

## Original Article

# Effectiveness of Ponseti technique in management of arthrogryptic clubfeet - a prospective study

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**Abstract:** Background: Clubfoot constitutes roughly 70 percent of all foot deformities in arthrogryposis syndrome and 98% of those in classic arthrogryposis. Treatment of arthrogryptic clubfoot is difficult and challenging due to a combination of factors like stiffness of ankle-foot complex, severe deformities and resistance to conventional treatment, frequent relapses and the challenge is further compounded by presence of associated hip and knee contractures. Method: A prospective clinical study was conducted using a sample of nineteen clubfeet in twelve arthrogryptic children. During weekly visits Pirani and Dimeglio scores were assigned to each foot followed by manipulation and serial cast application according to the classical Ponseti technique. Mean initial Pirani score and Dimeglio score were  $5.23 \pm 0.5$  and  $15.79 \pm 2.4$  respectively. Mean Pirani and Dimeglio score at last follow up were  $2.37 \pm 1.9$  and  $8.26 \pm 4.93$  respectively. An average of 11.3 casts was required to achieve correction. Tendoachilles tenotomy was required in all 19 AMC clubfeet. Result: The primary outcome measure was to evaluate the role of Ponseti technique in management of arthrogryptic clubfeet. The secondary outcome measure was to study the possible causes of relapses and complications with additional procedures required to manage clubfeet in AMC an initial correction was achieved in 13 out of 19 arthrogryptic clubfeet (68.4%). Relapse occurred in 8 out of 19 clubfeet. Five of those relapsed feet were corrected by re-casting  $\pm$  tenotomy. 52.6% of arthrogryptic clubfeet were successfully treated by the Ponseti technique in our study. Three patients failed to respond to Ponseti technique required some form of soft tissue surgery. Conclusion: Based on our results, we recommend the Ponseti technique as the first line initial treatment for arthrogryptic clubfeet. Although such feet require a higher number of plaster casts with a higher rate of tendo-achilles tenotomy but the eventual outcome is satisfactory. Although, relapses are higher than classical idiopathic clubfeet, most of them respond to re-manipulation and serial casting  $\pm$  re-tenotomy.

**Keywords:** Arthrogryposis multiplex congenita (AMC), clubfeet, Ponseti technique, Pirani score, Dimeglio score, relapse, tenotomy

## Introduction

Arthrogryposis multiplex congenita (AMC) is a group of congenital conditions that present with joint contractures in two or more body locations. Although the exact cause is unknown, it may include genetic, parental, and environmental factors, as well as abnormalities during fetal development. Patients present reduced joint movement in the affected body regions, as well as muscular weakness.

This is clinically diagnosed as a non-progressive syndrome characterized by:

a) Joint contractures in at least two places of the body at birth.

b) Clinical evidence of a non-progressive neurologic disorder.

c) Fusiform joint configuration with diffuse muscle wasting.

d) Dimpling of the skin around sites of severe joint contractures.

e) Webbing.

f) Normal skin creases are absent [1].

The literature describes different subtypes of arthrogryposis as amyoplasia, distal arthrogryposis and syndromic cases [2]. Boehm reported that various subtypes of arthrogryposis behave differently in terms of the rigidity of feet, the

## Management of arthrogryptic clubfeet by Ponseti technique

ability to achieve correction and the recurrence of deformity [3]. Therefore, a differentiation of the subtype should be determined beforehand for providing optimum treatment. The classic form of arthrogryposis, also called amyoplasia, or arthrogryposis multiplex congenita (AMC), constitutes about 30% of all congenital contractures and usually presents as severe, multiple upper and/or lower extremity contractures with muscle weakness of variable intensity. Hands and feet are mostly affected in distal arthrogryposis, which is an autosomal dominant disorder without underlying general neuromuscular pathology contractures around the proximal joints are spared or restricted in their appearance [2]. Etiology of arthrogryposis is still unknown. Fetal akinesia is most commonly associated with it [4]. Arthrogryposis is likely to occur in conditions of myogenic abnormalities like congenital muscular dystrophy, neurogenic abnormalities like spinal muscular atrophy [5]. It is also commonly associated with pregnancies worsened by physical constraints like oligohydramnios, amniotic bands or other maternal factors during pregnancy like multiple sclerosis, diabetes mellitus and maternal myasthenia gravis [6].

