

1 Department of Orthopaedics, University Hospital, Patras Medical School, Patras,

2 Department of Orthopaedics, Royal London Hospital, London, U.K.

3 Department of Radiology, Patras University Hospital, Patras, Greece

American Journal  $\cap$ 

2020.06.06 Received: Accepted: 2020.10.07 Available online: 2020.11.02 Published: 2020.12.06

> Authors' Contribution: Study Design A Data Collection B Statistical Analysis C Data Interpretation D Manuscript Preparation E Literature Search F

A Novel Treatment of a 65-Year-Old Woman with a Neglected Type IIIB Open Fracture of the Tibia with Inadequate Soft Tissue Coverage and Periosteal Stripping Requiring an Ilizarov Approach to Bone and Soft Tissue Lengthening and Reconstruction: A Case Report and Review of the Literature

Greece

AEF 1 Ioannis N. Vrachnis EF 2 Alexandros Vris **B 3 Petros Zampakis** F 1 Zinon T. Kokkalis **D 1** Andreas Panagopoulos Funds Collection G **Corresponding Author:** Antonios Kouzelis, e-mail: athkouzelis@gmail.com **Conflict of interest:** None declared Patient: Female, 65-year-old **Final Diagnosis:** Open tibia fracture with bone loss

A 1 Antonios Kouzelis

Symptoms: Pain • osteomyelitis **Medication: Clinical Procedure:** Specialty: **Orthopedics and Traumatology • Plastic Surgery Objective:** Challenging differential diagnosis Background: Type IIIB open bone fractures include loss of soft tissue with periosteal stripping and need rapid surgical repair. The Ilizarov method of bone lengthening and reconstruction offers limb salvage as an alternative to amputation. **Case Report:** We report a case of a neglected type IIIB open fracture of the tibia with inadequate soft tissue coverage and periosteal stripping requiring an Ilizarov approach for limb salvage in a 65-year-old woman. Surgical debridement resulted in a large bone deficit of 13 cm. Acute shortening facilitated wound closure, and the remaining skin defect was treated with skin grafting. Bone transport and limb lengthening techniques, in addition to skin expansion and support, were used to restore the length of the tibia. The regenerated bone had to be fused with the talus since the tibial plafond was excised during debridement. The external fixator was removed after 643 days of treatment. An external fixation index of 49.6 d/cm was estimated. Excellent bone and good functional results were obtained according to the criteria of the Association for the Study and Application of the Method of Ilizarov. Skin invagination, bone translocation, and pin tract infection were the major postoperative issues. Conclusions: This case showed that a multidisciplinary approach may be required for the successful management of neglected open fractures of the tibia and that the Ilizarov approach to both bone and soft tissue lengthening and reconstruction should be considered to ensure limb salvage and improve the final cosmetic appearance. **MeSH Keywords:** Fractures, Open • Ilizarov Technique • Limb Salvage • Reconstructive Surgical Procedures • Tibia Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/926622 - 12 I <u>1</u>2 3 2 2010 2 40



e926622-1

## Background

The soft tissue coverage in the lower third of the tibia is poor, making the tibia susceptible to open fractures when injured. Open fractures may account for as much as 24% of all tibial fractures [1]. Mistreated or neglected open fractures not only result in bone necrosis, but also involve large soft tissue deficits. This kind of injury requires combined treatment by both orthopedic and plastic surgeons [2]. The Ilizarov method is a type of circular external fixation used in orthopedic surgery to lengthen or reshape limb bones. It works as limb-sparing technique to treat complex and/or open bone fractures and cases of infected nonunion of bone that are not amenable to treatment with other techniques. It is named after the orthopedic surgeon Gavriil Abramovich Ilizarov from the Soviet Union, who pioneered the technique [3,4]. Since its origination in the mid-1900s, the Ilizarov method has advanced greatly and has become a viable method for bone lengthening, severe deformity correction, and defect management. As the reported studies show, it remains one of the most used tools for bone reconstruction [5-9]. Poorly perfused soft tissues may not be suitable for reconstruction by the more sophisticated techniques of plastic surgery. In such cases, medical and ethical dilemmas arise because a choice must be made between reconstruction and amputation [10]. Limb reconstruction always appears to be the better choice, but achieving the optimum result is a lengthy and difficult process.

