Current Medicine Research and Practice 9 (2019) 176–181

Contents lists available at ScienceDirect

Current Medicine Research and Practice

journal homepage: www.elsevier.com/locate/cmrp

Original article

Management of neglected, recurrent, and resistant congenital talipes equinovarus by controlled differential fractional distraction using the Joshi's external stabilization system

Manish Dhawan*, Nipun Rana

Department of Orthopaedics, Sir Ganga Ram Hospital, New Delhi, India

ARTICLE INFO

Article history:
Received 19 November 2018
Accepted 23 August 2019
Available online 29 August 2019

Keywords:
Clubfoot
External fixators
Ligamentotaxis
Distraction
Neglected

ABSTRACT

Background: Clubfoot is a common congenital foot anomaly that requires patients to be presented to the orthopedic surgeon.
Objectives: This study was conducted with the aim of assessing the efficacy in terms of the clinicoradiological outcome of the neglected, recurrent, and resistant congenital talipes equinovarus (CTEV) by ligamentotaxis using Joshi external stabilization system (JESS) fixators and evaluating the factors affecting the same.
Material and method: A total of 30 children (34 feet) were evaluated after correction with differential distraction using the JESS.
Outcome measures: All were evaluated clinically and radiologically and scored as per the Catterall-Pirani scoring system, both before and after correction of the deformity.
Results: All patients achieved good clinical results as per the Pirani score. Radiologically, all patients achieved the normal range of values. All findings were clinically significant.
Conclusion: JESS is a useful technique in neglected, resistant, relapse cases of CTEV or for any severity in patients aged between 1 and 10 years after a fair trial of Ponseti serial casting.

1. Introduction

The congenital talipes equinovarus (CTEV) deformity of foot or famously called clubfoot is a common foot problem seen at birth by pediatricians and pediatric surgeons with an incidence of 5–6 per 1000 live births, varying with race and geography.[1] When inadequately or not treated by various conservative measures such as manipulations, taping, serial and plaster cast techniques, the foot usually turns further deformed and turns resistant to simple physical corrective measures. Apart from the late presenters with neglect in the treatment, female sex, hereditary disorders, severity of deformity, associated cavus, clawing of toes, and small heel are considered as poor prognostic factors in CTEV management. Resistant, relapse, and neglect cases pose a serious problem in our country with surgical options such as soft-tissue and bony release, advocated by Carroll and McKay.[2–4] Extensile approaches to recently popularized mini-incision and percutaneous approaches are used to counter the complications of surgeries such as wound healing issues, scarring, and stiffness. The Ilizarov fixator has also been used for CTEV correction using the fractional distraction histiogenesis.[2] A combination of principles were laid down by Ponseti manipulative cast techniques and external fixator applications. In 1990, Dr. B. B. Joshi originally described the controlled differential fractional distraction with a mini-external fixator. This study was conducted to assess the efficacy in terms of the clinicoradiological outcome of the neglected, recurrent, and resistant CTEV by ligamentotaxis using Joshi's external stabilization system (JESS) fixators and evaluate the factors affecting the same.

2. Material and methods

This retrospective observational study was conducted at our institute on all the children of either sex aged between 1 and 10 years, with neglected, relapse, or resistant clubfoot since September 2005 to November 2013. Cases with the following characteristics were included in the study: idiopathic CTEV, first time, but late presentation (>9 months after birth) in outpatient department (OPD) without any prior treatment, previously

* Corresponding author.
E-mail address: drmanishdhawan@gmail.com (M. Dhawan).

