



Infected tibial nonunion in children: Is radical debridement mandatory?



Gamal Ahmed Hosny, Abdel-Salam Abdel-Aleem Ahmed*

Benha University, Faculty of Medicine, Egypt

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ABSTRACT

Nonunion is a devastating complication of tibial fractures with a debilitating effect. Several studies reporting tibial nonunions in adults are available; however, the reports on infected nonunions in children are scarce. The purpose of this study was to evaluate the results of treating paediatric infected tibial nonunion using Ilizarov fixator without radical resection.

Materials and methods: The study included 36 cases with infected nonunion of the tibia with 31 males and 5 females, and an average age of 11.4 years. The mean number of previous operations was 3.5. The duration before presentation averaged 10.4 months. At presentation, 24 cases were draining nonunions, while 12 patients had quiescent sinuses. Preoperative shortening was evident in 31 patients ranging from 1 to 12 cm. Six cases had equinus foot deformity. Stiffness of the ankle and/or knee was present in 11 cases. Angular deformity of more than 10° was evident in 13 nonunions. The monofocal compression-distraction was used for 10 cases managed by bloodless technique, and four draining cases managed by limited debridement and compression distraction. Limited debridement and bone transport was used in the remaining cases including proximal chondrodiastasis (two cases), proximal tibial corticotomy (17 cases), and partial osteotomy (three cases).

Results: The mean follow-up duration was 51.9 months. The external fixation period averaged 5.3 months. Delayed union occurred in three cases treated by repeating compression-distraction. Eventually, union was achieved in all cases. Two cases suffered refracture. Four cases had residual angular deformity of about 5–7°. Infection recurred in one case. ASAMI bone results were excellent in 29 patients, good in three patients, fair in two patients, and poor in two patients. The ASAMI functional results were excellent in 27 cases, good in 5 cases, fair in two cases, and poor in two cases.

Conclusion: The Ilizarov method provides a viable treatment method for treating paediatric infected nonunions in a single stage of management. Limited debridement was enough to control infection and achieved good results without radical resection.

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Introduction

Tibial fractures represent about 13% of paediatric fractures. Open fractures comprise approximately one-third of these fractures. Most paediatric tibial fractures heal uneventfully with adequate treatment, and nonunion is rare in children [1]. Nonunion is a devastating complication of tibial fractures with a debilitating effect on the quality of life. Its impact on physical health was reported to be worse than the impact of congestive heart failure, and comparable to that of end-stage hip osteoarthritis [2]. Infected tibial nonunion is a

complex challenging problem. The presence of nonunion, deformity, limb-length discrepancy (LLD), disuse osteoporosis, and scars of initial open fractures or from previous surgery render conventional reconstruction more difficult [3]. Therefore, below the knee amputation had been suggested as a treatment option for cases with recalcitrant infection and nonunion of the tibia [4].

The treatment objectives are union in addition to management of the coexisting difficulties. Surgical procedures were described including single and multiple-staged approaches [5]. Whereas several studies of management of tibial nonunions in adults are available, the reports on infected nonunions in children are scarce and mixing the results of septic and aseptic cases together or presented as sporadic cases in series of adult patients [6]. Consequently, it is hard to settle a standard management protocol. The Ilizarov fixator and method provide a multipurpose solution and can be considered as a limb salvage procedure. Mechanically,

* Corresponding author at: Department of Orthopaedic Surgery, Benha Faculty of Medicine, Farid Nada Street, Kalyubia, Benha, 13518, Egypt.

E-mail addresses: gamalahosny@yahoo.com (G.A. Hosny), Abdelsalam.Youssef@fmed.bu.edu.eg (A.-S.A.-A. Ahmed).

its stability permits early weight-bearing with progressive functional use of the limb. Biologically, the vascularity of bone and soft tissues is promoted by corticotomy which is also the key factor for bone transport in treating bone defects and restoring length. Moreover, it can be applied in the presence of active infection [7,8]. However, most authors considered radical resection

of the infected area in adults is the key to successful treatment. Children bones are different from adult one due to the remarkable biologic activity.

This study evaluates the results of treatment of infected nonunion of the tibia in children by Ilizarov circular fixator without radical resection.