We report a case of a neglected type IIIB open fracture, according to the Gustillo-Anderson classification (extensive soft tissue damage with periosteal stripping and bone exposure usually severely contaminated and comminuted, with flap coverage being required to provide soft tissue coverage). The patient was a 65-year-old woman whose tibia had inadequate soft tissue coverage and periosteal stripping. Treatment required an llizarov approach to both bone and soft tissue lengthening in different planes and reconstruction [11].

### **Case Report**

A 65-year-old woman with a medical history of hypertension was referred to our hospital, after experiencing a type IIIB Gustillo-Anderson open fracture in her left distal tibia. The fracture was complicated by deep infection. The fracture, classified as 43A3 according to AO classification [12], occurred after a fall from a height. (Müller AO classification of fractures is a system for classifying bone fractures, which was initially published in 1987 by the AO Foundation as a method of categorizing injuries according to the prognosis for the patient's anatomical and functional outcome. AO is an initialism for the German Arbeitsgemeinschaft für Osteosynthesefragen, the predecessor of the AO Foundation.) The patient was initially treated at the local hospital with intravenous antibiotics and stabilization of the fracture with a transarticular external fixation. Wound closure at the medial side was not possible. In the following days, the patient's condition deteriorated due to the fracture becoming complicated by a deep infection, and the patient was admitted in our hospital.

Bone necrosis was obvious and aggressive debridement was necessary. Bone transport was planned for treating the resulting bone loss. The existing hardware and the devitalized soft tissue and bone had to be excised. To obtain bleeding bone, 13 cm was removed from the distal tibia, including the joint surface. Specimens of bone and tissue from multiple sites were obtained for bacterial cultures.

A preconstructed classic Ilizarov ring fixator (PITKAR®) consisting of 2 supporting rings proximally, 2 movable rings at the level of the transporting bone, and a foot plate was used (Figure 1). Corticotomy by drilling multiple holes and chiseling took place at the tibial proximal metaphysis. Primary wound closure was not feasible, so the limb was acutely shortened by about 3 cm to facilitate soft tissue closure [13]. Wound coverage was achieved from surrounding muscles, while a 6×6-cm skin defect remained. We planned to cover this defect with a split-thickness skin graft after soft tissue survival was ensured. All bone surfaces were covered to avoid necrosis and osteomyelitis. The vascular status of the foot had to be monitored during shortening by Doppler ultrasound. The vessel loop shoelace technique was used to gradually approximate the edges of the wound [14]. The same loops, in addition to many others, were used to expand and support the skin over the tibia throughout the transportation period (Figure 2). The split-thickness free graft, with donor area from the ipsilateral femur, was applied 2 weeks later by the plastic surgeon. No complications occurred during the operation. Enterococcus faecalis and Staphylococcus epidermidis grew in the intraoperative cultures, and the patient was treated with intravenous antibiotics for 8 weeks.

Bone transport was initiated 10 days postoperatively at a rate of 0.75 mm/d [15,16]. This rate had to be reduced when pain was not well tolerated [17]. Partial weight bearing was allowed 2 weeks after the operation.

