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Treatment of complex foot deformities with hexapod external fixator in growing children and young adult patients

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ABSTRACT

Background: Treatment of complex foot deformities in growing children and young adult patients is challenging. The traditional approach consists of extensive soft tissues releases, osteotomies and/or arthrodesis. More recently, distraction osteogenesis has been proposed as an alternative strategy. The aim of this study was to describe our treatment strategy and report clinical outcomes of the patients affected from complex foot deformities treated by distraction osteogenesis and hexapod external fixator.
Materials and methods: We retrospectively reviewed 10 consecutive patients with complex foot and ankle deformities treated from 2014 to 2016 at our unit. A TrueLok external fixator system was used in all patients. Final outcome was classified as good, fair and poor according to the criteria indicated by Paley and Ferreira. The results were also evaluated by the pre-operative and post operative American Orthopedic Foot and Ankle Score (AOFAS) and The Manchester-Oxford Foot Questionnaire (MOXFQ).
Results: A plantigrade foot was obtained in eight patients at the end of treatment, while in two patients a recurrence of the deformity was noted. Result was classified as good in 6 patients, fair in 2 patients, and poor in 2 patients. The AOFAS score improved from 33.9 ± 21.2 pre-operatively to 67.25 ± 15.1 post-operatively ($p = 0.005$). A statistically significant improvement was observed for the MOXFQ score as well (from 60.6 ± 23.3 to 33.0 ± 25.2, $p = 0.020$).
Conclusions: Our study shows that the TrueLok hexapod external fixator is a safe and effective tool in treatment of complex rigid foot deformities. Nevertheless, deformity recurrence can be observed in some cases and treatment remains challenging. Distraction osteogenesis should be reserved as a salvage solution for particularly complex cases and should be performed at dedicated specialized centers.

1. Introduction

Complex foot deformities are defined as multiplanar rigid deformity of the foot with or without foot shortening. They can be the end result of congenital abnormalities of the foot (*e.g.* clubfoot), or acquired conditions later in life (*e.g.* trauma, poliomyelitis, osteomyelitis, burn contractures, or other neuromuscular disorders) [1]. In all cases the goal of treatment is to achieve a plantigrade, pain free foot, with a size similar to the contralateral foot [2,3].

The classic approach to the treatment of complex foot deformities involves a single-step surgical correction with multiple osteotomies and/or wedge resections, soft tissue release, and arthrodesis [4]. The main drawback of this approach is the resulting shortening of the foot due to the large bone resection needed for correction [5,6]. Furthermore, it is an invasive surgery prone to several complications including wound healing problems, neurovascular injury, and incomplete deformity correction [1,3,6,7].

Distraction osteogenesis according to Ilizarov's principles represents an alternative approach [8]. The main advantage of distraction osteogenesis is to allow correction with lengthening rather than shortening. Furthermore, correction forces can be adjusted dynamically during treatment based on the response of the deformity to the correction [2,9]. The disadvantages are similar to those of any other prolonged treatment with an external fixator, including prolonged discomfort for the patient, pin track infection,

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and joint contractures/stiffening [10,11]. Furthermore, in patients older than 8 years of age, one or more osteotomies are often needed before starting lengthening treatment with the external fixator [2]. Although several studies have shown the feasibility of using Ilizarov's external fixator to treat complex foot deformities, there is a paucity of data on the advantages offered by newer hexapod external fixator systems. These systems allow simultaneous three-dimensional correction of the deformity using the same frame throughout the whole treatment, making correction easier and more comfortable for the patient [12].

We have been using the TrueLok Hexapod (TL-HEX) external fixator system at our unit since 2012. The aim of this study is to review our results with the correction of complex foot deformities using distraction osteogenesis in young adults and the TrueLok hexapod system.

2. Materials and methods

Following Institutional Review Board (IRB) approval, we retrospectively reviewed all consecutive cases of complex foot deformity cases treated with the TL-HEX external fixator system at our unit. All patients gave their written informed consent at the enrolment and were included into a prospective observational database.

2.1. Inclusion/exclusion criteria

All patients treated from January 2014 to December 2016 were included in this study. Indication for surgical treatment by distraction osteogenesis and hexapod external fixator was a painful non-plantigrade foot due to a multiplanar deformity that did not respond to conservative, orthotics, physiotherapy, and/or previous surgical treatment [1]. All patients had rigid joint deformities not responsive to passive foot stretching/manipulation. Inclusion criteria for enrolment in this study were: age at surgery $\leq$20 years, and follow-up after removal of the external fixator $\geq$6 months. All data were collected by an independent investigator not involved in the surgical management of the patients.

