, approximately 530 scientific publications regarding the topic have been published in the last 5 years alone. Here, we discuss important new developments from the last 3 years in the study of idiopathic clubfoot.

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✉ Coleen S. Sabatini
Coleen.sabatini@ucsf.edu

Ryan M. O'Shea
ryan.michael.oshea@gmail.com

¹ San Francisco Orthopaedic Residency Program, 450 Stanyan Street, San Francisco, CA 94117, USA

² UCSF Benioff Children's Hospital Oakland, 747 52nd Street, OPC 1st Floor, Oakland, CA 94609, USA

Epidemiology, etiology, risk factors, and associations

Idiopathic clubfoot is bilateral in approximately 50 % of cases and is reported to affect males more than females. Although it is accepted that males are affected more than females, there is not yet consensus on whether there is a difference in severity of disease attributable to gender [1, 2•].

Multiple hypotheses exist regarding the etiology of idiopathic clubfoot. It is generally believed that the condition has a multifactorial etiology that includes genetic and environmental factors [3]. Recently, associations between clubfoot and four single nucleotide polymorphisms (SNPs) have been identified. Variations in the promoter regions of HOXA9, TPM1, and TPM2 alter muscle development and function in the embryo and have been implicated as a genetic risk factor for clubfoot [4]. Environmental factors are also thought to contribute to the etiology of clubfoot. A study through the Norwegian national cohort analysis demonstrated that smoking during pregnancy increased the risk of a child having clubfoot. The same study counters previous suggestions that season of birth and parental education level are risk factors, as these were not found to increase risk in the study population [5]. Another recent study challenged the idea that developmental hip dysplasia was associated with clubfoot deformity [6]. In fact, multiple recent studies agree that children with clubfoot do not require routine hip screening with radiographs or ultrasound [7•, 8].

International adoption of the Ponseti method

The Ponseti method has become the gold standard for treatment of idiopathic clubfoot across the world. The Ponseti method, developed by Dr. Ignacio Ponseti, consists of a series of manipulations to the foot with application of a long leg (toes to groin) cast that is well molded around the foot. The

cast is changed every 5–7 days and the majority of clubfeet correct after 5–7 weeks of casting. Syndromic or atypical clubfeet may require more casts to obtain acceptable correction, and are outside the scope of this review article. After the cavus, adductus, and varus have been corrected, the majority of feet (approximately 90 % of patients) require a heelcord tenotomy to correct the equinus deformity. After tenotomy, the foot is positioned in maximum dorsiflexion and abduction and casted in another long leg cast for 3 weeks, after which the child is transitioned to a foot abduction orthosis. There are many variations of braces available and utilized throughout the world. The brace is set to 60–70° abduction on the affected side, and 40° for the normal side if the clubfoot is unilateral. The brace is worn full-time for at least 3 months, and in many practices until the child starts pulling to stand. The brace is then worn overnight (12 h) and during naptimes (2–4 h) until they are 4 years old. The optimal age to stop bracing continues to be a topic of research.

In 2014, an estimated 113 of 193 United Nations member states had adopted the Ponseti method [9]. Numerous publications in the last several years have been published that demonstrate the success of programs based on the Ponseti method in low- and middle-income countries, with reported recurrence rates from 6 to 21 % at intermediate follow-up [10•, 11, 12].

International adoption of the Ponseti method has been successful for a variety of reasons, including (1) the abundance of literature supporting its efficacy in treatment of clubfoot, (2) the availability of instruction through the Internet, downloadable textbooks, and training sessions at international courses [9], (3) non-governmental organizations (NGOs), international charitable organizations, and prominent physicians collaborating to train local providers in this treatment modality and advocating for its use [10•, 13], and (4) the relatively low cost of treatment [14]. While focused treatment of clubfoot with the Ponseti method for individual patients has been hugely successful, multiple barriers to care still exist from a larger public health perspective [13, 15•].

Variability in the Ponseti technique

When Dr. Ponseti initially described his technique, he reported that 95 % of cases could be treated with his method, without significant surgical intervention. However, there has been great variability in the literature over the last several years in the degree to which this success rate has been substantiated. In recent studies, the need for surgical intervention (beyond initial heelcord tenotomy) after “failure” of casting has been reported in a range between 10 and 40 % [16, 17•]. It is recognized that deviations from the originally described method of Ponseti may account for significant variation in

treatment outcomes between institutions [16]. For instance, below-the-knee casting led to 37.5 % failure rate [18].

