Case Series

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A study on functional outcomes of 'Joshi's external stabilization system distractor' in the correction of deformities in neglected, resistant or relapsed cases of congenital talipes equinovarus

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ABSTRACT

Early intervention by an optimal treatment modality in the management of congenital talipes equinovarus (CTEV) or club foot is of paramount importance in order to preserve the functional efficacy of foot. Neglected, resistant, or relapsed cases of clubfeet are even more challenging to treat, as with time the deformities become fixed and the feet develops secondary adaptive bony changes. Joshi's external stabilization system (JESS) distractor is an effective treatment modality for such kind of deformities with a precise viability and efficacy. This case series study was conducted to evaluate the functional outcomes of JESS distractor in the correction of the deformities in neglected, resistant or relapsed cases of CTEV and to assess the complications associated with the procedure. Total 22 club feet (15 patients) included in this case series considering inclusion and exclusion criteria. JESS distractor was applied and distraction done as per distraction schedule, followed by its removal and corrective cast application. Functional outcome was based on the Pirani score with one year follow-up. The statistically significant (p value <0.05) improvement was found in all clinical and radiological parameters after the procedure in all the post-operative patients. Pirani score improved to good score (0-2) in comparison to its pre-operative poor score (5-6) without any significant complication. Considering the parents compliance, JESS distractor is a good alternative for neglected, resistant or relapse cases of club feet, as it avoids soft tissue as well as bony procedures to correct the deformity of CTEV while preserving the normal foot anatomy.

Keywords: Congenital talipes equino varus, Club foot, Joshi's external stabilization system distractor, Pirani score

INTRODUCTION

The commonest congenital deformity of foot is known as congenital talipes equino varus, also known as club foot, occurring in one per thousand live births. Male children more generally affected by this deformity. The occurrence of the deformity represents bilateral in about 50% of the cases. The term talipes equino varus is originated from latin -talipes-a combination of words "talus"(ankle) and "pes"(foot), equinus means "horse like" (the heel in planter flexion) and varus means adducted and inverted position of the foot. Both the principle and the methods of treatment

of club foot deformity are still debatable and the new management methods are still emerging. It's difficult to assess the results of various treatment modalities of congenital talipes equinovarus (CTEV), as there is no uniformly acceptable classification or grading system for this deformity. However, Dimeglio's and Pirani's are the ones which are most commonly used classification systems for grading the severity of clubfoot.^{2,3}

In the developed world clubfoot deformity is usually corrected in early life leaving little or no residual deformity.⁴ On the other hands, In the developing

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countries it's common to see late, neglected clubfoot, with relapse or recurrence of deformity.^{5,6} Although initial conservative treatment with manipulative reduction and CTEV plaster casting described by Ponseti et al, is widely accepted as an effective treatment method, but due to lack of understanding of anatomy, pathophysiology and kinematics of clubfoot among treating clinicians introduce gross errors in the treatment of clubfoot and thus creating space for invasive and minimally invasive surgical techniques.^{7,8-11}

As per available literature, the most appropriate age for soft tissue surgery for a clubfoot patient is recommended by many surgeons. Dimeglio et al and Pous et al in 1978 recommended the clubfoot surgery during neonatal age of 1-3 weeks. ¹² Lovell et al, in 1970 described that if desirable correction could not be obtained conservatively within 3 months, then operative procedure should be attempted. ¹³

Turco et al, has concluded that it's easier and appropriate to operate in a little older child, as the anatomical structures are easier to identify on after 6 months to 1 year of age. Turco et al, suggested posteromedial soft tissue release (PMSTR) surgery for clubfeet that need surgical correction. Since then, PMSTR has almost become the procedure of choice for this deformity. 14,15

Unlike that of virgin cases, relapsed or neglected clubfeet are even more challenging to treat, as with time the deformities become fixed and the feet develops secondary adaptive bony changes. Soft tissue release procedures alone are not sufficient to correct these late presenting clubfeet, and often the bony procedures are significantly needful as well.

Although, as the deformed foot is already smaller in size, bony procedures (closing wedge osteotomy, arthrodesis etc.) may lead to further shortening of clubfoot.

