

Original Research Article

Assessing the factors that influence a recurrence of congenital talipes equinovarus in children treated with the Ponseti method

Rahul Kumar Goyal¹, Manoj Kumar Meena¹, Mohit Agarwal¹, Naveen Sathiyaseelan^{2*}

¹Department of Orthopaedics, ESIC Medical College and Hospital, Alwar, Rajasthan, India

²Department of Orthopaedics, Saveetha Medical College and Hospital, Chennai, Tamil Nadu, India

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*Correspondence:

Dr. Naveen Sathiyaseelan,

E-mail: spnaveen17@gmail.com

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ABSTRACT

Background: Congenital talipes equinovarus, also known as clubfoot, is one of the most common congenital malformations in pediatric orthopedics. With a checkered treatment history extending from Hippocrates' bandages to Kite's serial plaster cast application to surgical treatment, no single modality can claim to be a complete treatment.

Methods: In a combined prospective and retrospective study at the Department of Orthopaedics of St. Stephen's Hospital, 192 patients with 292 clubfeet who met the inclusion criteria and received ethics committee approval were included. The purpose of the research was to identify early recurrence characteristics, undertake the Pirani scoring, and identify additional clinical characteristics to evaluate recurrence predictors.

Results: In our study, out of 192 patients, 188 were successfully treated with the Ponseti casting technique combined with tendoachilles tenotomy; tenotomy was required in 177 patients (93%). Four patients were lost to follow-up. Nine out of sixty-one children in the six-month-to-one-year age group showed the maximum rate of recurrence (15%). The mean number of cast applications was 7.23. In all, 90% of children required casts ranging from four to eight in number. The syndromic association was seen in 10.4% of cases with club feet. Most common among them were neural tube defects (4.2%) and arthrogyryposis (3% of cases). Fourteen out of 154 (or 9%) of the non-operated idiopathic patients had lateral border recurrence during an average follow-up of two years. According to the Pirani score, recurrence was seen in eight out of twenty (or 40% of) syndromic patients and five out of fourteen (or 35% of) operated cases. In 37% of recurrences, dynamic supination was observed.

Conclusions: Even though Ponseti's approach to treating idiopathic clubfoot is widely regarded as the gold standard, recurrences are known to occur even in the best of circumstances. The most common cause of recurrence was poor brace compliance, with families citing a variety of reasons.

Keywords: Congenital talipes equinovarus, Clubfoot, Ponseti technique, Pirani score, Serial cast

INTRODUCTION

Congenital talipes equinovarus (CTEV), also known as clubfoot, is among the most common congenital deformities in pediatric orthopedics.¹ One in every one thousand live births is its reported incidence rate. Each year, there are around one lakh new instances of clubfoot reported worldwide.^{2,3}

The term CTEV is an acronym: C: Congenital, T: Talipes, meaning "pes" (foot) and "talus" (ankle), E: Equinus, meaning "horse" (foot in plantar flexion), V: Varus, meaning "inverted and adducted".

Equinus

The ankle joint is where this deformity commonly arises; the foot is fixed in plantar flexion.

Inversion

This deformity occurs primarily at the subtalar joint (i.e., the talocalcaneal joint).

Forefoot adduction

This deformity occurs at the mid-tarsal joint, mainly at the talo-navicular joint.

The frequent complications of the surgical procedures (soft tissue release), including recurrence, overcorrection of the deformity, stiffness, and pain, have fueled an interest in the nonsurgical management of this deformity.⁴

A plaster cast is used after each manipulation in the Ponseti technique of treatment to preserve the degree of correction and soften the ligaments.

As a result, the misaligned bones gradually shift back into place while maintaining congruency on their joint surfaces. After casting treatment for two months, the foot seems to have somewhat overcorrected. However, the foot becomes normal after some weeks in splints.²

Three weeks after the tenotomy, the cast is removed, and a foot abduction brace (Steenbeek's brace) is applied to prevent a recurrence. To keep the feet in dorsiflexion in unilateral situations, the brace is placed at 60–70 degrees of external rotation on the clubfoot side and 30–40 degrees on the normal side.³ For the first three months after the removal of the final cast, the brace should be worn continuously (twenty-three hours/day). Following that, the child must wear the brace for a total of 14–16 hours every 24 hours, including twelve hours at night and two to four hours during naps. This practice is followed up until the child is three to four years old.²