The transported bone segment reached the docking surface of the talus 210 days postoperatively. The tibial plafond had already been excised, and the descending bone fragment was to be fused with the talus. The skin that was entrapped between the transported bone and the talus had to be excised to avoid skin necrosis, and the tibial edge was revitalized. Callus consolidation was achieved after 14 months. The frame was dynamized when 3 cortices were evident on the X-ray. It was removed in the operating theater under anesthesia 4 weeks

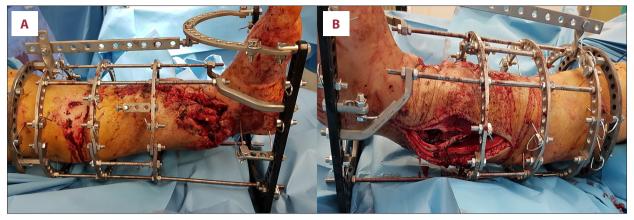


Figure 1. Pre-constructed classic Ilizarov ring fixator application. (A) Medial view and (B) lateral view.

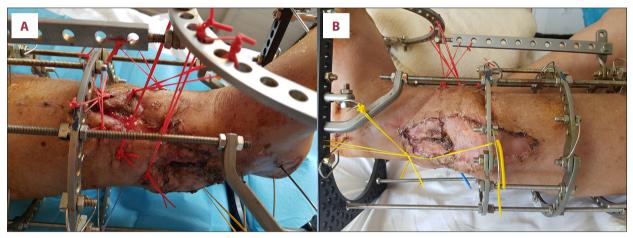


Figure 2. The vessel loop shoelace technique. (A) Red loops were used to support the skin over the tibia throughout the transportation period. (B) Yellow loops were used to expand the intact skin in order to gradually approximate the edges of the wound.

later. The regenerated bone was evaluated both clinically (Figure 3A, 3B) and radiologically (Figure 3C, 3D).

When the frame was removed, both fusion of the ankle and regenerated bone consolidation had been achieved. The results were evaluated according to the criteria reported by Paley et al. [18] and the Association for the Study and Application of the Method of Ilizarov (ASAMI) score [19]. The knee exhibited a range of motion from 0° to 125°. Axial deformity was not observed. The ankle was fused at a neutral position. The limb length discrepancy was less than 10 mm, with the injured limb being shorter. Total time in frame was 643 days. The external fixation index (time spent in an external fixator) was estimated to be equal to 49.6 d/cm. The patient was able to return to her previous level of activity during 6 months following the fixator removal. No significant pain was present when the treatment was completed. Clinical, laboratory, and radiological testing did not show any active infection. According to the ASAMI scoring system, the bone results were excellent, but because ankle had to be fused, the functional results were only scored as good.

Taking into consideration the length of time the patient had to be in frame, pin tract infection was unavoidable. The most severe incident was when an infection was complicated by cellulitis and classified as grade II pin tract infection according to Paley [17]. Intravenous antibiotic treatment had to be administrated and the Kirschner wires placed in the affected area had to be relocated. Oral antibiotics were prescribed for minor pin tract infections (grade I) twice.

#### Discussion

Emergency debridement, stable fixation, and early wound coverage are the basic principles in open fracture treatment [20]. The wound has to be covered in the first 72 h [21]. Patients should be treated in trauma centers where both plastic and orthopedic surgeons are available [2]. The time interval between injury and admission to a trauma center for definite treatment seems to be a more important prognostic risk factor for infection, even compared with early debridement [22]. In the present case, the patient remained exposed in a highly



Figure 3. Clinical and radiological appearance of the leg after removal of the Ilizarov frame. (A) Anteroposterior clinical view. (B) Lateral clinical view. (C) Standard anteroposterior xray of the regenerated bone and ankle fusion and (D) standard lateral xray of the regenerated bone and ankle fusion.

e926622-4

contaminated environment, and when she was finally admitted in our trauma center, aggressive debridement was the only solution to prevent impending sepsis. However, the excision of necrotized soft tissue and bone resulted in large deficits, making the reconstruction even more demanding.