https://doi.org/10.1016/j.cmrp.2019.08.002
2352-0817/

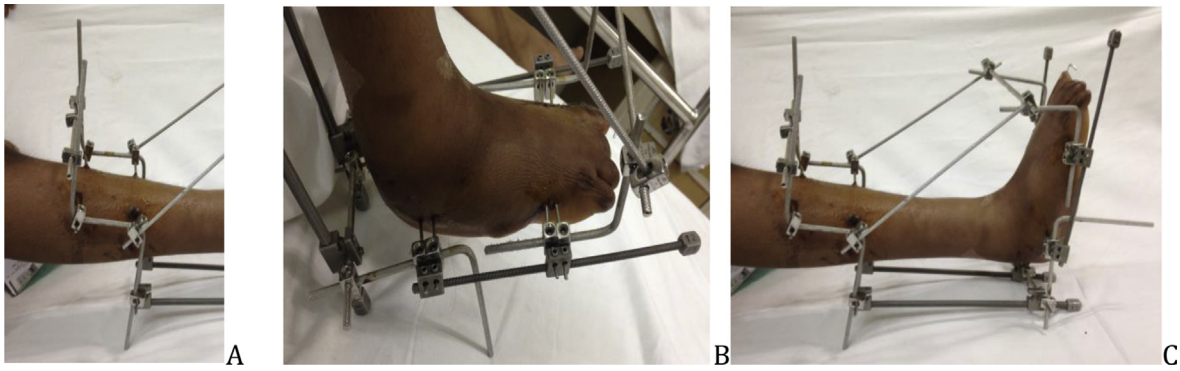

Fig. 1. (A, B) Showing JESS assembly application in leg and foot and the final construct (C) with two pairs of distractors for fractional differential distraction schedule.

managed cases of CTEV with full correction but insidious relapse of the deformities or rigid cases not correctable by conservative cast measures. We excluded the patients with secondary causes to CTEV such as arthrogryposis, meningomyelocele etc.

All the included patients were assessed clinically and radiologically. Clinically, the Catterall-Pirani scoring system and clubfoot assessment protocol were used to assess the severity of deformity and also to assess the final correction achieved.[5] Clinically the deformity, passive range of motion, muscle function, dynamic function, and pain were assessed. Radiographs were studied for talocalcaneal angle, talo-first metatarsal angle, talo-fifth metatarsal angle (on an anteroposterior view), talocalcaneal angle, tibiocalcaneal angle, and calcaneal pitch (on a lateral view). All the cases had already been subjected to a 4-week trial of serial Ponseti cast without any benefit and hence were included in the list for surgery with JESS. The mean follow-up duration of cases was 4.9 years.

We operated all the cases under general anesthesia. Two K-wires each were inserted in the tibia, calcaneum, and distal ends of metatarsals, followed by correcting the deformity by the standard Ponseti method and thereafter maintaining the reduction by connecting the inter-connecting straight, Z or L rods of the JESS fixator. A pair of distractors were placed on the medial and lateral sides between the tibiocalcaneal and calcaneal-metatarsal attachments. If needed, calcaneal percutaneous release of the plantar fascia was performed for cavus deformity and cuboid nuclectomy was performed in the same sitting (Fig. 1). After preliminary correction of the deformity of the foot, the anterior tension rods between the tibial and forefoot attachments were tightened. The differential distraction of the distractors was begun after the 3rd postoperative day. The standard technique was to distract 0.25 mm four times a day on the medial side and 0.25 mm twice a day on the lateral side. We continue this gradual distraction until clinical overcorrection is achieved, thus bringing the dynamic phase to an end. Thereafter, we keep the frame in situ in state of clinical overcorrection for twice the period of dynamic phase (static phase). After this, the JESS fixator was removed and an above-knee plaster of Paris (POP) cast was applied for 6 weeks in overcorrected position. Later, standard orthosis were used and children were followed up 6 monthly for clinical and radiological workup, especially until the deformities became static for 2 years, and thereafter, they were followed up annually. Parents were explained the need for strict follow-up and continuous use of braces/orthosis without any failure and were asked to inform the surgeon immediately in case of any relapse of deformity. Cases were considered as failure if there was no or incomplete clinicoradiological correction or if complications such as joint subluxation and rocker bottom foot occurred.[6]

3. Observations and results

We managed 34 feet in 30 patients aged between 1 and 10 years with neglected, relapsed, or resistant CTEV with prior failed trial of Ponseti method. We observed 10 of 13 relapse cases were managed with prior soft-tissue release surgery with scarring. Most cases were male children with right-sided unilateral involvement, with mean time to correction of 26 days. There were 20 male and 10 female patients. The minimum age was 1 year, and the maximum was 9 years (mean age, 4.2 years). Patients were assessed for clinical outcomes using the Pirani scoring system, and all the complications were noted. A total of 10 feet were managed elsewhere with posteromedial soft-tissue release surgery, 6 cases were managed previously with the POP cast, and the rest were not given any treatment. To start with, 32 of 34 feet had severe deformity with poor Pirani scores (Table 1).

