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ORIGINAL ARTICLE

Predictors of Success in Ponseti Casting, A Single Center Cross-sectional Study

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Abstract

Background: Congenital talipes equinovarus (CTEV), also known as club foot deformity, is a common deformity with an estimated incidence of around 1 in 1000 live births. The deformity consists of four components: Ankle equinus, hindfoot varus, forefoot adductus, and midfoot cavus. Various approaches exist to correct CTEV, with most authorities advocating a conservative closed reduction approach with serial casting.

Objective: Pirani scoring has been shown to correlate with the severity of CTEV and has been verified to track response to casting treatment. This study aimed to identify the relationship between initial presenting Pirani scores and other factors on the final Pirani score and the need for surgical intervention for patients managed with Ponseti casting in a single-center observational study design.

Methods: A total of 24 patients were followed across the duration of the study and serially assessed with the Pirani scoring system. The scores were charted and studied to determine patterns that predict the success of Ponseti casting in those children.

Results: The data showed that tenotomy is most likely corrective of the deformity in those with higher PS. While the age of diagnosis and age of casting were not significantly impactful, careful follow-up and discussion of goals with the family members are essential for expected outcomes, especially for those hesitant to undergo surgical interventions and those with higher Pirani Scores.

Keywords: Clubfoot; Humans; Child; Congenital equinovarus; Foot Deformities, Acquired; Foot Deformities

Introduction

Congenital talipes equinovarus (CTEV), also known as club foot deformity, is a common deformity with an estimated incidence of around 1 in 1000 live births.[1,2] The deformity consists of four components: Ankle equinus, hindfoot varus, forefoot adducts, and midfoot cavus.[3,4] Various approaches exist to correct CTEV, with most authorities advocating a

conservative closed reduction approach with serial casting.[3]

While most cases respond favorably to a conservative approach, severe cases of CTEV remain challenging and often require operative intervention to address various issues ranging from outright relapses to partial correction of the deformity in various degrees.[5,6] While various studies have confirmed the success of the Ponseti method for correction of CTEV and its relative superiority in comparison to other methods of corrections, i.e. (Kite's method, French physical therapy method),[7] clubfoot relapses have been reported following correction of CTEV with the Ponseti method nonetheless.[8–10]

Various factors have been identified and documented to increase the risk of recurrence of CTEV. The factors can be attributed to those pertaining to the patient, the physician, or the caregiver.[11,12] Identifying those factors and careful attention to addressing them early in the process of treatment of a patient with CTEV lowers the risk of needing various interventions and revision treatment modalities such as more casts, repeated tenotomy, revision surgical intervention that would lead to long-term reduction of the function of the patient.[12] While some variables affecting the success of therapy are non-modifiable, such as high grade of the deformity, ethnicity, and parental education level, arguably the most critical factor in the success of therapy is compliance with follow-up and physiotherapy with practicing stretching exercises can be detected early on and therapy modified to place particular emphasis on it. Thus, a physician managing CTEV cases must be familiar with factors that may complicate the course of the therapy. Identifying them early would lead to adopting a more cautious approach and possibly improve long-term outcomes.

Providing parents with valuable information about the likely course and outcome of treatment with Ponseti casting serves to increase the compliance of the parents with the treatment course and to set appropriate expectations of improvement depending on variables available on the initiation of treatment.

Pirani scoring has been shown to correlate with the severity of CTEV and has been verified to track response to casting treatment.[13,14] This study aims to identify the relationship between initial presenting Pirani scores and other factors on the final Pirani score and the need for surgical intervention for patients managed with Ponseti casting in a single center prospective observational study design.