Limb reconstruction is a challenging procedure and many treatment methods have been developed, each with advantages and disadvantages. Autologous bone grafting is indicated for defects less than 5 cm [23]. This procedure is accomplished in 1 stage, but it is accompanied by donor site morbidity [24]. Further, the quantity of the available graft is limited. Free vascularized fibula transfer from the contralateral limb is effective for restoring large bone and even soft tissue deficits when osteocutaneous flaps are used. Microsurgical skills and a considerable amount of operating time are required, and blood loss is larger than with the Ilizarov method. Another disadvantage of the free vascularized fibula transfer is the donor site morbidity [25,26]. However, it is a valid choice when large bone deficits are being treated. In the long run, stress fractures may complicate cases of fibula transfer [27]. The induced membrane (Masquelet) technique seems to be as effective as distraction osteogenesis in bone defect restoration [28,29]. The autograft requirements and the subsequent donor site morbidity have to be taken in consideration in the case of massive defects.

Distraction osteogenesis has been used to successfully treat bone defects up to 24 cm [30]. The use of an external fixator makes the technique less invasive, with less blood loss [31]. There is no need for a graft, and donor site morbidity is not a factor. Thus, the Ilizarov method has been the criterion standard for treating segmental tibial defects since the early 1990s [32,33]. When this method is combined with the circular ring fixator, maximum stability with minimum exposure to hardware is obtained. The length of the treatment is the main disadvantage of the method because it increases the probability of complications and the psychological burden of the patient.

Bone transport is performed when a large defect is present, and the deficit is filled while the transported segment is moving [34]. Since the bone loss in our case was around 13 cm, acute shortening would not suffice to achieve the contact of the bone edges and bone transport was selected as the treatment method. In the present case, acute shortening served the purpose of wound closure, with the applied technique being more of a bone transport than a bifocal compression distraction osteogenesis. Both bone transport and distractioncompression osteogenesis require a similar time in frame and show excellent results [35]. A lengthening of 3 cm after the end of the bone transport was necessary to restore the initial length of the limb. Soft tissue management is of equal importance to bone defect repair. Many reconstructive options are available, including free or local flaps and grafting. In 1982, Mathes et al. [36] introduced the reconstructive ladder concept in plastic surgery. According to this concept, the simplest of the effective methods of treatment should be used, and 4 decades later, the concept remains valid [37]. Distraction osteogenesis has been reported to combine successfully even with the more complex techniques of plastic surgery, such as the free vascularized flaps [38]. On the other hand, the use of the llizarov method may help to restore soft tissue defects without the need for the most demanding techniques of plastic surgery [39]. Soft tissue healing may benefit from distraction osteogenesis because it has been proved to increase the vessel density in the distraction area [40].

Our novel approach was initially to expand and eventually elevate the soft tissues of the tibia from the ring fixator frame using vessel loops. In this way, a suspension mechanism was created and was used throughout the transportation period. The vessel loops were attached to the tibia with metal surgical clips.

In writing this report, we hope to give new ideas to our colleagues in dealing with these difficult open, contaminated fractures, using the concept of the Ilizarov method and avoiding limb amputation with all its complications.

As far as we know, this case was the first time that this method was used to expand and support the soft tissues in a vertical plane during a bone transport.

# Conclusions

The case presented here showed that a multidisciplinary approach may be required for the successful management of neglected open fractures of the tibia and that the Ilizarov approach to both bone and soft tissue lengthening and reconstruction should be considered to ensure limb salvage and improve the final cosmetic appearance.

### Statement

Investigation performed at the Departments of Orthopaedic and Radiology of Patras University Hospital, Patras, Greece

### **Conflict of interest**

None.

