Centralization of the foot in tibial and fibular hemimelia: preliminary results

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Purpose
To assess the midterm results of acute centralization of the foot in cases of tibial and fibular hemimelia associated with severe foot deformity and ankle dislocation.
Patients and methods
We treated 11 limbs in 10 patients: eight patients with tibial hemimelia and two patients with fibular hemimelia. All cases were associated with severe foot deformity (equinovarus in tibial hemimelia and equinovalgus in fibular hemimelia), dislocated ankle, and tight tendoachilis. There were seven boys and three girls, with a mean age of 2.35 years.
In tibial hemimelia, two limbs were type Ia, four limbs were type II, and three limbs were type IV according to Jones classification. In fibular hemimelia, all cases were type II according to Achterman and Kalamachi classification.
The mean preoperative limb-length discrepancy was 5.4 cm (range, 4.5–7 cm). We had acute centralization of the foot by means of calcaneofibular or tibiocalcaneal arthrodesis in cases of tibial or fibular hemimelia, respectively, and talectomy, fibular shortening, and tibiofibular synostosis were done. Transcalcaneal fibular Kirschner wire was inserted to maintain the foot position. First-stage lengthening by Ilizarov fixator was done, with 5-cm gain of length in seven cases, and second-stage lengthening was done in two cases.
Results
The mean follow-up period was 50 months. We achieved foot correction and centralization of the foot, which was stable in all cases, except one with failed tibiofibular synostosis in Jones type IV.
No neurovascular complications occurred. Wound dehiscence occurred in five limbs, unstable knee was seen in two cases, recurrent equinus of more than 20° was seen in three cases, and two cases had poor regenerate potential. All patients and families were satisfied with the foot procedure and were not satisfied with knee centralization procedure owing to knee instability.
Conclusion
Acute centralization of the severely deformed foot in cases of tibial and fibular hemimelia by calcaneofibular or tibiocalcaneal arthrodesis, respectively, can correct severe foot deformity, achieve plantigrade foot, and preserve the patient but was associated with many secondary procedures.
Level of evidence
Level IV.

Keywords:
centralization of the foot, fibular hemimelia, tibial hemimelia

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Introduction

Tibial hemimelia is a rare congenital anomaly, occurring in 1/1 000 000 live births [1].

The deficiency in tibial hemimelia is a spectrum of pathology, ranging from a congenital short tibia with relative fibular overgrowth to complete absence of the tibia. It is commonly associated with rigid equinovarus deformity of the foot and medial ray defects [2].

Jones *et al.* [3] classified tibial hemimelia into five types:

(1) Absent tibia with hypoplastic distal femoral epiphysis.
(2) Cartilaginous proximal tibial anlage with normal distal femoral epiphysis.
(3) Absent distal tibia.
(4) Absent proximal tibia.
(5) Hypoplastic distal tibia with distal tibiofibular diastasis with proximal displacement of the talus.

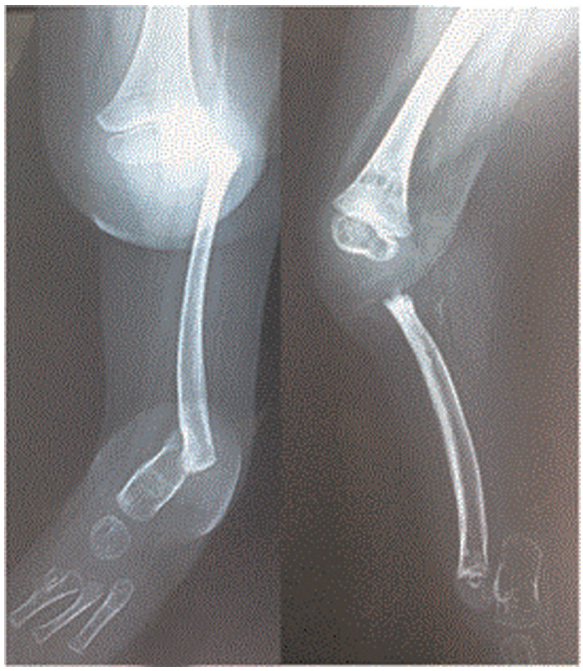
Amputation is recommended by many authors [1,4–9] as it is only one surgery with subsequent prosthetic fitting, but this concept is unaccepted in most middle-east countries because of different cultural beliefs; however, multiple reconstructive surgeries aiming to correct foot and knee surgeries and to equalize limb-length discrepancy are very difficult and challenging [10].

