ORIGINAL PAPER



Paediatric infected femoral nonunion; mid-term results of a rare problem with a single-stage treatment and up to eleven and half years follow-up

Gamal Ahmed Hosny¹ · Abdel-Salam Abdel-Aleem Ahmed¹

Received: 3 October 2019 / Accepted: 6 December 2019 / Published online: 16 January 2020 \odot SICOT aisbl2020

Abstract

Purpose Nonunion of femur fractures is a devastating disabling complication which is rare in children. The purpose of this study was to report the outcomes of treating infected femur nonunions in children by the Ilizarov fixator in one stage.

Patients and methods The study included 13 patients with unilateral infected nonunion of the femur with an average age of 9.1 years. The nonunion duration averaged 10.69 months. Ten cases were draining nonunions, and three patients had quiescent sinuses. Associated problems include shortening in all cases (mean 3.5 cm), joint stiffness (9 cases), and angular deformity (7 cases). The quiescent cases were treated by bloodless monofocal compression-distraction. Four draining cases were treated by debridement and compression with relengthening through nonunion site. The remaining six cases were treated by blocal technique.

Results The mean follow-up duration was 60.15 months. External fixation period averaged 5.3 months. Successful union was achieved in all patients. Recurrences of infection occurred in two cases including one with refracture and another one with late pathological fracture. Other complications included pin tract infections, one delayed union, two residual angular deformities, and 6 cm residual shortening in one patient. ASAMI bone results were excellent (8 patients), good (3 patients), fair (one patient), and poor (one patient). The functional results were excellent (9 cases), good (3 cases), and fair (one case).

Conclusions The Ilizarov method provided a viable treatment option for treating paediatric infected femur nonunions in single stage of management with infection control in most cases and satisfactory outcomes.

Keywords Nonunion · Paediatric femur · Infection · External fixator · Ilizarov

Introduction

The incidence of paediatric femoral shaft fractures is about 20 per 100,000 with bimodal age distribution at two years and 17 years. Most of these fractures are closed with traditional treatment by closed methods. Treatment has evolved gradually towards operative modalities for rapid recovery and avoiding the drawbacks of prolonged immobilization.

 Abdel-Salam Abdel-Aleem Ahmed salam_ilizarov@yahoo.com; ABDELSALAM.YOUSSEF@fmed.bu.edu.eg

Gamal Ahmed Hosny gamalahosny@yahoo.com

¹ Department of Orthopaedic Surgery, Faculty of Medicine, Benha University, Farid Nada Street, Kalyubia, Benha 13518, Egypt Different methods were advocated including intramedullary nailing, plating, and external fixation [1].

Nonunion of femur fractures in children is a rare problem that is devastating and disabling to the patient and challenging for the orthopaedic surgeon. In a recent study of 237,033 paediatric long bone fractures, the risk of nonunion was 0.2% in the group aged zero to six years and roughly eight fold higher (1.63%) in the older cohort (12–17 years old), and risk factors were similar to those of the adult nonunion [2]. The occurrence of an associated infection makes the treatment even more challenging. Moreover, the situation is harder with the superadded complications such as limb length discrepancy (LLD), deformities, disuse osteoporosis, and scars of the initial open wounds or previous surgical interventions [3].

Paediatric femoral nonunions are scarce especially the infected cases. The available literature include case reports [4–6], or cases presented among different bones in small case series mixing septic and aseptic nonunions [7–12]. Because of this rarity, there is no consensus on a standard method of treatment for such cases. The purpose of this study was to report the radiological and functional outcomes of treating infected femur nonunions in children by the Ilizarov fixator in one stage.

Patients and methods

This is a retrospective study that was conducted following the approval of Ethical Committee of the University. The inclusion criteria included infected femur nonunions in skeletally immature patients treated by Ilizarov circular fixator. Diagnosis of nonunion was done when there was failure of progression to union or there was no potential for more healing without further intervention [13]. The exclusion criteria included noninfected nonunions, bone defects (secondary to acute trauma, tumor resection, or congenital conditions), and patients with incomplete follow-up data. From May 2005 to September 2015, 13 patients with unilateral (9 right, and 4 left) infected nonunion of the femur were treated by Ilizarov fixator including 11 males (84.6%) and 2 females (15.4%) with an average age of 9.1 (range 2–14.5; SD 3.96) years (Table 1). The cause of nonunion was a pathological fracture complicating osteomyelitis (5 cases; 38.5%), following open reduction and internal fixation (ORIF) (5 cases; 38.5%), and open fracture (3 cases; 23.1%). The patients had previous surgery including drainage and debridement, ORIF, implant removal, and external fixation with an average number of 3.3 (range 2-5; SD 1.11) procedures. The duration of nonunion before presentation averaged 10.69 (range 8-16; SD 1.97) months. Ten cases (76.9%) were draining nonunions and

