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Management issues of congenital talipes equinovarus in the neonatal intensive care unit:
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Journal

Foot and Ankle Surgery, 27(5)

ISSN

1268-7731

Authors

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Whitaker, Amanda T

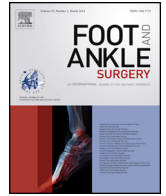
Publication Date

2021-07-01

DOI

10.1016/j.fas.2020.07.005

Peer reviewed



Review

Management issues of congenital talipes equinovarus in the neonatal intensive care unit: A systematic review

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ARTICLE INFO

Article history:

Received 6 May 2020

Received in revised form 17 June 2020

Accepted 10 July 2020

Keywords:

Congenital talipes equinovarus

Clubfoot

Neonatal intensive care

Ponseti method

Atypical clubfoot

Syndromic clubfoot

ABSTRACT

Background: The Ponseti method is the standard of care for managing idiopathic congenital talipes equinovarus (clubfoot) in the outpatient setting, but there are no clinical guidelines for inpatient treatment. Children in the neonatal intensive care unit (NICU) with clubfoot often delay treatment initiation due to medical reasons.

Methods: We systematically reviewed literature related to the treatment of clubfoot in the NICU, non-idiopathic clubfoot, and older infants, as well as barriers to care.

Results: In a mixed NICU population of syndromic and idiopathic clubfoot, the Ponseti method has good functional outcomes with minimal interference with medical management. The Ponseti method has good functional outcomes with reduced need for extensive surgical procedures in non-idiopathic clubfoot and idiopathic clubfoot with delayed presentation (under one year of age).

Conclusions: It is possible to begin Ponseti treatment in the NICU without compromising medical management. It is not clear if this confers an advantage over waiting for outpatient casting.

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1. Introduction

Clubfoot is one of the most common orthopedic anomalies present at birth, in which the ankle is in equinus, the hindfoot is in varus, and forefoot is adducted. It occurs in 1–2 infants per 1000 live births and is twice as common in males. Clubfoot is bilateral in 30–50% of children. Presentation varies from mild postural forms to severe rigid deformities [1]. Clubfoot is typically idiopathic but can present in conjunction with congenital syndromes such as arthrogryposis, myelomeningocele, and amniotic band syndrome [2].

The Ponseti method is the global standard for treating idiopathic clubfoot [3,4]. While the Ponseti method has been studied extensively in the outpatient setting, there is little data on its use in the neonatal intensive care unit (NICU) [5]. There are no clinical guidelines for managing clubfoot in NICU patients with complex medical needs. These children typically start clubfoot treatment at a later age, often after discharge [6]. Clubfoot in NICU patients can be idiopathic, syndromic, or neuromuscular in origin, which makes it challenging to study. There is little work that directly addresses inpatient management of clubfoot and the effect of treatment in this specific population [5].

2. Methods

This is a systematic review of clubfoot treatment in the NICU using PRISMA guidelines. We conducted a search using medical subject headings (MeSH) terms ‘clubfoot or congenital talipes equinovarus,’ ‘Ponseti treatment/method,’ ‘intensive care unit, neonatal,’ and ‘treatment outcomes/results.’ This search yielded one unique result. The search was repeated replacing ‘intensive care unit, neonatal’ with ‘inpatients’ which yielded no results. The search was then broadened by removing both ‘intensive care unit, neonatal’ and ‘inpatients’ (Fig. 1). This yielded 548 results on Cochrane Library, CINAHL EBSCOhost, Google Scholar, and PubMed. The titles, abstracts, and full texts to determine relevance were reviewed. We included reviews, randomized control trials, comparative studies, and case series written in English that discuss outcomes and management of NICU patients, non-idiopathic clubfoot, and older infants under one year of age. Abstract-only publications were excluded. The references of selected articles to identify additional relevant studies were reviewed. Of the relevant citations in the English literature as of February 2020, 35 articles met our inclusion criteria and are discussed.

