IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 23, Issue 5 Ser. 11 (May. 2024), PP 37-42
www.iosrjournals.org

The Utility Of Multiple Serial Casting As A Treatment Of Clubfoot In Amc Using Pirani Scores

Dr. Mohammad Iftekhar Alam[1*], Dr. Md. Abdullah Al-Maruf[2],
Dr. Nabil Zunayed Sidny[3], Dr. Md. Assaduzzaman[4]

[1]*assistant Professor, Department Of Orthopaedic Surgery (Ex), Ad-Din Women's Medical College, Dhaka, Bangladesh*
[2]*assistant Professor, Department Of Orthopaedic Surgery, Marks Medical College Hospital, Dhaka, Bangladesh*
[3]*assistant Professor, Department Of Orthopaedic And Trauma Surgery, Shaheed Monsur Ali Medical College Hospital, Dhaka, Bangladesh*
[4]*assistant Professor, Department Of Orthopaedic, Bikrompur Bhuiyan Medical College & Hospital, Dhaka, Bangladesh*

Abstract
Introduction*: Arthrogryposis multiplex congenita (AMC) is a group of over 400 conditions characterized by severe joint contractures in two or more body regions. Managing clubfoot in patients with AMC is particularly difficult, and it is more likely to recur compared to idiopathic clubfoot. The Ponseti technique can be used to treat idiopathic clubfoot, which avoids or delays more invasive procedures.*
Objective*: The objective of this study is to evaluate the effectiveness of multiple serial casting as a treatment for clubfoot in Arthrogryposis Multiplex Congenita (AMC) patients, using Pirani scores as an objective measure of deformity.*
Methods*: This study was conducted at the Department of Orthopaedic Dr. Sirajul Islam Medical College Hospital and IBN Sina Diagnostics, Dhaka. The aim of this retrospective study was to analyze the effectiveness of Pirani scores in treating clubfoot. The study retrospectively collected data from 17 AMC patients who had a total of 25 clubfeet, and who underwent treatment between December 2021 and December 2023. Patients who had received a minimum of three casting series were included in the study. Pre-treatment and post-treatment deformity scores were examined across casting series using analysis of variance (ANOVA) statistical analysis.*
Results*: The Pirani scores before treatment in the first series improved from 4.80±1.54 to 1.68±1.48 ($p<0.001$). In the second series, the scores improved from 4.23±1.03 to 2.72±0.916 ($p<0.001$), and in the third series, the improvement was from 3.87±1.07 to 2.82±1.02 ($p<0.001$). There was a significant decrease in Pirani scores from the first series to the second ($p=0.001$) and third ($p<0.001$). Additionally, it was found that the number of casting days significantly affected the change in scores during the third series ($p=0.038$).*
Conclusion*: The multiple series casting aka Ponseti technique can be effective improvement of clubfoot in AMC as measured by the Pirani score. Data shows that early intervention yields better results, with a diminished yet effective ability to elicit change over time.*
Keywords*: Ponseti, arthrogryposis multiplex congenita, pirani, casting, treatment, clubfoot, arthrogryposis*.

Date of Submission: 21-05-2024 Date of Acceptance: 31-05-2024

I. Introduction

Treatment of idiopathic clubfoot with the Ponseti method has been shown to produce high success rates compared to early and more aggressive surgical approaches [1]. Compared to idiopathic clubfoot, non-idiopathic clubfoot is usually more severe, stiffer, and often associated with syndromic conditions. Non-idiopathic clubfoot was initially thought to be untreatable by the ponseti technique; however, subsequent studies have documented the utility of this technique for more complex deformities [2-7]. Patients with arthrogryposis multiplex congenita (AMC) and clubfoot have a type of clubfoot known as non-idiopathic clubfoot. Non-idiopathic clubfoot is generally more difficult to treat compared to idiopathic clubfoot. In patients with AMC and clubfoot, the condition is often bilateral and more rigid than idiopathic clubfoot and has a much higher rate of recurrence [4-7]. The extent of treatment may depend on the patient's ability to walk, and any remaining deformity may be acceptable depending on the patient's walking ability and the family's goals. The aim of treating clubfoot in arthrogryposis multiplex congenital (AMC) patients is to achieve a foot that can be placed flat on the ground and used with a

