Congenital pseudoarthrosis of the tibia: role of the fibula

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Objective
Congenital pseudoarthrosis of the tibia is still a mystery and a great challenge to pediatric orthopedic surgeon. Owing to the different morphology of congenital pseudoarthrosis of the tibia, there are many treatment options with many debatable points. The fibula is a main strut that protects the tibial union and has implications on the orientation of the ankle.
Patients and methods
Nine patients had congenital pesudoarthrosis of the tibia, and their average age at the time of operation was 3.8 years; four of these nine patients had intact fibula (group 1), whereas five had associated pseudoarthrosis of the fibula (group 2). The treatment method was decided depending on the condition of the fibula, whether it was intact or affected by pseudoarthrosis. In cases with intact fibula, tibiofibular fusion was done. However, in cases with associated fibular pseudoarthrosis, the pathologic pseudoarthrotic tissues were resected and the tibia and fibula were fixed with intramedullary wires and Ilizarov ring fixator.
Results
A union rate of 100% was achieved without a refracture till the last follow-up. Good tibial alignment was achieved in all cases, and wide cross-sectional area at the pseudoarthrosis site was obtained in all cases. One case with fibula plus +1 position was complicated with a varus deformity of the ankle.
Conclusion
Addressing the fibular problem safeguards against refracture and protects against ankle joint deformity. Achieving tibial union with wide regenerate or wide fusion mass, good tibial alignment and fibular union is mandatory for preventing refracture. The article was level of evidence IV.

Keywords:
congenital pseudoarthrosis of the tibia, fibula, pseudoarthrosis of the fibula, refracture

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Introduction

Congenital pseudarthrosis of the tibia is a rare entity [1]. Pseudarthrosis of the fibula is associated with congenital pseudoarthrosis (CPT) in one-third of the cases [2].

There are different treatment options of CPT, including different bone-grafting techniques, the Ilizarov method [3], intramedullary rodding [4–7], and combined external fixation and intramedullary (IM) rodding [3,8,9].

Union in CPT has been achieved with the use of Ilizarov ring fixator [9] and intramedullary fixation [10]. However, complications most often occur like limb-length discrepancy (LLD), foot deformity, and ankle valgus deformity [11]. Refracture is considered the most serious complication [5,9].

Neurofibromatosis, surgery before age of 3 years, and failed previous surgery are negative prognostic factors.

However, this is not always the role; the consideration of NF1 as a negative prognostic factor for bone union in CPT is debatable, as comparable rates of union can now be achieved in these patients [12].

Young children are more liable to refracture owing to the small diameter of the bone; however, some authors found that the patients younger than 3 years had better results compared with those in older groups regarding the union rate and the residual abnormalities [13].

A failed previous surgery has an adverse effect owing to unfavorable scarred soft tissue with avascular bed that shows marked osteoclastic activity with potentially retained diseased periosteum and possible retained hardware and marked atrophic bone ends with poor bone quality [8]. Failure of bony union after three surgical attempts was considered an indication for amputation [14], but this indication is not valid nowadays [15].

The association of persistence of the fibular pseudarthrosis and tibial refracture and valgus deformity of the ankle is well reported [15,16].

In this study, the treatment method of congenital pseudarthrosis of the tibia by either tibiofibular synostosis or Ilizarov bone transport was decided according to the fibular condition, either normal or pseudoarthrotic, respectively.

Patients and methods

This study was approved by the university review board, and confirmed consent was obtained from the parents of all patients. This study was conducted at Mansoura university hospitals in the period between May 2009 and March 2014. Nine patients with congenital pesudoarthrosis of the tibia were included, comprising seven male and two female, with the average age at the time of operation being 3.8 years (range: 2.5–5 years). Of these nine patients, four had intact fibula (group 1), whereas five had associated pseudoarthrosis of the fibula (group 2).

All cases had unilateral involvement; the right side was involved in five (55.6%) patients and the left in four (44.4%). All patients had neurofibromatosis type 1, but neurofibromas were not found at the site of pseudarthrosis in any of our patients as evidenced by pathological examinations which revealed fibrous hamartomas rather than neurofibromas. All patients in this study had a history of a failed previous surgery.