Clubfoot constitutes approximately 70 percent of all foot deformities in arthrogryposis syndrome and 98% of those in classic arthrogryposis [7, 8]. The treatment of clubfoot in AMC is difficult and challenging due to a combination of factors like the stiffness of ankle-foot complex, severe deformities or resistance to conventional treatment, frequent relapses and the challenges are further compounded by presence of associated hip and knee contractures. The challenge of frequent relapses is further compounded by presence of associated hip and knee contractures. Various treatment options have been tried in the past for arthrogryptic clubfoot deformity. Soft tissue releases from basic tendo achilles tenotomy to radical soft tissue releases showed variable outcome ranging from 21% to 100% with the idea that better results can be obtained by operating at a younger age and using more aggressive methods [9]. Tarsal decancellation showed a potential drawback with collapse of the tarsal bones which disrupts the anatomy and congruency of adjacent joints, leading to severe osteoarthritic changes in the long run [10]. Talectomy is an ablative operation which cannot fix fore-

foot abnormalities and raises the risk of arthritic alterations in the future [11]. Multiple operational treatments to repair arthrogryptic clubfeet can cause stiff deformities, and various osteotomies may address their recurrence with variable success rate [12]. Choi used the Ilizarov technique to treat recurrent arthrogryptic clubfoot in children. Ten out of 12 feet were plantigrade after a 35-month follow-up [13].

In the study conducted by Lloyd, he reported an initial correction with Ponseti Technique in arthrogryptic clubfoot, however pointed out that complete correction was difficult to achieve [1]. To the best of our knowledge, there are limited studies in the literature, which have evaluated the outcomes of conservative management in arthrogryptic club foot deformities. With the premise that early intervention in idiopathic clubfoot deformities with Ponseti technique manipulation and casting can play a successful role in getting full correction, we decided to expand our area of interest to include arthrogryptic clubfeet instead of immediately pushing patients for extensive surgery. We conducted a clinical study of clubfoot management in AMC as there are only a few published studies of the management of arthrogryptic clubfeet using the Ponseti technique. The primary outcome measure was to evaluate the role of Ponseti technique in management of arthrogryptic clubfeet. The secondary outcome measure was to study the possible causes of relapses and complications with additional procedures required to manage clubfeet in AMC.

### Methods

#### *Study design*

This was a prospective clinical study conducted at a tertiary care centre from November 2019 to October 2022. The study was conducted after approval from institutional ethical committee (D.No-188/FM). An informed written consent was obtained from the parents of all patients.

#### *Case definition of arthrogryptic clubfoot*

Arthrogryptic children with two or more joint contracture along with clubfoot deformity were included in our study.

## Management of arthrogryptic clubfeet by Ponseti technique

**Table 1.** Laterality for children with clubfoot associated with arthrogryposis

Demographics	No. of patients	%
Bilateral	7	58.33
Unilateral	5	41.67
Total	12	

### *Inclusion and exclusion criteria*

Patient satisfying criteria of the case definition of arthrogryposis with clubfoot disorder were included in this study. Patients with classical idiopathic clubfeet and other non-idiopathic causes of clubfeet were excluded.

All study participants were thoroughly examined as per the study protocol to assess all their deformities, history of past treatment with casts or manipulation, and any family history of related malformations. A thorough examination of the patient was performed, including general-systemic and local examinations to diagnose Arthrogryposis multiplex congenita. Paediatric opinion was also sought to rule out any other syndromic association.

### *Study population and demographic characteristics*

Nineteen arthrogryptic clubfeet in twelve patients were included in our study. Manipulation and serial casting by Ponseti technique was initiated in all 19 AMC clubfeet. These cases were selected based on history and clinical examination. The proportion of patients with AMC clubfeet presenting to our institute was 4.76% (The total number of patients with clubfoot was 252, out of those, 12 patients had AMC clubfoot). Out of 12 AMC patient 4 were male and 8 were female. Five patients presented before 3 months of age and rest of them presented at later age. Mean age at presentation was 9.25 months ranging from 0 to 36 months. Seven out of twelve (58%) AMC clubfeet patient presented with bilateral clubfeet deformity. In a unilateral presentation left side was more common (**Table 1**).