Recurrence rates following the Ponseti casting have been reported to be between 10% and 30%; nevertheless, many recurrent abnormalities may be effectively treated with repeat casting, whether or not Achilles tenotomy or anterior tibial tendon transfer are also used. According to several authors, the most important factor in preventing recurrent deformity is patient compliance with the postoperative brace regimen. The degree of deformity, the patient's age at the start of medication, or prior medication was not found to have a significant influence on the likelihood of recurrence.⁵

Maintaining compliance with orthosis wear may be challenging. Because failure to comply with brace wear is the main cause of deformity recurrence and failure of this treatment approach, every effort should be made to support families in this effort.⁶

A third of the feet have a partial recurrence of the clubfoot deformity, which is often caused by poor adherence to the abduction orthosis or a delay in starting therapy. Early recurrence of deformity (within the first year) can be effectively treated by repeatedly manipulating the affected

area and applying casts to stretch and correct any residual deformity. It can be difficult to cast older newborns, but abduction orthosis can correct and then maintain a surprising amount of residual deformity. Again, compliance with orthosis wear is mandatory for a successful outcome. When newborn deformities reoccur and do not improve with repeated casting, surgical intervention may be necessary. It is uncommon to need much posterior medial and lateral release.³

Anterior tibialis tendon medial overpull may cause a dynamic swing phase supination deformity to appear in elderly individuals. The navicular tendon's function is changed from a foot dorsiflexor to a foot supinator when the navicular is incompletely reduced onto the head of the talus. This may cause hind-foot varus to reoccur if left untreated. After four to six weeks of recurrent casting, the anterior tibialis is transferred to the third cuneiform in these individuals. The tendon should be left beneath the ankle's anterior retinaculum to avoid bowstringing. At the time of anterior tibialis transfer, it is crucial to check for any recurring equinus deformity that can call for Achilles tendon Z-lengthening.³

The effectiveness of Ponseti's approach to therapy is determined by the length of follow-up as well as the outcome measures used to assess outcomes. Most current studies report a high degree of success over the short term. Longer-term studies reveal that roughly one-third of feet have deformity recurrence.^{7,8}

The differences in long-term foot function between feet treated with this method and those treated through significant surgical release are not well understood. Primarily, no prospective randomized studies comparing these two groups with long-term follow-up exist. It is also important to understand that, with this approach, one-third of patients could eventually need tendon transfer, tendon lengthening, or selective release. As a result, it would be wrong to compare only those who avoided surgery with those who tried alternative non-operative measures but were unsuccessful and needed posterior medial and lateral release. Nevertheless, it is logical that feet treated with fewer surgical procedures will operate better over time with less pain and more strength, mobility, and functionality.³

Recurrences are known to occur after conservative and operative treatment of clubfoot.

METHODS

Study design

A combined prospective and retrospective research approach, including 192 patients with 292 clubfeet, was carried out in the Department of Orthopaedics at St. Stephen's Hospital. Participants were tracked for a total of twenty-eight months. Most of the cases were idiopathic. Others had syndromic associations such as

meningomyelocele, arthrogryposis, Streeter's dysplasia, syndactyly, and developmental dysplasia of the hip.

Methodology

Before achieving full correction of the deformity with the help of the Catterall-Pirani scoring system and other clinical features, such as degree of external rotation, degree of dorsiflexion, and dynamic supination, there was initially a weekly follow-up for all of the children.⁹

Inclusion criteria

All children with congenital talipes equinovarus illnesses were treated using the Ponseti method. After getting complete deformity treatment, all children returned for follow-up, and parents allowed their children to be a part of the study.

Exclusion criteria

Children with established recurrence, children receiving any other type of treatment apart from the Ponseti technique, those not willing to be part of a study, and those who had typical clubfeet were excluded from this study.

Statistics of the study

For the sample size calculation, we expected the early recurrence rate to be about $p=10\%$ at a 5% level of significance, and the precision sample size was calculated using the following formula: $(Z^2 \times p \times (1-p)/d^2)$.

RESULTS

Age and sex distribution

There were 192 patients included in this research, of whom 138 (71.9%) were males, and fifty-four (28.1%) were females. The largest group among these children, i.e., sixty-one (31.8%), belonged to the one-to-six-month age range, whereas the lowest group, twelve (6.25%), belonged to the age range of over five years.

Laterality

A total of 192 individuals had clubfeet, of whom one hundred (52%) had bilateral deformities and ninety-two had unilateral ones, with fifty-one (27%) having a right-side deformity and forty-one (21%) having a left-side deformity. Thus, a total of 292 clubfoot cases were investigated.