The treatment method was decided depending on the condition of the fibula, either it was intact or affected by pseudoarthrosis. In cases with intact fibula, tibiofibular synostosis proximally, distally, and at the pseudoarthrosis site (tibia-fibula one mass fusion) was done for group 1 (four cases) and splinted with a long leg cast till tibiofibular fusion was achieved.

However, in cases with associated fibular pseudoarthrosis (group 2, five cases), the pathologic tibial and fibular pseudoarthrotic tissues were resected proximally and distally till reaching the healthy bone. The tibia and fibula were fixed with intramedullary wires inserted retrograde from distal to proximal and Ilizarov ring fixator to achieve fixation and alignment preservation. Bone transport through distraction osteogenesis is going on at the proximal metaphysis to restore the bone length and compensate for the excised bones. The Ilizarov fixator was maintained till the regeneration tissue was consolidated. A long leg cast was applied for six weeks after Ilizarov fixator removal, and ankle-foot-knee brace was applied thereafter.

The patients were followed up at 2 weeks postoperatively, then monthly till union, and every 6 months till the last follow-up for clinical and a radiographic assessment.

The mean preoperative LLD was 3.6 cm (range: 2–5 cm).

The patients were classified according to El-Rosasy-Paley, Boyd, and Crawford classifications [8,17,18]. All nine cases were El-Rosasy-Paley type II CPT (i.e. atrophic bone ends on radiographies, mobile, with previous surgery) as well as Crawford type 4 with discontinuity of the tibia in the initial radiographies, and four were Boyd type 4, with pseudarthrosis, sclerotic segments, and fracture tibia and the other five patients were Boyd type 5 with associated dysplastic fibula.

Radiographs were done to assess ankle joint mortise. The fibular leveling was evaluated according to Elgohary and Elmoghazy scale, a modification of the Malhotra scale [19] (Fig. 1), where

Zero position (0) is the normal position, with the distal fibular growth plate and the talar plateau leveled.

The fibula minus 1 position (fibula-1), where the distal fibular growth plate lies between the top of the talus and distal tibial growth plate.

The fibula minus 2 position (fibula-2), where the distal fibular growth plate is leveled with the distal tibial growth plate.

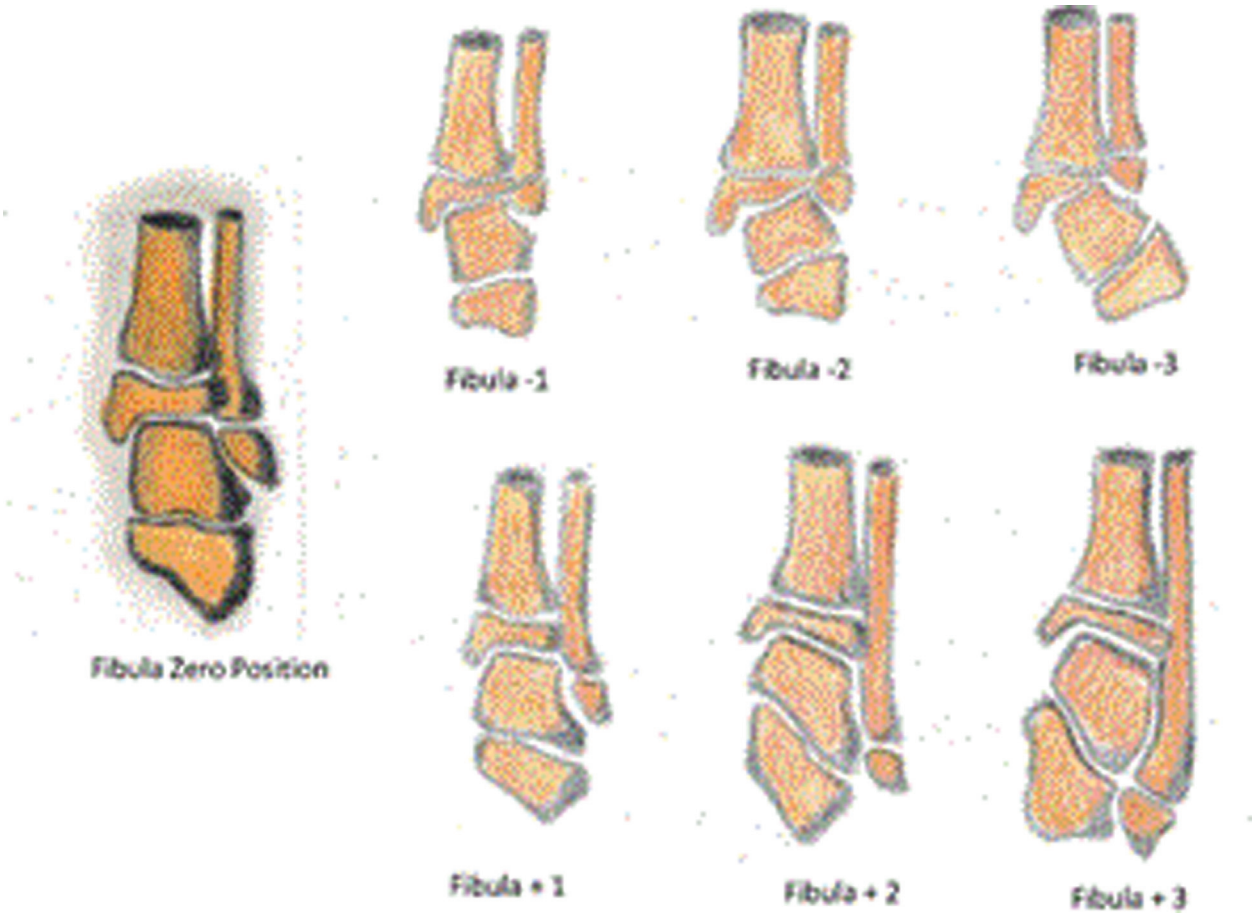
The fibula minus 3 position (fibula-3) where the distal fibular growth plate lies proximal to the distal tibial growth plate.