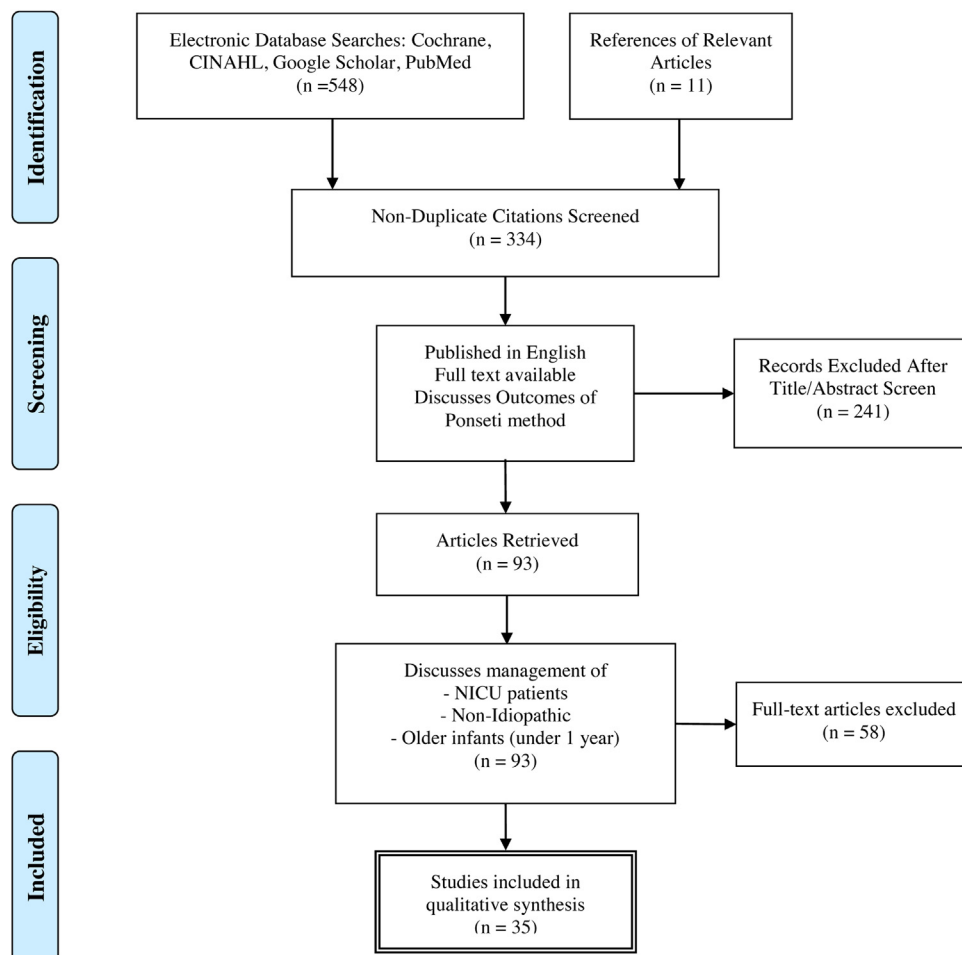


Figure 1. Methods of Systematic Review of Clubfoot Management in the NICU. This diagram outlines the search and selection process applied in our systematic review [54].

3. Results

3.1. Clubfoot treatment

3.1.1. Ponseti method

The Ponseti method is the standard of care for idiopathic clubfoot due to its high initial correction rate and excellent outcomes in long-term follow-up [7]. The clubfoot is gradually corrected by weekly manipulation and above-the-knee cast application usually beginning in the first month of life [4]. Cavus is corrected first, followed by adductus and varus, then equinus. Most clubfeet achieve correction after 4–6 casts [1,7,8]. An accelerated schedule with casts applied 3 times per week has been shown to achieve the same level of correction in an average of 16 days [1,7,8]. A percutaneous Achilles tenotomy is needed to achieve full correction of equinus in over half of clubfeet [1]. Feet not corrected by casting and tenotomy require additional soft tissue releases in the foot and ankle. To maintain correction, children with clubfeet wear a “boots and bar” abduction brace on a 23-hour wear schedule for three months or until they begin to stand, and then during nights and naps until age four [1,2,7,9]. Relapse in idiopathic clubfoot is generally attributable to brace noncompliance [1,7,10]. Relapses are treated with a second round of casting and/or surgical correction [1,7,11].