brace for walking. In some cases, there may be a temptation to perform surgery earlier than necessary due to the challenge of achieving and maintaining a successful correction [7]. Past literature has recommended early aggressive surgical intervention for non-idiopathic clubfoot deformities due to concern regarding the high relapse rate [4-7]. However, research has demonstrated that non-idiopathic clubfoot has a high relapse rate regardless of initial treatment [8-10]. In cases where the initial intervention was surgical, additional procedures were often necessary. Each additional surgical intervention can lead to increased scar formation, stiffness, and contracture, which can make treatment after subsequent relapses more challenging. Furthermore, there is no consensus on defining failure versus acceptable relapsed deformity. There is a growing body of literature supporting the idea of delaying or avoiding surgery during years of maximal childhood growth and instead using serial Ponseti-style casting to maximize the chances of successful outcomes [4-7, 11]. Church et al. specify the age range of 4.0-6.9 years as a period of rapid growth and a target age range for continuing serial casting [11, 12]. To address relapses of clubfoot in Arthrogryposis Multiplex Congenita (AMC), casting and Achilles tenotomies can be used as necessary. This approach helps to postpone further surgical procedures, as invasive interventions may not be as effective in this group until patients reach skeletal maturity. The objective of the treatment is to achieve a pain-free, plantigrade foot. While some practitioners utilize this technique, there is currently no literature supporting the long-term outcomes of this method for this particular population. Therefore, the target of this study was to describe the mid-term outcomes of serial Ponseti-style management of clubfoot in AMC. Ethical clearance and written consent were taken from the respective authorities.

II. Objectives

- *General objective:* The objective of this research is to study the effectiveness of multiple serial casting in the management of clubfoot.
- *Specific objective:* This study aims to evaluate the efficacy of multiple serial casting in the management of clubfoot in AMC using Pirani scores

III. Methodology

In this retrospective case series, we identified 15 patients (8 male, 7 female) with AMC and clubfoot deformities who underwent multiple casting series from January 2021 to January 2023 at Dr. Sirajul Islam Medical College Hospital and IBN Sina Diagnostics pediatric population. Long-leg plaster casts were applied using a Ponseti-style technique. The series of cast applications were planned in reverse order from the surgery date to ensure all casting was completed on time. After surgery, patients were cast for a minimum of three weeks until braces were manufactured, or longer, depending on concurrent orthopedic procedures. Patients were fitted with ankle-foot orthoses (AFOs) or knee-ankle-foot orthoses (KAFOs) for walking during the day. Bracing was continued until skeletal maturity to prevent or slow progression. Pirani scores were measured by the senior author in pediatric orthopedics. "Pre-treatment" measurement refers to the foot appearance at the initial presentation, "pre-series" measurements were those done just before starting a cast series, and "post-series" measurements were the first scores recorded after the last cast of a series was removed. Ordinal composite values for Pirani scores were calculated for initial deformity and the final result after each series. Statistical analysis was performed using repeated measures analysis of variance (ANOVA) with Greenhouse-Geisser correction where appropriate and post-hoc Bonferroni correction for multiple comparisons. Linear regression was used to examine the effects of casting days and the number of casts on improvement in Pirani scores.

- *Inclusion criteria:* Patients who were less than 5 years of age and guardians who provided written informed consent and understood the details of the study were included.
- *Exclusion criteria*: Patients who had incomplete measurements or treatment were excluded from the study.

Statistical analysis was performed using SPSS Statistics Version 25.0 (IBM Corp, Armonk, NY). The ethical review committee of Holy Family Red Crescent Medical College Hospital has approved the study. A well-informed written consent paper was signed by the parents.

IV. Result

A total of 15 treated clubfeet (8 male patients and 7 female patients) met inclusion criteria and were retrospectively reviewed. In Table-1, the initial Pirani score was 4.8±1.09 in series 1 which became 3.9±1.07 in series 3. The range was found to be (-0.5-3.5) from series 1 to series 3. The mean number of cast was 7.1 which took 61 days. The mean Pirani score improved by 1.50±1.06 from the start of the second series (4.23±1.03) to post-treatment (2.72±0.916) ($p<0.001$). The mean Pirani score improved by 1.05±1.00 from the start of the third series (3.87±1.07) to post-treatment (2.82±1.02) ($p<0.001$). There was also a significant difference between the pre-treatment Pirani scores before the first series and the post-treatment scores at the conclusion of the third series (4.80±1.54 vs. 2.82±1.02, $p<0.001$) [Table-2]. There was less improvement in the second and third series compared to the first (first to second $p=0.001$; first to third $p<0.001$) [Table-3]. Figure-1 shows the AFO used for

training after treatment. Figure-2 illustrates the Pirani score chart and figure-3 shows the outcome of multiple casting series according to the Pirani score.

