Original Article

Is Ponseti Casting Effective in Neuromuscular Clubfoot? A University Hospital Experience from Eastern Province in Saudi Arabia

Ammar K. Alomran, Dalal A. Bubshait, Bandar A. Alzahrani, Abdulaziz A. Almazrua[1], Mohammed A. Alyaqout[1]

Department of Orthopedic Surgery, College of Medicine, Imam Abdulrahman Bin Faisal University, [1]College of Medicine, Imam Abdulrahman Bin Faisal University, Dammam, Saudi Arabia

Abstract

Objectives: Clubfoot deformity occurs in approximately one in 1000 newborns. In patients with neuromuscular disease, the clubfoot deformity differs markedly from that in idiopathic clubfoot with respect to its severity and higher rate of complications. We aimed to compare the effectiveness of the Ponseti serial casting technique in treating idiopathic and neuromuscular clubfoot. **Methods:** We performed a retrospective study of patients with clubfoot identified from a pediatric orthopedic clinic database, who were treated at our hospital from 2004 to 2018. The follow-up period for patients in both groups ranged from 8 to 12 years. The information obtained included age at presentation and number of casts required for deformity correction. We measured the severity of the clubfoot using the modified Pirani score before and after Ponseti treatment. **Results:** The initial number of participants was 117; upon applying our inclusion and exclusion criteria, only 39 patients from the idiopathic group and 10 from the neuromuscular disease-associated clubfoot group were included. The average age in weeks of the participants in the idiopathic group and the neuromuscular disease-associated clubfoot group at diagnosis was 1.79 and 1.80, respectively. Both groups had a similar mean modified Pirani score before and after the Ponseti treatment. **Conclusions:** Ponseti casting is an effective primary modality of management for clubfoot associated with neuromuscular diseases, as it is in idiopathic clubfoot, to achieve functional pain-free feet and reduce the number of surgical interventions needed.

Keywords: Clubfoot, congenital foot deformities, neuromuscular disease, Ponseti, talipes equinovarus

Introduction

Congenital talipes equinovarus or clubfoot is defined as a foot with forefoot adduction, midfoot cavus, supination, and varus with inward inclination. The incidence of clubfoot was reported to be one in 1000 newborns in the English population, and it is one of the most common congenital foot deformities worldwide.[1,2] Clubfoot deformity is common in patients with neuromuscular disease. The clubfoot deformity in these patients differs markedly from that of idiopathic clubfoot with respect to stiffness, severity, rigidity, relapse rate, and the rate of complications.[3,4] The prevalence of neuromuscular diseases associated with clubfoot varies in the literature depending on the population group of focus. Bakalis *et al*. studied the prevalence of clubfoot in neuromuscular patients and found that the incidence was much higher (48.6%) on prenatal ultrasound than in postnatal examinations.[5] In contrast, a study conducted in Australia in 2005 used birth documents to demonstrate a 40% prevalence of clubfoot in neuromuscular patients.[6] The aim of orthopedic management is to achieve a functional plantigrade, painless, and braceable foot.[7] Idiopathic clubfoot has traditionally been managed with extensive soft-tissue releases, with multiple reported complications of skin breakdown, recurrence, and the need for revision and surgical procedures.[3,5,8] Subsequently, in 1963, Ponseti developed a noninvasive management technique for

Access this article online

Quick Response Code:

Website: www.journalmsr.com

DOI: 10.4103/jmsr.jmsr_73_19

Address for correspondence: Dr. Ammar K. Alomran, Department of Orthopedics, Imam Abdulrahman Bin Faisal University, Dammam, Saudi Arabia.
E-mail: aomran@iau.edu.sa

Received : 24-08-2019 **Revised :** 04-11-2019
Accepted : 22-11-2019 **Published Online :** 03-01-2020

How to cite this article: Alomran AK, Bubshait DA, Alzahrani BA, Almazrua AA, Alyaqout MA. Is ponseti casting effective in neuromuscular clubfoot? A university hospital experience from eastern province in Saudi Arabia. J Musculoskelet Surg Res 2020;4:31-5.

clubfoot composed of manipulation, casting, Achilles tendon release, and bracing. It takes approximately 30–45 days to achieve full correction.[4] The Ponseti method of casting is now believed to be the standard primary treatment for idiopathic clubfoot and is also useful in rigid clubfoot.[3,9] Many papers have studied the effectiveness of the Ponseti casting technique in nonidiopathic clubfoot and the results were not satisfactory among most of these studies.[10,11] However, only a few studies have been published on the effectiveness of the Ponseti casting technique in the management of neuromuscular disease-associated clubfoot.[9,10] The present study sought to determine the efficacy of using the Ponseti casting technique as a primary line of treatment in children with neuromuscular disease-associated clubfoot relative to that in a group with idiopathic clubfoot.