2.2. Pre-operative planning

A thorough clinical examination was performed in all patients in order to exclude any other associated lower limb deformities. Patients' gait pattern was also analyzed to exclude the presence of any muscle imbalance. A standard standing antero-posterior and lateral X-ray of the feet was used to confirm the diagnosis and assess bone anatomy. A template of the final external fixator frame was used in clinic before surgery to make sure that there will be no contact areas with skin (Fig. 1).

2.3. Frame set up

A modified long bone type frame consisting of a reference ring connected to the moving rings by 6 μm struts was used in all patients. In case of a V osteotomy approach (described below), the reference ring was connected to the tibial ring by two blocked rapid struts and to the two moving rings by 6 μm struts each (Fig. 2). With a supramalleolar osteotomy approach (described below), a similar frame was used but with two couples (ankle and forefoot deformity correction, respectively) of reference and moving rings connected by blocked rapid struts.

2.4. Surgical technique

The patient is placed supine on a radiolucent table with a soft pad under the ipsilateral buttock. The entire lower limb is prepped and draped, a tourniquet is placed at the thigh and inflated before starting osteotomy.

- *Step 1: fixation of the proximal ring.* A 1.8 mm lanceolate Kirschner wire tensioned at 110 N is positioned perpendicular to the tibial longitudinal axis at its middle third and is used to fix the

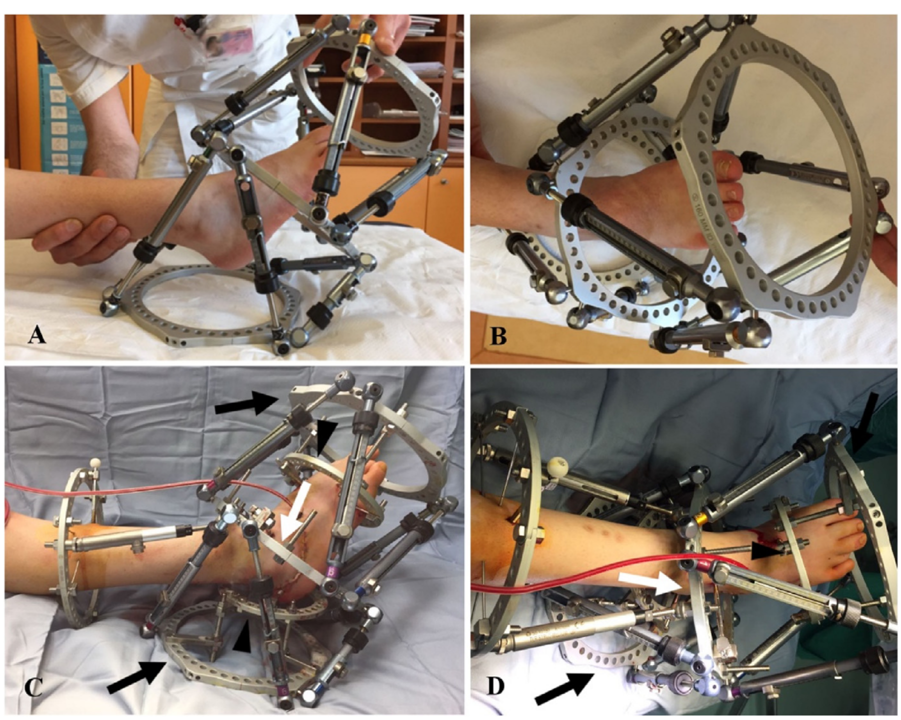

Fig. 1. (A, B) Pre-operative trial of the TL-HEX external fixator, (C, D) post-operative set up of the TL-HEX: moving rings (black arrows), reference ring (white arrow), and connecting rings (arrow heads).