So other than bony procedures, ring external fixator system based on principle of differential fractional distraction histogenesis of Ilizarov et al, has emerged as another treatment option for late presenting or after relapse cases of CTEV. Ilizarov ring fixator was successfully used by Grill et al, Franke et al, and Paley et al to correct complex three-dimensional deformities of CTEV. 16,17

However, use of llizarov ring fixator is questionable in very young child aged below 3 years. As there is insufficient strength in cartilaginous anlage of tarsal bones, the tensioned wired of llizarov ring fixator can't be applied successfully in this age group.¹⁸

Joshi's external stabilization system (JESS) is the treatment modality to answer the all these queries. Based on the principle of differential distraction, JESS was developed by Dr B. B. Joshi et al, for the correction of clubfoot deformities. JESS doesn't use tensioned wires, so it can be used even in children below three years of age.

The precise viability and efficacy of JESS for the correction of clubfoot deformities from the age 3 months to adulthood has been satisfactory presented by many studies mentioned in the literature. ^{19,20} The present case series study was conducted at our tertiary care centre to evaluate the functional outcomes of JESS distractor in the correction of deformities in neglected, resistant or relapsed cases of CTEV, and to assess the procedure related complications as well.

CASE SERIES

A hospital based prospective interventional case series study was conducted at Govt. Medical College and D.B. Hospital, Churu (Rajasthan) between November 2022 to January 2024. Total 22 feet in 15 patients (8-unilateral; 7-bilateral) were included in this case series for the management of old neglected, relapsed or resistant cases of CTEV foot by JESS.

Both genders with age group between 3 to 15 years; Old neglected, relapsed or resistant cases of CTEV foot; Both unilateral and bilateral cases were included in this case series, whereas neurogenic and syndromic patients; Patient's age <3 years & >15 years, unfit patients or parent's refusal for surgery were excluded.

On admission of the patient a careful detailed history was obtained from the parents /attendants regarding the birth history, duration and previous treatment of the deformed foot. The clinical examination included.

General examination

Gender, general examination, respiratory, CVS, alimentary, locomotor and nervous systems. Special emphasis put on ruling out spina bifida, meningocele, cerebral palsy, ataxia, CDH, constriction bands or arthrogryposis.

Local examination

Unilateral and bilateral presence of deformity, Previous scar, Skin condition, Constriction bands, Size of the limb (circumferential), Size of the foot, Medial and lateral border, Presence of skin creases over medial and posterior aspect of affected foot.

Investigations

All the patients, included in the study, underwent routine blood tests, chest X-rays, etc. for pre anaesthetic purpose and once surgical fitness obtained from anesthesia side, were taken up for surgical correction with JESS distractor.

Radiological evaluation

AP and stress dorsiflexion views of foot and ankle done for radiological evaluation of each patient. X rays were studied for talo-first metatarsal angle, talocalcaneal angle, and talo-Vth metatarsal angle (all in AP view); Tibiocalcaneal angle, talocalcaneal angle and Calcaneal pitch (all in lateral view).

Surgical technique

Surgery was performed under GA or spinal anaesthesia. We used power drills in older children, while hand drills were used in smaller children. The procedure involved two major steps: Installation of K-wires, and Creation of hold and connection between the hold. Figure 1 shows the parts of the JESS distractor, used as fixation device for the correction of the deformity.

Installation of K-wires

Tibial K-wire installation: On the operating table, patient's affected lower limb kept extended in supine position and two parallel K-wires passed one finger breadth below the tibial tuberosity in the proximal tibial diaphysis from lateral to medial direction and run parallel to the axis of the knee joint (Figure 2). The distance in between two K-wires was kept about 2 to 4 cm, according the length of the middle segment of Z rod. To increase the stability, 3 K-wires were passed in older children instead of 2.

Calcaneal K-wire installation

Two parallel K- wires were passed through the calcaneus from medial to lateral side avoiding any injury to neurovascular bundle on the medial side, as shown in Figure 2. To improve axial stability, one additional half pin K-wire was passed from the posterior aspect of the calcaneus along the long axis. The entry point for calcaneus wire was below the insertion of the tendoachilles in the midline to allow fractional TA lengthening.

