Biomedical and Case Reports Open Access Open Journal

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Case Report

An Unusual Case of an Extensive Proximal Tibia Bone Defect Secondary to Complicated Cellulitis Managed by Unifocal Bone Transport Using an Ilizarov Fixator

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Received: May 15th, 2021; **Accepted:** May 19th, 2021; **Published:** May 20th, 2021

Citation: Venkataram. V, Manoharan M. An unusual case of an extensive proximal tibia bone defect secondary to complicated cellulitis managed by unifocal bone transport using an ilizarov fixator. Biom Case Rep Open A Open J. 2021; 2(2): 66-70. doi: 10.33169/biomcase.BACROAOJ-2-118

ABSTRACT

Introduction

Bone defects occur following bone loss secondary to trauma or due to infection. Extensive bone defects are usually seen in the former. It is unusual to have large defects in isolated osteomyelitis without prior history of any trauma. We are reporting a case with a bone defect of around 17 cm due to infectious aetiology and its management by unifocal bone transport using the Ilizarov ring fixator.

Case Report

The present report describes a 45-year-old gentleman with newly diagnosed diabetes mellitus who was initially admitted under general surgery for management of cellulitis and was later admitted under orthopedics for suspected osteomyelitis of left proximal tibia and septic arthritis of left knee. Serial debridement, prolonged antibiotic therapy and skeletal stabilization by a temporary joint spanning external fixator was applied to control the infection. The infection finally resolved after 6 weeks with a bone defect of approximately 17 cm. Options for reconstruction were discussed with the patient along with their pros and cons. Unifocal bone transport by ilizarov ring fixator was finalized with the patient's consent with the aim of achieving a functional limb. Bone healing index was 1.17 months/cm. ASAMI scoring showed excellent bone result and good functional result. LEFS was 55/80. The lower scores were because of the fused knee.

Conclusion

To the best of the authors' knowledge this is the largest defect secondary to infection without history of trauma which has been successfully treated by unifocal bone transport.

Keywords: *Bone defect; Unifocal bone transport; Ilizarov fixator; Osteomyelitis; Cellulitis.*

INTRODUCTION

Bone defects secondary to osteomyelitis pose comprehensive challenges in management. A combination of compromised soft tissue and an active infection makes skeletal stabilization and reconstruction difficult. Various treatment modalities have been suggested like the Masquelet technique, vascularized fibula graft, Limb Reconstruction System (LRS) and Ilizarov ring fixator.[1-4]

Infection related defects have been reported extending up to 13.5 cm in literature.[3] Bone transport is indicated to address such defects with segmental transport for defects exceeding 10 cm.[5] Based on reported literature, the longest lengthening achieved using a single level corticotomy is 24.5 cm in tibia.[6] Prior to that a lengthening of 14.5 cm was achieved in tibia by unifocal lengthening.[7] Both of these cases were due to bone loss secondary to trauma related complications.

The present case is unusual in that it occurred as a complication of a mismanaged case of cellulitis of the leg leading to osteomyelitis and sep-

tic arthritis. It resulted in massive bone loss of around 17 cm involving the entire proximal tibia including the articular surface.

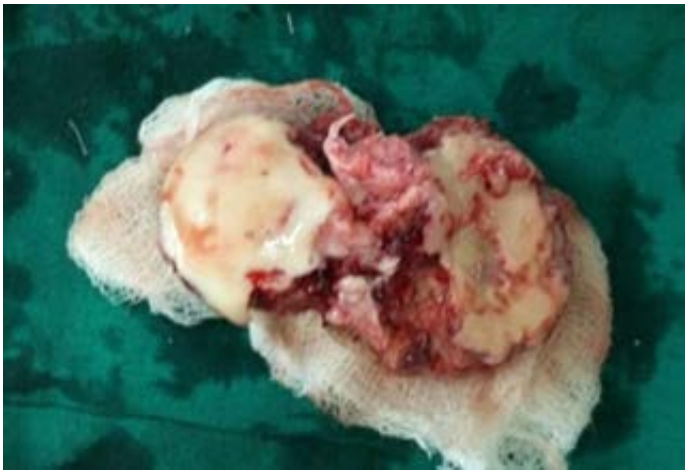
CASE PRESENTATION

In August 2015, a 45-year-old man was admitted under general surgery ward for management of cellulitis of the left leg. He was diagnosed with Diabetes Mellitus at that point. He received intravenous antibiotics and underwent debridement for uncontrolled infection. Non-healing ulcer had developed over the anteromedial aspect of the proximal tibia which was managed by a gastrocnemius flap.

He was then referred to orthopaedics after three months with pus discharge and knee swelling. On examination he was found to have discharging sinus from his surgical scar. There was severe tenderness over the proximal tibia. The knee joint was found to be swollen, warm and range of movements were painful and restricted. X-ray of the leg and USG of the knee were done to assess the extent of involvement. Pus was sent for culture and sensitivity. He was then diagnosed to have chronic osteomyelitis of tibia and septic arthritis of the knee.