Tibial hemimelia is commonly associated with equinovarus deformity of the foot, which could be severe and rigid and associated with proximal migration of the foot and ankle dislocation (Fig. 1) and may require foot centralization by means of calcaneofibular or talofibular arthrodesis [3,4,8,10–17].

Centralization of the foot can be done gradually by means of gradual distraction of the foot relative to the fibula by external fixator (Fig. 2) to relocate the foot under the fibula, and then either talo or calcaneofibular fusion is done [2,11] or can be done acutely by means of acute relocation of the calcaneus under the fibula with calcaneofibular arthrodesis with fibular shortening and talectomy to facilitate reduction and to relieve tension on the neurovascular structures with great advantages [3,4,8,10,12–17].

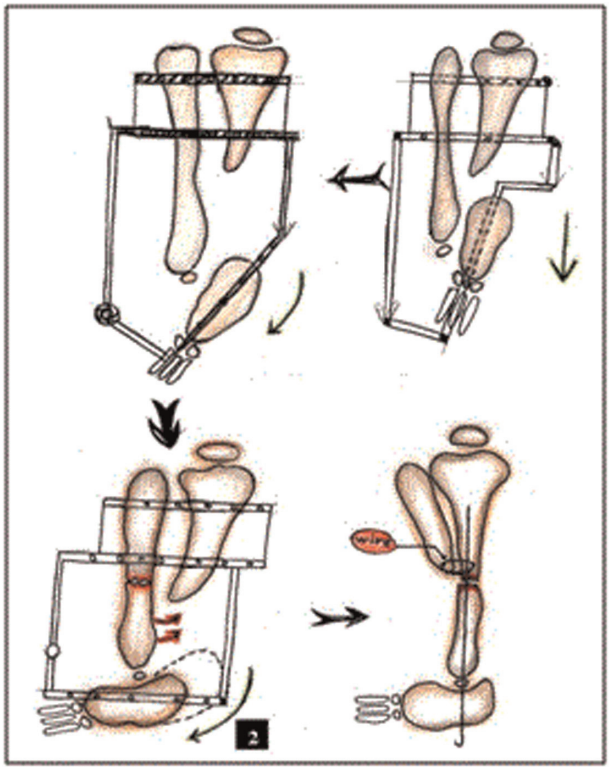
In severe types of fibular hemimelia (type II) according to Achterman and Kalamachi [18], the fibula is completely absent and associated with ACL deficiency, equinovalgus deformity, tarsal coalition, and absent lateral rays [19]; however, equinovalgus deformity may be severe, rigid, and associated with posterolateral dislocation of the ankle (Fig. 3), which