Methods

The medical records of patients who underwent serial casting with the Ponseti method in Salmaniya Medical Complex, Bahrain, were collected over 3 years from 2020 to 2023 and evaluated with serial Pirani scoring by a consultant pediatric orthopedist and a specialist pediatric orthopedist with a unified scoring method agreed on before data collection to minimize variations in scoring. The sample size was calculated to be 28, and the study aimed to collect around 30 cases. Sample size calculation was done by an independent statistician and was determined according to the following formula:

$$n = \frac{z^2 \times p \times (1-p)}{e^2}$$

Where z=1.645 is the standard normal value corresponding to a 90% confidence interval, p equals 0.001 is the proportion of patients born with congenital talipes (CTEV),[15] and the margin of error and is assumed to be 0.01. Therefore, the sample size is

$$n = \frac{1.645^2 \times 0.001 \times (1-0.001)}{0.01^2} = 2$$

The inclusion and exclusion criteria were set as follows: Patients must have CTEV and be managed with serial Ponseti casting, and cases must have minimum follow-up and scoring that includes all casting weeks and pre-tenotomy follow-up. Patients with secondary CTEV and those with loss to follow-up with incomplete scoring records throughout the weeks were excluded from the study.

The patient's information was recorded on each casting visit. The data included the age, the duration of treatment, age at initiation of treatment, gender, contralateral limb involvement, demographics, and surgical interventions in the form of Achilles tendon tenotomy, along with a score of the cast done after tenotomy. All patients were assessed by a consultant pediatric orthopedic surgeon and a

specialist pediatric orthopedic surgeon who agreed on a unified scoring method based on the Pirani scoring system.

A total number of 32 cases were collected, and 8 cases were excluded. The sample size for the study was 24 patients after exclusions. Details on excluded cases are outlined in the limitations section.

SPSS 26 was used for data entry and analysis. Frequencies and percentages were computed for the categorical variables, while mean, standard deviation (SD), median, and interquartile range (IQR) were computed for the quantitative variables. Mann-Whitney test was used to determine whether there is a significant difference in means between two independent groups. In contrast, the Kruskal-Wallis test was used to determine whether there is a significant difference in means between more than two independent groups. Fisher's exact test was used in the 2 × 2 contingency table to test whether there is a significant relationship between two categorical variables if at least one of the expected values is less than 5. Spearman's Correlation Coefficient was used to measure the correlation between two quantitative variables. A p-value of less than 0.05 was statistically considered significant in all statistical tests.

Results

Data Description

This study included a total of 24 patients, of which thirteen patients had bilateral CTEV for a total number of affected lower limbs of 37. Only four patients had left CTEV, and seven had right CTEV. Eighteen of the patients were diagnosed with CTEV at birth, while six patients had delayed diagnoses, which we considered to be any diagnosis after the first day of birth. Casting was performed in less than seven days for eleven (45.8%) patients, while thirteen patients (54.2%) were cast after seven days. Of the 24 patients, seven of them (29.2%) were syndromic, while seventeen (70.8%) had no associated syndromes or conditions. Sixteen patients required tenotomy (66.7%), while eight patients did not require tenotomy upon completing the casting regimen (33.3%). Pirani Scores were measured at 1 week, 2 weeks, 4 weeks, and 5 weeks pre and post-tenotomy during casting (Tables 1 and 2).

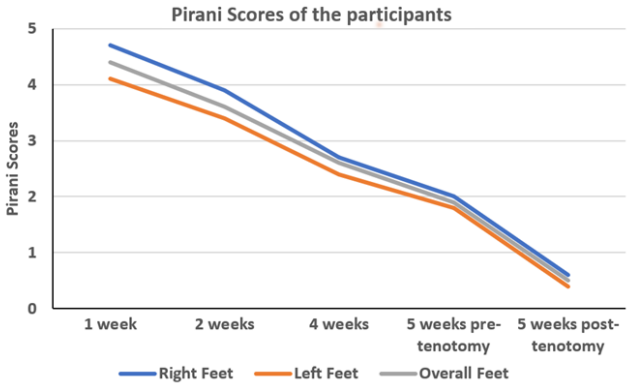

Figure 1: Pirani Scores over time

The mean Pirani Score (PS) at one week was 4.4 with a standard deviation (SD) of 1.6 for overall limbs, with a slightly higher score for right limbs of 4.7 (SD=2.3) in comparison to only left limbs of 4.1 (SD=2.7). This trend held for the duration of follow-up and even after surgical intervention, with PS at 5 weeks after tenotomy measuring at 0.5 (SD=0.8) for overall limbs and right 0.6 (SD=0.8) and left 0.4 (SD=0.4) (Figure 1).