The fibula plus 1 position (fibula+1) where the distal fibular growth plate lies between the talar plateau and the middle of the lateral surface of the talus.

The fibula plus 2 position (fibula+2) where the distal fibular growth plate lies between the middle of the lateral surface of the talus and above the level of talocalcaneal joint.

The fibula plus 3 position (fibula+3) where the distal fibular growth plate lies at or distal to the level of talocalcaneal joint.

Figure 1



Illustrations showing Elgohary and Elmoghazy scale, a modification of the Malhotra scale.

Surgical technique

Patients were under general anesthesia and placed on a radiolucent table in the supine position. The lower limb was prepared and draped. A tourniquet was placed on the thigh and inflated after the limb is elevated and exsanguinated.

For group1 with intact fibula

A longitudinal anterior incision was made over the pseudarthrosis. The diseased thick periosteum, hamartomatous tissue, and sclerosed bones of the tibia were resected. The fibrous tissues at the non-union site of the tibia was carefully debrided, protecting the anterior and the posterior tibial neurovascular bundles, and careful resection of the bone ends was done till healthy ends were reached. Detach the periosteum and interosseous membrane on the lateral surface of the tibia and the medial surface of the fibula for several centimeters above and below the pseudarthrosis site and shingle the opposing cortical surfaces of both tibia and fibula. Cancellous and corticocancellous bone grafts from the ilium are collected to fill the space between the tibia and the fibula. The graft was maintained in position through the help of screws transfixing the fibula to the tibia. Distal fibular epiphysiodesis was done in cases with fibular over length. An above-the-knee cast was then applied till the fusion was evidenced radiologically. Full weight bearing in the cast was allowed after about 2 months.

For group 2 with pseudarthrotic fibula

An anterior longitudinal incision at the site of pseudarthrosis was done for the tibia and another lateral incision for the fibula. The diseased thick periosteum, hamartomatous tissue, and sclerosed bones of the tibia and fibula were resected. The fibrous tissues at the non-union site of the tibia and fibula was carefully debrided protecting the anterior and the posterior tibial neurovascular bundles, and careful resection of the bone ends was done till healthy ends were reached.