Metatarsal K-wire installation

One transfixing metatarsal K-wire inserted from lateral to medial direction through the necks of first and fifth metatarsal in such a fashion that this K-wire engaged 2-3 metatarsals. Two supplementary parallel K-wires were inserted from either side fascinating three metatarsals each so that the third metatarsal had occupying half pins from either side through it. The space between the metatarsal wires on either side was kept 2-3mm more than the gap between the two holes of the distractor (Figure 2).

Creation of holds and connecting between the holds

Two 'Z' rods were connected to the tibial pins, one on each side. The wires were pre-tensioned before the link joints were tightened. Two transverse rods were connected to the 'Z' rods, one anteriorly and one posteriorly. Two 'L' rods were applied to calcaneal K-wires and two more 'L' rods were applied to the metatarsal K-wires one on each side with the arms of the 'L' rods facing posteriorly and inferiorly/superiorly (Figure 3).

Calacaneo-metatarsal distractors were then connected to the K-wires on either side of the foot over L rods. One posterior transverse rod was also connected to the posterior calcaneal half pin and the posterior arms of the 'L' rods. Tibio-calcaneal distractors were applied, one on either side attaching the corresponding transverse rods posteriorly. Two another transverse rod was attached to the inferior/superior arms of the 'L' rods which could deliver dynamic traction and support to toes. It prevents further flexion contracture of the toes as the club foot deformity was being corrected during distraction.

All four distractors were distracted intraoperatively till optimal resistance was felt. Additional lengths of the K-wires were cut, and no stress was given to K-wires. The static tibio-metatarsal connecting rod was attached on each side by the transverse anterior rod of the tibial hold and the metatarsal hold. Tension force was generated by this entire system and thus the anterior portion of the ankle joint kept open. The articular cartilage was preserved from crushing. An effective gliding mechanism to the talus was presented by this system while correcting the equinus deformity (Figure 4).

Our average surgical time was one hour for each foot. At the pin entry sites appropriate skin release was created and haemostasis was achieved. After cleaning of pin entry wounds, dry dressing was applied and the pointed cut ends of the K wires were covered enough to prevent further injury.

Distraction schedule

As per our post-operative protocol for hospitalized patients, fractional calcaneo-metatarsal distraction was initiated from third post-operative day at the rate of 0.25 mm. On medial side differential distraction was performed twice the rate than that on the lateral side (0.25 mm every 6 hours medially and 0.25 mm every 12 hours laterally). Whereas, parents did the distraction at the rate of 1 mm/day on medial side and ½ mm/day on lateral side as per our instructions, in non-hospitalized patients.

With the help of calcaneo-metatarsal distraction, we obtained correction of forefoot adduction at tarso-metatarsal joints, and reduction of calcaneocuboid joint by stretching the socket for head of talus. The tibio-calcaneal distraction was done in two positions. Initially, the distractors were applied between the posterior limbs of the calcaneal 'L' rods and inferior limbs of the 'Z' rods. The distractors lied just posterior to the transfixing calcaneal wires and parallel to the leg.

The distraction was carried out at the rate of 0.25 mm every 6 hours over medial side and 0.25 mm every 12 hours over lateral side and the end-point assessed clinically. Distraction in this position corrected equinus and varus deformity of the hindfoot.

After that, the tibio calcaneal distractors were transferred posteriorly and applied above to the transverse bar joining

the posterior limbs of 'Z' rods and below to the posterior calcaneal bars joining the posterior limbs of 'L' rods and axial calcaneal pin. The distractors applied on the both sides of the axial calcaneal pin. Distraction in this position gave thrust force to stretch posterior structures and corrected hind food equinus deformity at the ankle joint and subtalar joint. Both distractions were carried out at the rate of 0.25 mm every 6 hourly and the end point decided clinically. During the distraction phase, correction of deformities was noted visually. In most of the cases, full correction was obtained at the end of 5 to 6 weeks. Following the correction, JESS distractor was held in static position to allow soft tissue maturation in elongation position for almost double the time required for correction (Figure 5).