An initial correction rate of 90% has been reported in idiopathic clubfoot treated prior to one year of age with relapse in 4–27% [1,4,10,12–16]. The Ponseti method has been very effective for outpatient treatment of idiopathic clubfoot, but more work is needed on use in atypical populations [1].

3.1.2. Surgical

Historically, clubfoot was managed with surgical release of any number of the posterior, posteromedial, or posterolateral soft tissue structures in the foot and ankle [3,17]. Correction was completed prior to walking age when the foot was deemed large enough. The procedure was preceded by stretching and application of below-the-knee casts [18]. Surgical management has an initial correction rate of 70–90% [7,17]. There are similar rates of relapse between clubfeet treated surgically and with the Ponseti method, but initial Ponseti treatment leads to significantly fewer and less extensive revision surgeries [3,18–20]. Long-term outcomes of surgically-corrected clubfoot are challenged by overcorrection, stiffness, and pain later in life [3,7,17]. Those treated with the Ponseti method exhibit significantly greater range of motion of the ankle joint, greater strength, lower incidence of osteoarthritis, and higher self-reported physical function and quality of life compared to those managed surgically [21,22].

3.2. Measurement of treatment outcomes

Clubfoot outcomes are described inconsistently throughout the literature. Some studies rely on validated scoring systems to assess foot morphology and function. Scores are compared at initial presentation and follow-up to measure treatment effectiveness [23]. The Pirani scoring system characterizes clubfoot deformities based on hindfoot and midfoot morphology [24,25]. There is a positive non-linear relationship between the initial score and number of casts required for correction [24–26]. The Dimeglio scoring system assesses mobility and morphology [27]. Lower initial scores are associated with plantigrade feet following casting and tenotomy [25]. Other validated scoring systems assess morphology, range of motion, function, and patient satisfaction [9,17,22].

Many studies provide their own definition of a “corrected” foot. A corrected idiopathic clubfoot is considered to be pain-free and plantigrade with no residual forefoot adductus, cavus, or hindfoot

varus [11,12,14,28–30]. In non-idiopathic clubfoot, the definition of “corrected” is limited to pain-free, plantigrade and the ability to fit into orthotics or walk independently [5,16,31–35].

Some studies measure outcomes in terms of relapse [3,18,32,36]. Relapse is defined as recurrence of deformity after initial correction that requires additional casting and/or surgery [3,11,14,18,30,36]. Number of casts and need for tenotomy are also typically reported [1]. Inconsistent outcomes measures complicate comparison across studies.

3.3. Management of clubfoot in the NICU

The primary focus of treating NICU patients is addressing their serious medical needs, although other conditions (like clubfoot) may be present during the acute phase of care [6]. There is limited evidence for management of orthopedic conditions in the NICU [5,6].

Lebel et al. examined the use of Ponseti casting in a mixed population of 20 children with idiopathic and non-idiopathic clubfoot (30 feet) [5]. Ten children were diagnosed with an identifiable syndrome [5]. Treatment began as soon as medically possible, which was in the first week of life for 80%. Three of 75 casts (4%) required removal due to leg edema or need for venous access. Length of stay was unrelated to clubfoot management. At 9 years, 50% were independently ambulatory. Non-ambulatory status was attributable to underlying diagnoses. All surviving children had plantigrade, braceable feet. These results are limited by a small population and lack of control group but demonstrate clubfoot treatment can be initiated in the inpatient setting with minimal complications [5].