Figure-1: AFO used after treatment

Figure-2: Pirani score chart

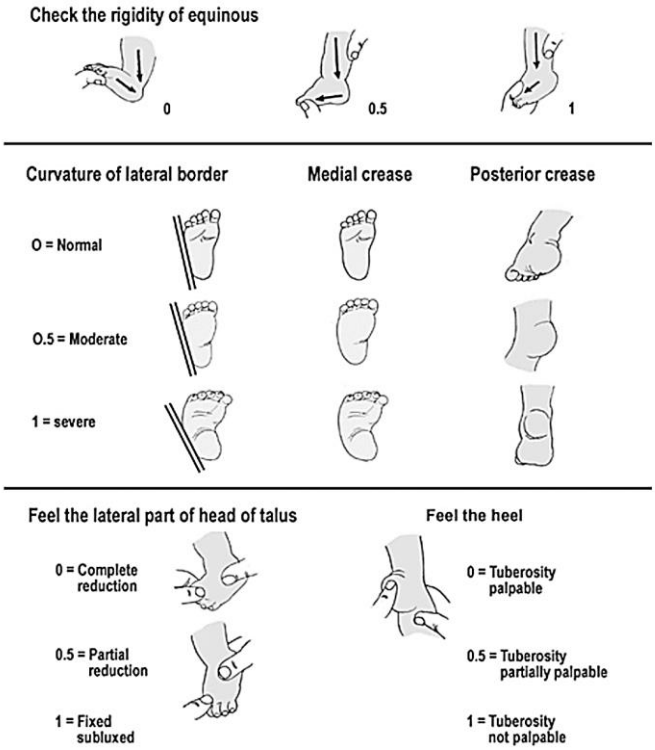

Figure-3: Pirani score for casting series

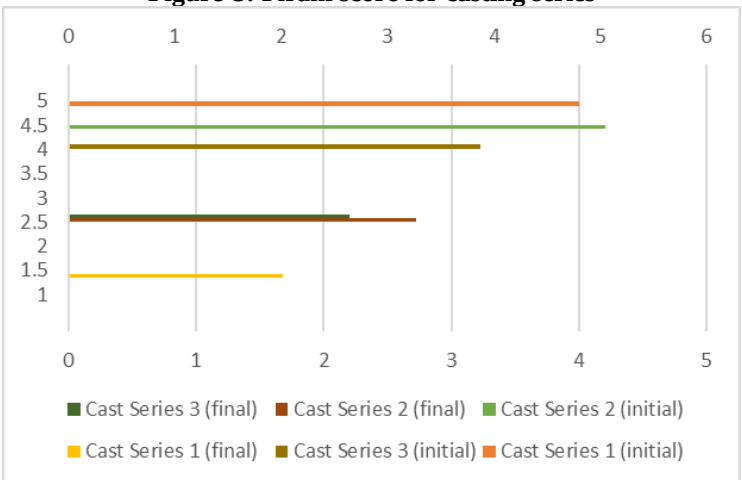

Table-1: Pirani score, number of casts, and casting days across multiple clubfoot casting series in AMC

Pirani score	Series 1			Series 2			Series 3		
	Mean	Range	SD	Mean	Range	SD	Mean	Range	SD
Initial Pirani score	4.8	(2.5-6)	1.09	4.2	(2.4-6)	1.03	3.9	(2-6)	1.07
Final Pirani score	1.7	(0-5)	1.49	2.7	(1-4.5)	0.92	2.8	(0.5-5)	1.02
Change in Pirani score	3.1	(-0.5-6)	1.69	1.5	(-0.5-4)	1.06	1.1	(-0.5-3.5)	1.00
Number of casts	7.1	(3-17)	3.82	6.7	(3-14)	2.98	5.1	(2-8)	1.68
Number of days	61.4	(14-133)	30.49	49.2	(14-150)	39.57	34.9	(7-70)	18.13

Table-2: Pre-treatment and post-treatment Pirani scores differences for each series

	Pirani score difference	SD	95% CI lower	95% CI upper	P-value
Series 1 pre-treatment vs. series 1 post-treatment	3.12	1.69	2.49	3.75	<0.001
Series 2 pre-treatment vs. series 2 post-treatment	1.50	1.06	1.10	1.89	<0.001
Series 3 pre-treatment vs. series 3 post-treatment	1.05	1.00	0.68	1.42	<0.001
Series 1 pre-treatment vs. series 3 post-treatment	1.98	1.30	1.50	2.47	<0.001

Table-3: Pairwise comparisons of change in Pirani score for each series

		Series Difference between change in Pirani scores	SD	95% CI lower	95% CI upper	P-value*
1	2	1.620*	0.385	0.627	2.613	0.001
	3	2.067*	0.370	1.111	3.022	0.000
2	1	-1.620*	0.385	-2.613	-0.627	0.001
	3	0.447	0.211	-0.099	0.992	0.137
3	1	-2.067*	0.370	-3.022	-1.111	0.000
	2	-0.447	0.211	-0.992	0.099	0.137

V. Discussion

Approximately 25% of all clubfoot deformities are non-idiopathic [14, 15]. One study found that a cohort of 357 clubfoot patients included 24% with non-idiopathic etiologies, with AMC and myelomeningocele being the most common underlying diagnoses [14]. The high percentage of non-idiopathic clubfoot patients shows the need to establish personalized treatment guidelines. Just as the treatment of idiopathic clubfoot has evolved, severe deformities in AMC were historically treated with surgical correction, while casting was only used to "buy time." [16]. Talectomy has even been recommended as a primary procedure for severe clubfoot in AMC in some of the research [17, 18]. Long-term follow-up studies have shown that people who have undergone primary talectomies are generally dissatisfied with the results [19]. Others have suggested early posterior-medial release, but subsequent studies have shown high relapse rates and low long-term success [8, 16, 20, 21]. Authors have recently reported success in avoiding surgical intervention by serially casting clubfeet in AMC [3, 5-7].