After inclusion in the study, Pirani [15] and Dimeglio [16] score was assigned to each foot at every visit followed by manipulation and serial cast application according to the Ponseti technique at a weekly interval. Clinical improve-

ment in deformity (Pirani and Dimeglio score) and complications were monitored and recorded at each visit. Observation indicators in this study includes the pre and post correction Pirani and Dimeglio score, the pre and post correction grading of the foot, the average number of casts required, the number of feet requiring Achilles tenotomy and the number of feet achieving successful plantigrade correction. A successful outcome was defined as a plantigrade foot with a straight lateral border during weight bearing, ability to comfortably wear regular shoes. Lyold Roberts and Lettin described the goal of treatment of Arthrogryptic clubfoot is "to convert a deformed rigid foot into a plantigrade platform" [1]. Record of relapse and failure of Casting were noted and any need of additional surgeries for successful correction.

### *Manipulation and serial casting with Ponseti technique*

The Ponseti technique of manipulation and serial casting was applied to all the patients to start with and were regularly monitored. No modification to the classical Ponseti technique was made. After the correction of feet, maintenance of correction was done using SFAB (Steenbeek Foot Abduction Brace) was given to all the patients 3 weeks after tenotomy to maintain correction. It was applied 23 hours a day for first 3 months and then at night/nap time for another 3 years. **Figures 1** and **2** depicting the pre-treatment and post-treatment clinical photograph of the two patients.

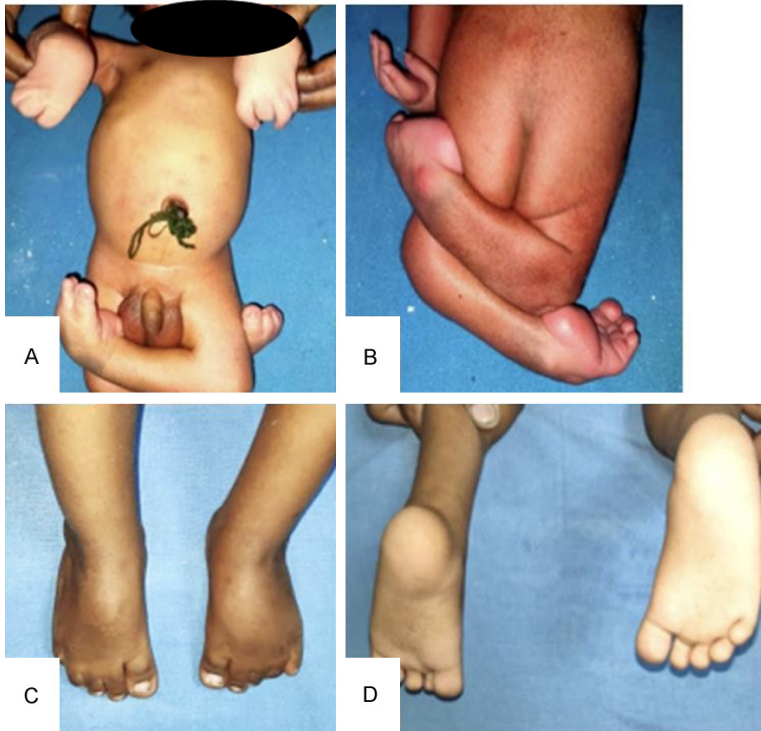
### *Management of relapses and failures*

Patients presenting with relapse were given repeat manipulation and Ponseti casting trials. Failures or non-responders to the classical Ponseti technique were subjected to additional procedures (soft tissue surgeries like plantar fascia release, posterior soft tissue releases and ligamentotaxis utilizing the Joshi's External Stabilizing System-JESS).

### *Statistical analysis*

The categorical variables were presented in the form of numbers and percentages. Descriptive statistics such as mean, median, mode, standard deviation, frequency and percentage were used to describe the data. Inferential statistics such as the Wilcoxon signed rank test was used

## Management of arthrogryptic clubfeet by Ponseti technique



**Figure 1.** A, B. Pre-correction bilateral clubfeet in AMC child. C, D. Post correction image showing plantigrade feet after Ponseti technique.



**Figure 2.** A, B. Pre-correction bilateral clubfeet in AMC child. C, D. Post correction plantigrade feet after Ponseti technique.

to analyze the difference in means before and after intervention. The data was entered into a

Microsoft EXCEL spread sheet, and the final analysis was performed using IBM's Statistical Package for Social Sciences (SPSS) software (Chicago, USA), version 21.0. A *P*-value of less than 0.05 was considered statistically significant.