Table 1 Demographics and results of patients

three patients (23.1%) had quiescent sinuses. Knee stiffness was present in seven cases (53.8%) and associated with ankle stiffness in another two cases (15.4%). Pre-operative shortening was present in all patients with a mean of 3.5 (range 1–12; SD 3.08) cm. Five cases (38.5%) had valgus deformity and one case (7.7%) had varus deformity. Group meeting with patients having Ilizarov fixator was arranged for better explanation and awareness of the procedure. An informed consent was obtained from the guardians of patients included in this study.

Surgical technique

The surgery was done under general anaesthesia and supine patient positioning on a radiolucent table. After scrubbing and draping of the whole lower limb, the procedure started by adequate debridement of cases having active draining sinuses with excision of all infected and necrotic tissues and bone till having punctuate bleeding of bone. Samples were obtained from the infected tissues for culture and antibiotic sensitivity testing. The opposing bone ends were reshaped to be transverse permitting adequate compression. Ilizarov fixator rings and arches were applied and fixed as guided by the atlas of the Association for the Study and Application of the Method of Ilizarov (ASAMI) [14] with fixation by olive and plain wires for the distal femur and half-pins proximally. The knee was flexed when the wires pass through the quadriceps to reduce its checkrein effect on knee flexion. Bone grafting was not used in any case.

Younger children with small femur were treated by the monofocal technique including the quiescent cases and four of the draining cases. The monofocal frame consisted of one distal ring and one proximal arch with or without a middle

| Case | Gender | Age | Side | No. of previous operations | Preop shortening (cm) | Nonunion duration | Follow-up (months) | Ext. fix period (months) | ASAMI bone result | ASAMI functional result |
|------|--------|------|------|----------------------------|--------------------------|-------------------|-----------------------|--------------------------|-------------------|----------------------------|
| 1 | М | 11 | Rt | 4 | 4 | 10 | 42 | 6 | Е | Е |
| 2 | М | 2 | Rt | 2 | 1 | 8 | 138 | 3.5 | G | Е |
| 3 | М | 10 | Lt | 4 | 1.5 | 11 | 72 | 4.5 | Е | Е |
| 4 | F | 7.5 | Rt | 3 | 12 | 16 | 51 | 9 | Е | G |
| 5 | М | 10.5 | Rt | 2 | 2 | 11 | 39 | 5 | Р | F |
| 6 | М | 9.5 | Lt | 5 | 2 | 10 | 51 | 4.5 | Е | Е |
| 7 | М | 13.5 | Rt | 4 | 3 | 9 | 75 | 4.5 | Е | Е |
| 8 | М | 4.5 | Rt | 5 | 2 | 12 | 54 | 5 | G | G |
| 9 | М | 11.5 | Rt | 3 | 8 | 10 | 40 | 7.5 | Е | Е |
| 10 | F | 3 | Lt | 2 | 3 | 12 | 42 | 6.5 | F | G |
| 11 | М | 8.25 | Rt | 2 | 2 | 9 | 61 | 4 | Е | Е |
| 12 | М | 14.5 | Rt | 4 | 2.5 | 11 | 36 | 4.5 | G | Е |
| 13 | М | 13 | Lt | 3 | 3 | 10 | 81 | 5 | Е | Е |

M male, *F* female, *Rt* right, *Lt* left, *Pre-op* pre-operative, *Ext. fix* external fixation, *ASAMI* Association for the Study and Application of the Method of Ilizarov, *E* excellent, *G* good, *F* fair, *P* poor

ring. The three quiescent cases were treated by the bloodless technique of monofocal compression-distraction without debridement (Fig. 1). Four draining cases were treated by debridement and monofocal compression and relengthening through the nonunion site (Fig. 2). The remaining older children (6 cases) had active draining infection and were treated by bifocal technique with debridement and bone transport after distal femoral corticotomy that was done using predrilling and an osteotome (Fig. 3). The frame in these cases consisted of one distal ring block formed by one ring and one 5/8 ring for better knee motion. The middle transported bone segment was attached to one ring and the proximal segment was fixed by an arch.