Materials and Methods

We performed a retrospective study of patients with clubfoot identified from the pediatric orthopedic clinic records in King Fahd hospital and we sought to evaluate the response to Ponseti serial casting technique, need for surgical intervention, and complications for patients with clubfoot who were treated at our hospital from 2004 to 2018. Patients with clubfoot were categorized as either having neuromuscular-associated clubfoot or idiopathic clubfoot for comparative purposes. The patients included were those diagnosed with idiopathic or neuromuscular disease-associated clubfoot who had been managed using the Ponseti casting technique treatment and followed up in our hospital. We excluded patients who either did not complete their treatment program at our hospital or whose data extraction form was <50% filled because of poor documentation of valuable information that was required to understand the patient's health and development with respect to the treatment program provided to them. Standard data included age at initial presentation, gender, prenatal history including the mode of delivery, family history, diagnosis, laterality, and rigidity. We recorded the number of casts needed for correction, recurrence rate, need for tibialis anterior transfer (TAT), and any other invasive surgical intervention performed for each patient. Age at last follow-up, any residual deformity, pain, gait abnormality, and whether the patient still uses a brace were also documented. Recurrence was defined as any residual deformity that required further casting and/or surgery to retain the optimal position in a child with clubfoot. We measured the severity of the clubfoot before and after Ponseti treatment using a modified Pirani score. The Pirani scoring is a system used to assess the severity of the clubfoot deformity scored by the treating physician ranging from 0 to 6, where 0 is normal and 6 is the most severe form of deformity.[12] A decrease in the score correlates with an improvement of the deformity. Failure of Ponseti casting was defined as a clubfoot that failed to be corrected with serial casting.[4,13] There were four treating physicians and their experience with pediatric orthopedics ranged from 10 to 20 years. All available clinical records, including the clinical charts, electronic charts, and operative notes, for all patients in these groups were reviewed. Data extraction was performed with the aid of forms designed to meet the specific requirements of this study. Ninety-five percent confidence intervals (CIs) were used. $P < 0.05$ was considered as statistically significant. The analysis was performed using the Statistical Package for the Social Sciences (SPSS) version 19 software (SPSS, Inc., Chicago, IL, USA).

Results

One hundred and seventeen patients with clubfoot treated from 2004 to 2018 were identified in our database; 67 patients were in the idiopathic group and 25 patients in the neuromuscular-associated group. The other 25 patients were not classified into any group either because the health status or the patient's condition was not documented fully in the database. Upon applying our inclusion and exclusion criteria, only 49 patients who were managed at our hospital were included in this study. Thirty-nine patients had idiopathic clubfoot and ten had neuromuscular disease-associated clubfoot. There were five different pathologies recorded in the patients included in the neuromuscular disease group: spinal muscular atrophy (30%), muscular dystrophy (20%), congenital myopathy (20%), unknown syndrome (20%), and Charcot–Marie–Tooth syndrome (10%). The number of participants in the idiopathic group who were managed using the Ponseti casting technique alone was 34. The number of male and female participants in the study's idiopathic group was 18 and 21, respectively. The demographic data for the idiopathic and neuromuscular groups treated with the Ponseti casting technique are summarized in Table 1. In the idiopathic group, female patients were more likely to have received Ponseti treatment than male patients, with an odds ratio of 1.85. Furthermore, patients who were vaginally delivered responded better to the Ponseti casting technique and demonstrated better prognoses compared to those delivered via caesarian section. In addition, a positive family history was neither predictive nor assumptive for congenital clubfoot in the idiopathic or neuromuscular groups. When the perinatal status of the mother was normal, there was a better response to the Ponseti casting than the surgical method and better prognosis compared to mothers with a complicated perinatal status. Furthermore, patients who were delivered normally had a better response to the Ponseti casting technique and better prognoses compared to those delivered through a cesarean section. Female patients with neuromuscular disease were more likely to undergo Ponseti casting than males, with an odds ratio of 1.7. The success of the Ponseti casting technique was more recognizable in babies with flexible clubfeet than rigid clubfeet. All patients with neuromuscular disease who were included in this study had bilateral clubfeet. The treatment results for both groups, including age at diagnosis, modified Pirani score before/after treatment, and the mean number of casts applied to achieve correction are listed in Table 2. The mean age of the patients at presentation in weeks in the idiopathic and neuromuscular groups was 1.79 and 1.80, respectively. We measured the effectiveness of the Ponseti casting technique in the treatment of clubfoot in both groups by comparing the results before and after Ponseti treatment