Figure 1



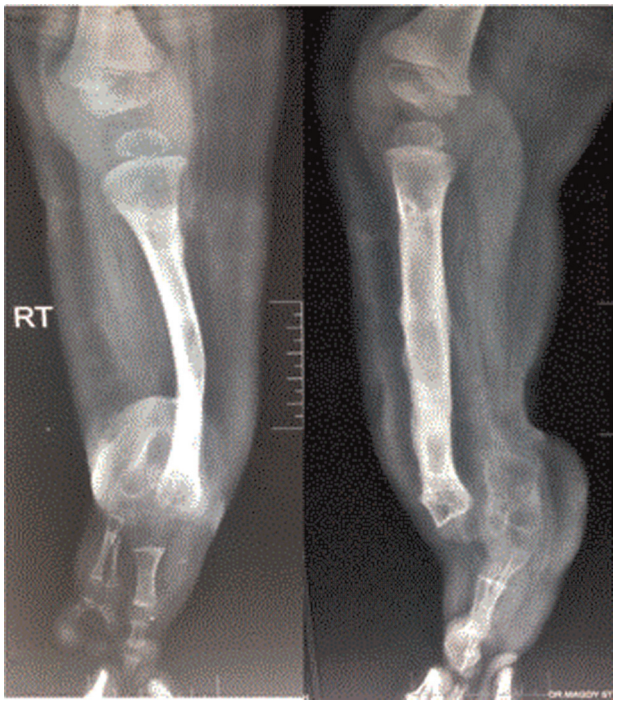
Complete dislocation of the ankle and knee in tibial hemimelia Jones type Ia.

Figure 2



Gradual centralization of the foot by external fixator.

Figure 3



Complete ankle dislocation in fibular hemimelia Kalamachi type II.

required centralization of the foot by means of calcaneotibial arthrodesis.

The aim of this prospective study is to assess the midterm results of acute centralization of the foot in case of tibial and fibular hemimelia associated with severe foot deformity and ankle dislocation.

Patients and methods

Between 2005 and 2015, acute foot centralization procedures were done in eight patients with tibial hemimelia and two patients with fibular hemimelia. The study was approved by the ethical committee of Mansoura University. All patients were operated upon in Mansoura University Hospital, after they have signed an informative consent. There were seven boys and three girls, with an average age of 2.35 years (range, 1.5–3 years). Right limb was affected in four, whereas left limb was affected in six, and one case had bilateral affection.

In tibial hemimelia cases, all feet had severe equinovarus deformity with subluxed or dislocated foot with very tight tendoachillis.

According to Jones classification [3], two limbs were type Ia, four limbs were type II, and three limbs were type IV.

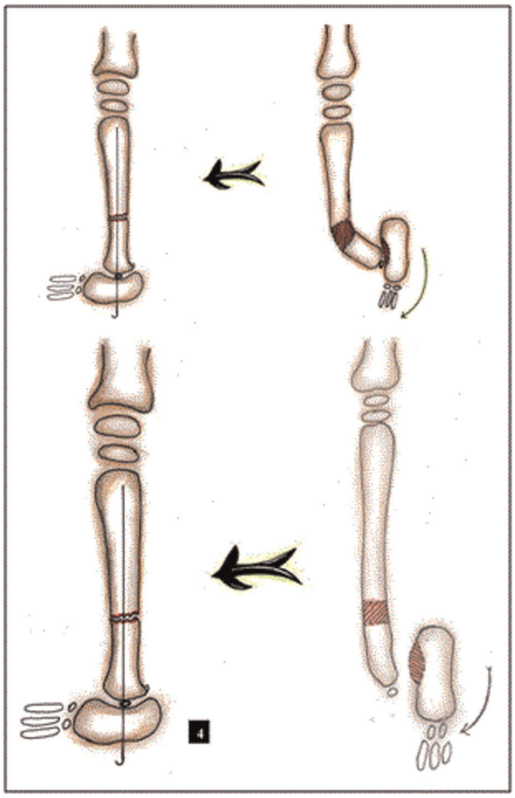
In fibular hemimelia cases, they are type II according to Achterman and Kalamachi [18] and had severe equinovalgus with dislocated foot with history of failed superankle procedure in one case [19].

Absent two rays were seen in six patients, and the mean initial limb-length discrepancy was 5.4 cm (range, 4.5–7 cm). The mean predicted limb discrepancy was 13 cm (range, 10–15.6 cm) according to the multiplier method [20,21], which guided us regarding the number of lengthening procedures.