Recurrence

Nine out of sixty-one children in the six-month-to-one-year age group showed the maximum rate of recurrence (15%). After that, it was for the two-to-five-year-old age group, which was 12.5% (three out of twenty-four).

Number of cast applications

The mean number of cast applications was 7.23. In all 90% (171) of the children required casts, ranging from four to eight in number. Children made up the majority in first- and second-order distribution. The first order comprised 112 children, or 58% of the population, followed by the second order (sixty children) at 31% and the third order (sixteen children) at 8%, with the remaining (four children) having higher-level contributions, totaling only 1%.

Previous treatment

Ninety-two (47.9%) out of the 192 patients at our hospital were untreated when they arrived. Sixty-eight (35.4%) children had already undergone casting alone. Fourteen (7.3%) children had been treated surgically with soft tissue release and had scars at the posteromedial aspect of the leg and foot from a previous residual deformity. Eighteen children (9.4%) had undergone a tenotomy.

Associated with other congenital anomalies

Notably, a syndromic association was seen in 10.4% of the cases with clubfeet; the most common among them were neural tube defects (4.2%) and arthrogryposis (3%). Out of these patients, eight had meningomyelocele, three of them (37.5%) developed recurrences at follow-up visits, and three out of four children (75%) with arthrogryposis developed recurrence, which was specified in the form of equinus at the ankle. One child with Streeter's dysplasia out of three (33%) experienced recurrence in the form of deepening of the posterior crease and reappearance of the equinus deformity.

Assessment of the Pirani Score in idiopathic, non-operated, syndromic, and operated groups

Lateral border

A total of 154 patients in the idiopathic, non-operated group had an average recurrence rate of 8%, compared to 29% for the operated group and 25% for the syndromic group during a mean two-year follow-up period. Of the total 192 patients, twenty belonged to the syndromic group, twenty to the previously operated group, and four were lost to follow-up (Table 1).

Medial crease

Because a medial crease is a relatively fixed variable after initial deformity repair, it was not altered in patients with deformity recurrence (Table 2).

Talar head coverage

With the Ponseti casting technique, 146 out of 154 idiopathic children had a score of zero. The frequency of occurrence among them was originally relatively high.

In further follow-up visits, it was shown to be higher. In terms of talar head covering, syndromic and operated patients were well controlled following rectification (Table 3).

Table 1: Assessment of lateral border (Pirani score).

Assessment of lateral border (n=192)	Operated group			Idiopathic non-operated			Syndromic group		
	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %
At presentation	0	14	-	41	113	-	5	15	-
At brace prescription	12	2	-	148	6	-	20	0	-
3 months post-correction	8	3	25	141	13	5	16	4	20
6 months post-correction	6	4	33	124	30	16	14	7	28
1-year post-correction	8	4	33	132	22	11	14	6	33
2-year post-correction	9	3	25	142	12	4	15	4	20

The recurrence rate in the idiopathic non-operated group was significantly lower than that in the operated group (8 percent vs. 29 percent, p=0.007) and the syndromic group (8 percent vs. 25 percent, p=0.002), whereas it was comparable between the operated group and the syndromic group (29 percent vs. 25 percent, p=0.697).

Table 2: Assessment of medial crease (Pirani score).

Assessment medial crease (n=192)	Operated group			Syndromic group			Idiopathic non-operated		
	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %
At presentation	5	9	-	2	18	--	7	147	-
At brace prescription	14	0	-	16	4	--	139	15	-
3 months post-correction	13	1	-	15	5	--	141	13	-
6 months post-correction	14	0	-	14	6	--	136	18	-
1-year post-correction	14	0	-	15	5	--	138	16	-
2-year post-correction	14	0	-	15	5	-	136	18	-

Since the medial crease is a relatively constant variable after the initial repair of the deformity, it was not changed in situations where the deformity recurred.

Table 3: Assessment of talar head coverage (Pirani score).

Assessment talar head coverage (n=192)	Operated group			Syndromic group			Idiopathic non-operated		
	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %
At presentation	6	8	-	2	18	--	0	154	-
At brace prescription	12	2	-	14	6	--	146	8	-
3 months post-correction	13	1	-	15	5	--	152	2	-
6 months post-correction	12	2	-	12	8	--	150	4	-
1-year post-correction	11	3	-	13	7	--	141	13	-
2-year post-correction	11	3	-	12	8	-	143	11	-

With the Ponseti casting method, 146 out of 154 idiopathic children had a score of 0. They originally had quite a bit of recurrence. In further follow-up visits, it was shown to be higher in terms of talar head covering, and syndromic and operated patients were well-controlled following rectification.