IM Kirschner wires were inserted retrograde for both the tibia (transcalcaneal) and fibula to maintain alignment, and the Ilizarov fixator was then applied with two rings proximal and one ring

Figure 2

(a) Radiographies at presentation for a female child aged 2.5 years with congenital pseudoarthrosis tibia and fibula with previous surgery of nonvascularized fibular graft. (b) A photograph of the child with shortening of left leg by 3 cm. (c) AP and lateral radiographies with the Ilizarov circular external fixator in place after consolidation of the regenerate and bone graft for the docking site with intramedullary K-wires. (d) 4 years of follow up with union of the pseudoarthrosis and bone remodeling with mild procurvatum of the distal epiphysis. (e) Photographs at the last follow-up with 3-cm limb-length discrepancy (shortening) and good range of motion of ankle and knee.

distal to the pseudarthrosis site with the fibula fixed proximally and distally to proximal and distal rings and another calcaneal 5/8 ring was added for better stability. Metaphyseal corticotomy was done between the proximal and second rings, and bone transports starts from proximal to distal till reaching the docking site, and then lengthening continues to correct LLD. The docking site was attacked and a large amount of cancellous bone graft from the iliac bone was placed around the pseudarthrosis site of the tibia and fibula and enveloped by free periosteal graft, which was harvested from the internal side of the iliac bone.

We encouraged the patients to partial weight bearing through the help of walking ring, which was connected to the 5/8 calcaneal ring.

Radiographs for the leg from the knee to ankle were done in each follow-up visit to assess the regenerate, bone union, and complications if any.

Removal of the Ilizarov fixators was done under anesthesia in the operating theater after manual testing for union and bone consolidation becomes certain.

Results

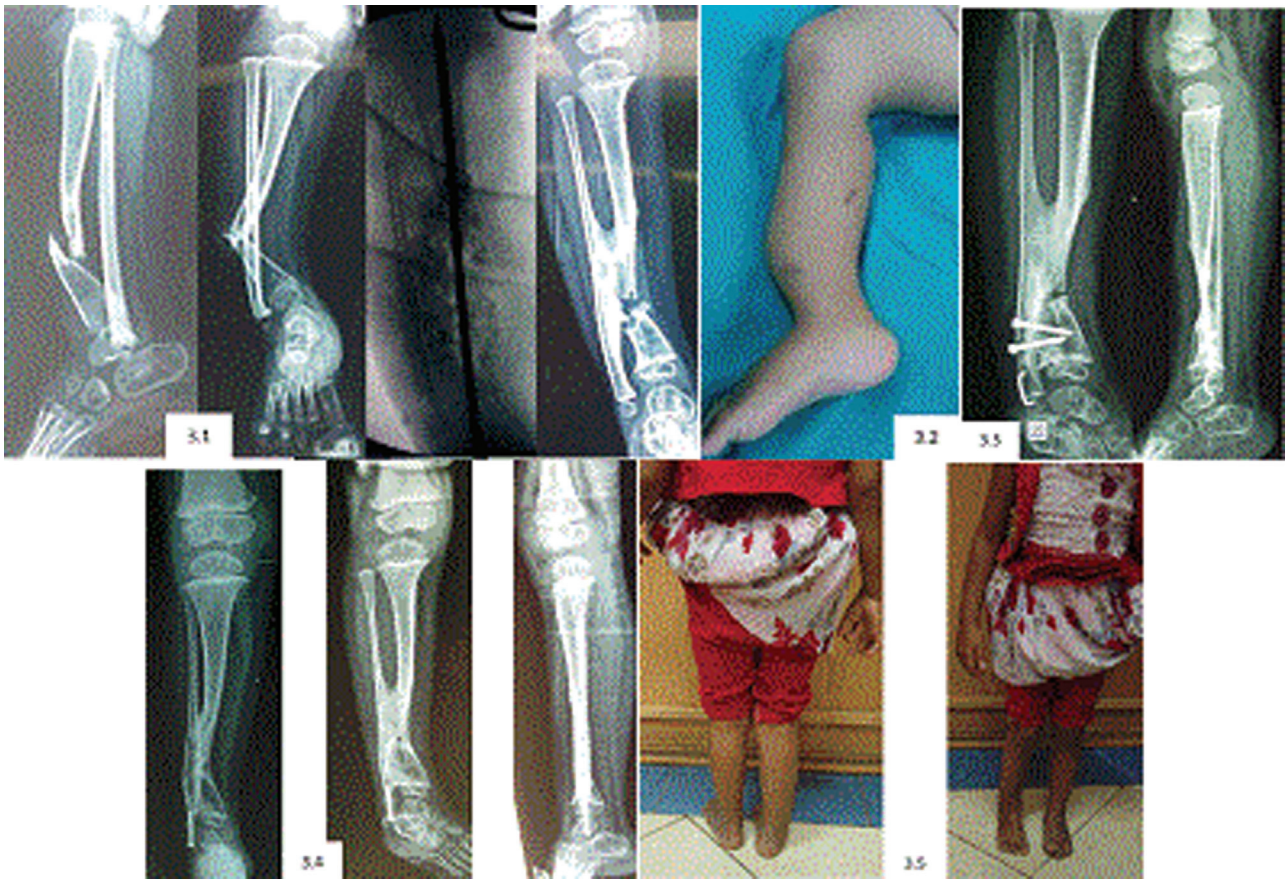
All patients in this study had a history of failed previous operation: five of them had histories of failed Masquelet technique, two had failed nonvascularized fibular graft (from mother), and the remaining two patients had failed previous resection of the pseudoarthrosis and fixation with Ilizarov ring fixators elsewhere. The patients were followed up for a mean period of 3.7 years (range: 2–6.2 years). In six patients, the pseudoarthrosis sites were in the middle lower third, and in the other three patients, they were in the extreme distal tibia (Figs 2–4).