Operative technique

Tibial hemimelia types I and II: in type I tibial hemimelia, through a curved posterolateral incision, a full-thickness anteromedial flap is created and refashioned. Tight tendoachillis was lengthened, and the ankle capsule was opened completely. The distal fibula was exposed, and the distal articular surface was reshaped to match with the trough created in the posterior calcaneus with preservation of the distal fibular physis for future fibular growth (Fig. 4).

Figure 4

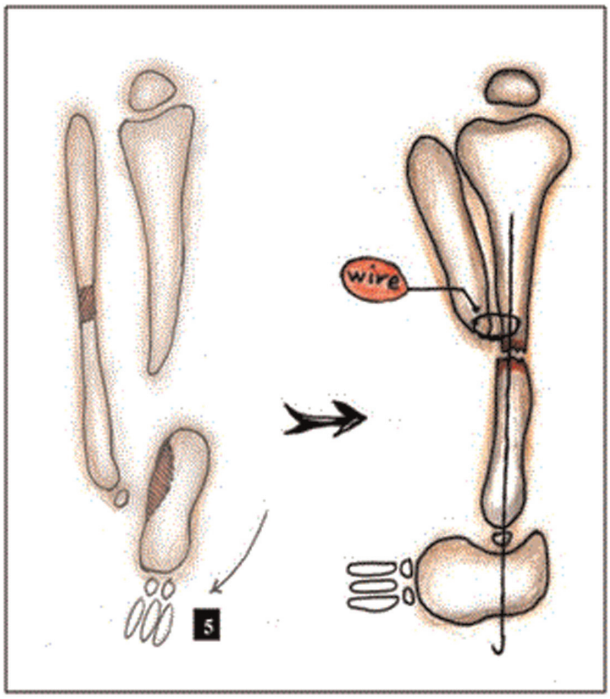


Acute centralization of the foot in type I tibial hemimelia and type II fibular hemimelia with fibular shortening and talectomy.

Talectomy or even excision of the talar part of the talocalcaneal coalition was done, and then excision of 1-cm fibular segment from the fibula was done to relieve tension of the neurovascular structures and enable reduction and centralization of the calcaneus under the fibula, and then transcalcaneal fibular K-wire was inserted with the foot in mild equinus position. If remnant of the proximal tibial metaphysis (type II) was present, a fibula protibia technique was done. Repair of the lengthened tendoachillis and capsule was done, and excision of the redundant skin was performed, with closure of the wound over a drain. Plaster cast was applied for 6 weeks. Weber patellar arthroplasty (22) was done in two cases with type Ia, where the fibula was transfixed to the patella.

Tibial hemimelia type IV (diastasis type): through a curved posterolateral incision, tight tendoachillis was lengthened, and the ankle capsule was completely opened. After excision of the fibular segment of 1 cm and talectomy, we did concomitant centralization of the foot and tibiofibular synostosis, which was fixed by a circulage wire, creating a Y-shaped configuration (Fig. 5). Two subtypes were seen: one with absent distal tibial physis, where synostosis was done and centralization of the calcaneus under the fibula and

Figure 5



Acute centralization of the foot in type IV tibial hemimelia with fibular shortening, talectomy, and tibiofibular synostosis.

fixed with K-wire, and the other with preserved distal tibial physis, where shortening of both tibia and fibula with tibiofibular synostosis and centralization of the calcaneus under both tibia and fibula were done.

Fibular hemimelia

Curved posterolateral incision was done, superankle procedure [19] was done with excision of the fibular anlage, lengthening of tendoachillis and peroneal tendons, opening of the capsule then talectomy and excision of tibial segment, where the tension over the neurovascular structure was relieved and centralization of the calcaneus under the tibia became easy. Transcalcaneal K-wire was inserted, followed by repair of lengthened structures and lastly, a plaster cast was applied for 6 weeks (Fig. 4).