## **References:**

- Court-Brown CM, McBirnie J: The epidemiology of tibial fractures. J Bone Joint Surg Br, 1995; 77: 417–21
- 2. Naique SB, Pearse M, Nanchahal J: Management of severe open tibial fractures. J Bone Joint Surg Br, 2006; 88-B: 351–57
- Aktuglu K, Erol K, Vahabi A: Ilizarov bone transport and treatment of critical-sized tibial bone defects: A narrative review. J Orthop Traumatol, 2019; 20(1): 22
- 4. Lasanianos NG, Kanakaris NK, Giannoudis PV: Current management of long bone large segmental defects. Orthop Trauma, 2010; 24(2): 149–63
- Gubin A, Borzunov D, Malkova T: Ilizarov method for bone lengthening and defect management review of contemporary literature. Bull Hosp Jt Dis, 2016; 74(2): 145–54
- Chan JK, Harry L, Williams G, Nanchahal J: Soft-tissue reconstruction of open fractures of the lower limb: Muscle versus fasciocutaneous flaps. Plast Reconstr Surg, 2012; 130(2): 284–95
- Reddy RS, Kumar CY, Shah HM et al: Evaluation of tibial condyle fractures treated with Ilizarov fixation, a prospective study. J Clin Diagn Res, 2014; 8(11): LC05–7
- Yin P, Zhang Q, Mao Z et al: The treatment of infected tibial nonunion by bone transport using the Ilizarov external fixator and a systematic review of infected tibial nonunion treated by Ilizarov methods. Acta Orthop Belg, 2014; 80(3): 426–35
- 9. Spiegelberg B, Parratt T, Dheerendra SK et al: Ilizarov principles of deformity correction. Ann R Coll Surg Engl, 2010; 92(2): 101–5
- Ilizarov GA, Ledyaev VI, Golykhovsky V: The replacement of long tubular bone defects by lengthening distraction osteotomy of one of the fragments. Clin Orthop Relat Res, 1992; 280: 7–10
- 11. Gustilo RB, Anderson JT: Prevention of infection in the treatment of one thousand and twenty-five open fractures of long bones: Retrospective and prospective analyses. J Bone Joint Surg Am, 1976; 58: 453–58
- Müller ME, Nazarian S, Koch P: Classification AO des fractures. Tome I. Les os longs. Berlin, Springer-Verlag, 1987
- Sen C, Kocaoglu M, Eralp L et al: Bifocal compression-distraction in the acute treatment of grade III open tibia fractures with bone and soft-tissue loss: A report of 24 cases. J Orthop Trauma, 2004; 18(3): 150–57
- 14. Berman SS, Schilling JD, McIntyre KE et al: Shoelace technique for delayed primary closure of fasciotomies. Am J Surg, 1994; 167(4): 435–36
- Alkenani NS, Alosfoor MA, Al-Araifi AK, Alnuaim HA: Ilizarov bone transport after massive tibial trauma: Case report. Int J Surg Case Rep, 2016; 28: 101–6
- Catagni MA, Azzam W, Guerreschi F et al: Trifocal versus bifocal bone transport in treatment of long segmental tibial bone defects. Bone Joint J, 2019; 101-B(2): 162–69
- 17. Paley D: Problems, obstacles, and complications of limb lengthening by Illizarov. Clin Orthop Relat Res, 1990; 250: 81–104
- Paley D, Catagni MA, Argnani F et al: Ilizarov treatment of tibial nonunions with bone loss. Clin Orthop Relat Res, 1989; 241: 146–65
- Shahid M, Hussain A, Bridgeman P, Bose D: Clinical outcomes of the Ilizarov method after an infected tibial non union. Arch Trauma Res, 2013; 2(2): 71–75
- National Institute for Health and Care Excellence (NICE): Fractures (complex): assessment and management: NICE guideline [NG37]. 2016. https:// www.nice.org.uk/guidance/NG37/chapter/recommendations
- 21. Gopal S, Majumder S, Batchelor GB et al: Fix and flap: The radical orthopaedic and plastic treatment of severe open fractures of the tibia. J Bone Jt Surg, 2000; 82(7): 959–66