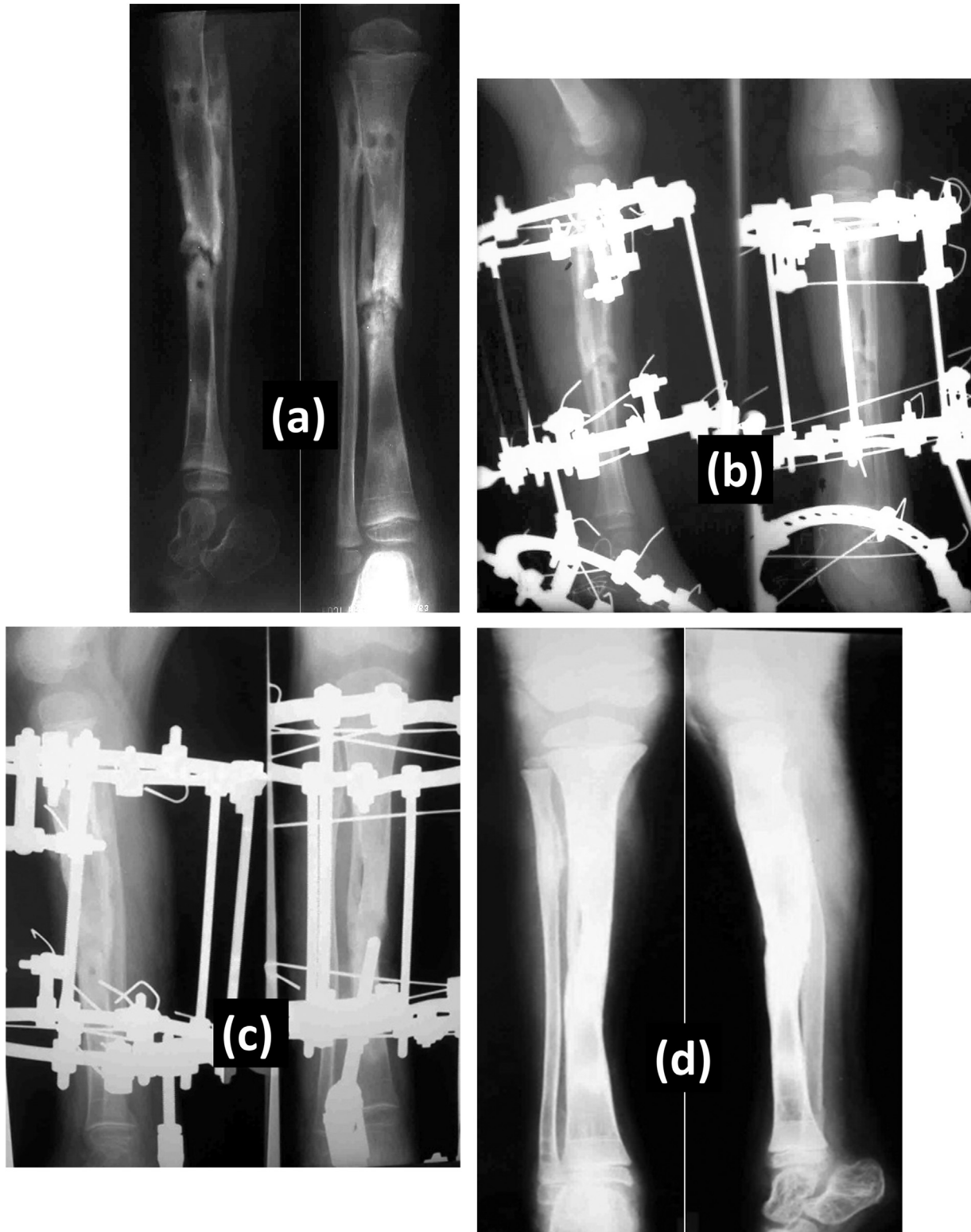


Fig. 1. A case of infected tibial nonunion complicating an open fracture previously treated by a unilateral fixator. This case was treated by the bloodless technique with cycles of compression-distraction. (a) Preoperative radiographs. (b) Radiographs after Ilizarov fixator application. (c) With progression of consolidation. (d) After sound union.

Materials and methods

This retrospective study was conducted after approval of the Ethical Committee of the University. The study inclusion criteria were infected nonunions of tibial fracture treated by Ilizarov fixator in patients younger than 16 years old. Nonunion was diagnosed when the fracture failed to progress to union or has little or no potential for further healing without additional intervention [9]. The age limit of 16 years was selected because the epiphyseal

plate at this age is still open and growing in many boys and some girls [6]. Aseptic nonunions, and congenital tibial pseudo-arthritis were excluded from the study. Four cases were also excluded due to inadequate follow-up data. From January 2004 till December 2015, 36 cases with infected nonunion of the tibia were referred to our centre including 31 males (86.1%) and 5 females (13.9%). The right side was affected in 28 (77.8%) cases, and the left in the remaining cases (22.2%). The age of patients at operation averaged 11.4 (Range 1.5–15; SD 2.83) years.