Table 4: Assessment of rigid equinus (Pirani score).

Assessment rigid equines (n=192)	Operated group			Syndromic group			Idiopathic non-operated		
	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %	Score 0	Score 0.5/1	Recurrence %
At presentation	0	14	-	2	18	-	0	154	-
At brace prescription	11	3	-	18	2	-	141	13	-
3 months post-correction	9	5	19	6	14	33	129	25	9
6 months post-correction	8	6	31	5	15	28	134	20	5
1 year post-correction	7	7	43	3	17	17	132	22	13
2 year post-correction	8	6	31	4	16	22	126	28	11

None of the idiopathic children who were not operated on had a score of 0. 141 out of 154 idiopathic children had a score of 0 after finishing their therapy. The observed average recurrence rate was 9.5%. This was 25% higher in the syndromic group and 31% higher in the operated group.

Rigid equinus

None of the idiopathic children who were not operated on had a score of zero. One hundred forty-one out of 154 idiopathic children had a score of zero after finishing their therapy. The observed average recurrence rate was 9.5%, higher in the syndromic group at 25% and 31% in the operated group (Table 4).

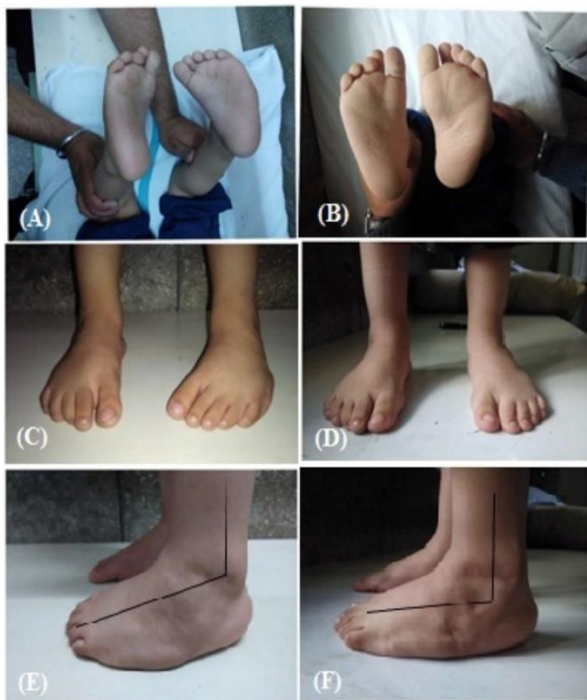


Figure 1: Previously fully corrected clubfeet in a 2-year-old child came with deformity managed with restart casting in maximum correction. A) curved lateral border, B) correction achieved after redo casting, C) forefoot adduction, D) correction achieved after strict brace protocol, E) early equinus and F) equinus corrected after casting.



Figure 2: A 5-year-old child with persistent dynamic supination, treated with tibialis anterior tendon transfer. A) persistent dynamic supination, B) visible scar mark of tendon transfer, C) reduced supination.



Figure 3: A 3-year-old child with dynamic supination was treated with tibialis anterior tendon transfer. A) curved lateral border, B) dynamic supination, C) heel varus, D) after correction.

DISCUSSION

To prevent the deformity from recurring, patients must be monitored over an extended period and kept in a brace. Before the 1980s, the recurrence rate in Ponseti's series was up to 48% in the year after deformity repair.¹⁰ Only when the brace regimen was adjusted to allow for use for three to five years did these numbers start to improve.

After the first year of life, the foot's rate of development slows down, and after five years, it significantly slows down. Relapses happen more slowly in older babies and more quickly in preterm newborns. Relapses are less frequent and less severe in children with loose ligaments and moderate clubfoot, where there is little to no fibrosis. They take place because the causes of the deformity are still present. Relapses are uncommon beyond the age of four.¹¹

In our study, a total of 192 patients (138 males and 54 females) were included based on predefined inclusion criteria after an explanation of the research protocol to the parents and obtaining written consent.

Out of these, 188 patients were treated successfully with the Ponseti casting technique combined with a tendoachilles tenotomy. Tenotomies were required in 177 patients (93%). Four patients were lost to follow-up.

Age and sex distribution

The total number of male children was 138, whereas females numbered 54, for a ratio of 2.5:1, which was comparable with the study conducted by Siapkara et al.¹² There could be a gender bias with parents bringing in more male children than females. This could be due to a difference in case prevalence between male and female children; however, this is statistically insignificant.

There are various factors responsible for parents bringing in children at a later age. A large number of children are referral cases from another hospital where they have been treated partially or incompletely in the form of casting or surgery.