The mean duration of treatment with the Ilizarov fixators was 5.6 months (range: 4–8 months).

Limb lengthening was achieved in five patients (group 2) who had associated pseudoarthrosis of the fibula and fixed with Ilizarov fixator. All those five achieved union at the docking site. No case was complicated by a refracture by the last follow-up.

Average limb length discrepancy for the fouor patients in group 1 was 2.1 cm (range: 1.5–3 cm). The mean length of the defect was 3.2 cm (range: 2.5–4 cm). The average postoperative limb length discrepancy was

Figure 3



(a) Preoperative radiographies of a 4-year-old female child with congenital pseudoarthrosis of the tibia at the lower third with intact fibula and distal tibiofibular diastasis with history of failed Masquelet technique. (b) A preoperative photograph showing the site of pseudoarthrosis. (c) Radiographies showing fusion mass between tibia and fibula extended to middle and lower thirds of tibia and fibula (with back out of the stable). (d) Fusion mass is consolidated and mature. The screws, stable at the fusion site, are removed and an intramedullary screw was inserted for epiphysiodesis to stop distal fibular growth. (e) A last follow-up photograph with mild varus ankle.

1.3 cm (range: 1–2 cm), and the average LLD at the last follow-up was 2 cm (range: 1.5–2.5 cm).

The average preoperative limb length discrepancy for the five patients in group 2 was 4 cm (range: 3–6 cm). The average postoperative limb length discrepancy was 2 cm (range: 1.5–4 cm). The mean length of the defect was 3.5 cm (range: 4–6 cm), and the average LLD at the last follow-up was 3.5 cm (range: 2–5 cm),

The final LLD was compensated with shoe lifting and was accepted by the parents, except for two cases with LLD of more than 3 cm that were scheduled for another stage of lengthening.

The union of tibial pseudarthrosis was achieved at a mean time of 5.1 months (range: 3–8 months). Fibular union was achieved in all patients. The tibial and fibular intramedullary K-wires were removed in all patients after solid union to avoid stiffness of the subtalar and ankle joints.

Patients in group 1 had achieved the tibiofibular fusion in an average period of 4 months (average: 3–5 months).

Good tibial alignment was achieved in all cases. Wide cross-sectional area at the pseudoarthrosis site was obtained in all cases as observed on the anteroposterior and lateral radiography views by comparing the width of the bone at the site of pseudoarthrosis to the bone proximal and distal to it in serial radiographs at different stages of follow-up. Narrowing of the regenerate was not found in any case, indicating adequate resection of the pathologic tissues.

Two cases in group 1 had a fibula plus (fibula +2), and distal fibular epiphysiodesis was done for both of them, where one case was corrected to normal zero position by the last follow-up and the other case was corrected to fibula plus +1 position, but it was complicated with a varus deformity of the ankle. Table 1 summarizes the distal fibular position in this study.

Figure 4

(a) Radiographies showing congenital pseudoarthrosis tibia and fibula in a female child aged 3.5 years with history of failed Masquelet technique. (b) A photograph at presentation with limb-length discrepancy of 5 cm. (c) Ilizarov circular external fixator with proximal bone transport over IM wire, the transported segment reached docking site. (d) Last follow-up with consolidation and remodeling of the regenerate, and union of both tibia and fibula with good alignment. (e) Last follow-up photographs with limb-length discrepancy of 5 cm.