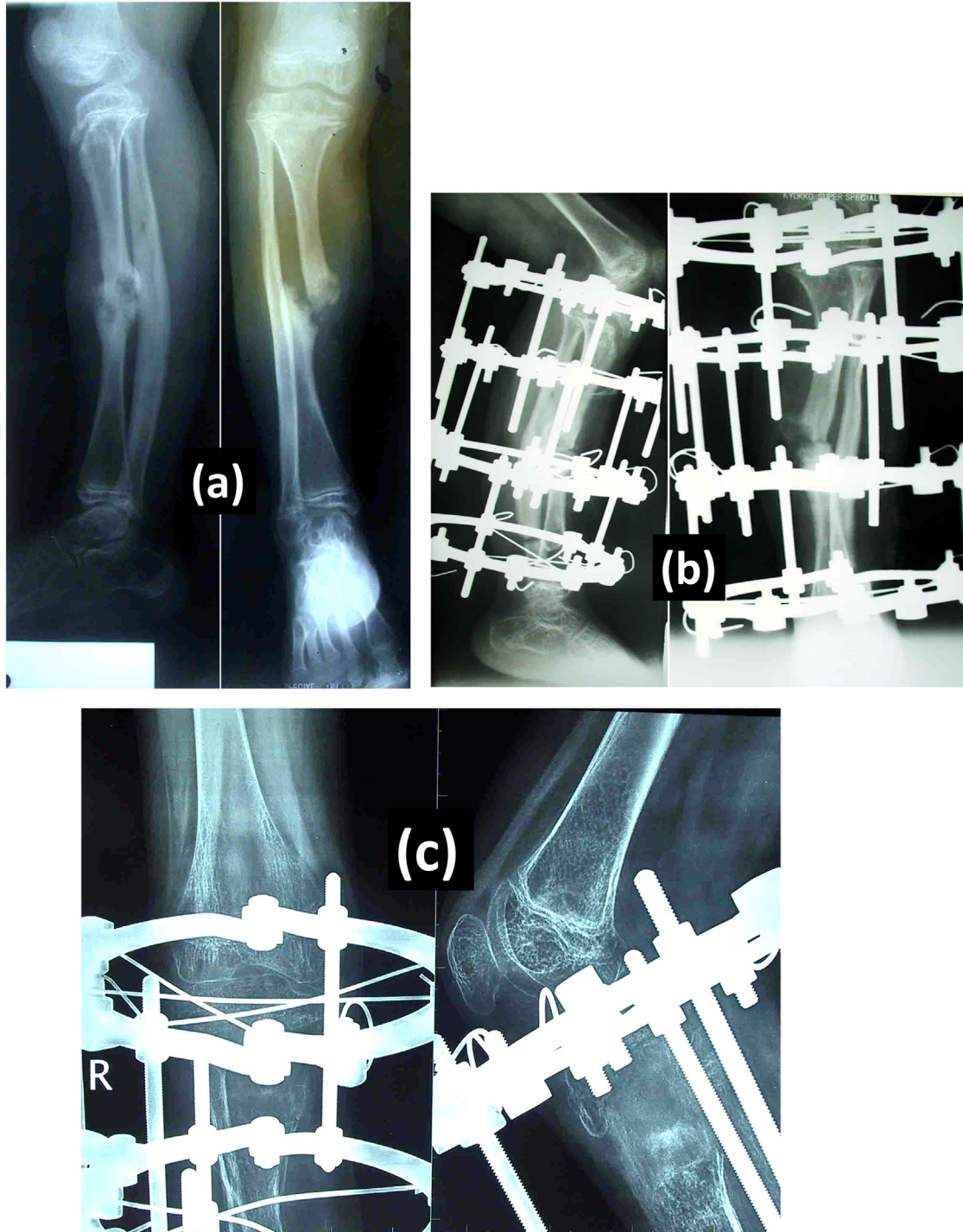


Fig. 2. An infected tibial nonunion treated by limited debridement and chondrodiastasis. (a) Preoperative radiographs. (b) Radiographs showing the initial distraction of the proximal tibial growth plate. (c) With progressive distraction. (d) Radiographs showing consolidation of the distraction zone. (e) After sound healing. (f) Radiographs at 12 years of follow-up.

The nonunions were subsequent to open fracture (18 cases; 50%), acute haematogenous osteomyelitis (4 cases; 11.1%), and open reduction and internal fixation (14 cases; 38.9%). All cases had previous surgeries e.g. ORIF, external fixation, implant removal, and repeated debridement. The number of previous operations ranged from 1 to 6 (Mean 3.5, SD 1.25). Twelve nonunions were in the distal third, and the remaining cases were in the middle third. The duration from trauma till presentation averaged 10.4 (Range 7–16; SD 1.93). At presentation, 24 cases (66.7%) had nonunion with active draining sinuses. The wounds in these cases were highly variable from tiny draining one or more sinuses to bigger wounds of about 7 cm in diameter with bone exposure. The remaining 12 patients (33.3%) had no signs of active infection despite presence of quiescent sinuses. Preoperative shortening was evident in 31 patients (Range 1–12; Mean 3.6; SD 2.71) cm. Six cases had equinus foot deformity. Stiffness of the ankle and/or knee was present in 11 cases (30.6%). Angular deformity of more than 10° was evident in 13 nonunions (36.1%).