Table 1: Frequency and percentage distribution of demographic characteristics, Laterality, Syndromic, and Tenotomy of the participants (Total = 24)

Variables	n (%)
Age On diagnosis[1]	
1 day	18 (75)
>1 day	6 (25)
Age on casting[2]	
<7 days	11 (45.8)
≥7 days	13 (54.2)
Number of siblings[3]	
None	15 (62.5)
≥1	9 (37.5)
Educational level	
Secondary or below	8 (33.3)
University or above	11 (45.8)
Unknown	5 (20.8)
Laterality	
Left	4 (16.7)
Right	7 (29.2)
Bilateral	13 (54.2)
Syndromic	
Yes	7 (29.2)
No	17 (70.8)
Tenotomy	
Yes	16 (66.7)
No	8 (33.3)

Median (IQR) = 1 (0.5); 2. Median (IQR) = 7 (5); 3. Median (IQR) = 0 (1).

Table 2: Pirani Scores of the participants (Total = 24)

Pirani Scores	Mean ± SD		
	Right	Left	Overall
Pirani Score (1wk)	4.7 ± 2.3	4.1 ± 2.7	4.4 ± 1.6
Pirani Score (2wk)	3.9 ± 2.0	3.4 ± 2.3	3.6 ± 1.5
Pirani Score (4wk)	2.7 ± 1.8	2.4 ± 1.9	2.6 ± 1.5
Pirani Score (5wk) pre-tenotomy	2.0 ± 1.6	1.8 ± 1.6	1.9 ± 1.4
Pirani Score (5wk) post-tenotomy	0.6 ± 0.8	0.4 ± 0.4	0.5 ± 0.8

Mean ± SD was computed out of 6.

Risk Factors

Several risk factors were collected for analysis, encompassing the following: Age on diagnosis (AOD), Age on casting (AOC), Number of Siblings, education level of primary caretaker, and Associated Syndromes. On statistical analysis using Mann-Whitney testing, the following P values were calculated: AOD (P= 0.362), AOC (P= 0.679), Number of Siblings (P= 1), Educational level (P= 0.262), Associated Syndromes (P= 0.352). Thus, the data suggested that none of the aforementioned risk factors were significantly relevant. The same risk factors were also analyzed using Fisher's Exact test for a relationship with tenotomy or surgical intervention rates, and no significance was noted. Of the aforementioned risk factors, PS at 5 weeks pre-tenotomy was statistically significant, with higher scores of 2.5 ± 1.2 in the group casted before 7 days when compared to after 7 days of 1.1 ± 1.4 (P= 0.025) and higher Pirani scores at 5 weeks pre-tenotomy of 2.5 ± 1.4 in patients with University Graduates as compared to lower levels at 1.3 ± 1.0 (P= 0.038). This difference between the two groups was no longer significant post-tenotomy. In the post-tenotomy PS at 5 weeks, AOD was significant in that a delay of more than one day led to higher average scores of 0.8 (P=0.040) (Tables 3 and 4).

Correlation rates

Spearman's correlation testing was used to compare primary scores PS at 1 week and PS at 5 weeks both before and after tenotomy. A positive correlation coefficient of 0.575 (P= 0.003) was measured for right limbs, pre-tenotomy, while the correlation was insignificant post-tenotomy. A similar finding was

Table 3: Relationship between risk factors and Pirani Score (5wk) pre and post-tenotomy

Risk factors	PS (5wk) pre-tenotomy	PS (5wk) post-tenotomy
	Mean ± SD	Mean ± SD
Age on diagnosis		
1 day	2.2 ± 1.3	0.4 ± 0.8
>1 day	1.1 ± 1.4	0.8 ± 0.1
P-value	0.093	0.040
Age on casting		
<7 days	2.5 ± 1.2	0.5 ± 0.3
≥7 days	1.4 ± 1.4	0.6 ± 1.1
P-value	0.025	0.407
Number of siblings		
None	1.7 ± 1.3	0.4 ± 0.4
≥1	2.2 ± 1.6	0.7 ± 1.2
P-value	0.471	0.909
Educational level		
Secondary or below	1.3 ± 1.0	0.3 ± 0.4
University or above	2.5 ± 1.4	0.6 ± 0.9
P-value	0.038	0.559
Syndromic		
Yes	2.8 ± 1.6	0.6 ± 0.3
No	1.5 ± 1.1	0.5 ± 0.9
P-value	0.090	0.068
Laterality		
Left	1.2 ± 0.2	0.4 ± 0.5
Right	1.1 ± 0.8	0.7 ± 1.3
Bilateral	2.5 ± 1.5	0.5 ± 0.4
P-value[1]	0.063	0.753