The entire clinicoradiological parameters improved in the patient statistically. Before the surgery, the feet had varus, equinus, cavus and adduction deformity. The mean preoperative equinus deformity was 54°, heel varus deformity was 40.2°, and forefoot adduction deformity was 28°. The mean postcorrection values for varus were 2° and for abduction was 4°. The cavus deformity was corrected in all the cases. The mean postcorrection dorsiflexion achieved was 17.3°. The calf and foot size remained unaffected by the procedure, although the foot was found suppler after the procedure than their preoperative state in rigid feet. The radiological correction of angles was found to be significant after the end of treatment (Table 2).

The cases were evaluated using the Pirani scoring system, grading them from good to poor. A score of 0–2 was regarded as a good clinical outcome. All the patients could gain a score between 0 and 2 (mean 0.8) from a mean preoperative score of 5. Therefore, the feet could gain flexibility and remained flexible until their latest follow-up. Clinicoradiological illustrations are shown in Figs. 2 and 3.

Table 1
Number and type of patients/feet under study.

Number of patients (n)	30
Number of feet	34
Number of neglected	18
Number of resistant	13
Number of relapsed	3

Table 2
Radiographic parameters of foot in AP and lateral view.

Radiological angles	Normal	Pre-operative	At fixator removal	Final follow-up
Radiological index				
Talocalcaneal	30-35	6	20	26
Lateral	25-50	25	36	40
Talo-first metatarsal	0 to −15	−27	−13	−10
Talo-fifth metatarsal	0 or +	−12	20	26
Talocalcaneal index*	>40	31	56	Not taken

AP, anteroposterior.
All figures are expressed in degrees.
* Addition of talocalcaneal angle in AP and lateral view.

4. Discussion

CTEV deformity of the foot is a common congenital problem seen after birth. The standard management protocol being followed consists of conservative management with serial manipulations and corrective cast by the Ponseti method, with excellent results in a toddler. Surgical management has been mainly advocated for the late, neglected, and relapsed cases, where the conservative trials fail.[7–10] Many publications have now been made where a complete reversal of this treatment protocol has also shown distinct advantages. A conservative trial in late neglected cases and less-extensive surgical soft-tissue release in early cases have shown variable success rates. Khan and Kumar[8] evaluated the efficacy of the Ponseti technique in 25 neglected clubfoot in children older than 7

Fig. 2. Clinical photographs showing the frame assembly and correction after ligamentotaxis. CTEV, congenital talipes equinovarus.