3.4. Non-idiopathic clubfoot

Approximately 11–48% of clubfoot exists as part of a congenital syndrome or neuromuscular disorder [10,13,16,21]. A portion of these patients will be seen in the NICU. Non-idiopathic clubfoot is challenging to treat due to the inherent stiffness of the foot and ankle, frequent relapses, and musculoskeletal and medical comorbidities [1,14,15,34]. A systematic review conducted in 2014 found a substantial dearth of high or moderate quality evidence for the treatment of relapsed, neglected, complex, and non-idiopathic clubfoot [7]. This is in part due to the relatively small number of cases (Table 1). Although the Ponseti method has been extensively studied in idiopathic clubfoot, research on use in non-idiopathic clubfoot is less abundant. Ponseti management can produce good functional outcomes in non-idiopathic clubfoot despite increased relapse rates (15–44%) and greater need for additional surgical procedures compared to idiopathic clubfoot (3% vs. 37%) [1,13,15,16,34].

3.4.1. Arthrogyposis

Arthrogyposis is a group of congenital contracture syndromes involving the upper and lower limbs. Severe rigid clubfoot is present in 78–90% of children with arthrogyposis [37]. Clubfoot in arthrogyposis was historically treated with extensive release that often required revision surgeries [31,33]. Compared to surgical release, the Ponseti method can increase function, reduce revision surgeries, and lessen complications of clubfoot treatment in distal arthrogyposis [31,33,38]. Arthrogyptic clubfeet require more casts to achieve correction than idiopathic clubfeet (average 7–9, range 4–12), which is positively correlated with the severity of the deformity [28,29,31–33,38]. The relapse rate is significantly higher (25–90%) and not solely due to brace noncompliance [28,29,31–33,38]. A modified Ponseti method with tenotomy performed before and after casting has been successful in treating more severe clubfoot associated with classic

Table 1

Available case studies and retrospective reviews of non-idiopathic clubfoot treated with the Ponseti method.

	Study	No. of patients (no. of feet)	Treatment setting ^c	Average follow-up (in years) ^d	Level of evidence
Mixed non-idiopathic	Gurnett et al. [10]	84 (147)	Outpatient	≥2	II
	Janicki et al. [14]	23 (40)	Outpatient	2.8 (1–5)	II
	Matar et al. [34]	16 (28)	Outpatient	7 (4–12)	IV
	Moroney et al. [15]	29 (43)	Outpatient	3.2 (1–5.3)	III
	Richards et al. [16]	47 (N/A)	N/A	≥2	IV
Arthrogryposis	Boehm et al. [28]	12 (24)	Outpatient	N/A	IV
	Kowalczyk et al. [31]	9 (18)	N/A	7.3 (5–10)	III
	Kowalczyk et al. [32]	5 (10)	N/A	3 (2–3.7)	IV
	Matar et al. [33]	10 (17)	Outpatient	5.8 (3–8)	IV
	Morcuende et al. [29]	16 (32)	N/A	N/A	IV
	Van Bosse et al. [38]	10 (19)	Outpatient	3.2 (1.1–5.8)	IV
Amniotic band syndrome ^a	Agarwal et al. [40]	3 (4)	N/A	≥1	IV
	Carpiaux et al. [36]	12 (21)	N/A	3.9 (0.8–10)	IV
	Zionts et al. [30]	5 (6)	N/A	2.7 (1.8–4.1)	III
Neural tube defects	Abo El-Fadl et al. [42]	24 (48)	N/A	2.3 (2–2.8)	IV
	Arkin et al. [41]	17 (26)	Outpatient	5.4 (1.8–7.8)	III
	Gerlach et al. [12]	16 (28)	N/A	2.8 (2.1–3.6)	II
	Jackson et al. [43]	8 (12)	N/A	2	II
	Matar et al. [35]	11 (18)	Outpatient	4.5 (3–9)	IV
NICU ^b	Lebel et al. [5]	20 (30)	NICU	(2–9)	III

^a Also referred to in the literature as congenital annular band syndrome and congenital constriction band syndrome.