### Results

Initial correction was achieved in 13 out of 19 AMC clubfeet (68.4%) with an average duration of follow-up of 1.83 years, ranging from 9 to 30 months. Mean pre correction Pirani score in our study was  $5.23 \pm 0.5$  (range from 4.5-6) and mean post correction Pirani score at last follow-up was  $2.37 \pm 1.9$  (range from 0.5-5.5). Mean pre correction Dimeglio score in our study was  $15.79 \pm 2.41$  (range from 12-19) and mean post correction Dimeglio score at the last follow-up was  $8.26 \pm 4.93$  (range 2-17). On presentation majority of feet (11/19, 58%) were Grade IV according to Dimeglio criteria. Post Ponseti casting the majority of feet were either Grade I (4 out of 19 AMC clubfeet) or Grade II (8 out of 19 AMC clubfeet) as shown in **Table 2**. The change in mean score post intervention was found to be statistically significant (**Table 2**; **Figures 5 and 6**).

Despite initial major rigid deformities, these arthrogryptic clubfeet responded to Ponseti's early manipulations and casting with total or partial deformity correction. An average of 11.3 casts was required to achieve correction. Tendo-achilles tenotomy was required in all 19 AMC clubfeet (100%) (**Table 3**). Average number of casts required to achieve the initial correction

clubfeet (100%) (**Table 3**). Average number of casts required to achieve the initial correction

## Management of arthrogryptic clubfeet by Ponseti technique

**Table 2.** Pre and Post correction Pirani scores (PS) & Dimeglio Scores (DS) by using Ponseti technique

	Mean	SD	Wilcoxon signed rank test	P-value
Pre correction PS	5.23	0.5	-3.84	0.001
Post correction PS	2.37	1.9		
Pre-correction DS	15.71	2.41	-3.83	0.001
Post-correction DS	8.26	4.93		

PS: Pirani Score, DS: Dimeglio Score.

increased with increase in age of presentation and the severity grade of clubfeet at presentation.

### Relapses and failures

We observed relapse in 8 out of 19 AMC clubfeet (42.1%). Five of these relapsed cases responded to re-casting ± tenotomy, based on the classical Ponseti technique. Three of them required some form of additional procedures for achieving full correction (**Table 4; Figure 3**). Successful correction was achieved in total 10 out of 19 AMC Clubfeet (52.6%) by Ponseti technique. Those AMC clubfeet which failed to get corrected by the Ponseti technique required some form of surgical intervention; JESS (**Figure 4**) in 2 feet, plantar fascia release and posterior release in 3 feet (**Table 5**). Along with clubfeet deformity in Arthrogryptic child, other deformity like knee flexion and extension contracture were also corrected either by stretching and casting or by tendon release or osteotomy to make child ambulate independently.

### Discussion

The purpose of this study was to evaluate the role of classical Ponseti technique in management of an arthrogryptic clubfoot. The treatment of such feet is difficult owing to the nature of deformity and its association with hip and knee joint contractures. Children with arthrogryptic clubfoot often had many surgeries in the past because it was thought they would be resistant to non-operative treatment. The most frequent foot deformity linked with arthrogryptosis is a severe, resistant clubfoot. Its treatment has always been complex and prone to complications and relapses. Extensive soft tissue and bony procedures were carried out in the past which often leads to stiff and painful feet with high recurrence rates. The disappoint-

ing results of primary aggressive surgery suggested that the approach of primary aggressive surgery is not proficient enough to provide successful outcome, regardless of the short term result may look satisfying [17, 18]. Several studies have reported on the use of Ponseti approach for these difficult feet, citing the poor results of major surgery and the spike in interest in conservative ways of clubfoot therapy during the last several decades [3, 19-21].