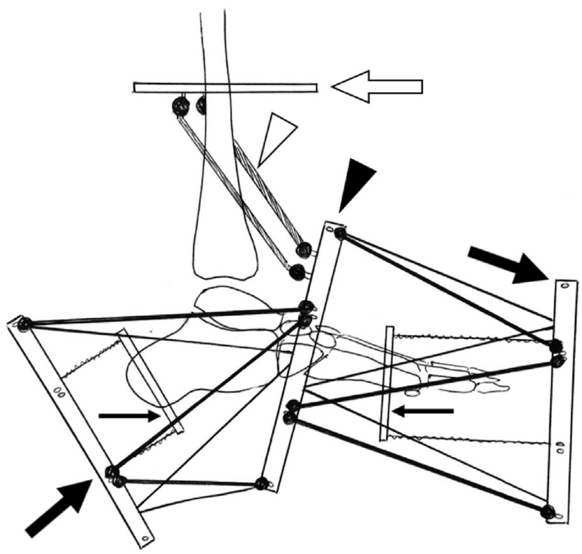

Fig. 2. Standard TL-HEX set up for correction of complex foot deformities. Tibial ring (white arrow), rapid struts (white arrowhead), moving rings (black arrows), reference ring (black arrowhead), connecting rings (little black arrows).

proximal ring. Two 4–5 mm screws are then added to further stabilize the ring to the bone;

- *Step 2: fixation of the reference ring.* The reference ring is fixed to the talus and to the anterior part of the calcaneus with one Schanz screw each;

- *Step 3: fixation of the moving rings.* The posterior moving ring is fixed to the moving part (posterior half) of the calcaneus by two screws; the anterior moving ring is fixed to the metatarsal bone by one screw fixed to the first metatarsal and two tensioned (90 N) K-wires passed in divergent pattern in order to avoid flattening of the plantar arch as previously described by Young et al. [13];

- *Step 4: osteotomies execution.* The chosen osteotomy technique is performed at this stage. In order to do this, one or two struts may be removed and positioned back at the end of the procedure;

We have been using two types of osteotomies depending on the deformity pattern: the V-osteotomy (VO) and the supramalleolar osteotomy (SO) (Fig. 3).

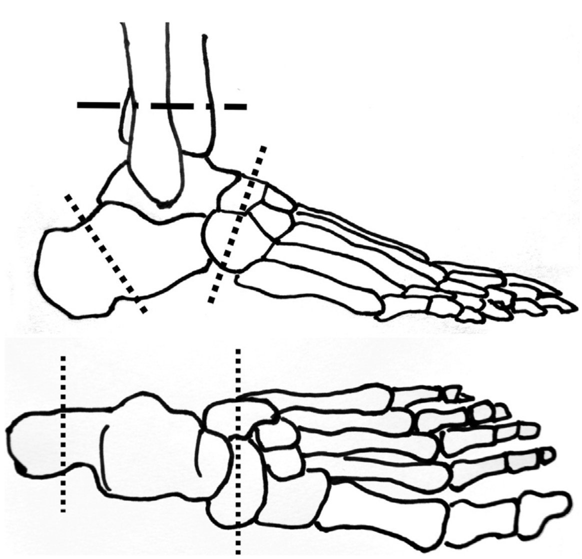

Fig. 3. The dash and dotted line shows the level of supramalleolar osteotomy, while dotted lines show the level of V-osteotomy.

The VO is performed through an Ollier approach to the lateral foot at the level of the midfoot (through cuboid and navicular bone) and hindfoot (calcaneus) with plantarly directed apex. VO allows independent correction of forefoot and hindfoot, therefore it is indicated in cases where the deformity if completely inside the foot. VO makes use of two center of rotation of angulation (CORA) for correction, one being located between the leg and hindfoot and a second one located between forefoot and hindfoot. VO is particularly suited for correction of calcaneus varus or valgus, prono-supination deformities, cavus deformities, plantar flextion deformities of the forefoot, abductus/adductus forefoot deformities, and foot lengthening [2].

The SO is used when the main deformity is located around the ankle joint. SO can be used to correct plantar flextion deformity due to distal tibia or talus deformity or to correct tibial torsion and limb length discrepancy [2]. The SO can be combined with the anterior part of the VO when a hindfoot or forefoot deformity is also present. The SO is performed through a standard anterior ankle approach. A drain is placed at the end of the procedure and removed on the second postoperative day.