Informed consent was obtained from the guardians of all patients included in the study after explaining the procedure in detail. To facilitate familiarity with the frame, these children were allowed to meet other patients with Ilizarov fixator.

Surgical technique

The surgical management was done in a single stage under general anaesthesia with supine patient positioning on a radiolucent table. The whole lower limb was scrubbed and draped in the usual way. An osteotomy was done for the fibula between the distal third and middle third. Suitable size rings were fixed to the tibia by wires guided by the goniometric atlas of the Association for the Study and Application of the Method of Ilizarov (ASAMI) [10]. Each bone segment was attached to one or two rings depending on the size of that segment. Each ring was fixed by two or three wires tensioned to 120–130 kg. No half pins used in this series. A drop wire was used to enforce the single ring fixation. Opposing olive

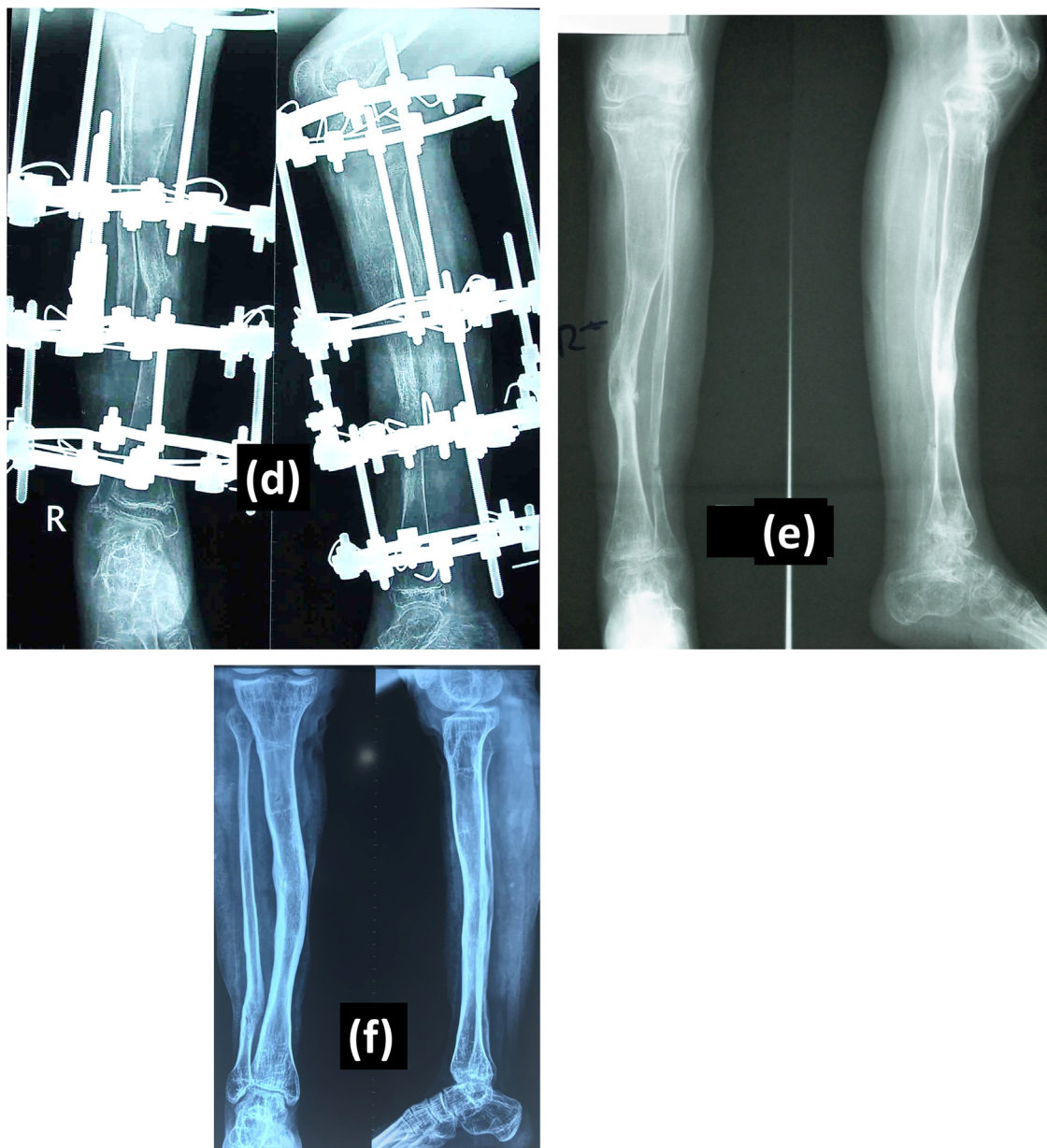


Fig. 2. (Continued)

wires were used to stabilize short segments. Rotational flap was performed in 4 cases.