Whole assembly of JESS distractor was removed in single stage under general anaesthesia. After removal of the assembly, below knee plaster in well molded position was applied in maximum correction just after removal of the assembly. The child was asked to walk with full weight bearing position in the plaster. After that, a short plaster boot was given to the child which acted as an orthotic device and allowed mobilization of the ankle joint, strengthening of tendo-achilles as well. To achieve maximum dorsiflexion of the foot, we encouraged the child to sit frequently in squatting position.

At an interval of 15 days, plaster was changed for 2-3 times, followed by night splints and CTEV shoes. The bracing protocol and its importance were explained to the parents and advised to follow it strictly. After that, a regular clinical assessment of the child was carried out at the end of every month. At the end of one-year, radiological assessment was done and analysed. The parents of all the operated children were asked to report, if they noticed relapse of any deformity. A case was enlisted as failure case if there was no or suboptimal clinicoradiological correction or rocker bottom deformity, joint subluxation like complications occurred during any follow-up.

Outcome measures

Statistical analysis

Online statistical software GraphPad and EpiInfo were used for calculating p values. We used Pirani scoring

system in the present case series study to evaluate the severity of deformity and to assess the final correction achieved after complete casting. The significance of difference between preoperative and postoperative values were computed using the student paired t-test. P value of <0.05 was taken statistically significant. The statistically significant results were within a 95% Confidence Interval.

Demographic data

In this case series, 50% of the patients were in the age group of 3-7 years, whereas 35% and 15% were found to be distributed in the age ranges 8-11 years and 12-15 years respectively. 15 feet were male, and remaining 7 feet were female. So, males predominated the study constituting 67% of cases, while male female ratio was almost 2:1. Total 22 feet in 15 patients (8-unilateral; 7-bilateral) were included in the study. Bilateral involvement was seen in 64% of patients; while in unilateral cases, 25% right side alone and 11% left involvement was seen.

Out of 22 feet, 10 feet were managed earlier by corrective manipulations with serial casting, 3 feet were treated earlier with soft tissue release (PMSTR) procedure at somewhere else and rest 9 had never received any kind of treatment.

All 22 feet had severe clinical deformities preoperatively (clinical grade III, Pirani score 5-6 and Foot and Bi-Malleolar Angle <65 degrees). There was statistically significant (p-value <0.05) improvement in all clinical and radiological parameters in all the post-operative patients. The study results were statistically significant within a 95% confidence interval (Table 1, Figure 6).

The average time taken for correction with JESS distractor was 4.18 weeks, highest being 8 weeks and least 2 weeks with a standard deviation of 2.583 weeks. The clinical evaluation was done using Pirani score graded as good to poor outcome.

Pirani score of 0–2 is denoted as good clinical outcome. In this case series, we achieved good clinical correction (Pirani score 0–2) and more flexible foot in all the cases, that was a good outcome. By the end of one year follow-up, the flexibility of the corrected feet remained unchanged.

Table 1: Assessment of radiological and clinical parameters.

S. no.	Measurement unit with normal value	Preoperative score	Postoperative score	P value
1.	Talo-calcaneal index (> 40°)	21.90°±5.61° (range 12°-31°)	55.61°±9.51° (range 37°-69°)	< 0.05
2.	Talus first metatarsal angle (0°-20°) (an angle above 0° denotes forefoot adduction)	25.27°±6.86° (range 8°-38°)	2.63°±6.21° (range 4°-24°)	< 0.05
3.	Tibio-calcaneal angle (60°-90°)	114.02°±10.22° (range 92°-139°)	85.97°±6.89° (range 71°-102°)	< 0.05
4.	Mean FBM angle (82.5°) (foot and bimalleolar angle)	61.31°±2.73° (range 59-64)	79.74°±1.57° (range 78-81)	< 0.05
5.	Mean pirani score	5.0	0.5	< 0.05



Figure 1: Components of JESS distractor framework.



Figure 2: Tibial, calcaneal & metatarsal K-wires placement.



Figure 3: Placement of 'Z' rods & 'L' rods.



Figure 4: Placement of calacaneometatarsal & tibiocalcaneal distractors.