Table 1: The demographical data of the idiopathic group and neuromuscular group who treated successfully with Ponseti casting

Parameters	Gender		Mood of delivery		Perinatal status		Family history		Rigidity	
	Male	Female	Normal	c-section	Normal	Complicated	Positive	Negative	Rigid	Flexible
Idiopathic	15	19	24	10	30	4	3	31	7	27
	Odds ratio of 1.85, *P*=0.36		Odd ratio of 0.77 *P*=0.74		Odd ratio of 1.59, *P*=0.57				Odd ratio of 1.2, *P*=0.75	
Neuromuscular -associated clubfeet	5	8	3	5	4	4	0	8	2	6
	Odd ratio of 1, *P*=0.73		Odd ratio of 6, *P*=0.26		Odd ratio of 2.67, *P*=0.5				Odd ratio of 2, *P*=0.59	

Table 2: Result of treatment with Ponseti casting in idiopathic and neuromuscular group

Parameters	Number of patients who were included	Age of the patients at presentation in weeks	Modified Pirani score before treatment	Modified Pirani score at last follow-up	Number of casts applied for achievement of correction	Follow-up period
Mean of the parameters						
Idiopathic	39	1.794	5.1	0.39	6.1	6.1
Neuromuscular-associated clubfoot	10	1.80	5.6	0.41	5.7	10.2

Table 3: Comparison of treatment requirements, failure, and recurrence rates and shoe wear type

	Idiopathic	Neuromuscular
Complain after treatment		
Yes	6 (17.6)	1 (12.5)
No	28 (82.4)	7 (87.5)
Pain		
Yes	4 (11.7)	1 (12.5)
No	30 (88.3)	7 (87.5)
Shoe wear		
Normal	30 (88.3)	7 (87.5)
Medicated	4 (11.7)	1 (12.5)

Table 4: Comparison between neuromuscular-associated clubfeet patients who were included in this study

	Neuromuscular disease	Age at diagnosis, week	Age at 1st cat, week	Number of cast needed	Follow-up periods, years	Procedure	Additional procedures	Outcome
1st	spinal muscular atrophy	1	1	7	11	No	No	Good
2nd	spinal muscular atrophy	1	1	6	12	No	No	Good
3rd	spinal muscular atrophy	2	2	6	10	No	No	Good
4th	Muscular dystrophy	1	1	5	8	No	No	Good
5th	Muscular dystrophy	1	1	4	10	No	No	Good
6th	Congenital myopathy	3	1	7	12	No	No	Good
7th	Congenital myopathy	1	1	6	9	No	No	Good
8th	Unknown syndrome	1	1	7	10	TAT	No	Good
9th	Charcot marry tooth syndrome	4	1	5	9	TAT	Osteotomy	Good
10th	Unknown syndrome	3	1	4	11	TAT	Osteotomy	Good

TAT: Tibialis anterior transfer

using a modified Pirani score. The mean modified Pirani score before treatment was 5.1 in the idiopathic group and 5.6 in the neuromuscular disease group. Post-Ponseti treatment, both groups had a similar mean modified Pirani score (0.39 in the idiopathic group and 0.41 in the neuromuscular group). The follow-up period for patients in both groups ranged from 8 to 12 years. These results indicate the effectiveness of the Ponseti treatment in achieving correction in both groups. The idiopathic group had a slightly higher average of 6.19 casts needed for initial correction. Presumably, it is because the number of eligible patients included in the study was higher in the idiopathic group than in the neuromuscular disease clubfoot group, which created an opportunity for a higher deviation from the mean. Table 3 shows the comparison between neuromuscular disease-associated clubfoot and idiopathic clubfoot regarding treatment requirements, failure, recurrence

rates, and shoe wear type. Patients in both groups were less likely to have any complaints after treatment, with an odds ratio of 1.32 (CI: 0.3–5.8). Moreover, they were more likely to wear normal shoes after casting compared to those treated with a surgical method. Data on the ten patients in the neuromuscular group, including the patient's neuromuscular disease, age at diagnosis, age when the first cast was applied, mean number of casts needed for correction, follow-up period, any additional procedures performed, and outcomes of treatment are illustrated in Table 4. Neuromuscular disease-associated clubfoot patients required a mean of 5.7 casts for initial correction, and a TAT was performed on three out of ten patients. TAT was successful in achieving a plantigrade foot in one patient, but two out of the three patients had to undergo additional surgery (osteotomy) for foot correction. The achievement of the initial correction with the Ponseti casting technique with TAT was possible for one patient (10%) in the neuromuscular group and five patients (14.7%) in the idiopathic group. The indication for surgery was failure of casting to achieve correction in two patients, one of whom had an unknown syndrome and the other one had Charcot–Marie–Tooth syndrome; both required osteotomies.