- *Step 5: additional surgical steps.* Depending on the presence of associated contractures, soft tissue procedures can be added including Achilles tendon lengthening or Standler fasciotomy. Extra bone procedures are also sometimes added to correct associated bone deformities (*e.g.* proximal osteotomy for correction of a rotational malalignment or leg length discrepancy,Fig. 4);

- *Step 6: hexapod external fixator set up.* At the end of the procedure all struts are assembled in the final position and numbered according to manufacturer's instructions. The length of each strut is recorded (Fig. 1).

2.5. Postoperative care and correction

Standard daily pin care was started on postoperative day 1, whereas lengthening/correction program was started on post-operative day 3–5. At discharge from hospital, patients and their caregivers were trained on how to perform daily lengthening and pin care at home. The first post-operative X-ray was performed on post-operative day 7–10. After that, patients were monitored in clinic every two weeks until removal of the external fixator. Following this, patients were immobilized in cast for 4 weeks with no weight bearing allowed. A second weight bearing cast was prescribed after this for 4 more weeks.

2.6. Primary and secondary outcome parameters

Standard demographics and past medical and surgical history was collected in all patients. The primary outcome of the study was defined as good, fair, and poor according to previously described criteria: (1) plantigrade foot, (2) absent or limited pain (VAS score <2 after activity); (3) no recurrent deformity at follow-up; (4) patient satisfaction (yes/no) [2,7,14]. When all 4 criteria were met, the result was classified as good; when 3 out of 4 criteria were met, the result was classified as fair; when $\leq$2 out of 4 criteria were reached the result was classified as poor. Secondary outcome measures of the study included the American Orthopedic Foot and Ankle Score (AOFAS) and the Manchester-Oxford Foot Questionnaire (MOXFQ) [15,16]. Complications were recorded in all patients and classified as previously described by Paley as problems, obstacles, and complications [17].

2.7. Statistical method

Data are described as absolute and relative frequencies for categorical variables, while means, standard deviation (SD), medians, and range are used for continuous variables.

Fig. 4. Case example (patient #4). (A, B) Pre-operative clinical pictures demonstrating a left complex foot deformity associated with tibia intra-torsion, (C) the TL-HEX fixator was extended to the proximal tibia for intra-torsion correction. (D) Final clinical outcome.

Non parametric analysis (Wilcoxon test) for continuous variables and the Chi square or Fisher's exact test for categorical variables were used. A p-value less than 0.05 was considered statistically significant; all p-values were based on two-tailed tests. Statistical analysis was performed using SPSS for Windows (SPSS Inc., Chicago, Illinois, USA).

3. Results

A total of 10 patients (7 males, 3 females) were enrolled for this study, none of them was lost at follow-up at the time of this study. Median age at surgery was 14 years (range 13–16.5) and median of follow-up after removal of the external fixator was 13,5 months

Table 1
Demographic data and clinical data of our patient sample.

#	Age (years)	Sex	Side	Etiology	Previous treatments
1	12	M	Right	Arthrogryposis	P.S.T.R.; TA
2	13	M	Left	Clubfoot	P.S.T.R.; T.I.C.
3	12	F	Left	Arthrogryposis	P.S.T.R.; Jones procedure
4	18	M	Left	Myelomeningocele	T.A.T.L.; A.T.L.; T.A.; S.F.
5	18	M	Left	Arthrogryposis	P.S.T.R.; A.T.L.
6	14	M	Left	Clubfoot	P.S.T.R.; A.T.L.; T.I.C.;
7	15	F	Left	Clubfoot	P.S.T.R.; T.A.
8	14	F	Left	Myelomeningocele	S.F.; T.P.T.T.; L.S.C.O.
9	17	M	Left	Clubfoot	P.S.T.R.; A.T.L.
10	13	M	Left	Arthrogryposis	P.S.T.R.; A.T.L.; S.F.; D.C.O.; L.M.O.

P.S.T.R. = Posteromedial Soft Tissue Release; T.I.C.= Tibial Intrarotation Correction, T.A. = Triple Artrhodesis; T.A.T.L. = Tibialis Anterior Tendon Lengthening, ATL = Achilles Tendon Lengthening; S.F. = Steindler Fasciotomy; T.P.T.T. = Tibialis Posterior Tendon Transposition; C.R. = Capsular Release; T.T.L. = Tendon Transposition or Lengthening; O.A. S. = Osteo-Articular Surgery; L.S.C.O. = Lateral Sliding Calcaneal Osteotomy; D.C.O. = Dwyer Calcaneal Osteotomy; L.M.O. = Lelievre Metatarsals Osteotomies.