^b Mixed idiopathic and non-idiopathic population.

^c Setting in which treatment was initiated (i.e. NICU, inpatient, outpatient/clinic, not available).

^d Presented as average (range).

arthrogryposis [38]. The majority of these feet have satisfactory functional outcomes at 8 years [29,33,38].

3.4.2. Amniotic band syndrome (ABS)

ABS is a congenital malformation arising from a separation between the amnion and chorion in utero. This produces fibrous bands that encircle limbs, causing damage during development [36]. Clubfoot is present in 12–56% of children with ABS [37]. Clubfoot in ABS is rigid, complicated by limb amputations, constrictions, nerve palsy, and visceral comorbidities [37,39]. This was historically treated with band release and extensive surgical correction of the clubfoot [39]. In preliminary trials, the Ponseti method in ABS lead to fewer extensive surgical interventions (5–16% of clubfeet) [30,36,40]. Despite a higher rate of relapse (33–83%) than in idiopathic clubfoot, most children exhibit supple plantigrade feet at 10 years [30,36,40].

3.4.3. Neural tube defects

Neural tube defects encompass all conditions where the neural tube fails to close. The most serious of these is myelomeningocele, where neural tissues herniate from within the tube. Foot deformities are present in 60–80% of infants born with neural tube defects, and clubfoot is seen in 30–50% [27,37]. Higher level spinal lesions are associated with more rigid deformities [37,41]. The rate of initial correction is similar to that of idiopathic clubfoot. There are mixed results regarding the number of casts needed to achieve initial correction in this population [12,35,41–43]. Despite relapse in 33–68% of clubfeet, most have good functional outcomes [12,35,41,43]. At 9 years, 83–86% of clubfeet are mobile, plantigrade, and able to fit into orthotics; this is necessary to ensure future independence [12,27,35,37].

In very severe deformities, Ponseti casting with open tenotomy increases the rate of initial correction to 42% in children with neural tube defects and leads to improvement in 90% of clubfeet at 3-year follow-up [42]. Both initial percutaneous (vs. open) tenotomy and higher level spinal lesions are associated with higher rates of relapse [12,41]. Treatment is complicated by insensate skin that can break down in casts and delay wound healing [27,37,41,42].

3.5. Treatment of older infants

Children with clubfoot in the NICU typically start treatment at a later age, often after discharge. Treatment can also be interrupted by medical or social issues [6]. Substantial evidence exists for the successful treatment of idiopathic clubfoot in delayed presentations before walking age [1,2,9,11,44–48]. There is mixed evidence regarding the influence of age on the number of casts required to achieve correction (range 3–12 casts) [9,11,44–46,48–50].

Several studies have shown no correlation between age at start of treatment and relapse, need for open surgical release, or functional outcomes in non-ambulatory children with idiopathic clubfoot under 1 year of age [9,11,44,46,49,50]. Outcomes are comparable to those of infants receiving treatment in the first month of life. This suggests the Ponseti method can be successfully applied even after delayed presentation in idiopathic clubfoot [49]. However, the severity of deformity in the delayed treatment group is often unclear. No study has examined the effect of delayed treatment in the NICU population.

4. Discussion

There is limited literature that addresses how clubfoot is managed in the inpatient setting in medically complicated children (Table 1). The study by Lebel et al. is the only study found that explicitly addresses initiation of clubfoot treatment in an inpatient setting [5]. All other studies included in this review state that the standard of care (outpatient casting) was followed or make no reference to the treatment setting. Lebel et al. examined long-term functional outcomes of inpatient clubfoot treatment, suggesting that the Ponseti method could produce satisfactory outcomes with minimal complications [5]. The main concern regarding casting in the NICU is interference with medical management. Casting can interfere with weight evaluation, which is crucial for monitoring growth. It can also cause skin breakdown, leading to infection. This is of particular concern in children with myelomeningocele and other neurologic deficits. The application and removal of casts can be distressing for the infant and should be approached cautiously. The low reported

rate of unplanned cast removal illustrates that a judicious approach to casting can minimize interruptions in medical care [6].