Variable faces of applications of Ilizarov principles were used according to the characteristics of nonunion cases. The monofocal compression-distraction was used for cases without bone defects or shortening more than 2 cm including 10 cases managed by bloodless technique (i.e. without open debridement or corticotomy) in quiescent nonunions (Fig. 1), and four active draining cases managed by monofocal debridement and compression distraction. Debridement and bone transport was used in the remaining cases viz. cases having bone defects or more than 2 cm shortening including two quiescent cases and 20 draining nonunions. Removal of implants, if any, was done. Further debridement was limited to removal of infected and necrotic soft and bone tissues till the appearance of punctuate bone bleeding while obtaining transverse surfaces suitable for adequate compression. Radical or aggressive debridement with removal of healthy tissues was avoided. Samples from the infected soft tissues and bone were taken and sent for culture and antibiotic sensitivity testing.

Bone transport was achieved by proximal chondrodiastasis (i.e. distraction through the upper tibial growth plate) in two cases (Fig. 2). Proximal tibial corticotomy was done in 17 cases by the sequence of the corticotomy–first technique. After initial draping, the first step involves isolating the draining infected nonunion site by an extra draping followed by application of two-rings Ilizarov frame to the intact longer bone segment and corticotomy through a

1.5 cm incision using an osteotome after pre-drilling. The second step was debridement and completing the frame application.

In the remaining three cases, partial osteotomy (hemi-corticotomy) was done in the anterior half of the proximal tibial shaft aiming for closure of an anterior bone defect postoperatively (Fig. 3). This partial corticotomy was done after pre-drilling in L-shaped fashion followed by fixation of the transport fragment by two olive wires. Then the corticotomy was completed by an osteotome, and a transport ring was fixed to two olive wires. None of the cases had bone grafting. Finally, the alignment was checked under image intensifier, frame stability was assessed, any skin tenting around the wires was released, and dressings were applied followed by crepe bandage.

Postoperative care

The guardians were instructed about range of motion (ROM) exercises for knee and ankle, pin site care, and gradual weight-bearing. Antibiotic therapy continued for 6 weeks guided by the culture-sensitivity results. Wounds were followed-up weekly till complete healing. Cases of equinus were handled by intensive supervised physiotherapy program. The monofocal management cases were treated by repeated cycles of 1mm-daily compression and re-distraction. Bone transport cases started distraction after 7–10 days postoperatively and continued by 1 mm per day with modification depending on the consolidation progression. This was continued until bone defect closure, compression of the docking