To better understand the observed variability in treatment success with the Ponseti methods, Miller et al. recently compared two institutions’ clubfoot programs. One institution had multiple providers using a non-uniform Ponseti treatment approach, and the second institution had only cases with strict adherence to Ponseti principles. At the first institution, 46 % of clubfoot cases required major surgery after failure of the Ponseti method, compared to only 1.7 % at the second institution. The authors suggest that deviation from the Ponseti method increased the likelihood of treatment failure. Presence of a single provider coordinating a clubfoot program, with strict adherence to Ponseti principles, can result in low rates of recurrence and decrease the chance of requiring major surgical intervention [17•].

A separate retrospective 5-year follow-up study demonstrated that a dedicated Ponseti clinic, as opposed to a general clinic, improved the outcomes of clubfoot treatment [19]. A study by Dunkley demonstrated that a dedicated Ponseti treatment program led by a physiotherapist who did the casting, with the supervision of an orthopedic surgeon, was successful with a recurrence rate of only 6 % in idiopathic clubfeet at an average 4.6 year follow-up [20]. These recent studies provide evidence that there are improved outcomes, with lower rates of surgical intervention and recurrence, when children are treated in dedicated clubfoot clinics [17•, 19, 20]. Adherence to the Ponseti principles minimizes risk of failure.

Achilles tenotomy

With the Ponseti method, the vast majority of patients require Achilles tenotomy to correct residual equinus after casting. Multiple techniques have been described for tenotomy, each with their own risk profile. Some advocate for an open procedure performed in the operating room while others perform the procedure in clinic. MacNeille et al. recently shared the results of their series of 41 patients where Achilles tenotomy was performed in the operating room with a mini-open technique. All patients were discharged home the same day; none experienced adverse effects of anesthesia or complications from the procedure [21]. Authors purport that doing the procedure in the operating room provides advantages of direct tendon visualization, while minimizing the risk of incomplete tenotomy and neurovascular injury. Another recent article described a technique they called the “posterior to anterior controlled technique,” which is similar to the technique used in the MacNeille paper—with the use of a mosquito clamp to isolate the tendon for sectioning. These tenotomies were performed in a clinic setting under local anesthetic, rather than under general anesthesia. The authors advocate for this

technique to minimize risk to adjacent neurovascular structures while avoiding a general anesthetic [22].

With a strictly percutaneous technique performed in a clinic, there is a relatively high rate of incomplete tenotomy. Karami et al. looked at the unintentional effects of partial Achilles tenotomy on outcomes after treatment with the Ponseti method using ultrasound to evaluate the tendon before and after the procedure [23]. Surprisingly, the tendon was completely cut in only 44 % of patients, and the mean percentage of tendon cut was 77 %. However, no correlation was observed between the amount of tendon cut, mean dorsiflexion achieved after tenotomy, and rate of recurrent deformity. The authors concluded that if the tenotomy is incomplete and sufficient dorsiflexion is achieved, the final result will be satisfactory [23, 24].

Regardless of the technique used to perform tenotomy, correction of the equinus deformity is critical to prevent relapse. Residual equinus deformity after Achilles tenotomy is associated with a high rate of recurrent deformity and increased need for future surgical procedures [25].

Recent studies have used ultrasound to evaluate the Achilles tendon healing after tenotomy. Niki et al. performed ultrasounds of the Achilles tendon at several intervals up to 2 years after tenotomy to evaluate gap healing. Continuity and gliding were noted within 4 weeks of tenotomy. The tendon appeared to remodel for upwards of 1 year, and at 2-year follow-up, a slight irregularity of the internal tendon structure persisted when compared to the unaffected contralateral Achilles tendon [26]. In a separate study, Niki et al. examined the effect of Achilles tenotomy on associated calf muscle atrophy. The diameter of calf muscles with unilateral idiopathic clubfoot was measured before tenotomy and followed for a mean of 27 months after tenotomy. Differences between the affected and unaffected sides were examined. While there were significant differences in the diameters of either side (the unaffected side was larger than the affected side), there was no significant difference in the rate of change over time. Thus suggesting that Achilles tenotomy has no short-term effects on calf-muscle atrophy [27].

New techniques without tenotomy

New techniques that are variations of the Ponseti method may one day provide an alternative to Achilles tenotomy for correction of equinus deformity. In one study, a dynamic dorsiflexion splint was applied to correct equinus after completion of serial manipulation and casting. Mean dorsiflexion improved nearly 15° at final follow-up with the use of a dynamic splint; however, 10 % of patients dropped out of the study group secondary to poor compliance and/or skin complications secondary to the splint [28]. Another non-surgical method of equinus correction involves injection of Botox into

the gastrosoleus complex after serial manipulations and casting. Ultrasound of the gastrosoleus and Achilles tendon 6 weeks after injection demonstrated lengthening of the Achilles tendon with a relative decrease in muscle mass [29]. It is suggested that avoiding an Achilles tenotomy preserves the viscoelastic properties of the native tendon and may avoid potential complications of surgery. However, these techniques are in their infancy and require further research to demonstrate non-inferiority compared to current standards.