### Effectiveness of classical Ponseti technique in arthrogryptic clubfeet

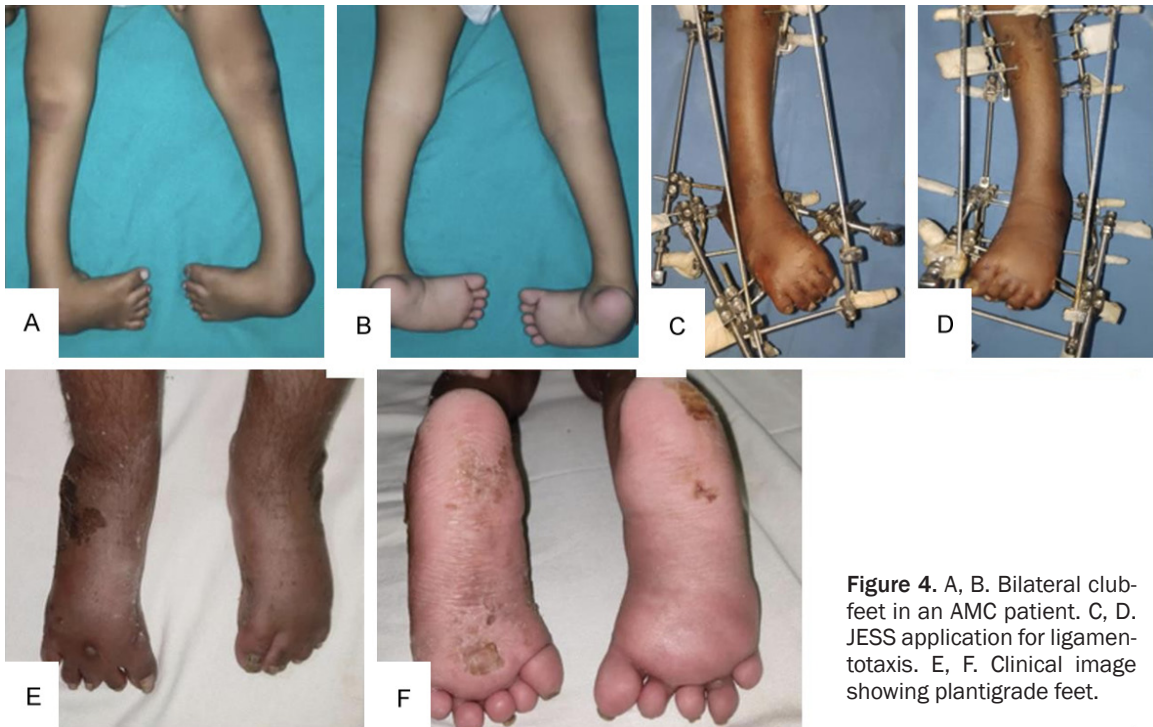
We studied 19 arthrogryptic clubfeet, this sample size is comparable to the study done by Boehm (24 feet) [3], Kowalczyk (18 feet) [22], and Matar (17 feet) [19]. Initial correction was obtained in 13 out of 19 feet accounting for 68.4% of initial correction by the Ponseti method. Our initial correction of AMC clubfeet was lower than other studies. This could be due to the higher mean age of presentation of AMC clubfeet child in our study as compared to other studies. Mean follow up duration was 1.83 years (range from 9 to 30 months) which is less than other studies for example Morcuende (4.6 years) [20], Kowalczyk (3 years) [22] and Janicki (2.6 years) [23]. This could possibly be due to the fact that most of the other studies in management of AMC clubfeet are retrospective studies. In our opinion a shorter follow-up is not enough time to comment on the frequency of relapses and its pattern which arthrogryptic clubfeet develops over a period of time.

In our study, the successful correction (painless plantigrade feet) by Ponseti method was 52.6% which is comparable to study done by Avi Shah (56%) [24], Janicki (50%) [23], Mulder (58%) [25]. The successful correction achieved in our study is comparable to other studies, regardless of the high relapse rate and higher number of casts. Some of our patients were advised orthosis other than the standard foot abduction orthosis due to co-existing deformities of the hip and knee joints and/or inability to abduct foot to 70° including Ankle Foot Orthosis (AFO) and Knee Ankle Foot Orthosis (KAFO) as per the clinical indication. Even when tolerated, a foot abduction brace may need to be turned out by not more than 30-40°, unlike the 70° external rotation recommended for idiopathic clubfeet. Eight of these nineteen AMC clubfeet

## Management of arthrogrypotic clubfeet by Ponseti technique



**Figure 3.** A. Clinical image showing calcaneo-valgus foot on right side and equino-varus foot on left side in an AMC child. There was no significant correction despite Ponseti casting. B. Intra-operative image of showing soft tissue releases. C-E. Post correction image showing plantigrade feet from front, side and back.