The knee range of motion (ROM) was evaluated with skin and soft tissue release around the wires to facilitate knee motion. Lastly, the stability of the frame was checked and

Fig. 1 An example of the bloodless monofocal technique. **a** Pre-operative radiographs. **b** Radiographs after frame application with better healing after repeated compression and redistraction. **c** Radiographs with sound union. **d** Radiographs after fixator removal. **e** Radiographs after 8 years of follow-up alignment was seen under image intensifier. Dressings and crepe bandage were applied.

Post-operative care

Gradual weight-bearing was permitted as tolerated. Pin sites care and regular knee and ankle exercises were emphasized on. Antibiotic administration followed the results of the culture and sensitivity tests and continued for six weeks. The patients were followed-up clinically with evaluation of the frame stability, progress of monofocal or bifocal strategies, condition of wounds and pin sites, knee and ankle ROM, and weight-bearing state. Physiotherapy program was followed for the cases having associated joint stiffness. Monofocal management was performed by repeated cycles of 1 mm daily compression followed by redistraction after adequate





Fig. 2 An example of monofocal management with using the nonunion site for the lengthening. **a** Pre-operative radiographs. **b** Radiographs after Ilizarov fixator application with compression and redistraction for lengthening through the nonunion site. **c** Radiographs showing the progression of the regenerate of lengthening. **d** Radiographs showing union and the consolidation of lengthening. **e** Radiographs after fixator removal. **f** Radiographs after 10 years

compression. This was delayed after ten post-operative days in cases treated by debridement and monofocal compression and relengthening through the nonunion site. Similarly with bone transport, distraction of corticotomy site began after seven to ten days post-operatively with 1 mm daily. The rate was modified according to the progress of consolidation.

Besides post-operative images, radiographs were taken every two weeks during distraction and compression, every month until removal of Ilizarov fixator, every three months in the first year, and lastly every year. Ilizarov fixator was



Fig. 3 An example of bifocal management with nonunion debridement and bone transport through a distal femoral corticotomy. **a** Radiographs showing Ilizarov fixator after implant removal and debridement of nonunion site. **b** Radiographs showing closure of the nonunion defect and regenerate of the restored length. **c** Radiographs showing the progression of nonunion site healing and consolidation of the regenerate. **d** Radiographs after 2 years

removed after obtaining union evidenced in orthogonal radiographs by the presence of crossing trabeculae and at least three cortices of bone. Physiotherapy continued after removal of the frame till improvement of the joints ROM. The evaluation tools were the bone and functional results criteria of ASAMI [15]. The statistics were performed using IBM SPSS Statistics for Windows, Version 22.0 (IBM Corp., Armonk, NY, USA), with a significance level at p < 0.05.

Results

The mean days of hospitalization was 8.08 (range 3–17; SD 4.73) days. The culture results showed *Staphylococcus aureus* in most cases, mixed infection in two cases, and no growth in two cases. The patients were followed-up for an average of 60.15 (range 36–138; SD 27.63) months. The period of external fixation averaged 5.3 (range 3.5–9; SD 1.53) months. Successful union was achieved in all patients with infection

control in 11 cases. Three recurrences of infection occurred in two cases. One developed after 12 months with a refracture and was treated by debridement and reapplication of Ilizarov fixator and bone transport. Infection recurred twice in another case. The first developed after five months of fixator removal and was treated by debridement. The second recurrence was after ten years with a pathological fracture and was treated by debridement and bone transport with a second application of the frame.

Superficial pin tract infection occurred in nine cases and was treated by oral antibiotics. There was no need to change any wire or half-pin. One patient had delayed union and was treated by repeating compression-distraction. Residual angulation developed in two cases including one varus and one procurvatum that improved with growth. Three patients had residual shortening including 6 cm in one case treated by another lengthening procedure with Ilizarov fixator and anothertwo cases with 2 cm LLD. Occasional pain was reported by three patients. Knee ROM showed some restriction in all cases immediately after fixator removal. However, this was improved with supervised physiotherapy except in two cases where residual stiffness with a flexion range of about 90° and 75°, respectively. All patients were capable of full extension. ASAMI bone results were excellent in eight patients (61.5%), good in three patients (23.1), fair in one patient (7.7%), and poor in another patient (7.7%). The functional results were excellent in nine cases (69.2%), good in three cases (23.1), and fair in one case (7.7%). The bone and functional results were significantly correlated (Spearman's rho = 0.629, p = 0.021).