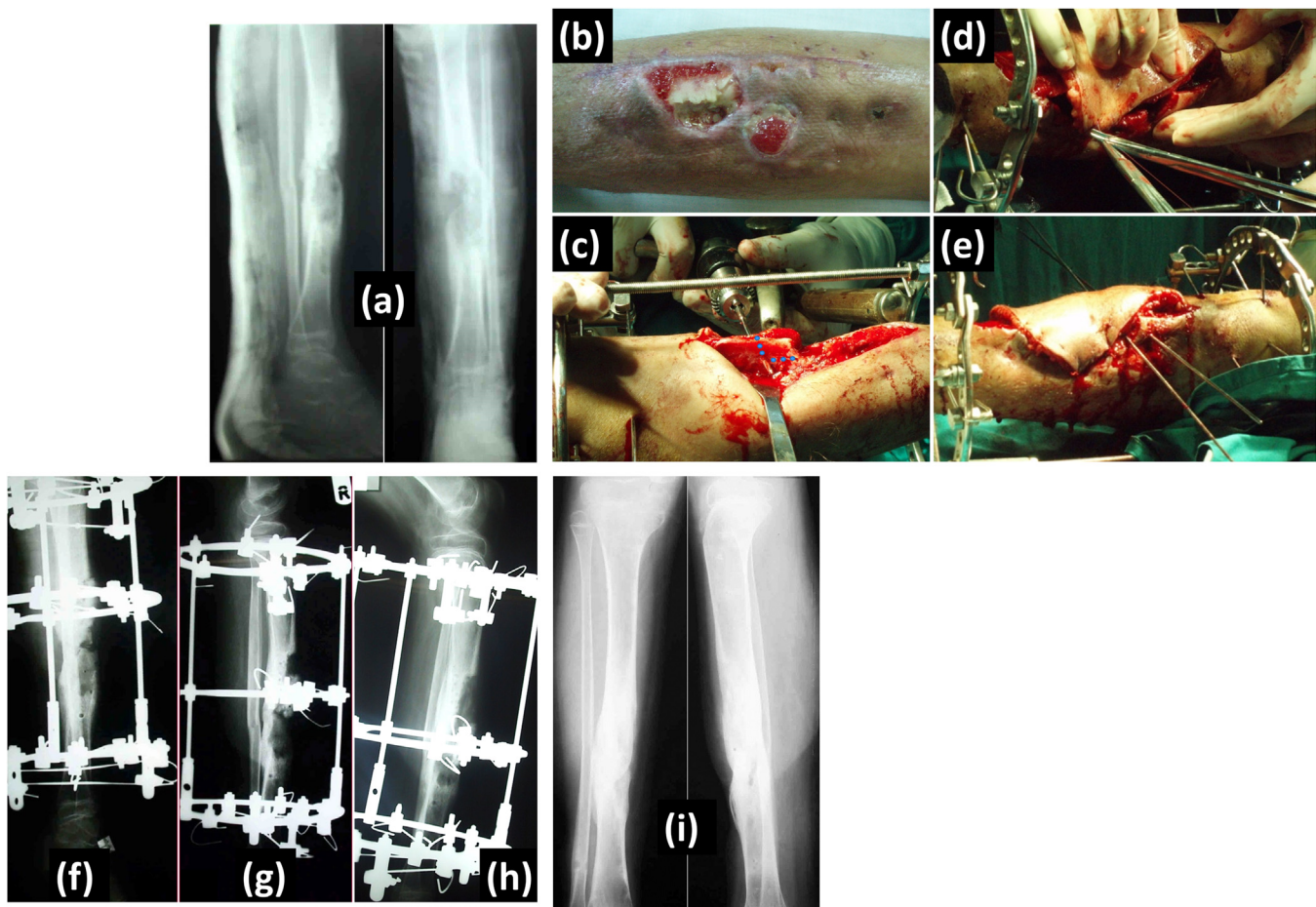


Fig. 3. An infected nonunion treated by partial corticotomy (hemi-corticotomy). (a) Preoperative radiographs. (b) Preoperative clinical photo. (c) Multiple drilling of the planned area for partial corticotomy. (d) Rotational flap performed. (e) Application of 2 olive wires in the transport segment and completing the corticotomy by osteotome. (f) Lateral radiographs showing the partial corticotomy before distraction. (g) With continued distraction. (h) With signs of consolidation. (i) Radiographs after sound union and closure of the anterior bone defect (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

site, and restoration of equal leg lengths. Radiographs were taken bi-weekly during bone transport or compression distraction, every month until fixator removal, at 3-months intervals for a year, and every year after that. Follow-up assessment included wound status, pin-site care, frame stability, ROM, and progression of union. The fixator removal was done after sound union seen in the orthogonal radiographs by crossing trabeculae with lack of radiolucent lines in the nonunion part, and at least three cortices in the lengthening part

The efficacy measures assessment included the bone and functional results based on the criteria of the Association for the Study and Application of the Method of Ilizarov (ASAMI) [8]. The safety criteria included evaluation of both intraoperative and postoperative complications. The statistical part of the study was done by IBM SPSS Statistics for Windows, Version 22.0 (IBM Corp., Armonk, NY, USA), with setting the significance level at $p < 0.05$.

Results

The bacteriological testing revealed the presence of *Staphylococcus aureus* in 14 cases including 3 MRSA infections, *Staphylococcus epidermidis* in 5 cases, *Klebsiella pneumonia* in 3 cases, *Pseudomonas aeruginosa* in 3 cases, mixed infection in 4 cases, and no growth in 7 of the quiescent nonunions. Postoperative hospitalization ranged from one to three days (mostly one day). Patients were followed-up for a mean of 51.9 (Range 24–144; SD 23.99) months. The external fixation period (EFP) averaged 5.3 (Range 2.5–12; SD 2.07) months. EFP was correlated with the age (Spearman's $\rho = .684$, $p = .000$), preoperative shortening (Spearman's $\rho = .910$, $p = .000$), number of previous surgeries (Spearman's $\rho = .464$, $p = 0.004$), and duration of nonunion (Spearman's $\rho = .662$, $p = .000$). Two patients had delayed ossification of the regenerate treated by slowing the distraction rate. Delayed union occurred in three cases treated by repeating compression-distraction. Bone graft was added in two of them. Eventually, union was achieved in all cases; however, two cases (5.6%) suffered refracture in the nonunion site rendering their bone and functional results poor. They healed following cast treatment.

Superficial pin track infection was reported in 19 patients (52.8%) and treated by pin site care with oral antibiotics. No case suffered neurovascular complications. Infection recurred in one case (2.8%) after fixator removal and treated by debridement. Restriction of sport activities was noted in two patients. Four patients had some exertional pain in the nonunion site. Restriction of joint motion was reported in the ankle (two cases) and the knee (one case). Recurrence of equinus deformity occurred in one case. Four cases (11.1%) had residual angular deformity of about 5–7°. Two patients had residual shortening of about 2 cm. ASAMI bone results were excellent in 29 patients (80.56%), good in three patients (8.3%), fair in two patients (5.6%), and poor in two patients (5.6%). Whereas, the ASAMI functional results were excellent in 27 cases (75%), good in 5 cases (13.9%), fair in two cases (5.6%), and poor in two cases (5.6%). The bone and functional results were correlated (Spearman's $\rho = .509$, $p = .002$). There was no significant correlation between the functional results and age of patients (Spearman's $\rho = -0.190$, $p = 0.268$).