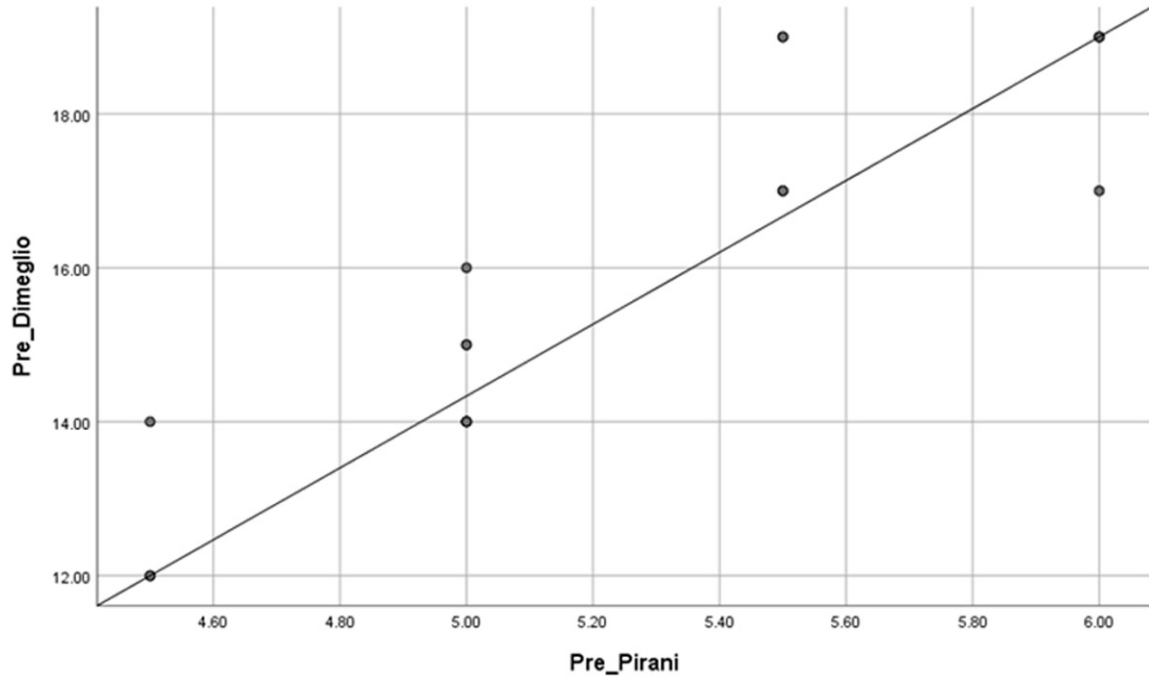


**Figure 4.** A, B. Bilateral clubfeet in an AMC patient. C, D. JESS application for ligamentotaxis. E, F. Clinical image showing plantigrade feet.

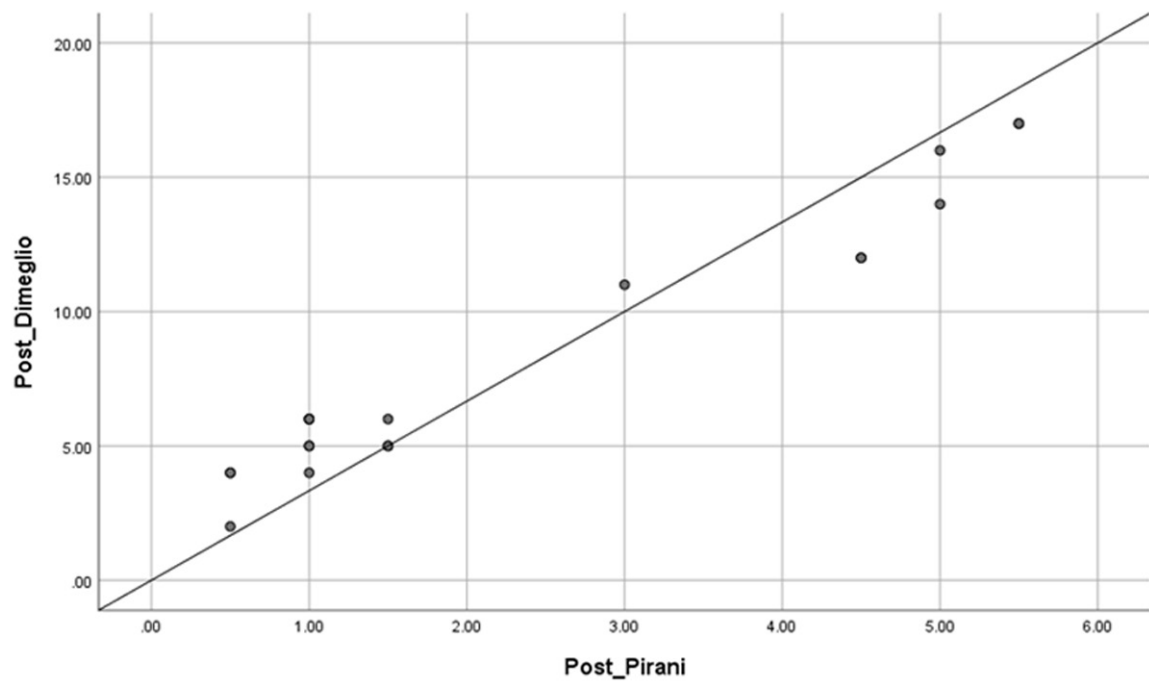
(13 feet got initial correction) relapsed, accounting for 42% of relapse cases. This is comparable to the study done by T. De Mulder (35%) [25] and Derevyanko (55%) [26] (Table 6). Despite 8