(range 7–27). All patients presented with a complex rigid foot deformity, the most common diagnosis was arthrogryposis and idiopathic clubfoot sequelae (Table 1). All patients had been treated with previous surgery consisting of capsular release, tendon lengthening or other bone procedures with unsatisfactory results. The average time from last surgical procedure before start of distraction osteogenesis was 48 ± 30 months.

In 9 patients a VO was used for deformity correction. One patient (case #3) required a SO for the correction of a rigid plantar flexion deformity due to tibio-talar malformation in addition to VO (forefoot cut only) for correction of an associated metatarsus adductus deformity and supination of forefoot.

A fully plantigrade foot was achieved in 8 patients (80%) at the end of treatment. Six patients also reported a significant improvement of pain with VAS score ≤ 2 (good outcome), whereas 2 patients still presented some pain with walking and activity (fair outcome). In the remaining 2 patients a recurrence of the deformity occurred at the end of treatment, and a triple arthrodesis was needed to correct their deformity (poor outcome) (Table 2).

Mean hospitalization time was 24 days $\pm$ 9.11. The mean latency time (TL) was 5 ± 1.49 days; the mean distraction time (TD) was 58 ± 19.78 days, and the mean consolidation time (TC) was 61 ± 36.57 days, and the mean time with the TL-HEX external fixator (TF) was 119 ± 36.39 days. The mean lengthening at the end of treatment was 34 ± 7.45 mm. The AOFAS and the MOXFQ scores are reported in Table 3. A statistically significant improvement was noted in all subdomains of the scores (Table 3).

We observed 1 problem (toes flexion which did not require any treatment), and 3 obstacles: 2 metatarsophalangeal joint subluxations (which required tenotomy of the flexor tendons and temporary arthrodesis by pinning), and 1 consolidation delay (treated with autologous bone graft from the iliac crest) which, however, occurred at a proximal tibial osteotomy site and was not related to foot deformity correction.

4. Discussion

Treatment of complex rigid foot deformities remains a difficult aspect of orthopedic practice. Correction by means of multiple osteotomies and arthrodesis is associated with several complications, including skin and wound healing problems, incomplete correction, and shortening of long axis of the foot [1,12]. Distraction osteogenesis allows a gradual and continuous control of the correction, this permits lengthening of the foot rather than shortening and a dynamic adaptation of the correction plan to patients' needs. Historically, the Ilizarov's frame has been used to treat this type of deformities [1,2,18]. However, the Ilizarov's frame has the disadvantage of needing multiple frame adjustments during treatment. Newer systems, like the TrueLok hexapod system, allow three-dimensional correction with a single frame, which does not require multiple adjustments during treatment [12]. Some of the available hexapod external fixator systems also have dedicated frames and software for foot deformities correction [11,19].

Although only a few studies with limited number of patients have been published, favorable results of distraction osteogenesis have been reported. Ferreira et al. [14] in a study based on thirty-eight feet with neglected congenital clubfoot deformity and treated by Ilizarov's method showed good results in thirty feet, fair in three, and poor in 5 feet. In a more recent study using Taylor

Table 2
Treatment data.

#	Osteotomy	Additional surgery	L	LT	CorT	ConT	MT	Outcome	Complications	FU
1	V	None	40	6	62	80	142	Good	M.P.J.S. (O)	27
2	V	None	29	3	72	61	133	Poor	None	23
3	S.O. + F	A.T.L.	30	6	42	127	169	Good	None	18
4	V	T.I.C. + A.T.L.	22	6	52	122	174	Good	T.C.D. (O)	17
5	V	T.T.A.	40	5	86	38	124	Fair	M.P.J.S. (O)	10
6	V	None	35	6	78	30	108	Fair	None	9
7	V	None	45	5	74	33	107	Good	T.F. (P)	10
8	V	T.T.A.	40	3	47	36	83	Poor	None	8
9	V	None	32	7	32	38	70	Good	None	7
10	V	None	25	3	30	48	78	Good	None	25

Osteotomy: V = V-osteotomy, SO + F = Supramalleolar Osteotomy + Forefoot V Osteotomy; **Additional Surgery**: ATL = Achilles Tendon Lengthening, TTA = Temporary Toes Arthrodesis, TL = Tibial Lengthening, TIC = Tibial Intrarotation Correction; **External Fixator Parameters**: L = Lengthening (mm), LT = Latency Time (days), CorT = Correction Time (days), ConT = Consolidation Time (days), MT = Mounted TL-HEX Time (days); **Complications**: MPJS = Metatarso-Phalangeal Joint Subluxation, TCD = Tibial Consolidation Delay, TF = Toes Flexion, O = obstacle, P = Problem, FU = Follow-Up (months).