Figure 5: Follow up at 2 weeks.





Figure 6: Comparative clinical evaluation: preoperative club feet (a) and postoperative clinical outcome (b).

Complications

During this case series study, we noticed that pain was the most common complication that was encountered during distraction process, and removal of JESS distractor. Pin tract infection was another common complication. Only 4 (18.2%) feet developed superficial pin tract infection (not severe enough compelling any active intervention) in our

study. Other less common complications seen were 1st and 5th MTP subluxation, flexion contracture of toes, Skin necrosis, intermittent swelling, occasional intraoperative pin prick injuries to surgeon and assistant. Most of the above-mentioned complications resolved spontaneously after JESS distractor removal. In some patients, pain was persisted on weight bearing. Although there was no pain on resting condition and it was also subsided spontaneously in next few months follow-ups. Recurrence of the deformity due to non-compliances of the parents was a dreadful issue. In this study only 6 (27.3%) feet presented with relapsed forefoot adduction (corrected by manipulations and retention by plasters in all cases) and all returned to orthosis. No open correction of any component of deformity in any case at any stage was done in the study.

DISCUSSION

The present case series study comprised of a total 22 clubfeet in 15 patients presenting in the Department of Orthopedics, P.D.U. Medical College and D.B. Hospital, Churu (Rajasthan). The age range of the patients of our study sample was 3 years to 15 years. The majority of the patients were in the age group of 3 years to 10 years. Out of the 22 clubfeet studied, 15 (68%) were male patients, and 7 (32%) were female patients. All these findings are comparable with the studies conducted by Grill et al, 11 boys and 7 girls and Joshi, 14 males and 6 females. Bilateral involvement more commonly seen in the present study (64% bilateral involvement).^{6,19} Turco et al, treated 100 patients (200 clubfeet) with bilateral involvement out of 273 clubfeet treated (73%).¹⁵ So, this finding of our study is also comparable with reports in literature.

The minimum duration to achieve correction was 15 days and the maximum duration was 57 days. About 48% of the patients required almost 4 weeks of distraction. The mean duration of distraction was 29.4 days in our study. Cantin et al and Fassier et al, described that the patients required on an average 7 weeks to overcome the deformity. Brandish et al, Noor et al, described 4 to 8 weeks as the mean period required to distract the deformity (age group 6-11 years). Our results are comparable with these studies. Wherever possible, we maintained the distractor in static phase for a minimum of double the time required for distraction of deformity. The average duration of static phase in our study group was 53.7 days. The duration of static phase ranged from 30 days to 100 days. 50% of cases required less than 6 weeks of static phase.

Joshi et al, recommended maintaining the fixator on static phase for double the period of distraction, which we also followed for our study. Paley et al, recommended maintaining the static phase for at least for 6 weeks. To maintaining the correction, we applied walking corrective cast for 6 weeks. Although, in case of severe deformity, it was applied for longer period. Joshi et al, kept his patients in walking cast for 6 weeks and later changed it to a boot allowing ankle movements and maintained it for another 6 weeks. After stopping corrective casts, all the parents

were taught foot stretching and ankle mobilization exercises. They were also asked to motivate the child to walk. During the follow-up, one 15 years old female patient asked to wear the AFO (ankle and foot orthosis) throughout the day and night but due to cosmetic reasons, she refused to wear it. Further we advised her to wear lateral shoe raise which also she didn't follow. However, the correction stayed maintained till last follow up Joshi et al, used an AFO made of a thermoplastic material which allowed for a minor adjustments and appropriate corrective shoes for long term use Paley et al, advocated an AFO to be maintained for full time for 6 months after removal.^{7,19}

CONCLUSION

Although, small size of the study and short duration of follow up are certain limiting factors in this case series. However, based on the findings of this case series study we can conclude that JESS distractor is a good alternative for neglected, resistant or relapsed cases of club feet. JESS distractor can avoid soft tissue as well as bony procedures to correct the deformity of CTEV while preserving the normal foot anatomy. The recurrence due to noncompliance can be prevented by counselling the parents about the importance of follow up till the child achieves a plantigrade foot and through the maintenance.

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