Discussion

In our study, the initial correction of neuromuscular disease-associated clubfoot using the Ponseti casting technique with/without TAT was achieved in eight patients. The surgical correction techniques were successful on those for whom the Ponseti treatment was first used but failed to correct the foot; one patient out of the two needed surgical correction. Neuromuscular disease-associated clubfoot has been reported to be difficult to manage nonoperatively; surgical management is generally required.[13] Many studies have examined the effectiveness of surgical management and have confirmed the lack of satisfactory results in treating nonidiopathic clubfoot through conservative management only. Hennigan and Kuo noted positive results in 62% of patients with clubfoot associated with constriction band syndrome.[10] Furthermore, de Carvalho Neto *et al*. reported that only 63% of patients with clubfoot associated with spina bifida who were operatively managed improved.[14] Furthermore, Flynn *et al*. reported positive results in 61% of patients with clubfoot associated with myelodysplasia who were managed surgically in their study.[3] Widmann *et al*.[15] reported that only 50% of arthrogrypotic patients had a satisfactory outcome in their study, while Chang and Huang[16] reported that 68% exhibited excellent outcomes. There was a notable difference between the outcomes of these studies. When we compare the effectiveness of the Ponseti method in neuromuscular disease-associated clubfoot with our series of casting for idiopathic clubfoot, the Ponseti casting technique was equally successful in both groups. The idiopathic and neuromuscular clubfoot groups required an equal mean number of casts (6.11 vs. 5.7) to achieve optimal correction. The failure rate of Ponseti casting technique to achieve initial correction was not significantly different between the two clubfoot groups: five patients out of 39 in the idiopathic group and two patients in the neuromuscular group. In the group of patients with neuromuscular disease-associated clubfoot in this retrospective study, the Ponseti casting technique was satisfactory in 80% of patients, and they did not require surgical intervention. Our results for the management of clubfoot associated with neuromuscular disease surpassed those reported in the studies published by Dobbs *et al*., which showed a failure rate of 6% in the idiopathic group and 33% in patients with clubfoot associated with myelodysplasia.[17] Boehm *et al*. showed a failure rate of 8% in the idiopathic group and 50% in clubfoot associated with arthrogryposis.[1] Postcorrection bracing strategy is a significant factor that affects the achievement of optimal correction.[15,18,19] We could not estimate brace compliance from our review of the health documentation system for our patient groups. Many studies have reported that suboptimal brace compliance is a crucial factor that leads to high recurrence rates, as well as the need for surgical releases in patients with neuromuscular clubfoot; this should be considered in future studies.[20,21] This study had two major limitations that could be addressed in future research. First, the number of patients who were eligible for the study was small. Second, some patients were lost to follow-up, so it is possible we did not capture all Ponseti treatment failures in the neuromuscular group in our study population. Despite these limitations, the success of the Ponseti method of treatment for patients with neuromuscular disease-associated clubfeet in our hospital surpassed that reported in the few previously published reports. We recommend the establishment of a national registry for clubfoot cases in the country so that we can disseminate our successful experience in the treatment of clubfoot on an international level.

Conclusions

The Ponseti casting technique is an effective primary modality of treatment for patients with clubfoot associated with neuromuscular diseases, as it is in idiopathic clubfoot, to achieve functional pain-free feet and reduce the number of surgical interventions required. The Ponseti treatment method for patients with neuromuscular disease-associated clubfoot in our institution performed better than that in the very few previously published reports.

Ethical considerations

Ethics approval was provided by the Ethical Committee, University Hospitals, Imam Abdulrahman bin Faisal, December 2017.