Patient was taken up for staged debridement. The entire proximal tibia was found to be sequestered. Arthrotomy revealed destruction of articular cartilage (Figure 1). Hence the entire proximal tibia was removed and the limb was stabilized temporarily with a knee spanning external fixator. Post debridement the defect was found to be approximately 15cm. Intra-operative and pre-operative cultures showed growth of Methicillin Resistant Staphylococcus Aureus (MRSA) sensitive to vancomycin and linezolid. Intra-venous vancomycin was given for two weeks followed by oral linezolid for four weeks.

Figure 1: Intraoperative picture showing the destroyed articular cartilage of the proximal tibia

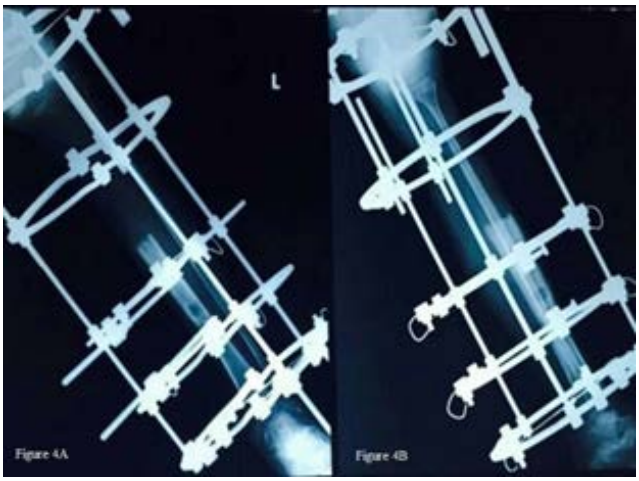


At the end of six weeks, he was reassessed. The soft tissue had healed adequately and the infection had settled down which was confirmed by relevant clinical and haematological parameters (Figure 2). After discussing all possible surgical options with the patient, we decided to go for Ilizarov ring fixator and bone transport.

During ilizarov application a further 2cm of bone was excised as the edges showed no bleeding and there was unhealthy granulation tissue. The bone was nibbled till fresh bleeding edges. Antibiotic bone cement beads were packed into the proximal part of the medullary cavity. Ilizarov ring fixator was applied spanning the knee (Figure 3).

Figure 2: Healed soft tissues and controlled infection before ilizarov fixator application

Figure 3: Immediate post-operative X-ray in AP and lateral views. AP – Anteroposterior



Corticotomy was done in the same sitting and bone transport was started at the end of five days. Distraction started initially at the rate of 0.5 mm per day. It was continued for three weeks and then increased to 0.75 mm per day at the end of three weeks. After six weeks the rate was increased to 1 mm per day.

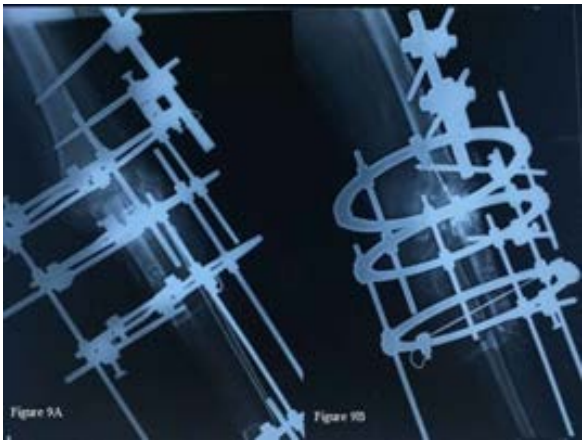
Regular X-rays were taken at the end of one month, two months, four months and six months to assess bone transport and regenerate formation.

At the end of six months the transported bone fragment began to push through the soft tissue defect at the proximal gap.

Patient was taken up for surgery. Acute docking and proximal fibula cuff resection were done (Figure 4). The resected fibula was fixed with the docked tibia using 3.5 mm screws to increase the surface area for fusion with the femoral condyles and to make load bearing more evenly distributed (Figure5).

Figure 4: Intraoperative picture showing acute docking of proximal tibia

Figure 5: Post-operative X-ray showing the docked tibia along with the fibula



The limb was found to be still shortened by 5 cm. Hence limb lengthening was continued for another two months until the length was equalized.

A total of 17 cm of regenerate was achieved over a period of eight months. The ilizarov ring fixator was continued for another one year after stopping the bone transport making the total duration of fixator application to an approximate time period of 20 months to allow time for regenerate consolidation and healing. The bone healing index was 1.17 months/cm.

During this time progressive loading was encouraged by loosening the fixator to promote fusion at the knee joint and to improve the consolidation of the regenerate. After removal of the fixator patient was put on an above knee fiberglass cast for another 6 months and allowed to weight bear fully.