Lengthening procedures by Ilizarov fixator was started at the age of 4 years with 5 cm gain of length in seven cases, whereas second stage of lengthening with 6 cm was done in two cases at the age of 8 years.

Results

We treated 10 limbs: eight with tibial hemimelia and two with fibular hemimelia with severe foot deformity. We acutely centralized the foot in all cases by means of calcaneofibular arthrodesis in case of tibial hemimelia and calcaneotibial arthrodesis in fibular hemimelia (Fig. 6). The mean follow-up was 50 months (range, 18–120 months) (Table 1).

We achieved correction in all cases in mild equinus, which were stable in all cases except one. Tibiofibular fusion occurred in all cases except one case with Jones type IV. No neurovascular complications were seen, but the most common complication is skin sloughing, which occurred in five limbs and treated appropriately by debridement and frequent dressing.

We achieved one-stage limb-lengthening procedure in seven limbs with a 5-cm mean length gained (range, 4–6 cm). Two cases did two-stage lengthening with 11-cm gained length. Two cases with poor regenerate needed autogenous bone graft.

All patients and parents were satisfied by the foot centralization procedure but were not satisfied with the knee centralization (two cases with type Ia) because of knee instability and progressive knee flexion contracture.

Residual deformity occurred in five limbs, which was treated in the next stage of lengthening. Unstable knee was seen in two cases with Jones type Ia, which needed bracing for walking. Dislocated knee was present in two cases, which were treated with an Ilizarov fixator and translation hinge, and both were corrected; one ended with a stiff knee and the other one was still unstable. Transient peroneal nerve palsy was seen in one case, which improved spontaneously. Superficial pin-tract infection occurred in most of the cases and treated appropriately by antibiotics. Recurrent severe equinus was seen in three limbs after 2 years and treated by supramalleolar osteotomy and posterior soft tissue release. The mean additional procedures were four per case (range, 2–9).

All cases except one walked independently with brace for limb-length discrepancy more than 4 cm and for unstable knee in two cases.

Discussion

Tibial hemimelia is commonly treated by amputation at different levels of either knee, Symes or Boyed amputation with subsequent prosthetic adaptation, with a good functional outcome [1,3–8,22,23], when amputation was done early it will be accepted by many children as congenital amputation [1]. However, this concept is not accepted in eastern countries because of different cultural beliefs and psychosocial factors, and the families push the orthopedic surgeons to

Figure 6

(a) Two-year-old boy with type II fibular hemimelia with severe equinovalgus deformity and failed superankle procedure. (b) Radiograph of type II fibular hemimelia with complete dislocation of the ankle with tarsal coalition and absent lateral two rays. (c) Acute centralization of the foot with fibular shortening, talectomy, and transcalcaneal K-wire. (d) Three-month postoperative follow-up. (e) Foot centralized with mild foot equinus. (f) Second stage lengthening of 6 cm with supramalleolar osteotomy to correct equinus ankle. (g) Equinus corrected and eight plate for correction of valgus knee. (h) Foot centralized with brace and shoe lift with 5-cm limb discrepancy.

reconstruct the limbs [2,11,24]. Limb lengthening and reconstruction to preserve the limb is difficult and challenging and requires multiple procedures and is more painful to the patients and families [10].

Tibial hemimelia associated with severe equinovarus deformity requires a reconstructive foot centralization procedure either talofibular arthrodesis [25], or calcaneofibular arthrodesis [4,5,7,8,12–16], with