Laterality of clubfoot

Out of the 192 individuals included in this research, one hundred were bilateral (52%), and ninety-two were unilateral (48%), with fifty-one having a deformity on the right side and forty-one having it on the left. As the studies conducted by Gray et al, Pavoneet al, and Ganesan et al.^{1,13,14} suggest, half of the children with clubfoot are known to have a bilateral deformity.

Birth order distribution

It was also noted that 58% of the children were of the first order of birth, while 31% were of the second order. Sixteen were of the third order. Three children were the fourth-

oldest siblings in their family, and one child was of the fifth order. The recurrence did not show significance in terms of the order of birth. There is no such correlation in the literature review either.

Assessment of the Pirani score

The Pirani score is validated only for younger children. However, for the sake of convenience and uniformity of evaluation, all children were evaluated using the six-point Pirani scoring.² Most of the feet-188 out of 192-were corrected with the Ponseti casting technique combined with tendoachilles tenotomy. Four patients were lost to follow-up during the study's average two-year duration.

Notably, lateral border and rigid equinus were the two most important components of the Pirani score, which significantly altered the recurrence of clubfoot deformity. The medial crease, Talar head coverage, posterior crease, and empty heel were not altered significantly in recurrent cases.

Among the 192 children, 158 were of idiopathic non-operated origin. Among them, 153 children had a score of zero after the Ponseti casting at the time of starting brace wear. This comparison is well in line with the published study conducted by Sachdeva et al.¹⁵ The number of previously treated cases was high in this series because the center gets a high number of referrals even though full correction was achieved in a good number of cases. The empty heel was found unreliable as the changes in the score were statistically insignificant.

On evaluating the rigid equines, a Pirani score of zero could be achieved in all cases of idiopathic non-operated clubfeet. However, compared to idiopathic cases, satisfactory dorsiflexion could be achieved with a greater number of casts in syndromic cases such as arthrogryposis and previously operated cases, whereas difficulty in brace wear and relapse were higher in these children.

Fourteen out of 154 (or 9% of) patients with idiopathic non-operated lesions had lateral border recurrence during an average follow-up of two years. Recurrence occurred in eight of twenty (40%) syndromic patients and five of fourteen (35%) operated cases. The earliest recurrence was seen six months after the brace prescription. The earliest recurrence noticed was in equines, followed by forefoot adduction in the form of a curved lateral border.

The most important factor that coincided with the recurrence of clubfoot was the patient's noncompliance with the brace protocol. Older children learn to undo the laces and take out the braces. Parents coming from faraway places develop a large interval period between two follow-ups because they become less motivated to follow the brace-wear protocol. Some children developed soreness at the margins of the brace, which was corrected using increased padding at the margins.

Signs of early relapse

Ankle dorsiflexion decreased to fewer than ten degrees, early heel rise (child moving away from the examiner), swing phase dynamic supination (child facing the examiner), and supple feet with a slight tendency of dynamic supination were in the early phases of recurrence of deformity. They have managed to ensure adequate and proper brace wear protocols. Steenbeek's brace was carefully examined in those children for bar length, angle of abduction, and dorsiflexion.

Automatic slippage of Steenbeek's shoes was one of the most common complaints made by parents as a reason for inadequate brace wear. Incomplete correction of deformity can lead to difficulty in brace wear.

Children having a significant recurrence in the form of decreased angles of dorsiflexion and external rotation with reversal of the Pirani score were put on the Ponseti maneuver and casting protocol again with frequent follow-up. We had to restart casting in twenty-one patients, and twelve needed a tenotomy redo after a few casts. Once the foot was corrected, the child was again put on the bracing protocol.

A tibialis anterior tendon transfer was done in two patients with persistent dynamic supination beyond the age of three years. After that, a child is kept in a short leg cast for six weeks. Significant improvement in gait was noticed in both children.

Limitations of the study included children who presented with recurrence and were observed to have less foot abduction. Before real equines occurred, limited external rotation was seen in several children. This was an observation that needs to be investigated and could be used as an early marker of recurrence. This aspect should be studied in detail because there is no literature to support this observation.

CONCLUSION

Even under ideal conditions, recurrence is known to occur, even though the Ponseti method is widely recognized as the gold standard for treating idiopathic clubfoot. The recurrence rate was much lower in the idiopathic non-operated group than in the operated group and the syndromic group, while it was equal in the syndromic group. The most frequent cause of recurrence was inadequate bracing compliance, with families providing a variety of explanations for this.

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