Acknowledgment

We would like to express our gratitude to Prof. Khalid Khoshhal, Pediatric Orthopedic Surgeon, Taibah University, for his expert advice, revision, and support throughout the writing of this article and Mr. Soban Qadir Khan, Lecturer of Biostatistics, College of Dentistry, Imam Abdulrahman Bin Faisal University, for his expert technical assistance in data analysis using SPSS.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

Authors' contributions
AKO and DAB designed the study and provided the research materials. BAZ analyzed, interpreted data, and wrote the initial and final drafts of the paper. AAM and MAY conducted research and collected the data. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

References
1. Boehm S, Limpaphayom N, Alaee F, Sinclair MF, Dobbs MB. Early results of the Ponseti method for the treatment of clubfoot in distal arthrogryposis. J Bone Joint Surg Am 2008;90:1501-7.
2. Gurnett CA, Boehm S, Connolly A, Reimschisel T, Dobbs MB. Impact of congenital talipes equinovarus etiology on treatment outcomes. Dev Med Child Neurol 2008;50:498-502.
3. Flynn JM, Herrera-Soto JA, Ramirez NF, Fernandez-Feliberti R, Vilella F, Guzman J. Clubfoot release in myelodysplasia. J Pediatr Orthop B 2004;13:259-62.
4. Jan WF, Jahan A, Dar MY, Bangroo FA. A study on management of congenital clubfoot by Ponsetti technique in a rural medical college in India. Int J Res Orthop 2018;5:172.
5. Bakalis S, Sairam S, Homfray T, Harrington K, Nicolaides K, Thilaganathan B. Outcome of antenatally diagnosed talipes equinovarus in an unselected obstetric population. Ultrasound Obstet Gynecol 2002;20:226-9.
6. Pavone V, Bianca S, Grosso G, Pavone P, Mistretta A, Longo MR, *et al*. Congenital talipes equinovarus: An epidemiological study in Sicily. Acta Orthop 2012;83:294-8.
7. Legaspi J, Li YH, Chow W, Leong JC. Talectomy in patients with recurrent deformity in club foot. A long-term follow-up study. J Bone Joint Surg Br 2001;83:384-7.
8. Sherk HH, Ames MD. Talectomy in the treatment of the myelomeningocele patient. Clinical orthopaedics and related research 1975;110:218-22.
9. Westcott MA, Dynes MC, Remer EM, Donaldson JS, Dias LS. Congenital and acquired orthopedic abnormalities in patients with myelomeningocele. Radiographics 1992;12:1155-73.
10. Hennigan SP, Kuo KN. Resistant talipes equinovarus associated with congenital constriction band syndrome. J Pediatr Orthop 2000;20:240-5.
11. Banta JV. Congenital Clubfoot: Fundamentals of Treatment. By Ponseti IV. Oxford: Oxford University Press. 1996. p. 140.
12. Smythe T, Chandramohan D, Bruce J, Kuper H, Lavy C, Foster A. Results of clubfoot treatment after manipulation and casting using the Ponseti method: Experience in Harare, Zimbabwe. Trop Med Int Health 2016;21:1311-8.
13. Janicki JA, Narayanan UG, Harvey B, Roy A, Ramseier LE, Wright JG. Treatment of neuromuscular and syndrome-associated (nonidiopathic) clubfeet using the Ponseti method. J Pediatr Orthop 2009;29:393-7.
14. de Carvalho Neto J, Dias LS, Gabrieli AP. Congenital talipes equinovarus in spina bifida: Treatment and results. J Pediatr Orthop 1996;16:782-5.
15. Widmann RF, Do TT, Burke SW. Radical soft-tissue release of the arthrogrypotic clubfoot. J Pediatr Orthop B 2005;14:111-5.
16. Chang CH, Huang SC. Surgical treatment of clubfoot deformity in arthrogryposis multiplex congenita. J Formos Med Assoc 1997;96:30-5.
17. Dobbs MB, Rudzki JR, Purcell DB, Walton T, Porter KR, Gurnett CA. Factors predictive of outcome after use of the Ponseti method for the treatment of idiopathic clubfeet. J Bone Joint Surg Am 2004;86:22-7.
18. Green AD, Lloyd-Roberts GC. The results of early posterior release in resistant club feet. A long-term review. J Bone Joint Surg Br 1985;67:588-93.
19. Grill F, Franke J. The Ilizarov distractor for the correction of relapsed or neglected clubfoot. J Bone Joint Surg Br 1987;69:593-7.
20. Yen CC, Huang SC. Surgical treatment of congenital convex Pes valgus. J Formos Med Assoc 1997;96:424-8.
21. Ganesan B, Luximon A, AlJumaily A, Balasankar SK, Naik GR. Ponseti method in the management of clubfoot under 2 years of age: A systematic review. PLoS One 2017;12:e0178299.