We presume children in the NICU are best represented in available literature through non-idiopathic clubfoot and management of clubfoot in older infants due to the high prevalence of syndromic clubfoot and delays in treatment initiation [5,6,10,13,16,21]. There is encouraging evidence for the successful outpatient treatment of non-idiopathic clubfoot and idiopathic clubfoot with delayed presentation using the Ponseti method. However, it is unclear if delayed treatment in the non-idiopathic or medically complicated child have worse outcomes.

Developing evidence-based practice guidelines for inpatient clubfoot management is complicated by conflicting evidence on the optimal timing of treatment. Iltar et al. found that children casted between one month and one year of age exhibited more correction, as demonstrated by lower final severity scores, than those casted in the first month of life [50]. These findings are corroborated by Liu et al., who found that children beginning treatment between 28 days and 3 months of life had fewer casts, fewer relapses, and better functional outcomes compared to children casted prior to 28 days of life or from age 3 months to 6 months [48]. Difficulty manipulating a small foot could contribute to these findings. Iltar et al. and Hemo et al. found that children with feet less than 8 cm in length at the start of casting required more casts to achieve correction and had worse severity score after treatment compared to those with feet longer than 8 cm [50,51]. Shorter foot length at treatment initiation has also been associated with an increased incidence of cast slippage and need for further surgical intervention for residual or recurrent deformity [52]. Advantages of early intervention have also been suggested. Neonates exhibit ligamentous laxity, which could improve mobility and amount of correction with each manipulation [33]. Syndromic clubfoot is often more rigid than idiopathic clubfoot, so prompt interventions may increase the effectiveness of casting [1,14,15,33,48]. Bone remodeling is also increased in neonates, which may account for good outcomes seen in severe clubfoot treated early [5,53].

Clinical guidelines are further complicated by inconsistent outcomes measures, resulting in an inability to aggregate data from multiple studies. Standardized use of validated scoring systems and functional measures are needed to facilitate evidence-based protocol development.

5. Conclusion

Children with clubfoot in the NICU have a variety of diagnoses that influence treatment decisions. They can generally be divided into those with idiopathic clubfoot and those with multiple congenital anomalies or an identifiable syndrome. Prognosis and probable course of medical treatment must be weighed when deciding if and when to initiate casting. The presence of congenital anomalies may also impact the severity of deformity and response to treatment. In premature infants, care is further complicated by small foot size and serious medical concerns. Addressing clubfoot may not be considered a priority when facing acute and life-threatening medical problems, but many children in the NICU grow up to be independent adults. The available literature suggests it is possible to begin Ponseti treatment in the NICU without compromising medical management. However, it is unclear if this confers an advantage over the current model of outpatient treatment. Further study is needed to examine the outcomes and optimal timing of Ponseti treatment of clubfoot in the NICU.

Funding

This work was supported in part by The Ohio State University College of Medicine Roessler Research Scholarship (SMT). The

sponsor had no involvement in study design; the collection, analysis, and interpretation of data; writing of the report; and the decision to submit the article for publication.

Conflicts of interest

The authors declare that they have no conflict of interest.

Ethics approval

Not applicable for this systematic review.

Availability of data and material

All data and material are cited, described, and available upon request.

Authors' contributions

Sally Trout, BS: Completed systematic review, organized the literature, drafted and edited the paper, approved final manuscript.

Amanda Whitaker, MD: Conceptualization of study, study design, organized the review and paper, assisted with drafting and revised the manuscript, approved final manuscript.

Consent to participate

Not applicable due to systematic review of available literature.

Consent to publish

Not applicable due to systematic review of available literature. No Copyrighted materials are included in this manuscript.

Acknowledgments

We would like to thank Dr. Christopher Iobst and Dr. Maurice Manring for their edits and suggestions. We would also like to thank The Ohio State University College of Medicine Roessler Research Scholarship (SMT).

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