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Management of Idiopathic Congenital Talipes Equinovarus by Ponsetis Technique-Outcome and Analysis

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Abstract

Congenital talipes equinovarus (CTEV), also known as clubfoot, is a common idiopathic deformity of foot presenting in neonates, characterized by cavus, adduction of forefoot, varus and equinus deformity of foot. Management can be conservative (stretching or splinting of foot) or surgical. If untreated, it causes permanent deformity and impairment of foot. To evaluate the effectiveness of Ponseti's method for treating idiopathic congenital talipes equinovarus. This prospective, observational study includes infants up to 12 months of age. Severity of CTEV was assessed using Pirani scoring system. Ponseti method of serial manipulation and casting was done in these patients. Clinical results were evaluated one year post follow-up period. In our study average number of casts required by Ponseti technique for full correction of CTEV was 6.5. Out of 85 feet, per cutaneous Achilles tenotomy was required in 73 cases (85.88%) to achieve full correction. There is significant improvement in Pirani score from an average of 4.8 on initial presentation to 0.005 after complete casting. We conclude that, Ponseti's technique of CTEV casting having >90% success rate in avoiding comprehensive surgical release, being affordable, safe and effective procedure, providing painless, plantigrade, cosmetically acceptable foot with better functional outcome with minimal complications. Success rate is higher among infants who were compliant with the treatment protocol.

INTRODUCTION

CTEV also known as club foot is one of the most common congenital pediatric foot deformities with incidence of 1 per 1000 live births^[1]. Clubfoot can range in severity from a modest deformity that is noticeable at birth to an excessively rigid foot that is difficult to manipulate^[2]. DiMeglio et al. and Pirani Score is used to classify CTEV^[3]. Idiopathic clubfoot is isolated deformity of the foot and ankle complex, can be identifiable in utero and consists of four components, forefoot adductus, midfoot cavus, hindfoot varus, ankle equinus^[2,3]. Untreated child walks on dorsolateral aspect or lateral border of foot resulting in pain on walking, callus formation, can't wear normal shoes, predisposing the foot to skin and bone infections, callus formation, with persistence of deformities it becomes gradually rigid due to secondary changes in tarsal bones and joints^[4,5]. CTEV may associated with arthrogryposis, myelodysplasia, or multiple congenital abnormalities, but idiopathic variety is most common, which is isolated birth defect^[6,7]. The prevalence of additional congenital anomalies or chromosomal abnormalities in CTEV ranges from 24-50%^[2,3,8], of which disorders involving nervous system comprising greatest most known aetiologies are numbers. The myelomeningocele and distal arthrogryposis [2,3,8]. With different aetiologies, CTEV representing the final common pathway for disruption anywhere along the neuromuscular unit, including the brain, spinal cord, nerve, or muscles^[2,3,8].

For idiopathic club foot, many theories have been proposed, including in utero positioning, genetic factors, vascular deficiencies, abnormal muscle insertions and environmental factors [9]. Clubfoot is multi factorial in origin, genetic factors also play major role with 33% incidence in identical twins and 25% cases are familial [3,10] and with difference in prevalence of CTEV across ethnic population, highest in Hawaiians and Maoris (seven per 1000) and lowest in Chinese^[11]. Though multi factorial and polygenic causation have been proposed, the precise genetic mechanism behind clubfoot remains unknown[11]. Earlier studies using complex segregation analyses results showing a single incompletely dominant disease gene with unmeasured factors contributing to incomplete penetrance^[12]. In a study of Pacific and Maori people, segregation analysis indicated a recessive mixed model, while complex segregation analysis revealed a single dominant gene with 33% penetration. A polygenic threshold model, supported by the finding of the Carter effect, in which females require a greater genetic load to inherit the disorder than males^[13]. However, the physiological cause of this sex dimorphism, male, female ratio of 2:1, is currently unknown. Environmental factors also play role in some cases of clubfoot. Increased risk of CTEV was partially associated with amniotic fluid leakage,

suggesting oligohydramnios occurring at a critical gestational period may be detrimental to foot development. But among patients with idiopathic clubfoot, amniocentesis is a rare risk factor^[11]. Early amniocentesis (<13 weeks gestation) was associated with an increased risk in CTEV compared to midgestational amniocentesis or chorionic villus sampling^[11]. Unlike positional foot deformities, such as metatarsus adductus, which occur at increased frequency in twin pregnancies, there are little data to support an association of clubfoot with late gestational uterine compression^[11].