Table 3
AOFAS and MOXFQ scores (n = 10).

	T_0	T_1	*p*-value
AOFAS x ± SD	33.9 ± 21.2	67.2 ± 15.1	**0.005**
MOXFQ walk x ± SD	71.7 ± 26.4	42.5 ± 28.4	**0.020**
MOXFQ pain x ± SD	47.5 ± 20.0	26.0 ± 25.4	**0.040**
MOXFQ social x ± SD	58.7 ± 27.3	27.2 ± 25.2	**0.030**
MOXFQ summary x ± SD	60.6 ± 23.3	33.0 ± 25.2	**0.020**

Data are shown as mean ± SD (standard deviation). AOFAS = American Orthopedic Foot and Ankle Score; MOXFQ = Manchester-Oxford Foot Questionnaire.

Spatial Frame (TSF), Floerkemeier et al. [7] showed good results in 8 out of 9 patients with multiplanar foot deformities. Similarly, Waizy et at. [11] in a study on nine patients treated with TSF reported good results in 7 patients. Eidelman et al. [20] in their series of 11 patients suffering from residual clubfoot deformities and treated with TSF classified their results into excellent (3 feet), good (6 cases), and poor (1 case). Hassan and Letts [21] reported satisfactory results in 8 cases out of 9. Eidelman and Katzman [19] reported the outcomes of 15 feet (13 patient) with multiaxial deformity. Treatment goal was reached in 11 patients, while in 2 patients a mild residual deformity was noted at the end of treatment with TSF. The same authors [22] also reported their experience in 7 patients (10 feet) with arthrogryposis treated with TSF and showed good results in all patients. Although results of these studies are hardly comparable because of different outcome measures used, our study shows similar results. We achieved complete correction in 8 patients out of 10. Result was good in 6 patients, fair in 2 patients, and poor in 2 patients. The 2 patients with poor results had a recurrence of their deformity after treatment and eventually needed a triple arthrodesis.

The number of complications in our study was similar to previously reported figures from other authors [7,11,20,23]. We observed 2 metatarsophalangeal joint subluxations that required surgical treatment and one toes flexion contracture which was treated with external manipulation. Eidelman et al. [20,23] reported 1 metatarsophalangeal joint and 1 talus subluxation during correction of foot deformities in their study. To avoid dislocation or subluxation of metatarsophalangeal joint, Waizy et al. [11] recommended temporary arthrodesis of these joints during distraction phase. We do not routinely perform this procedure because not all patients will develop this complication and not all those affected will need surgery. In cases where this happens, surgical treatment of the metatarsophalangeal joint subluxation can be delayed until removal of the external fixator. Finally, we did not observe any case of tibiotalar joint dislocations in our patients, this may be due to the fact that we always fix the talus to the reference ring which is also fixed to the tibial ring.

Our study has several limitations. While we acknowledge the small number of patients included in this study, we also understand the relatively small number of patients with complex foot deformities requiring this kind of surgical treatment [7,11,20,23]. Median follow-up in after removal of the external fixator was 13,5 months (range 7–27). This represent a potential limitation in case of late recurrence of the deformity.

5. Conclusion

To the best of our knowledge, this is the first study to report outcomes of complex foot deformities treated with TrueLok hexapod external fixator with long bone setting and not using a dedicated foot frame with results similar to those reported in literature. Our study shows that this approach is feasible and safe. It allows correction of the deformity achieving a plantigrade foot. The main advantage of this treatment is the possibility of having a near normal length foot. However, our study also shows that this treatment is plagued by a 20% failure rate, with some patients still needing an arthrodesis procedure at the end of treatment. Patients should be carefully counseled about all available options before surgery and should be informed about the possibility of failure and prolonged treatment times that this type of surgery requires. Distraction osteogenesis is a useful option and should be considered as a salvage solution in complex cases but it should be performed by surgeons with considerable experience with external fixator correction technique.

Declarations of interest

None.

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None.

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