relapses, 5 of them responded to re-casting ± tenotomy as per classical Ponseti technique. Three patients whose clubfeet could not be corrected through the Ponseti technique went

## Management of arthrogryptic clubfeet by Ponseti technique



**Figure 5.** Significant positive correlation between initial Pirani Score (pre-treatment PS) and initial Dimeglio Score (pre-treatment DS).



**Figure 6.** Shows significant positive correlation between Pirani Score at SFAB (post-treatment PS) and Dimeglio Score at SFAB (post-treatment DS).

under surgical management for the correction of the deformity. In our study, no major bony surgery (talectomy or osteotomy) was carried out

to correct the deformity. In our opinion, the type and extent of surgical procedure were reduced due to the initial management of such clubfeet

## Management of arthrogryptic clubfeet by Ponseti technique

**Table 3.** Treatment requirement, failures and recurrence

	No. of feet
No. requiring Tendo-achilles Tenotomy	19 out of 19 feet
No. of initial success	13 out of 19 feet
No. of recurrence	8 feet
No. of correction after relapse by Ponseti technique	5 feet out of 8 relapsed
No. of failures in total	9 feet

**Table 4.** Management of relapses

Treatment of relapse	No. of feet
Re-manipulation and casting	5 feet had successful outcome out of 8 relapse
Repeat Tendo-achilles tenotomy	8 feet
Posterior soft tissue releases	1 foot
Plantar fascia release	2 feet

**Table 5.** Management of failure: six feet did not achieve initial correction by Ponseti technique, and were managed with surgical intervention

Treatment of failure	Number of feet
Posterior soft tissue release	3
Plantar fascia release	3
JESS application	2

with Ponseti technique which resulted in stretching of the soft tissues around the rigid ankle-foot complex. One of the crucial factors for the success of the Ponseti method is appropriate and adequate bracing with a Foot Abduction Brace (FAB).

In our clinical experience postoperative use of FABs remains difficult, with a high rate of brace non-compliance. This may be due to intrinsic factors in AMC clubfeet including poor fibrotic muscle development. Non-compliance to wear the brace may also be a factor in the failure of the full correction of a deformity as it is quite a rigid deformity. An under corrected foot with a residual deformity is likely to fit poorly in a foot abduction brace. Moreover, the majority of relapses occurred in first 9 months of achieving correction. Hence, the first year post-correction is the most crucial time to be vigilant against recurrences and relapses. A longer duration of bracing is required in arthrogryptic clubfeet to maintain the correction. We recommend bracing for at least 4 years to maintain correction. Bracing protocol should be supplemented with aggressive stretching exercises of ankle and

feet. It is essential to emphasize that these children have complex needs that necessitate a multidisciplinary approach in order to address them.

### *Complications*

No major complications of the Ponseti casting like rocker bottom foot or flat foot were noted. Tenotomy related complications like excessive bleeding or wound complications did not occur in any patient. Cast slip-

page and pressure sores were the most common complication encountered in our study (**Table 7**). Minor dermatological complications like abrasion, eczema and plaster sores were managed conservatively with emollients, topical steroids and extra padding over the lesion. For cast breakage, we re-enforced or reapplied the cast. For overcrowding of the toes, we trimmed the cast. Some of these complications are illustrated in **Figure 7**.