Environmental exposure of cigarette smoke in utero is independent risk factor for CTEV^[14]. In a study including more than 3000 individuals, Honein et al. found a correlation between smoking and clubfoot, with adjusted odds ratios of 1.34 for smoking alone, 6.52 for family history alone and 20.30 for smoking and family history combined^[7,14]. Genetic polymorphism responsible for the metabolism of tobacco, elevated maternal homocysteine, seasonal viral infections and methylenetetrahydrofolate reductase (MTHFR) gene polymorphism have also been associated with an increased risk of clubfoot^[7,14]. It's not yet clear to what degree CTEV result from, inheritance of common genetic variants (such as single nucleotide polymorphism which, by definition, are present at an allele frequency of >5%) or rare mutations in susceptibility genes^[13]. The homeodomain transcription factors HOXD12 and HOXD13, as well as a number of apoptotic genes such the caspase genes, have been linked to common genetic variations associated with CTEV through the use of candidate gene analysis. However, these genetic variants are all relatively small effect, contribute only an individual's risk of clubfoot and will require replication in larger cohorts to confirm their importance^[13]. With the use of huge sample sizes and an unbiased methodology, future genome-wide association studies will identify the genes responsible with clubfoot susceptibility and if major and minor susceptibility genes are present, it can identify them^[13].

MATERIALS AND METHODS

This prospective, observational, outpatient-based study, conducted at tertiary care medical teaching institute from February 2023 to February 2024. Infants from birth to 12 months of age with idiopathic clubfoot, were included in this study after obtaining informed consent from their parents. Children above 12 months of age, atypical or secondary club foot or past history of club foot surgery were excluded from study. Based on some inclusion and exclusion criteria's (Table 1), 50 infants with 85 feet of untreated idiopathic CTEV were selected for study. Infants, detailed history was taken and thorough general and local examination was done. CTEV deformity was scored according to Pirani scoring system^[15]. Ponseti's

technique of manipulation and serial casting was applied at weekly interval. Of the CTEV deformities, sequence of correction is cavus, followed by adduction and varus deformity. According to Pirani's scoring system, equinus deformity was corrected after midfoot deformity correction and usually require percutaneous Tendo-achillis tenotomy. Serial manipulation and corrective casting was done for six to eight weeks as per requirements and cases were regularly followed up at weekly interval. On every follow up before applying cast, improvement in CTEV deformity was documented using Pirani scoring system. Percutaneous Tendo-achillis tenotomy is planned when midfoot Pirani score came to zero after serial casting by Ponseti technique, baby taken under general anesthesia or alternatively oral sedative for the procedure and final cast was applied for three weeks. Then Dennis Brown splint was applied for three months. Children, who have started weight bearing on lower limbs, modified CTEV shoes were given to them. For three to four years night time bracing instructions was given to their parents. Parental education and their reassurance, compliance to follow up and long-term bracing protocol, helped in maintaining the deformity correction and preventing relapse.

Pirani Scoring System: For grading clubfoot, Pirani described a system in 1995^[15], which is composed of six different physical examination findings, three at midfoot level, three at hindfoot level, each scoring 0 for no abnormality, 0.5 for moderate abnormality, 1.0 for severe abnormality. Each foot was assigned, total Pirani score ranging from zero to six. Higher the score indicating more severe clubfoot deformity. Components of Pirani scoring system (Table 2). Ponseti's technique for CTEV aims to achieve biomechanical realignment of foot^[16-18]. Ponseti's technique can be divided into two phases, first being the treatment phase during which CTEV deformities are corrected, second being maintenance phase in which CTEV brace are used to prevent, recurrence of deformities. While applying ponseti's technique for CTEV it's important to note that heel is never directly manipulated, due to coupling of tarsal bones, correction of heel varus and ankle equinus takes place simultaneously.

Treatment Phase: Started as early as possible, if infants skin conditions permit, till that infant mother is taught to do regular corrective manipulation of foot after each feed.

First Cast: First cast of treatment phase, aims to correct cavus deformity (high medial arch), cause of which being pronated forefoot in relation to hindfoot, being corrected by supinating the forefoot by raising the first metatarsal and abducting the foot gently,

while stabilizing the talus by placing thumb over lateral part of head (fulcrum of rotation), followed by application of well-padded above knee plaster casting, while maintaining position and moulding it well. Parents are instructed to return after a week for next cast and complications which may happens are explained properly. (Fig. 1:). Fig. 1A is showing clubfoot with cavus deformity. Fig 1B is showing above knee cast in the corrected position.

Subsequent Cast: Severity of cavus deformity assessed using Pirani scoring system after removal of first cast and if cavus is not corrected, cast as applied first time was reapplied. If cavus get corrected by first cast, subsequent cast was applied, aiming adduction, heel varus and ankle equinus deformity. Thumb placed over lateral part of talar head, manipulating supinated foot in abduction then well-padded above knee plaster cast was applied. Complete correction anticipated after six to seven cast. (Fig. 2). Fig 2A is showing corrected cavus deformity. Fig 2B is showing above knee cast in the corrected position.