Table 1 Summary of the position of the distal fibula according to Elgohary and Elmoghazy scale

Case	Preoperative position	Postoperative position
1	–3	–1
2	–2	–1
3	2+	+1
4	+2	0
5	–2	–1
6	–2	0
7	–2	–1
8	–1	0
9	–2	0

Superficial pin-tract infections were encountered in four patients in group 2 (80%), which were resolved with thorough disinfection and antibiotic therapy according to the culture.

Malorientation of the tibial epiphysis was encountered in two cases: recurvatum of the proximal tibial epiphysis in one cases and procurvatum of the distal tibial epiphysis in the other, without affecting knee or ankle functions or ranges of motion or the overall alignment of the tibia (Fig. 2d and Fig. 4d). By the last follow-up, all patients in this study were able to walk unaided. For both groups by the last follow-up, the range of movement of knee and ankle was comparable on both sides.

Discussion

The natural history of CPT is a dilemma in itself; the pseudarthrosis is amenable to nonunion, progressive LLD, deformity, and refracture after union [12,17].

The goal of treatment is to achieve union and to maintain this union without bone or joint deformity and correction of LLD [4,5,8].

There are many surgical options for management of CPT like vascularized fibular grafting, IM stabilization, Ilizarov external fixation, and amputation [3,4,9,14,20].

In this study, all patients had neurofibromatosis; all of them had a history of a failed previous surgery.

Union was achieved in all patients in this study. Patients with pseudoarthrotic fibula were managed according to a technique recommended by Paley, where a free periosteal graft harvested from the iliac wing and wrapped around the area of pseudarthrosis after complete excision of the diseased bone and periosteum was used in association with Ilizarov ring fixator and intramedullary rodding [15].

In this study, the authors considered the fibula as the cornerstone to safeguard against tibial refracture and ankle valgus. Fibular pseudarthrosis should be attacked carefully aiming to achieve fibular union, and intact fibula should not be interrupted. For cases with intact fibulae, proximal and distal tibiofibular synostosis (tibia–fibula one mass fusion) was done, and this wide fusion mass between tibia and fibula protects against refracture (Figs 2–4).

For cases with fibular pseudarthrosis, we did resection of the diseased bone and periosteum. IM K-wires were

applied for both the tibia and fibula to maintain alignment and the Ilizarov fixator was applied. Proximal metaphyseal coticotomy was done for bone transport, and when reaching the docking site, autogenous cancellous bone graft and periosteal graft for both tibia and fibula were applied.

Patients in this study were followed-up for a mean period of 3.7 years. All the cases in group 1 had achieved union at the synostosis site, and all cases in group 2 had achieved union of both tibia and fibula. No case in this study was complicated by a refracture through the period of follow-up, and this is probably lower than the actual rate, as no case in this study had reached skeletal maturity during the period of follow-up.

Different studies used Ilizarov circular fixators, where success rate of union ranged from 75–100% and a refracture rate ranged from 19–44% [9,21,22], and intramedullary rodding, with 33–87% union rate and refracture rate up to 57% [3–7].

Thabet *et al*. [15] had 20 patients with CPT treated with combined IM rodding of the tibia and fibula and circular external fixation with additional periosteal and bone grafting, With a mean age at the index operation of 4.2 years, eight patients had a history of at least one failed operation. All patients (100%) achieved union. They found that a high probability of refracture was associated with fibular pseudoarthrosis as refracture occurred in eight of their patients, and six of them had fibular pseudarthrosis [15].

In our study, there was no case of refracture in spite of removal the intramedullary wires after solid union to avoid ankle stiffness. This may be due to the corrected alignment of the tibia, and wide bone section area at the pseudarthrosis site, and also because fibulae were united in all cases in our study, which adds to the stability and safeguarding against refracture.

Refractures are frequent after union. They can occur early as long as the medullary canal has not become permeable again and can be partly avoided by a protective brace [23].

There is a general consensus that proper realignment of the tibia is mandatory for union, and intramedullary fixation is the best method to correct and maintain alignment during growth and for guarding against refractures [23].

Many authors reported ankle joint stiffness after fixation through the ankle and subtalar joints [3,4,7,10,11].

The problem of refracture after rod removal to regain the ankle motion encouraged some authors to use interlocking nails or introduce the rods from the medial malleolus [8].