Discussion

Diaphyseal femur fractures constitute about 1.4 to 1.7% of paediatric fractures. In children, open femur fractures represent 3–4% of all open fractures [4, 16]. There is no standard definition of nonunion in paediatric literature [17]. Greenbaum et al. described delayed union as incomplete healing at ten weeks in children aged less than nine years and at 12 weeks in cases aged nine years or older [18]. Paediatric fracture nonunion is rare particularly in uncomplicated closed fractures owing to the more vascularized robust thick periosteum and the higher potentials of growth and remodelling [4, 11].

Risk factors associated with paediatric nonunion include severity of initial trauma, soft tissue interposition, soft tissue affection and periosteal stripping, fracture comminution and bone loss, infection, repeated fracture manipulation, and inadequate fracture stabilization [1, 12, 19]. These predisposing factors were found in patients of the present study.

Bone reconstruction and control of infection constitute a formidable therapeutic challenge in these cases [9].

Treatment of infected nonunions involves adequate debridement, stable fracture fixation, and bone grafting when needed. Single- and multiple-staged approaches have been described [20]. There is no standard fixation method for paediatric femoral nonunions. The ideal stabilization method involves a load-sharing device to reduce stress shielding [16]. In the current series, the Ilizarov fixator was used for treatment of 13 infected femoral nonunions in children with monofocal and bifocal strategies. The recently described corticotomy-first technique for treatment of infected nonunions [21] was not applicable in the present study owing to the short femur in children. Ilizarov circular fixator is a versatile tool providing a multipurpose solution for reduction of deformed fractures with compression of the fracture site and fine-tuning of alignment. The provided stability facilitates early weight-bearing with the advantage of functional use of the limb. Biologically, it is applied percutaneously with no extensive soft tissue stripping, and can be applied even in infected cases. Moreover, the vascularity is promoted by the corticotomy which is also the solution for bone transport to handle shortening and bone defects [3, 22]. On the other hand, it has known problems of complexity of application, risk of neurovascular injury, the bulky frame requiring patient compliance, and pin track infection [22].

Whereas the current series represents a nearly homogenous group of infected femoral nonunion in children treated by one method, some published cases of this problem were presented as case reports. Soldado et al. [5] reported one distal femoral infected nonunion associated with 18 cm shortening, stiff extended knee, and stiff equinus ankle. They treated this 11-year-old child by trans-tibial amputation and addressing the pseudoarthrosis by a vascularized tibial periosteal graft taken from the amputated part and fixation by a T-plate without bone grafting. Union was complete after two months and the patient ambulated in a femoral weight-bearing prosthesis. Zhen et al. [6] reported healing after ten weeks without deformity after using a conventional nail with vascularized iliac crest graft for infected femur nonunion in a seven year-old patient previously treated by plate fixation followed by external fixation.

Other published cases were included as a part of heterogeneous studies having different bones with different nonunions (septic and aseptic), unrelated aetiology (trauma, congenital, tumours), and variable treatment methods. Lewallen and Peterson [10] presented 30 nonunions in tibia, femur, ulna, humerus, radius, and fibula. The five femoral cases were treated by graft and plate (3 cases), brace (one case), and IMN and graft (one case). Yeo et al. [12] reported 16 paediatric long bone diaphyseal nonunions (6 femora, 5 tibiae, 2 ulna, 2 radius, one humerus) treated by bone grafting and internal or external fixation. Four of the six infected cases (2 tibial, 2 femoral) were treated firstly by debridement and antibiotic-loaded cement insertion, followed by osteosynthesis using a plate and/or bone graft. The other two femoral cases were treated by Ilizarov fixator. Union was obtained after 3.4 months in the septic cases and 2.9 months in the remaining. Residual LLD occurred in five patients and a varus deformity complicated one ulna.

Arslan et al. [7] treated 11 paediatric cases including four nonunions and seven traumatic bone losses in eight tibia and three femora with one infected femoral case. Bone transport was performed using unilateral fixator for the femoral cases and circular fixator for the tibial cases. Union was obtained in all cases with a mean external fixation time of 4.2 months without refracture. The complications included permanent ankle stiffness (2 patients), temporary knee flexion contracture (3 patients), and premature callus in one patient which needed recorticotomy.