Outcome was assessed by the ASAMI (Association for the Study and Application of the Method of Ilizarov) scoring system.[8] Bone result was found to be excellent with solid fusion (bony union), no infection or deformity with LLD (limb length discrepancy <2.5cm) as seen in Figures 6 and 7 respectively. Functional result was found to be good where patient could perform all his activities of daily living (ADL) with minimal difficulty.

Figure 6: Follow-up X-ray at 24 months showing complete fusion

Figure 7: Clinical picture at 24 months follow-up showing no active infection, with LLD < 2.5cm. LLD - limb length discrepancy

24 months after fiberglass cast removal, we performed a follow-up evaluation using Lower Extremity Functional Score (LEFS).[9] The LEFS was found to be 55/80. There were no adverse or unanticipated events during the entire treatment duration (Figure 8).

Figure 8: The total length of regenerate achieved and consolidated at the latest follow-up

The summary of all events has been briefly tabulated under Table 1.

Table 1. Summary of events

Time Frame	Important events
Day 1	Cellulitis of left leg
First week	Debridement and intravenous antibiotics
6 weeks	Gastrocnemius Flap for non-healing ulcer in proximal tibia
3 months	Presents to orthopedics OPD for the first time with discharging sinus and painful swollen knee. Taken up for debridement. 15cm bone defect.
4 and half months	Debridement and Ilizarov ring fixator application with distal corticotomy. Bone transport was started 5 days later.
10 months	Acute docking with cuff resection of proximal fibula and fibula grafting of proximal tibia. Around 5cm bone defect present. Lengthening continued.
12 months	Bone lengthening stopped.
24 months	Ilizarov ring fixator removed and above knee articast applied.
30 months	Articast removed and patient resumed normal activities.
OPD - Out-patient Department; LLD - Limb length discrepancy	

DISCUSSION

Bone defects secondary to infection are complex and difficult to manage. The dual challenges of controlling the infection and addressing the bone defect are why they are so troublesome to address. However, they have been successfully managed with the ilizarov ring fixator or by the LRS.[3,10,11]

Bone defects secondary to osteomyelitis usually occurs due to a combination of multiple factors. Infection and inflammation mediated damage to bones lead to osteolysis and bone destruction.[12] The process of surgical debridement also leads to further bone loss.

Our case is particularly unique because of its unusual onset as cellulitis and its rapid progression to extensive osteomyelitis with septic arthritis involving the knee joint within a short time period. We believe a combination of patient related factors like uncontrolled diabetes mellitus and under estimation of the extent of infection initially had led to his condition.

The initial goal of management was to control and eradicate infection before embarking on reconstruction. This was carried out by staged debridement and temporary skeletal stabilization by means of a joint spanning external fixator. The total defect post both debridement procedures came at around 17cm. Joint had to be sacrificed as the articular cartilage was destroyed due to the infection.

A defect of this size is best addressed by bone transport.[13] Moreover, it is also recommended to perform a trifocal bone transport to address such a large defect.[3,14] However, in our case the absence of the entire proximal tibia rules out such a transport.

The entire duration from distal tibia corticotomy to fusion with the distal femur and consolidation of the regenerate lasted for a period of 20 months in the ilizarov fixator. The patient was placed on an above knee articast for a further period of 6 months after fixator removal as an added precaution.

Fibula strut grafts harvested from the ipsilateral fibula was fixed with the transported distal tibia to ensure a more even load transmission from the distal femur and to augment the fusion. No cancellous bone grafts were used at the time of docking or later.

Many complications are reported in literature with prolonged use of ilizarov fixator for purpose of limb lengthening and bone transport.[15,16] Adequate diabetic control, diligent pin tract care and frequent monitoring ensured that our patient remained free of major complications during the bone transportation and consolidation phase. However, during bone transport, he developed clawing of the toes. It was managed by percutaneous Flexor Digitorum Longus tenotomy at the time of docking.

The relatively lower score in LEFS questionnaires were due to the absence of knee range of movement and the resulting functional limitations. The patient was nevertheless satisfied with his outcome as he had resumed most of his activities with respect to his day to day living.

CONCLUSION

Bone transport and limb lengthening is not possible without active participation from the patient. Credit needs to be given to the patient for his co-operation and willingness to come for regular follow-ups. Hence it is very important to have an open discussion before commencing the treatment enumerating the various pros and cons of the procedure. Patient expectations also have to be tailored in accordance with the expected outcome.

Our case report also reiterates the importance of aggressive management of soft tissue infections in diabetic patients and having a high index of suspicion of underlying bone infection in non-healing ul-

cers to prevent such a devastating complication.

CLINICAL MESSAGE

Bone and soft tissue infections require an aggressive management in Diabetics to prevent such a disastrous complication. Limb salvage using ilizarov ring fixator and bone transport is an option even in such extensive defects despite a compromised immunity.

CONFLICTS OF INTEREST

None.

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