Discussion

Fracture nonunion is rare in children with adequate treatment owing to the biologic factors like vital bone tissue and the thick periosteum with greater osteogenic power than in adults [11,12]. The often-held belief that paediatric closed or open diaphyseal fractures heal readily may result in a less aggressive approach, and consequently unacceptable morbidity with long treatment [13]. In a recent inception cohort study of 237,033 paediatric fractures in

18 bones, the incidence of nonunion was 0.85% with the group aged 12–17 years having a roughly 8-fold higher risk than those aged 0–6 years [14].

Despite rarity, nonunion of fractures in skeletally immature patients is complex and devastating complication especially when infected [2,13]. A systematic review of Gustilo grade IIIB paediatric tibial fractures reported delayed union in 22%, nonunion in 13% of cases, and deep infection in one-third of cases needing debridement and flap coverage but treated without flaps [15]. In addition to initial high-energy trauma and periosteal stripping, other predisposing factors of nonunion include: comminution, bone loss, infection, and lack of proper stabilization [13]. All these factors were present in cases of the current study.

Nonunion treatment by conventional methods is successful in non-infected cases without compromised bone vascular supply and integrity of soft tissues. Infection disturbs the biological environment resulting in delayed union, fixation loosening, and chronic osteomyelitis. Complex strategies are necessary for management of these infected nonunions taking into consideration control of infection, suitable fixation, alignment and length restoration, and rapid patient rehabilitation. The Ilizarov method is the modality of choice in these challenging situations [16,17]. The current study presents infected tibial nonunions in a series of 36 children treated by Ilizarov fixator in one stage with eventual union in all cases after a mean EFP of 5.33 months but with two (5.6%) refractures, one (2.8%) infection recurrence, four (11.1%) residual angular deformity, and three (8.3%) stiff joints. Even after successful healing of nonunion, children are inclined to have varying degrees of LLD and residual deformity because of the remaining growth potential [12].

Radical or aggressive debridement was not performed in the current study which was meant to question the use of minimal debridement limited to necrotic bone and soft tissues for treatment of paediatric infected tibial nonunion. The limited debridement was done by removal of the clinically-evident necrotic bone and soft tissues without excision of any healthy margin. Radical debridement is considered the key step to control bone infections and to treat infected nonunions [3,13,18,19]. Wide margin surgical debridement is considered fundamental to address both the offending biofilm and the vascular deficiency [18]. It is possible to achieve good results with the standard debridement especially when the infection is not very serious. But, with resistant infections, radical debridement will be necessary [20]. The Aggressive debridement eradicates infection more efficiently, but it creates a larger bone defect. Moreover, it is possible that this radical debridement may excise some bone unnecessarily [21]. Certainly, all necrotic tissues must be removed, however, as much of non-infected healthy tissues as possible have to be retained for better healing. The term 'radical' entails the liability of removing too much healthy tissue [22]. Simpson et al. [23] reported recurrence of infection in 28% of chronic osteomyelitis cases treated by marginal resection of less than 5 mm all of them were in type B hosts, and none had recurrence following wide resection. However, recurrence of infection may occur after radical debridement of infected nonunions with an incidence of 2–30% [3].

On the other hand, paediatric bones are different from adult bones with stronger osteogenic biological power [11,12]. The argument about the necessity of radical debridement for treating infected nonunion in children was not discussed before in the literature. The current study reports only one case of infection recurrence despite limited debridement.

A systematic review of 24 studies of adult infected tibial and femoral nonunions treated by Ilizarov methods reported a union rate of 97.5% and a mean EFP of 9.4 months with infected tibial nonunions. The rates of peroneal nerve palsy, malunion, refracture, infection recurrence, knee stiffness, and amputation were 13%, 7%,

4%, 5%, 12%, and 4% respectively. Pin track infection ranged from 10% to 100% [3]. Up to the authors' information, there is no similar systematic review about paediatric nonunions.

In one stage surgical procedure, the Ilizarov principles and the law of tension-stress were used in all cases of the current series with different modifications according to the nonunion personality including the bloodless technique, debridement and compression distraction, and debridement with bone transport (by chondrodiastasis, proximal tibial corticotomy, and hemi-corticotomy). For monofocal management cases, the fibular osteotomy was done to allow for cycles of compression and distraction. The fibula was also osteotomized in cases of lengthening and bone transport because of the associated fibular shortening and angular malunion. On the other hand, several authors recommended staged approaches for management of infected nonunions. Struijs et al. [24] reviewed 34 articles of infected nonunion of the long bones and reported 18 studies describing a two- or multiple-stage strategy for management.