Predicting treatment resistance and the role of imaging

Goldstein et al. reviewed 134 feet with a minimum of 3-year follow-up and analyzed factors that were predictive of treatment resistance and eventual need for surgical correction. Patients who were non-compliant with foot abduction orthosis were 7.9 times more likely to need surgery. Female patients were 5.4 times more likely to need surgery than male patients. Dimeglio score at presentation and number of casts required prior to the start of a foot abduction brace were also predictive for treatment-resistant cases [2•].

New physical exam findings have been identified to help predict resistance to clubfoot treatment. The “Samir-Adam sign,” which is a relatively extended big toe compared to the lesser digits, is highly predictive of a flexor digitorum accessorius longus muscle. The presence of this anomalous muscle may increase the likelihood of clubfoot being resistant to casting treatment [30]. Additionally, poor eversion muscle activity qualitatively assessed on physical exam has been demonstrated as a risk factor for relapse of clubfoot [31].

While evaluation of treatment is most frequently assessed by appearance, function, and range of motion, radiographic parameters have been suggested as well.

Obtaining radiographs for the diagnosis of clubfoot has not been shown to be necessary; however, some recent studies suggest they may reveal important prognostic information. The reliability of radiographic measures in infants with clubfoot deformity was recently demonstrated [32], and multiple studies have explored the utility of plain radiographs in predicting recurrent or treatment-resistant clubfoot. Shabtai et al. claim that on the lateral view with the foot in maximal dorsiflexion, a tibiocalcaneal angle greater than 77° and a talocalcaneal angle less than 29° are helpful in predicting functional outcome and need for additional surgery [33]. In a separate study, dorsiflexion less than 16° on lateral forced dorsiflexion pre-tenotomy radiograph was associated with an increased risk of recurrence in Ponseti-treated clubfeet [34].

The utility of advanced imaging studies in predicting Ponseti-resistant clubfoot was also recently studied. Moon et al. looked at the anatomic abnormalities on MRI in Ponseti-resistant clubfoot. In this study, the MRI of legs

affected by clubfoot was compared to normal contralateral legs. A significant reduction in total leg volume, muscle volume, and a relative increase in fat volume was appreciated in the affected leg of all clubfeet. Treatment-resistant clubfoot was associated with increased epimysial and intramuscular fat that was not present in patients that responded to Ponseti treatment. Additionally, distinct muscle hypoplasia was present in a subset of resistant patients. There was appreciable heterogeneity in treatment-resistant clubfoot anatomy and in the amount of fat infiltration, which likely represents a heterogeneous etiology [35•]. The clinical value of these findings and having this information for a given patient remains to be elucidated. Certainly, given the high success rates of the Ponseti method, it is not cost effective to obtain MRIs on idiopathic clubfoot patients. Whether this information is just of academic interest or if there is clinical application of this information for resistant clubfoot needs to be determined before we consider obtaining MRIs for children with clubfoot.

Tibialis anterior tendon transfer

Dynamic supination during gait and heel varus are common deformities that can occur after Ponseti casting and are usually attributed to poor brace compliance. Residual forefoot adductus after completion of casting [36] and higher initial Pirani scores at the beginning of treatment [37] increase the probability of recurrent deformity and subsequent surgical intervention.

The tibialis anterior tendon transfer (TATT) has been described as an effective procedure to address recurrent deformity. Holt et al. demonstrated the long-term effectiveness of the TATT. In this study, 25 Ponseti-treated clubfeet that developed recurrent deformity requiring TATT were followed for 37 to 55 years. The authors found no significant difference in range of motion, foot function, pain, and pedobarograph findings in patients treated with the Ponseti method and TATT compared to patients without recurrent deformity who did not require TATT. No patient in this group required subsequent surgical intervention for their clubfoot [38•]. These findings are further supported by other recent studies that demonstrate TATT improves pressure distribution through the foot during gait [39] and restores foot eversion strength [40]. While the data demonstrate TATT is a highly effective tool for preventing relapse of clubfoot, new literature suggests that a second recurrence after TATT can occur in as many as 20 % of patients [41]. Luckett et al. found that young age at the time of TATT and poor brace compliance were predictive of a second recurrence and recommended that these cases be monitored closely. Further research would be helpful to determine the ideal age range for TATT.