Table 1 Patients' data and results

Case	Age (years)	Sex	Side	Type	Follow up (months)	LLD (cm)	Predicted LLD (cm)	Joint instability	Brace	Number of procedure	Complications	Additional procedures
1	3	♂	L	Jones Ia	42	7	15.6	Knee	yes	5	–Equinus ankle – Unstable knee – Flex deformity knee	– Lengthening of fibula – Weber arthroplasty – Lengthening tendoachillis
2	2.5	♂	R	Jones II	36	5	12.1	No	Yes	2	– Transient peroneal. nerve palsy	– Tibiofibular synostosis – Lengthening of fibula
3	2	♂	Bil	Jones IV	24	No	No	Lt. ankle	No	2	– Skin slough – Failure of synostosis Lt. ankle – Unstable Lt. ankle	– Tibiofibular synostosis
4	2.5	♀	L	Jones Ia	18	7	15.5	Knee	Yes	2	– Unstable knee – Flexion. Def. knee	– Weber arthroplasty
5	2.5	♂	R	Jones II	48	6	14.5	No	Yes	3	– Equinus ankle – Angular deformity.	– Lengthening of fibula – Lengthening of tendoachillis
6	3	♂	L	Jones II	92	4.5	10	No	Yes	5	– Angular deformity. – Varus ankle	– Lengthening of fibula – Tibiofibular synostosis – Supramalleolar ost.
7	1.5	♀	L	Jones IV	36	6	13.4	No	Yes	3	– Skin slough	– Tibiofibular Synostosis
8	3	♂	R	Jones II	120	4	11.2	No	Yes	9	– Sublux knee – Angular deformity. – Stiff knee	– Lengthening of fibula – Bone graft
9	2	♂	R	Kalamachi II	46	5	13	No	Yes	4	– Skin slough – Delayed consolid – Valgus knee	– Superankle – Lengthening of tibia – Bone graft
10	1.5	♀	L	Kalamachi II	34	4.5	12.1	No	Yes	5	– Equinus and valgus ankle	– Lengthening of tibia – Supramallular ost.

add, additional; Def, deformity; length, lengthening; LLD, limb-length discrepancy; Ost, osteotomy; Tib-fib, tibiofibular.

the advantages of preservation of the original foot, provision of a wide landing area for ambulation of the foot, and maintenance of the bone growth of the distal fibula [15,16].

Centralization of the foot can be done gradually by initial distraction and relocation of the foot under the fibula by means of Ilizarov external fixator with changing of hinge placement or using Taylor Spatial Frame and then calcaneofibular arthrodesis is done, with satisfactory results [2,11,24], or can be done acutely by acute calcaneofibular arthrodesis with talectomy and fibular shortening, which is a challenging procedure for cases of severe equinovarus

deformity associated with ankle dislocation and tight posteromedial structures, where there is a great risk of neurovascular compromise, and this can be avoided by doing fibular shortening and talectomy.

Our goals of treatment are achievement of plantigrade foot, stable functioning knee joint, and acceptable leg length equalization.

Centralized foot was stabilized in mild equinus position, which allows both Chopart and Lisfranc joint to give a smooth step [11,16,17]. We achieved a stable centralized foot in all cases except one with type IV with failure of tibiofibular synostosis (Fig. 7), where the centralized fibula was displaced early and became unstable and needed bracing.

Residual equinus deformity more than 20° were seen in three limbs, which required posterior soft tissue release and supramalleolar osteotomy. Wound dehiscence is the most common complication encountered with our procedure and seen in four cases.

Yousef Ahmed *et al.* [10] in their report on eight patients with type II tibial hemimelia achieved satisfactory results. They did acute centralization of the foot followed by tibiofibular synostosis and later Ilizarov distraction lengthening to correct limb-length discrepancy. They reported no complication during foot centralization.

In types II and IV tibial hemimelia, we did both procedures (acute centralization and tibiofibular synostosis) at the same sitting without vascular compromise, whereas in type Ia tibial hemimelia, centralization of the fibula under the femur by either Brown or Weber patellar arthroplasty [14,22] should be done first followed by foot centralization at later stage. The poor results of Brown procedure [7,8,17,24,26,27] with progressive knee flexion contracture leads us to use the Weber arthroplasty, which was done in two cases but the knee was still unstable and required bracing.