In the present study, the corticotomy-first technique was used in active draining infected nonunions aiming at avoiding contamination and possible surgical site infection (SSI) in the corticotomy [25]. SSI is a dreaded complication in orthopaedic surgery and associated with prolonged hospital stay, reduction in quality of life, and increased health care costs by more than 300% [26]. Maksimovic et al. [27] reported SSI rate of 13.5% after clean orthopaedic surgeries, and 70% when the procedure was classified as infected. None of present series cases developed corticotomy infection. Rohilla et al. [28] reported one corticotomy site infection in a study of 70 infected adult tibial nonunion.

Besides bone transport, bone defects in children can also be treated by a variety of methods. Steinlechner and Mkandawire [29] used non-vascularised fibular grafts and intramedullary Kirschner wire for management of long bone defects of in children after sequestrectomy. One of the seven cases developed infected nonunion. Sales de Gauzy et al. [30] reported paediatric 27 bone defects in multiple long bones including seven infected nonunions. They used different techniques including; the induced membrane technique (10 cases), bone transport (7 cases), bone autograft (8 cases), a vascularized fibular transfer (one case), and no bone reconstruction in one case. Union was achieved in all patients, but with five cases of limited knee ROM, four cases of ankle equinus, five malalignments, and five patients with a protective brace. Gouron et al. [31] treated 14 children with long bones defects by the induced membrane technique. Bone union was obtained in 9.5 months. Complications included non-union in 35% of cases two cases of wound dehiscence, and one case of massive graft resorption.

Three cases of the present series were treated by hemi-corticotomy. Hemi-corticotomy was reported to close anterior tibial bone defects in chronic osteomyelitis in adult cases [32]. However, up to the authors knowledge, this was not yet reported in paediatric cases.

Chondrodiastasis or physal distraction is a method for lengthening or deformity correction by external fixation and progressive distraction of the growth plate [33]. Langlois and Laville [34] used chondrodiastasis in 20 cases (15 tibiae, 4 femora, one radius) by monolateral fixator or Ilizarov fixator and achieved a mean lengthening of 4.75 cm and mean angular correction of 22.2°. Zarzycki et al. [35] reported an average lengthening a mean of 4.6 cm and 4.7 cm in 24 femoral physal distractions and 16 tibial physal distractions respectively without premature physal cartilage fusion. Meanwhile, Bjerkrejms [36] reported this complication in all of his series of 10 proximal tibial physal distractions. None of the two cases of chondrodiastasis in the present study developed premature cartilage fusion. The use of chondrodiastasis by Ilizarov fixator in tibial nonunion was reported by Liow and

Montgomery [13] in one case of their series of 9 tibial cases (3 nonunions, 3 acute bone loss, and 3 anticipated nonunions). Other cases were treated by monolateral frames in two cases and circular frames in rest of cases including 3 bone transports, and one tibial lengthening with tibiocalcaneal fusion. Bony union was obtained in all their patients with a range of treatment time from 3 to 12 months. The complications included three knee fixed flexion deformities, one fused ankle, 3 LLDs, and 3 angular deformities.

Owing to rarity, there is paucity of literature reporting nonunited paediatric tibial fractures. Arslan et al. [36] treated 19 children with a mean age of 9.6 years (Range 1–15 years) having multiple long bones nonunions including 10 tibial and 14 infected cases. They used different treatment modalities including decortication with external fixator with or without autograft, circular fixator and bone transport, decortication with osteosynthesis and autograft, and decortication with autograft and cast. All cases healed except three nonunions that needed reoperation. Yeo et al. [12] presented a series of 16 diaphyseal nonunions (6 femoral, 5 tibial, 2 ulna, 2 radius, one humerus) with a mean age 11.1 years treated by internal or external fixation (including 2 cases by Ilizarov fixator) with or without bone grafting. All nonunions healed after 3.4 months in infected cases and 2.9 months in others. LLD was present in five patients and varus ulnar bowing was present in one patient.

Adequate soft tissue coverage is essential during management of infected tibial nonunions. Muscle transfers are beneficial for providing a new vascular supply, and consequently improving antibiotic dispersal and host immune system [9]. In the current study, Ilizarov fixator was not an obstacle for using muscle flaps in four cases of the series.

The strength of the present study lies in reporting a homogenous series of a relatively larger number (than reported) of paediatric infected tibial nonunions treated by the same fixation method with limited debridement. On the other hand, reported literature include case reports [11], studies combined adults with children [38], studies mixing infected and non-infected nonunions [6,12,37,39], studies including multiple long bone nonunions or bone defects [12,29–31,39] and variable fixation methods in the same study [6,12,13,37]. So, it is hard to fairly compare the current study with others. However, the study has the obvious limitations of the retrospective nature and lacking a control group.

Conclusion

The Ilizarov method provides a viable treatment option for treatment on infected nonunions in children in a single stage of management. Radical resection was not necessary and limited debridement achieved good results. There is no a single protocol of management, and the treatment should be individualized for each nonunion.

Conflict of interest

The authors declare that they have no conflict of interest.

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