Various techniques have been described for performing the TATT. A recent cadaveric study sought to compare techniques

by analyzing how these different techniques influence forefoot and hindfoot motion. Knusten et al. compared three popular procedures: (1) the two-incision technique favored by Ponseti and Smoley, which involves rerouting the whole tendon distally and anchoring it to the lateral cuneiform, (2) the three-incision technique proposed by Garceau and Palmer, which involves pulling the tendon proximally and reinserting it more laterally beneath the extensor retinaculum, and (3) the split transfer described by Hoffer which requires three incisions and reinserting half the tendon more laterally, which is usually reserved for spastic equinovarus foot. The authors concluded that all three techniques are useful and deliver varying degrees of increased forefoot pronation. The three-incision technique by Garceau and Palmer provided the most forefoot pronation and could potentially provide a more powerful correction. Small changes were seen in hindfoot motion with all three techniques. The authors suggested that the two-incision technique is good for patients with a supple dynamic forefoot supination, but the three-incision technique and split transfer better for more resistant feet [42•]. As this was a small cadaveric study, clinical studies would be helpful in elucidating if there are relevant clinical differences.

Other surgical treatments of idiopathic clubfoot

While the Ponseti method is now the more widely accepted intervention, additional studies have been conducted to compare the outcomes of casting versus early or primary surgical approaches. Smith et al. published a review comparing 42 adults (24 treated surgically and 18 treated with the Ponseti method as infants) to 48 health controls. Both the casting group and surgery group had diminished strength and range of motion compared to controls; however, the Ponseti group had significantly greater plantar flexion and eversion strength and a decreased incidence of osteoarthritis compared to the surgical group. Surgically treated patients had higher pain scores and lower physical function and quality of life scores [43•].

Additionally, it has been documented that the calf circumference of patients treated with surgical posterior medial release is significantly smaller than those of patients treated with Ponseti casting alone or with TATT. Pirani scores at the initiation of treatment did not have an influence on calf circumference, thus suggesting that calf circumference is directly related to the degree of surgical intervention performed. The authors concluded that diminished calf girth is directly related to the extent of surgical intervention and that this discrepancy persists with long-term follow-up. This study offers further evidence that the Ponseti technique is the treatment of choice resulting in improved cosmesis and function [44].

Long-term follow-up studies indicate that surgically treated clubfeet are more likely to be painful and stiff [45–47]. One

study suggests decreased walking capacity in surgically treated clubfeet compared to healthy age-matched controls [48]. Additionally, surgically treated feet are prone to develop bony deformities of the distal tibia and talus. Burghardt et al. analyzed the radiographs of 65 patients after surgical treatment of their clubfoot with an average of 10 years follow-up. The authors noted a significant talar flattening, distal tibia valgus, and anteflexion deformity in a large proportion of patients [47]. It is now widely accepted that primary surgical management of idiopathic clubfoot is no longer the standard of care. However, it has been suggested that in certain patients, “a la carte” surgical interventions may be useful in those for whom the Ponseti method does not result in adequate correction. A study by Mahan sought to compare a group of idiopathic clubfoot patients treated with a judicious posteromedial and lateral release to normal controls using patient-based outcomes. They found no significant differences between the groups—suggesting that surgical outcomes in children treated with this selective technique can have satisfactory results in experienced hands [49]. Bocahut et al. shared their results of a medial to posterior open surgical release in 199 feet after failure of the French functional method with a mean follow-up of 10.8 years. Twelve percent of patients required revision surgery; medial to posterior open surgical release achieved mostly good or very good results when assessed by the International Clubfoot Study Group (ICSG) outcome evaluation system [50].

Another recent study suggested that surgical outcomes for treatment of clubfoot might be improved with early physiotherapy. Previous studies have demonstrated markedly reduced ankle range of motion after posterior ankle and subtalar capsulotomy. Jauregui et al. presented a series of 20 feet that required posterior ankle/subtalar capsulotomy after treatment with the Ponseti method. After capsulotomy, a long leg cast was applied for an average of 26 days and after removal, parents initiated immediate home physiotherapy. The ankle range of motion improved dramatically. No significant reduction in these gains was observed at the final follow-up (average 62 months). Authors concluded that using a protocol for early mobilization after capsulotomies may protect against postoperative ankle stiffness [51].

Elgeidi published on double wedge tarsal osteotomy with transverse osteotomy to address persistent forefoot adduction and midfoot supination deformities. This technique involves removing a wedge from the cuboid and inserting it into the medial cuneiform as well as a transverse osteotomy. This procedure was performed on 35 feet in 27 children with at least a 5-year follow-up. The authors noted significant improvement in the sagittal and coronal alignment on follow-up radiographs. Clinical and functional results were not well described, however [52].