Tendo-Achilles Tendon Tenotomy: In majority of children, some equinus deformity still persist at the end of cast, which is corrected by per cutaneous surgical release of Achilles tendon, allowing dorsi flexion at ankle joint, timing of which is decided by Pirani score, when mid foot score becomes less than 1 and hind foot score become more than 1. After per cutaneous Tendo-Achilles tenotomy, final cast applied in 70 abduction of foot and 10-15 foot dorsi flexion for three weeks. (Fig. 3). Fig 3A is showing residual equinus at the ankle joint. Fig 3B is showing the procedure of tenotomy. Fig 3C is showing post-tenotomy cast in dorsi flexion at the ankle joint.

Brace: Brace maintain corrected position of foot, after removal of final cast. Brace contain, straight open toe shoes, which is attached with a bar, which has to be worn 23 hours a day for the first 3 months. After that, brace has to be worn for 14-16 hours, per day including 12 hours at night and 2-4 hours during day time, till child become three to four years of age. Relapse rate is more than 80%, if brace protocol not followed correctly. (Fig. 4). post-correction orthosis to maintain the correction. Fig 4A is showing Denis Browne (DB) splint. Orthopaedic CTEV shoes front view Fig 4B and bottom view Fig 4C. Clinical images of our case at the final follow-up after the end of the treatment phase with the continuation of the maintenance phase. (Fig. 5). Fig 5A is the view from the back side and Fig 5B is the front view.

RESULTS AND DISCUSSIONS

Fifty cases with 85 idiopathic club foot, were included in our study after well informed and written consent. 40 infants were male (80%), 10 infants were female (20%), with male to female ratio of 4:1. 70%

having bilateral involvement, 20% have right foot involvement, 10% having left foot involvement. All these cases were newly diagnosed, idiopathic club foot with no past history of conservative or operative treatment. Thirty five cases (70%) presented within first two month of age. 10 cases (20%) presented within 2-6 months and 5 cases (10%) presented after six months. 38 cases (76%) were first born infant, 12 cases (24%) were birth order second or above. Out of 50 cases, 45 cases (90%) were born from non-consanguineous marriage and 5 case (10%) born with consanguineous marriage. (Table 3). Mean initial pirani score was 4.8, maximum score was 6, minimum score was 2.5. In our study, complete correction of CTEV was achieved in 12 cases (14.11%) by Ponseti's technique and 73 cases (85.88%) in addition required per cutaneous Achilles's tendon tenotomy with casting. Number of Ponseti's corrective cast required in 51 cases (60%) were five to six, while in 34 cases (40%) six to seven casts were required. Average number of Ponseti's cast required for full correction was 6.5. Only 3 case (3.52%) developed superficial blisters due to casting, no complications encountered in rest 82 cases (96.47%). (Table 4). In our study final mean Pirani score was 0.055, with (p<0.001) being highly statistically significant, indicating Ponseti's technique for CTEV corrective casting is highly effective. (Table 5). (A p-value of less than 0.001 is highly significant and indicates the effectiveness of the Ponseti method in the treatment of congenital talipes equinovarus).

Clubfoot being complex musculoskeletal deformities of foot, that needs to be corrected with meticulous attention to detail by both surgeons and the parents. Excellent functional results have been achieved in 85-90% cases, who had strictly followed ponseti's technique protocol. 50 cases in our study were classified using Pirani scoring system. Dimeglio or Carroll classification system also present for CTEV but Pirani's scoring system also having prognostic importance and helps in deciding management. Out of 50 cases, 35 infants (70%) presented within first 2 month of life, 10 cases (20%) presented between third to six months, remaining 5 patients (10%) presented between seven months to one year of age, this suggests inadequacies in the referral system as well as ignorance on the part of parents and guardians. Our study showing male preponderance with male to female ratio of 4:1, out of 50 cases, 40 cases (80%) were male and 10 cases (20%) were female. Findings are similar to study done by Yamamoto [19] and Chesney et al.[20] suggesting male preponderance. Prejudices, biases and social customs that favor men in our community may be the cause of this. Our study showing, bilateral involvement in 35 cases (70%), 10 case (20%) having right side and 5 cases (10%) having left side involvement. Guruprasath et al. [21] study showing bilaterality in 57.89% of cases, right sided in 26.3% of cases and left sided in 15.78% of cases. Our study showing, 38 cases (76%) were first born

children, these finding is consistent with studies done by Pulak *et al.*^[22] and Yamamoto^[19]. This suggests that firstborn children had a higher prevalence of congenital clubfoot. None of the 50 cases in this study had any coexisting medical problems. In contrast, study



Fig. 1: Ponseti technique of ctev correction: the first cast for cavus correction