Masquelet et al. described the two-stage induced membrane technique for bone reconstruction [23]. Gouron et al. [24] used the induced membrane technique to treat 14 children having defects in different long bones with various causes (trauma, tumour resection, and congenital). They included four femora after malignant tumour excision. Bone healing occurred in 9.5 months after the second stage with nonunion in 35% of cases. The main complications of this approach include pseudarthrosis, graft resorption, wound dehiscence, and loosening of the stabilization device. Nonunion at the extremities is frequent and ranges from 30 to100% of cases. The significant volume of autograft needed for large defects is the limiting factor in children and adding allograft bone for graft expansion should not exceed 30% of the total amount. Moreover, weight-bearing is not permitted between the two stages [24, 25].

Sales de Gauzy et al. [11] treated paediatric 27 bone defects (including seven infected nonunions) in multiple long bones. Of these cases, nine femora were treated by bone auto-grafting (3 cases), induced membrane technique (3 cases), bone transport (2 cases), and a vascularized fibular transfer (one case). All 27 cases healed, however with limited knee ROM (5 cases), ankle equinus (4 cases), one claw toe, five malalignments, and five patients still using a protective brace.

Variable adjuvant treatment modalities were described. Hissnauer et al. [26] treated five non-infected femoral nonunions in patients with a mean age of 11 years using rhBMP-2 and a locking plate. Four cases healed in a mean of 12.1 months, and one developed infected nonunion. Boyette and Herrera-Soto [17] treated 22 nonunions complicating eight osteotomies and 14 fractures in 21 children by pulsed electromagnetic field alone in 17 limbs, and with additional bone marrow injection in five limbs. After initial treatment, 8 of 14 fracture nonunions (57%) healed at an average of 13.2 weeks, and seven of eight osteotomy cases united at an average of 18.8 weeks. However, the authors did not clarify the affected bones or the method of fracture stabilization. Soldado et al. [27] used vascularized fibular periosteal graft for 13 different bones (including 5 femora) in 12 children to prevent nonunion of bone allograft-host junction and treat recalcitrant nonunion with variable fixation methods in different bones with variable neoplastic, traumatic, and congenital aetiologies. Except in one case, union was achieved after an average of 2.8 months with metaphyseal junctions and 7.1 months for diaphyseal cases.

The current study presents a single-stage treatment method for these complicated cases. On the other hand, several authors reported two- or multiple-staged approaches [9, 12, 20, 24, 25, 28]. Patwardhan et al. [28] presented 26 children with gap nonunion after osteomyelitis of different bones including seven femora treated by initial stage of debridement. After control of infection, non-vascularized fibular grafting was applied with intramedullary K-wire fixation and occasional additional external fixation. Complications included delayed union (4 cases), infection recurrence (1 case), physeal arrest (3 cases), and limblength discrepancy (mean 3 cm) in all lower-limb cases.

In the current study, the ASAMI functional results were excellent to good in 11 cases (84.6%) and fair in one case (7.7%). Several authors did not report a functional scoring for assessments, perhaps, for reporting nonunion in variable sites necessitating different unrelated scoring systems [8, 11, 24, 26–28]. Arslan et al. [7], in their series of 11 nonunions, the functional results were described as perfect (9 cases), good (1 case), and insufficient (1 case).

The limitations of the present study are the retrospective design, small number of patients, and the lack of control group or randomization. These limitations are explained by the known rarity of the selected nonunions with strict inclusion criteria to obtain a homogenous series. The strengths of this study are presenting an almost homogenous group of patients, treatment by the same surgical approach in one stage, and the relatively long-term follow-up reaching up to 138 (mean 60.15) months. Moreover, up to the best knowledge of the authors, there are no available comparable published studies.

Future prospective multicenter studies of large number of cases with a control group might provide a more insight and better assessment.

Conclusion

Infected nonunion of femur is rare in children. The Ilizarov fixator and method provided a viable treatment option for these complex challenging problems in a single-stage approach. This method allowed for different protocols of treatment with monofocal (bloodless or open) and bifocal techniques according to the personality of nonunion. The bone and functional results were excellent to good in most cases.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. For this retrospective type of study formal consent is not required.

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