Management of neglected clubfeet

Traditionally, the Ponseti method is utilized in the infant population, but a growing body of evidence supports its use in older children as well [53, 54, 55]. Bashi et al. applied Ponseti cast treatment to 18 children with neglected clubfeet at an average age of 11.2 years old. The Ponseti technique was modified to utilize short leg casts instead of long and each cast was applied for 3 weeks. Patients required an average of nine casts to correct the deformity. An Achilles tendon lengthening and posterior tibiotalar release corrected equinus deformity at the end of casting. Plantar fascial release was required in three patients to correct residual cavus. To prevent recurrence, AFOs were worn for 3 months and then patients were instructed to wear high top shoes. 17/18 feet achieved a good outcome with mean follow-up of 15 months [53].

Multiple surgical approaches to neglected clubfeet have also been described. Cuboid subtraction osteotomy with a posteromedial release was demonstrated as an effective approach to manage stiff neglected clubfoot deformity in children older than 5 years old [56]. Multiple reports suggested the Ilizarov technique can achieve plantigrade feet and a good clinical outcome in children up to age 14 [57, 58].

Gait

Parents are naturally concerned about how clubfoot will influence their child's walking. Zionts et al. recently shared their experience evaluating motor milestones in patients with idiopathic clubfoot treated by the Ponseti method. The authors showed that children treated with the Ponseti method tend to achieve independent walking around 14.2 months, or approximately 2 months after their peers without foot deformity. Patients with a severe deformity or relapse usually achieve independent walking by 16 months [59].

Gait analysis of children treated with the Ponseti method has revealed multiple kinematic differences when compared to healthy controls. In particular, patients with clubfoot tend to have a decreased range of motion at the ankle, a decreased foot progression angle, and less power with ankle plantar flexion [60]. However, these gait disturbances did not diminish functional outcome nor parental assessment and satisfaction at 5-year follow-up [61]. Additionally, congenital clubfoot with satisfactory treatment does not significantly impair athletic performance [62].

Culturally sensitive care

A diagnosis of clubfoot deformity is understandably difficult for parents and many struggle with the burden of treatment. It is known that brace use is highly correlated with relapse, brace

wear rates decline over the first 3 months after initiation, and caregivers over-report the actual rate of wear [63]. Traditional dogma is that it is best to start casting as soon as possible after birth. A recent study by Zions however suggests that allowing adequate time for parental education and coping can positively affect treatment outcomes, and a delay “of a few weeks” was not shown to negatively impact outcome [64].

Recent research also suggests that respecting more subjective and emotional elements of care can improve compliance with bracing programs and prevent recurrence. Morin et al. suggest that a positive, culturally sensitive communication paradigm can reduce recurrence when treating children with clubfeet using the Ponseti method. Elements of this paradigm include using positive reinforcement, praise for compliance, and avoiding negative comments or threatening surgical intervention [65]. In a separate investigation, Malagelada et al. looked at the emotional and psychological impact of Ponseti treatment on parents and caregivers of affected children in the UK and South Africa. This study showed that serial casting and bracing has a significant impact on families, and utilization of coping strategies was different between the two countries. The authors recommend implementation of culturally sensitive protocols that support parents and improve the well-being of families [66].

Conclusion

Idiopathic clubfoot has a tremendous worldwide prevalence and if left untreated, the deformity has severe disabling effects on mobility and quality of life. The treatment paradigm of clubfoot has changed dramatically in the past couple of decades. The Ponseti method has replaced surgical intervention as the initial treatment of choice for idiopathic clubfoot. The Ponseti method is a highly effective, reproducible, and cost-effective technique. Its success had been demonstrated by its rapid international adoption. Achilles tenotomy at the end of Ponseti casting is frequently required, and multiple acceptable techniques for this procedure have been described. Variation in the rate of recurrent deformity after Ponseti treatment appears to be most attributable to deviation from Ponseti principles. Recurrence of deformity after correct Ponseti treatment is infrequent, but can be successfully treated with TATT or other surgical procedures. There are multiple factors that affect compliance with bracing, and there are still questions about how long bracing should be utilized. After treatment with the Ponseti method, there are subtle alterations in the gait of affected persons that seem not to impair functional mobility or athletic performance. Idiopathic clubfoot impacts a tremendous number of children worldwide—its importance is illustrated by the vast array of studies that are published on this condition every year. In this review, we have attempted to

highlight new work from the last 3 years on this important pediatric orthopedic condition.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Human and animal rights and informed consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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