P-values were computed by using the Mann-Whitney test; Mean ± SD was computed out of 6; 1. P-value was computed by using the Kruskal-Wallis test; PS = Pirani Score.

Table 4: Relationship between risk factors and Tenotomy

Risk factors	Tenotomy		P-value
	No	Yes	
	n (%)	n (%)	
Age on diagnosis			
1 day	13 (72.2)	5 (27.8)	0.362[a]
>1 day	3 (50)	3 (50)	
Age on casting			
<7 days	8 (72.7)	3 (27.3)	0.679[a]
≥7 days	8 (61.5)	5 (38.5)	
Number of siblings			
None	10 (66.7)	5 (33.3)	1..000[a]
≥1	6 (66.7)	3 (33.3)	
Educational level			
Secondary or below	5 (62.5)	3 (37.5)	0.262[a]
University or above	10 (90.9)	1 (9.1)	
Syndromic			
Yes	6 (85.7)	1 (14.3)	0.352[a]
No	10 (58.8)	7 (41.2)	
Laterality			
Left	4 (100)	0 (0)	Not computed
Right	5 (71.4)	2 (28.6)	
Bilateral	7 (53.8)	6 (46.2)	

a. P-values were computed by using Fisher's Exact test.

appreciated with left limbs, with a pre-tenotomy correlation of 0.760 (P=<0.001) and no significance in post-tenotomy correlation, and in both limbs, with a pre-tenotomy correlation of 0.528 (P=0.008) and no significance in post-tenotomy correlation. Suggesting a higher PS at 1 week was positively correlated with higher PS pre-tenotomy but not with Post-tenotomy PS (Tables 5,6, and 7).

Table 5: Spearman's correlation between right Pirani Score (1wk) and Pirani Score (5wk) pre and post-tenotomy

Pirani Scores	PS (1wk)	
	Correlation coefficient	P-value
PS (5wk) pre-tenotomy	0.575	0.003
PS (5wk) post-tenotomy	-0.104	0.748

PS = Pirani Score.

Table 6: Spearman's correlation between left Pirani Score (1wk) and Pirani Score (5wk) pre and post-tenotomy

Pirani Scores	PS (1wk)	
	Correlation coefficient	P-value
PS (5wk) pre-tenotomy	0.760	<0.001
PS (5wk) post-tenotomy	-0.448	0.167

PS = Pirani Score.

Table 7: Spearman's correlation between overall Pirani Score (1wk) and Pirani Score (5wk) pre and post-tenotomy

Pirani Scores	PS (1wk)	
	Correlation coefficient	P-value
PS (5wk) pre-tenotomy	0.528	0.008
PS (5wk) post-tenotomy	0.043	0.874

PS = Pirani Score.

Early Pirani Scores and Need for Tenotomy

The relationship between PS at one week and the need for tenotomy was calculated and revealed no significant relationship between scores and the need for tenotomy for right, left feet and overall (P=0.201, 0.572, 0.172).

Table 8: Relationship between Pirani Score (1wk) and Tenotomy

Pirani Scores	Tenotomy		P-value
	Yes	No	
	Mean ± SD	Mean ± SD	
Right Pirani Score (1wk)	4.3 ± 2.6	5.5 ± 1.1	0.201
Left Pirani Score (1wk)	4.0 ± 2.8	4.4 ± 2.7	0.572
Overall Pirani Score (1wk)	4.2 ± 1.5	4.9 ± 1.7	0.172

P-values were computed by using the Mann-Whitney test.