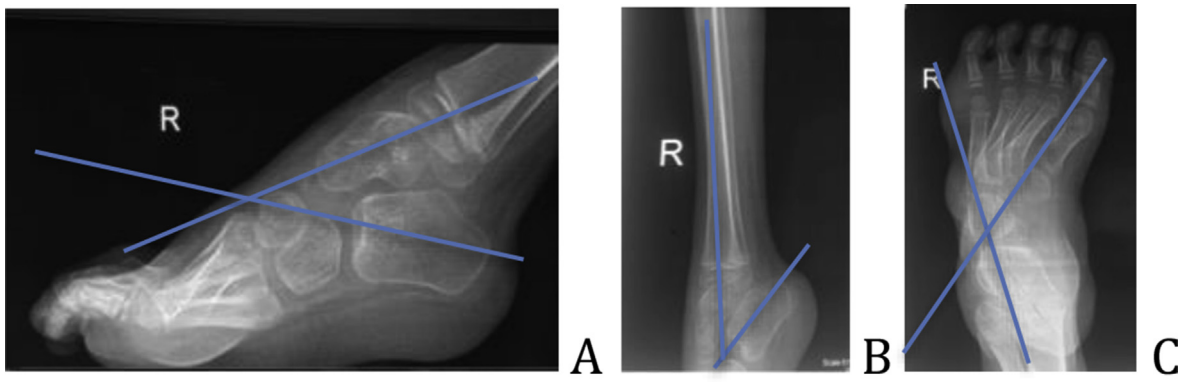

Fig. 3. Radiograph of foot and leg showing the angle measurements for assessment. Talo - Calcaneal angle (A)- longitudinal axis of talus and calcaneum bone in lateral view. Tibio Calcaneal angle (B)- longitudinal axis of tibia and calcaneum bone in lateral view. Talo - 1st Metatarsal angle (C)- longitudinal axis of talus and first meta-tarsal bone in AP view.

years (mean age, 8.9 years). The mean follow-up period was 4.7 years. The observed 85.7% of feet were fully corrected, with recurrence in 24% of feet.

The surgical principle behind treatment of clubfoot is based on the soft-tissue releases and reconstruction of the already deformed bones. Surgery performed at any stage of CTEV carries the problems such as potential wound healing complications, scarring of tissue causing secondary stiffness, and finally, giving a single chance to correct the deformity in the theater.

On the other end, the Ponseti principle is based on gradual transformation of the collagen matrix of tightened ligaments and repositioning of the bones around the talus. Many articles published now have shown high success rates with the Ponseti method alone in both early and late neglected and relapsed cases after prior surgery.[11–14] The prime philosophy behind the Ponseti technique is that the center of CTEV deformity lies with the head of the talus (with a medial talar spin), which explains the principles of this technique. The first cast is applied in further supination of the forefoot to allow the pronated forefoot to align in line with the calcaneum, measured by the foot bimalleolar angle (FBA). Second, the pronation force is never applied, as pronation can cause the calcaneum to get jammed under the talus, preventing the heel varus correction. Free movement of the calcaneum under the talus and that of the talus in the ankle mortise is achieved by only abduction of the forefoot with fulcrum over the talus head. Third, the cavus deformity is never corrected at the midfoot as it can cause rocker bottom foot deformity. Most workers would now opine on the trial of the Ponseti technique in all the cases of CTEV before reverting to any other conservative or surgical measure.

External fixators work on the similar principles as the Ponseti technique with manipulation of the deformed foot using the osseoligamentous hold of the wires. Ilizarov fixators have been used in treatment of complex CTEV with the principle of fractional distraction histiogenesis. With the same concept, many orthopedic surgeons are now using the JESS fixator, introduced by Dr. B. B. Joshi, with differential controlled fractional distraction histiogenesis. Fixators carry their own distinct advantages of a more physiological elongation of the foot by an instrumental manipulation, wherein time-to-time modulation is possible as needed. There is less scarring of tissue, no epiphyseal damage while correction, and minimal motor loss, providing more supple cosmetic foot with near-normal size. It has been labeled as an extended conservative management. Thirty-four cases of severe relapsed and neglected clubfoot deformity were treated with Ilizarov fixators.[12] Good results were achieved in about 58.8% of cases with recurrence in about 8.7%.

JESS fixator principles differ from the Ilizarov method in many ways: (1) Ilizarov method uses the axially tensioned wire, which can show a “cheese cutter effect” on soft osseocartilaginous bones of children. (2) Clubfoot is a spatial deformity in multiple planes;

Table 3
Comparative studies in JESS fixator used for CTEV.