Leg-length discrepancies are also a common problem that may be a part of the disease or may be iatrogenic [23].

The combination of an intramedullary nail and the Ilizarov technique increases stability, preserves long-term alignment, and corrects LLD, with less refracture rate [8].

The average age of the patients in our study was 3.8 years (range: 2.5–5 years). Many authors reported the association between good results and the older age of the patient [3,21,22].

There is consensus that surgery should be avoided before the third year of life, and if possible, it should be postponed until the age of five years [4,22].

The analysis of different therapeutic methods used by the European Pediatric Orthopedic Society members in a multicenter study on 340 patients with CPT showed a 75% union rate achieved with Ilizarov external fixation, and a refracture rate of 44%. The study recommended IM rodding as a guard against refracture. It found that the age older than 5 years was associated with the best results and that the optimal age for surgery was after 3 years [3].

However, Joseph *et al*. [24] using intramedullary rod placement and dual on-lay cortical bone grafting found that the patients younger than 3 years had better results compared with those in older groups regarding the union rate and the residual abnormalities.

The fibula has a great effect on the stability and preventing the complications in CPT. Dobbs *et al*. [4] reported a higher rate of union in patients in whom fibular pseudarthrosis was resected as compared with those in whom fibular pseudarthrosis was not resected.

All patients with fibular pseudarthrosis in our study achieved union. Many authors agree that persistence of pseudarthrosis of the fibula is associated with worse functional results and tibial refracture and that tibiofibular synostosis can limit the deformity [5,7,13].

Treatment of fibular pseudarthrosis is mandatory for optimal alignment and union, otherwise there will be an increased risk of valgus ankle and refracture [4,5,9].

Tudisco *et al*. [25] stated that a fibular pseudarthrosis is responsible for most of the poorest results, and Cho *et al*. [26] reported that the risk of re-fracture increased significantly with persistent fibular pseudarthrosis and union with a small diameter of the healed segment.

Choi *et al*. [27] classified fibula into two types: type A (mild-A1, moderate-A2) includes a fibula of normal integrity in the presence of established atrophic-type CPT, whereas type B (mild-B1, moderate-B2, and severe-B3) has atrophic-type CPT with concomitant fibular pseudarthrosis.

They advocated internal bone transport of the tibia for type A with large tibial resection gap, and circumferential on-lay bone grafting for small gaps. They recommended end-to-end osteosynthesis for type B1; 4-in-1 osteosynthesis for type B2, in which all four proximal and distal segments of the tibia and fibula are placed in one healing mass; and distal tibiofibular fusion for type B3 [27].

4-in-1 osteosynthesis has the merits of maximizing the cross-sectional area of healing at the pseudarthrosis level, widens the bone healing area, stabilizes the ankle while preserving its mobility, and guards against proximal migration of the fibula [27].

Valgus ankle is present in ~45% of patients, owing to proximal migration of the fibula. This deformity results in more stress on the lateral part of the tibial epiphysis leading to asymmetric growth. Valgus deformity can be corrected by epiphysiodesis of the medial malleolus [28].

Dobbs *et al*. [4] recommended placement of a syndesmosis screw to prevent valgus deformity of the ankle.

Although valgus ankle is a part of the natural history of the disease, achieving fibular union can alter the natural history. No cases were complicated by valgus ankle in this study, and the authors claimed that to the fibular union achieved.

The level of the lateral malleolus has its effect on the position of the heel. Among the nine case in this study (Table 1), there were seven cases with fibula minus and two cases with fibula plus. The position of the distal fibular physis was improved by the last follow-up in all cases, and this was accomplished through distal fibular epiphysiodesis for cases with fibula plus, and addressing the problem of fibular pseudoarthrosis and fibular union that was achieved in all cases preventing proximal fibular migration. In spite of the improved position of the distal fibular physis in all cases, there was still one case with fibula plus +1 position that was complicated with a varus deformity of the ankle (Fig. 3).

This study is limited by the small number of patients, short follow-up duration, and that none of our patients has reached skeletal maturity during the period of the study.

Conclusion

Addressing the fibular problem safeguards against refracture and protects against ankle joint deformity. Achieving tibial union with wide regenerate or wide fusion mass, good tibial alignment, and fibular union is mandatory for preventing refracture.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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