- Pollak AN, Jones AL, Castillo RC et al: The relationship between time to surgical débridement and incidence of infection after open high-energy lower extremity trauma. J Bone Jt Surg, 2010; 92(1): 7–15
- 23. Barlow BT, Smith W: Management of segmental abstract. J Am Acad Orthop Surg, 2015; 23(3): 143–53
- Loeffler BJ, Kellam JF, Sims SH, Bosse MJ: Prospective observational study of donor-site morbidity following anterior iliac crest bone-grafting in orthopaedic trauma reconstruction patients. J Bone Jt Surgery Am, 2012; 94(18): 1649–54
- El-Gammal TA, El-Sayed A, Kotb MM: Microsurgical reconstruction of lower limb bone defects following tumor resection using vascularized fibula osteoseptocutaneous flap. Microsurgery, 2002; 2295: 193–98
- El-Gammal TA, Shiha AE, El-Deen MA et al: Management of traumatic tibial defects using free vascularized fibula or Ilizarov bone transport: A comparative study. Microsurgery, 2008; 28(5): 339–46
- Chew WY, Low CK, Tan SK: Long-term results of free vascularized fibular graft. A clinical and radiographic evaluation. Clin Orthop Relat Res, 1995; 311: 258–61
- Masquelet AC, Begue T: The concept of induced membrane for reconstruction of long bone defects. Orthop Clin North Am, 2010; 41(1): 27–37
- Tong K, Zhong Z, Peng Y et al: Masquelet technique versus Ilizarov bone transport for reconstruction of lower extremity bone defects following posttraumatic osteomyelitis. Injury, 2017; 48(7): 1616–22
- Wen H, Yang H, Xu Y: Extreme bone lengthening by bone transport with a unifocal tibial corticotomy: A case report. BMC Musculoskelet Disord, 2019; 20(1): 555
- Cierny G, Zorn KE: Segmental tibial defects. Comparing conventional and Ilizarov methodologies. Clin Orthop Relat Res, 1994; 301: 118–23
- 32. Dagher F, Roukoz S: Compound tibial fractures with bone loss treated by the Ilizarov technique. J Bone Joint Surg Br, 1991; 73-B(2): 316–21
- 33. Xu K, Fu X, Li YM et al: A treatment for large defects of the tibia caused by infected nonunion: Ilizarov method with bone segment extension. Irish J Med Sci, 2014; 183(3): 423–28
- 34. Rees A, Saleh M: Bifocal surgery for deformity and bone loss lower limb fractures. J Bone Jt Surg Br, 1995; 77-B(3): 429-34
- Mahaluxmivala J, Nadarajah R, Allen PW, Hill RA: Ilizarov external fixator: acute shortening and lengthening versus bone transport in the management of tibial non-unions. Injury, 2005; 36(5): 662–68
- 34. Ilizarov GA: The tension-stress effect on the genesis and growth of tissues. Part I. The influence of stability of fixation and soft-tissue preservation. Clin Orthop Relat Res, 1989; 238: 249–81
- Sigmund IK, Ferguson J, Govaert GAM et al: Comparison of Ilizarov bifocal, acute shortening and relengthening with bone transport in the treatment of infected, segmental defects of the tibia. J Clin Med, 2020; 9(2): 279
- Mathes SJ, Nahai F, Louis ST: Clinical applications for muscle and musculocutaneous flaps. 2<sup>nd</sup> printing edition. Mosby Inc., 1982
- Soni SK, Shaikh MF, Rana B: Lower limb defects and effectiveness of step ladder pattern in its management in present time (2019). Int J Med Res Health Sci, 2019; 8(6): 17–21
- Mahajan RK, Srinivasan K, Singh M et al: Management of post-traumatic composite bone and soft tissue defect of leg. Indian J Plast Surg, 2019; 52(1): 45–54
- Rozbruch SR, Weitzman AM, Watson JT et al: Simultaneous treatment of tibial bone and soft-tissue defects with the Ilizarov method. J Orthop Trauma, 2006; 20(3): 194–202
- Matsuyama J, Ohnishi I, Kageyama T et al: Osteogenesis and angiogenesis in regenerating bone during transverse distraction: Quantitative evaluation using a canine model. Clin Orthop Relat Res, 2005; 433: 243–50