Wada *et al.* [17] recommended fibulofemoral fusion if the patella is absent and Weber patellar arthroplasty if the patella is present, but with unpublished results.

Paley and Chong [2] with their new classification and strategies for tibial hemimelia recommended correction of knee flexion contracture with frame distraction until fibula and femur become collinear and posterior capsulotomy before Weber patelloplasty and reattach the remnants of medial and lateral hamstring to the fibula at the end of the procedure, and also they recommended foot distraction relative to the fibula until it became centralized under the fibula.

In cases with absent patella, they recommended a preliminary step of distraction of the fibula relative to the femur and the foot relative to the fibula, and then they anchor the fibula to the distal femur by creating an interosseous ligament by a half of biceps tendon then the four muscles (half of biceps tendon, semitendinosus, tensor fascia lata, and adductor Magnus) attached to the quadriceps and then to the fibula to act as patellar tendon, and finally, the ankle is fused [2].

We had two cases of unstable knee after fibular centralization, with unsatisfactory outcome reported by the patient's family. We offered them two options, either amputation (knee disarticulation) or fibulofemoral fusion, which is more realistic for our patients.

Regarding the issue of limb-length discrepancy, the patients wore prosthesis to compensate for big discrepancy till multiple lengthening procedures (at least 3) were done, as the discrepancy exceeded 12 cm in most of the cases as estimated by the multiplier method [20,21].

Most complications that occurred during the lengthening procedures were minor and treated accordingly: pin-tract infection (antibiotics), poor regenerate (autogenous bone graft), transient peroneal nerve palsy (spontaneous recovery), residual angulation (correction by Ilizarov during next lengthening or hemiepiphysiodesis), and residual equinus more than 20° (posterior soft tissue release and supramalleolar osteotomy).

Most of literature describes the technique of foot centralization by calcaneofibular arthrodesis in tibial hemimelia, and to our knowledge, no publication describes such technique in fibular hemimelia with severe equinovalgus deformity and dislocated ankle to achieve calcaneotibial arthrodesis and stable plantigrade foot.