Discussion

Various studies have looked at risk factors for the success and failure of serial casting in the management of CTEV, most of which were conducted in different settings. To the best of the author's knowledge, this is the first study to look at the outcomes of Ponseti casting in the kingdom of Bahrain and the larger Arabian Gulf region. This study can serve as a reference for service providers when counseling parents of children with CTEV. It can further provide evidence to support using the Ponseti casting method in the local patient population.

In this single-center prospective study of patients with CTEV, a survey of the risk factors and associations was conducted and measured. Our data suggests that while early diagnosis is essential, a minor delay did not cause significant changes. The data indicates that a minor delay in casting the child after 7 days of diagnosis may result in lower pre-tenotomy scores (P=0.025). However, this difference is no longer noted post-tenotomy (P=0.407) (Table 3). The data also shows some effect of the age on diagnosis on Pirani scores post tenotomy with delays in diagnosis of more than one day resulting in higher Pirani scores post tenotomy (P=0.04). This suggests that while early diagnosis is essential in CTEV, early initiation of serial casting may not be as necessary, and a small gap in the initiation of casting may not result in adverse outcomes for patients. This is in keeping with the findings of a recent study that suggested that casting should probably be initiated after a brief delay of around a month or two, as they found that uncertainty was correlated with better outcomes.[16]

The study found no correlation between age on the cast and Pirani scores pre and post-tenotomy. This finding is in contention in the current literature, with recent articles viewing age as an indicator of the number of casts required to achieve optimal correction.[17]

Another significant result in the study was the positive correlation between PS at one week and PS at five weeks, pre-tenotomy in all limbs, and when measured in isolated cases (P=0.003) (Table 5). This suggests that higher PS early on would still result in higher PS at 5 weeks pre-tenotomy; this difference is no longer noted after tenotomy (P=0.748); this is in line with the previous finding of most predictors being of minor effect on the final score after tenotomy and suggests that the surgical tenotomy accounts for a significant amount of deformity correction. This is echoed in studies performed in other settings that found a correlation between the initial Pirani score and the number of casts required to achieve acceptable final deformity correction.[17] The data suggests that operative tenotomy compensates for any shortcomings in the management, whether due to risk factors related to the patient or those related to the technical considerations of the orthopedic provider; this is a finding that ought to be balanced by various reports in the literature of increased complication rates such as fibrosis, scarring and stiffness that is observed with surgical intervention.[18]

The data from this study reinforces pre-existing evidence of the efficacy and relative safety of Ponseti casting in managing CTEV. It also serves to add the growing evidence of the correlation between initial presenting score and final Pirani score that was alluded to in the work of Gupta et al. that described the relation between initial Pirani scores and number of castings required to achieve

satisfactory correction or satisfactory final Pirani score in other words.[17] The study also reinforces the findings of Vaishy et al. in the potential usefulness of delaying the initial cast for some time as that does seem to be correlated with better Pirani scores.[16] Finally, the study can be used to counsel anxious parents and to set expectations about the efficacy of casting and the possible need for tenotomy in more severe cases of CTEV. Parents can be reassured that a higher PS at 1 week does not necessarily correlate with the outcome after surgical tenotomy but is correlated with pre-tenotomy outcome.

Conclusion

While casting technique and follow-up are essential when treating congenital clubfoot, operative tenotomy remains the most corrective intervention for the deformity, especially in those with higher PS, as reflected in this study. While age of diagnosis and age of casting were not significantly impactful on final Pirani scores post-tenotomy, careful follow-up and discussion of goals with the family members are essential for expected outcomes, especially those hesitant to undergo surgical interventions and those with higher Pirani Scores. Parents and orthopedic providers can be reassured that minor delays in the initiation of casting may not result in adverse final Pirani scores post-tenotomy.

Ethical considerations and limitations

Ethical approval was obtained from the local research committee in Salmaniya Medical Complex; the authors did not receive any funding for publication or preparation of this study and have no conflict of interest to declare.

The small sample size limited the study due to the relatively low prevalence of the condition in the community. Statistical analysis considered the small sample size. Of the 32 patients in the study, 8 cases were excluded due to loss of follow-up, inability to continue casting due to various reasons, including relocation to the primary home country, death, or having more than one missing entry. A single missing value for one of the entries was filled to the best of the author's estimation post-operative tenotomy.

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