Fig. 2: Ponseti technique of ctev correction: subsequent casts



Fig. 3: Ponseti technique of ctev correction: achilles tendon tenotomy



Fig. 4: Ponseti technique of ctev correction: post-correction orthosis to maintain the correction



Fig. 5: Ponseti technique of ctev correction: clinical images at final follow-up

Table 1: Study inclusion and Exclusion Criteria's

Inclusion Criteria	Exclusion Criteria
Infant up to 12 months	CTEV due to Spinal defects or muscle imbalance
No previous history of CTEV treatment at other institute	CTEV with more than three anomalies AMC (Arthrogryposis multiplex congenita)
	Patients treated at other institutes and referred out to our institute
All genders	Relapses cases
Legal guardian given written informed consent	Resistant case
to participate in the study	Patient's refusal to take part in the study

Table 2: Pirani Scoring System

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Parameters	Mild	Moderate	Severe
Midfoot score			
Curved lateral border	0	0.5	1
Deep medial crease	0	0.5	1
Talar head coverage	0	0.5	1
Hindfoot Score			
Empty heal	0	0.5	1
Posterior crease	0	0.5	1
Rigid equinus	0	0.5	1

Table 3: Demographic variables of cases

Variables		No. of cases (%)
Sex	Male	40 (80)
	Female	10 (20)
Side of involvement (Out of 31)	Bilateral	35 (70)
	Right	10 (20)
	Left	05 (10)
Birth Order	Firstborn	38 (76)
	Second or above	12 (24)
Age at presentation (in months)	0-2 months	35 (70)
	2-6 months	10 (20)
	>6 months	05 (10)
Consanguinity	Non-consanguineous	45 (90)
	Consanguineous	05 (10)

Table 4: Treatment and complication-related variables

Variables		No. of cases (%)
Treatment Modalities	Casting only	12 (14.11)
	Casting and per cutaneous Achilles-tendon tenotomy	73 (85.88)
Number of casts required for full correction	5-6 casts	51 (60)
	6-7 casts	34 (40)
Complications	Superficial blister formation	03 (3.52)
	No complications	82 (96.47)

Table 5: Relationship between Pirani score at initial presentation and after final cast removal

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Pirani Score	Before treatment	After treatment	
Min-Max	2.00-6.00	0.00-1.00	
Mean±SD	5.012±0.94	0.102±0.24	
n-value	<0.001 (highly statistically significant)		

done by Guruprasath et al. [21] showing, 10.52% showing associated medical conditions, that includes cleft lip (5.26%), omphalocele (2.63%) and developmental dysplasia of hip (2.63%). Number of corrective Ponseti cast required in 85 feet in our study was, 5-6 cast in 51 cases (60%) and 6-7 cast in 34 cases (40%) and average number of casts being 6.5 for full deformity correction. Laaveg et al. [23] study showing average number of corrective casts being seven for deformity correction. Cast replacement at shorter interval with fewer cast per foot showing better results, therefore many orthopaedicians following this protocol. We observed that higher the initial Pirani score and older the age of initial presentation, required more cast. In our study, maximum initial Pirani score was 6, with minimum score of 2.5, with mean initial Pirani score of 4.8. Our study findings are comparable with study done by Syed et al. [24] and Pulak et al. [22].

Percutaneous Tendo-Achilles tenotomy performed after correction of forefoot adduction and heel varus. Out of 85 feet in our study, 73 cases (85.88%) required tenotomy. Pirani *et al.*^[25] study showing, 90% patient underwent tenotomy, while 91% patient underwent tenotomy in Dobbes *et al.*^[26] series. Our study finding

showing that, tenotomy usually required in patients, who had pirani score of 5 or more at presentation. In our study, out of 85 feet, 3 case (3.52%) developed superficial blister infection, which healed with adequate padding and dressing. Lehman^[26] study showing 10.2% complication rate. Guruprasath et al. [21] study showing 13.15% complication rate, that include superficial sore and crowding of toes, which was managed by soft cotton padding, providing enough space for toes, mainly dorsum of foot, for free toe movement. In our study final mean pirani score was 0.055, with (p<0.001) being highly statistically significant, indicating Ponseti's technique for CTEV is highly effective. In study done by Thacker et al. [27], at final follow up, final pirani score was 0.00, indicating Ponseti's technique highly effective for correction of clubfoot deformities.

CONCLUSION

We conclude that, Ponseti's technique of CTEV casting having >90% success rate in avoiding comprehensive surgical release, being affordable, safe and effective procedure, providing painless, plantigrade, cosmetically acceptable foot with better

functional outcome with minimal complications. Success rate is higher among infants who were compliant with the treatment protocol. Parents who receive continuous encouragement and support to accept long-term brace treatment are better able to sustain the correction and avoid relapses. The Ponseti method of serial cast correction is the best technique for correcting idiopathic CTEV deformity, based on our studies.

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