We recommended this protocol of management

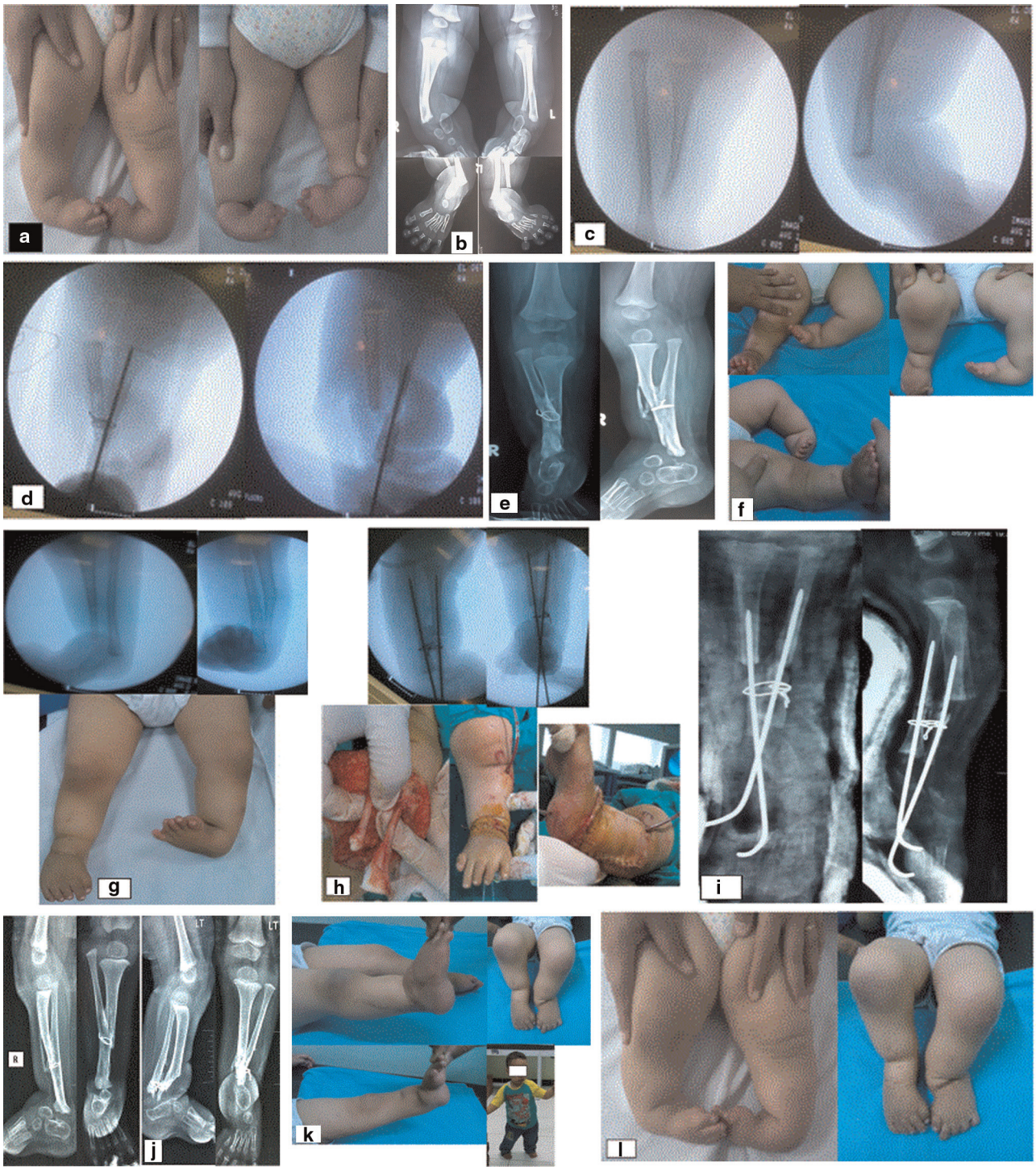
Before 1 year, gradual stretching of the foot deformity by gentile manipulation like Ponseti technique.

At age of 1 year, centralization of the fibula at the knee.

At age of 2 years, acute centralization of the foot and tibiofibular synostosis.

At age of 4 years, first lengthening.

Figure 7



(a) Bilateral tibial hemimelia with severe equinovarus deformity. (b) Bilateral radiograph of tibial hemimelia type IV. (c) Intraoperative radiograph of right tibial hemimelia type IV with short tibia and absent distal physis. (d) Intraoperative radiograph with foot centralization, fibular shortening, and tibiofibular synostosis (Y configuration). (e) Six months postoperative follow-up with centralized foot and tibiofibular fusion. (f) Right foot corrected and centralized. (g) Preoperative radiograph and picture of left foot with preserved distal tibial physis. (h) Intraoperative radiograph and picture of the centralized foot with correction of the left foot. (i) Three-month postoperative follow-up of the left side with transcalcaneal K-wire of both tibia and fibula. (j) Right side, centralized foot with sound tibiofibular fusion, left side, centralized foot with failed tibiofibular synostosis. (k) Both feet corrected and centralized and patient walks without braces independently. (l) Preoperative and final pictures of both sides.

At age of 6–10 years, second lengthening.

At age of 12–14 years, third lengthening.

Limitations of our study are the limited number of cases with different stages of affection and short follow-up.

We conclude that acute centralization of the severely deformed foot in cases of tibial and fibular hemimelia by calcaneofibular or calcaneotibial arthrodesis, respectively, can correct severe foot deformity, achieve plantigrade foot, and preserve the patient foot to be used as biological prosthesis but with many secondary procedures.

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Nil.

Conflicts of interest
There are no conflicts of interest.

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