S.No.	Study	Year	No. of feet	Average Follow-up	Excellent to good results	Tool
1	BB Joshi et al	2001	-	-	68% excellent, 16% good	-
2	Suresh S Ahmed et al	2003	34	2.8 yrs	77.3% excellent	**Hospital for joint diseases Orthopaedic Institute** functional rating system for clubfoot surgery
3	Anwar Marthya et al	2004	41	3.5 yrs	59.7% excellent	Caroll criteria for clinical assessment of severity of clubfoot
4	Mahendra et al	2008	106	6.5 yrs	63% excellent, 30% good	**Hospital for joint diseases Orthopaedic Institute** functional rating system for clubfoot surgery
5	Mandappa et al	2009	15	16 mths	14/15 feet showed satisfactory results	Simon's criteria for clinical assessment of outcome of clubfoot
6	Devados et al	2010	28	12 yrs	71.4% excellent	Dimeglio classification Pontesi scoring system
7	Ajai Singh et al	2011	33	4.7 yrs	100% excellent	Pirani scoring system
8	Present study	2014	34	4.9 yrs	76%	Pirani scoring system Clubfoot assessment protocol

hence, accurate placement of hinges for correction in foot is difficult. JESS is not a constrained fixator and allows correction of the deformity at natural foot joints in multiplanes. (3) JESS is based on differential distraction, with elongation of both the medial and lateral borders, without showing any convex side crushing effect on the bones as seen with the Ilizarov fixator.[15]

The possible drawbacks of JESS fixators seen in our series were unacceptability of the fixator on children by parents and by the child himself/herself because of pain and swelling of toes. Second, the pins prick and cause injury to the patient and the relatives while nursing (Fig. 1). Complications had no correlation with the age and the type of CTEV but a positive correlation with the initial severity of disease. We had 2 failures (5.8%) in our series, which continued to show poor Pirani scores. There were no relapses in our study, probably because most cases were older than 4 years. Generally, there is a tendency to relapse irrespective of the treatment, seen most commonly within 2 years and rarely after 4 years of age.[7]

As per our observations, radiological parameters return to normal range. Some of our patients were completing the 1st decade of life, when cartilaginous bone begins to mature and becomes stiffer. We observed the midfoot and hindfoot soft-tissue correction by stretching of the contracted ligaments gradually and differentially along with gradual remodeling of the deformed bones as well during the progressive histiogenesis. The difference in precorrection and postcorrection Pirani scores in these patients was found to be statistically significant ($P = 0.01$). Hence, in our study, we did not find a correlation between the age of patients and the initial severity of deformity and with potential for clinical correction of deformity. In a study by Singh et al.,[17] they excluded the cases older than 6 years, as by this age, significant bony changes may affect the outcome.

The overall results of the use of JESS fixators in our series were very encouraging with more than 90% with excellent Pirani scores. Most cases were with unilateral foot involvement, and they showed smaller feet than normal feet but were cosmetically acceptable to all parents.

In the present study, improvement in the medial border-to-lateral border ratio was observed in all subjects although we were not able to achieve complete reversal of the medial border-to-lateral border ratio, probably because the duration of observation was short. Radiological correction was observed in our cases but did not have any correlation with the clinical outcome. The radiological correction occurred even after JESS removal until the final follow-up, probably because of natural remodeling of bones and soft tissue once they have mechanically aligned by JESS. talo-calcaneal angle (TCA) and talo-calcaneal index (TCI) are simple parameters to compare preoperative and postoperative radiographs but cannot be used to comment on the clinical severity of each case.[16]

A study on 44 neglected clubfoot[13] managed by JESS distractors and followed up for a minimum period of 2 years had obtained about 90% excellent to good results. In a study,[14] 41 children with idiopathic neglected CTEV, residual CTEV, or recurrent CTEV were managed by JESS and followed up for 3.6 years. They obtained 59.7% excellent and good results. Many studies have been conducted to show the efficacy of JESS in CTEV in the past, but little has been mentioned regarding its use in relatively older children finishing their first decade of life except for the primary author himself (Table 3).

Although it is a very small series, by far, we are able to show excellent to good results in 76% of cases with complications of temporary consequences. The final outcome following JESS in problematic CTEV may be influenced by the duration of the static phase and type of CTEV but has no influence of the age of child and the initial severity of deformity. Hence, we may conclude that JESS is a very useful surgery in neglected/resistant/relapse cases of CTEV or any severity in children aged between 1 and 10 years after a fair trial of the Ponseti serial casting. JESS works on the principle of ligamentotaxis and is a patient- and surgeon-friendly procedure in many ways. But in this procedure, the active participation of the patients' attendants is one of the prime factors for the successful outcome. It is also urged that the motivated and compliant parents play a pivotal role in the success of surgery.