### *Relapses and failures*

Arthrogryptic clubfeet which did not respond to the classical Ponseti technique were managed with soft tissue releases or ligamentotaxis with JESS frame application. Although major corrective surgeries were performed by most previous authors on relapsed AMC clubfeet; we did not perform any major bony surgeries like osteotomy or talectomy. The surgical treatment of arthrogryptic clubfeet is usually complicated with high relapses which leads to salvage surgery in the end.

Though extensive soft tissue releases as a primary management of arthrogryptic clubfeet have been successful under the age of 1 year [27, 28], but in relapse cases due to extensive post-operative scarring of previous surgery repeat soft tissue surgery usually fails.

### *Strengths, limitations and future recommendations*

Our study evaluates the role of Ponseti technique in management of arthrogryptic club-



## Management of arthrogryptic clubfeet by Ponseti technique

**Table 6.** Comparison of various studies on management of arthrogryptic clubfeet by ponseti method

Author	No. of patient	No. of AMC clubfeet	Study design	Mean follow up	Mean No. of cast	Need for tenotomy	Successful treatment	Relapses
Morcuende [20]	16	32	Retrospective	4.6 years	7	--	22/32=68.7%	25%
Kowalczyk [22]	5	10	Retrospective	2.9 years	8.4	--	7/10=70%	90%
Boehm [3]	12	24	Retrospective	2 years	6.9	24/24	22/24=91.6%	25%
Janicki [23]	5	8	Retrospective	--	--	--	4/8=50%	25%
Derevyanko [26]	10	20	Prospective	--	7	14/20	9/20=45%	55%
Matar [19]	10	17	Retrospective	5.8 years	8	16/17	11/17=64.7%	-
T D Mulder [25]	--	67	Retrospective	--	7.2	64/67	39/67=58.2%	35%
Avi Shah [24]	--	25	Retrospective	5.8 years	--	--	14/25=56%	--
Our study	12	19	Prospective	1.83 years	11.3	19/19	10/19=52.6%	42%

**Table 7.** Complications of Ponseti casting in AMC clubfeet

Complication	No. of feet
Pressure sore	6
Cast slippage	6
Eczematization with scaling	2
Skin blister	3
Cast breakage	4
Dynamic supination	2
Fracture distal tibial metaphysis (forced dorsi-flexion)	1

feet. The strengths of our study includes its prospective nature compared to most of the other studies which are retrospective in nature. We strictly adhered to the Inclusion criteria of our case definition. Standard treatment protocol was followed in all our cases and there was no modification in the classical Ponseti technique as compared to some studies where some modification to the original technique was done. Short comings of this study include small sample size, short follow-up and lack of comparison group. We admit that the short follow-up in our study may not be enough to assess the relapse pattern and functional outcome of the foot when the patient becomes an adult.

For patient to ambulate independently we need to look at the patient as a whole and address their hip and knee contracture along with the clubfeet deformity. Hence future studies with a large sample size and long follow-up are required for the management of arthrogryptic children ambulating independently.

### Conclusion

The correction of arthrogryptic clubfeet is difficult and challenging. These difficulties and challenges are due to the lack of standard treatment protocol for these rigid clubfeet. Other associated deformities like hip and knee contractures interfere with the conventional treatment of these feet. Our study was an attempt to standardize the management of these difficult feet with good outcome using the classical Ponseti technique. Although relapses are quite high, these feet respond to re-manipulation and serial casting  $\pm$  re-tenotomy with early total or partial deformity correction. Furthermore, long-term negative implications such as foot discomfort, ankle and foot osteoarthritis, and low quality of life could be prevented in the future by treating them with Ponseti casting rather than invasive surgery. Therefore, based on our study results and the existing literature we recommend the classical Ponseti technique as the first line initial treatment for arthrogryptic clubfeet. Although these feet require higher number of plaster casts, higher rate of tendo-achilles tenotomy and higher relapse rate than their classical idiopathic counterparts, but the eventual outcome is satisfactory. Before starting treatment, advising parents of the need of higher number of casts, high relapse rate, realistic expectations and regular and long term follow-up is essential. Treatment should focus on effective manipulation and serial casting.

## Management of arthrogryptic clubfeet by Ponseti technique



**Figure 7.** A. Plaster cast slippage and breakage. B. Pressure Sore over Dorsal aspect. C. Sore over Dorsolateral aspect of foot. D. Eczematization with Scaling. E. Distal tibial metaphyseal fracture due to forced dorsiflexion.

Adequate compliance to bracing and regular follow up for early recognition of relapse should be emphasized.

### Disclosure of conflict of interest

None.

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