The use of serial Ponseti casting in treating clubfoot in AMC has been successful in improving foot morphology, allowing for comfortable bracing and stable weight-bearing. Pirani scores showed significant improvement at follow-up compared to pre-treatment; however, the degree of change decreased with each successive casting series. This trend is expected to continue with further casting series. Although the ability to correct clubfoot deformity decreased over time, it is interesting to note that the benefits of Ponseti casting can be successfully applied to older children. This is demonstrated by the fact that 16 out of 17 subjects were ambulatory at their most recent follow-up and had an average age of 6.04 years. One subject required further surgical intervention: a posterior soft tissue ankle release. Ponseti casting management for non-idiopathic clubfoot has been shown to approach a 90% success rate in some series, which is consistent with our findings [6, 14, 22].

The technique described by van Bosse et al. involves an optional initial Achilles tenotomy in addition to Ponseti casting, and has shown a 90% success rate [7]. However, other studies have shown less favourable long-term outcomes despite initially high success rates. These studies have reported a 45-50% recurrence rate, leading to surgical intervention in 20-25% of cases [4, 5, 23].

A study by Kowalczyk et al. compared early soft tissue release to Ponseti style management and found that while most subjects eventually required a soft tissue release, there were fewer complications when Ponseti casting was attempted first. This suggests that when initial intervention fails, early Ponseti management efforts may still lead to improved outcomes. Several studies have reported high success rates with the use of Ponseti casting in treating idiopathic clubfoot recurrence [24].

The results of this study may not directly apply to the AMC population, but they do provide a rationale for using Ponseti casting in cases of non-idiopathic clubfoot recurrence. Few studies have aimed to evaluate patient-reported outcomes after multiple Ponseti treatments for non-idiopathic clubfoot. Church et al. conducted a study comparing pediatric outcomes scores and gait analysis of clubfeet in AMC to those in cases of idiopathic clubfeet following Ponseti management. The study found that subjects with AMC generally had worse outcomes compared to those with idiopathic clubfeet [11].

Unfortunately, the outcomes reported by patients' scores were unavailable for all subjects included in this study. The current study had several limitations. One issue was the large standard deviation in the number of days included in each cast series. This was likely due to the nature of the institution, where many patients travelled long distances for treatment, leading to a casting schedule that was either compressed or stretched out for the convenience of the family. Additionally, the retrospective study series was small and underpowered to detect the effects of cast number and casting duration on each series. In the third series, there was a detectable effect of casting days, but it was not very strong. Since this series showed the least improvement, it's uncertain if there's a clinical correlation to this finding. Conducting further research with a larger group of people would help to confirm the associations found in this study and provide better guidance for treating patients with clubfoot in AMC.

The average age of the group at the start of the first series was earlier years of the study patients. Some patients started their initial series of Ponseti cast treatment after infancy, possibly due to referral patterns, the need for long-distance travel to the institution, or delays in diagnosis. According to some studies, patients with idiopathic clubfeet who received treatment between one and three months of age had lower relapse rates and better clinical outcomes compared to older age groups [25].

VI. Limitations

Some patients may have received treatment at other institutions before coming to the study institution. The study suggests that patients with arthrogryposis multiplex congenita (AMC) and clubfoot who underwent three or more rounds of serial casting likely represent the most challenging cases. The study did not include patient-reported outcome measures, so it's unclear how casting affected their functional capacity or quality of life. Despite these limitations, the data from the study supports the use of serial casting in patients with AMC and clubfoot. However, the authors suggest that a larger multi-center study is needed to determine if a specific subset of AMC patients may benefit from this type of treatment and if success rates vary with different syndromic diagnoses in this diverse population. Additionally, longer follow-up is necessary to determine the lasting success of the treatment.

VII. Conclusion

Clubfoot in arthrogryposis multiplex congenita (AMC) has a high likelihood of recurring. The Ponseti technique can be used to repeatedly improve recurrent deformities in this population, as measured by the Pirani score. Early treatment leads to better outcomes, although the ability to bring about change diminishes over time. Future studies may explore the concept of accepting residual deformity by comparing a group of patients who undergo dramatic interventions using patient satisfaction outcome measures.

Funding
Self-funded research.

Conflicts of interest: N/A

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