Conflict of interest

There is no conflict of interest.

Key message

What is already known?

JESS is a known procedure among orthopedic surgeons, both patient- and doctor-friendly, which allows correction of the deformity of the clubfoot by ligamentotaxis.

What this study adds

JESS is a worthwhile procedure in neglected, resistant, and relapsed cases of CTEV even after ossification of the cartilaginous bones of foot in children nearing their first decade of life by gradual remodeling of bones with time.

References

1. Carroll NC, McMurtry R, Leete SF. The pathoanatomy of congenital clubfoot. *Orthop Clin N Am*. 1978;9:225–232.
2. Dobbs MB, Nunley R, Schoenecker PL. Long-term follow-up of patients with clubfeet treated with extensive soft-tissue release. *J Bone Jt Surg Am*. 2006;88:986–996.
3. Khan SA. "Ponseti's manipulation in neglected clubfoot: are we joking?. In: *Proceedings of the Annual Meeting of the American Academy of Orthopaedic Surgeons*. vol. 63. 2006.
4. McKay DW. New concept of and approach to clubfoot treatment: section II–correction of the clubfoot. *J Pediatr Orthop*. 1983;3:10–21.
5. Andriesse H, Hägglund G, Jarnlo GB. The clubfoot assessment protocol (CAP); description and reliability of a structured multi-level instrument for follow-up. *BMC Muscoskelet Disord*. 2005;6:40.
6. Joshi BB, Laud NS, Warrier S, Kanaji BG, Joshi AP, Dabake H. Treatment of CTEV by Joshi's external stabilization system (JESS). In: Kulkarni GS, ed. *Textbook of Orthopaedics and Trauma*. first ed. New Delhi: Jaypee Brothers Medical Publishers; 1999.
7. Ponseti IV, Smoley EN. Congenital club foot: the results of treatment. *J Bone Jt Surg Am*. 1963;45:134–141.
8. Khan SA, Kumar A. Ponseti's manipulation in neglected clubfoot in children more than 7 years of age: a prospective evaluation of 25 feet with long-term follow-up. *J Pediatr Orthop B*. 2010;19:385–389.
9. Turco VJ. Surgical correction of the resistant club foot. One-stage posteromedial release with internal fixation: a preliminary report. *J Bone Jt. Surg*. 1971;53:477–497.
10. Radler C, Manner HM, Suda R, et al. Radiographic evaluation of idiopathic clubfeet undergoing ponseti treatment. *J Bone Jt. Surg*. 2007;89:1177–1183.
11. Marthya AH, Arun B. Short term results of results of correction of CTEV with JESS distractor. *J Orthop*. 2004;1:e3.
12. Garg S, Dobbs MB. Use of the Ponseti method for recurrent clubfoot following posteromedial release. *Indian J Orthop*. 2008;42:68–72.
13. Correll J, Forth A. Correction of severe clubfoot by Ilizarov method. *J Foot Ankle Surg*. 2003;2:27–32.

14. Suresh S, Ahmed A, Sharma VK. Role of Joshi's external stabilization system fixator in the management of idiopathic clubfoot. *J Orthop Surg (Hong Kong)*. 2003;11:194–201.
15. Ilizarov GA. The tension-stress effect on the genesis and growth of tissues. Part I. The influence of stability of fixation and soft-tissue preservation. *Clin Orthop Relat Res*. 1989;238:249–281.
16. Varun GBS, Muralidhar N, Bharathidasan K. Radiological outcomes of management of congenital clubfoot using JESS. *Int J Sci Study*. 2016;4:198–202.
17. Singh A. Clinical study - evaluation of neglected idiopathic Ctev managed by ligamentotaxis using Jess: a long-term followup. *Adv Orthoped*. 2011;2011:6. https://doi.org/10.4061/2011/218489.