



Case Report

Masquelet Technique for Management of Infected Tibial Diaphyseal Non-union in Ghana: A Case Report

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Abstract

Introduction: Infected non-union of the tibial diaphysis is a refractory disease with high recurrence rates. It may be complicated by persistent local infection, bone and soft tissue loss and limb deformity. The masquelet technique is cost-effective in managing bone loss in the treatment of infected non-union. It does not require specialized training and sophisticated instruments. **Case Report:** A 16-year old female with recurrent discharging sinuses on the right leg following open reduction and internal fixation of a tibial shaft fracture. There were shortening, muscle atrophy, discharging sinuses on the anteromedial right leg and stiffness of the right ankle joint. **Conclusion:** Infected tibial shaft non-union may run a protracted course with reduced productivity and missed work. It may be refractory to multiple surgical procedures and result in limb deformity. Operative treatment of the disease with the masquelet technique could be safe, effective and applicable in a limited-resource setting. Additionally, the masquelet technique improves bone union and limb function. Staged surgical treatment, involving thorough debridement, maximises treatment success. The masquelet technique is a viable choice for the management of infected tibial shaft non-union and improves bone union and limb function. Tri-calcium phosphate augmentation of autograft in the treatment of large long bone defects increases graft volume and could promote bone union. Staged surgical treatment of infected tibial diaphyseal non-union maximises treatment success.

Keywords: Masquelet; Infected non-union; Polymethyl Methacrylate Cement

Introduction

Infected non-union may be defined as failure of a fracture to adequately heal in the presence of progressive inflammatory destruction following bacterial seeding and may be associated with retained necrotic bone and/or colonized implant. The manifestations of the disease include local symptoms of pain, redness, swelling and persistent drainage of pus. In the disease process, bacteria form and remain hidden in a hydrophobic matrix -biofilm, which is avascular and out of reach of systemic antibiotics, making systemic antibiotic therapy ineffective. It is refractory and difficult to eradicate with reported recurrence rates of 20-30% [1,2]. The disease commonly results from high energy open fractures and the distal one-third of the tibia is the most commonly affected anatomic site. Open fractures of the tibial diaphysis are the most common predisposition, and the risk of the disease increases linearly with increasing degree of wound contamination and inadequate initial irrigation and debridement. Additionally, underlying immunosuppression such as diabetes, malnutrition, sickle cell disease or smoking history are known risk factors [3]. Infected non-union is a chronic and milder stage V chronic osteomyelitis and *Staphylococcus aureus* is the most commonly isolated organism.

Furthermore, tibial fracture nonunion may be complicated by persistent local infection or sepsis, bone and soft tissue loss and limb deformity [4]. A persistent infected non-union of the tibial diaphysis may lead to a below knee amputation. The condition poses a substantial management challenge to the orthopaedic surgeon and there is no consensus on the most appropriate reconstruction technique. The principles of treatment are to eradicate infection and achieve bone union. Treatment is fraught with prolonged, multiple and unsuccessful surgeries as well as protracted hospitalisations which may impact negatively on the quality of life of the patient and poses remarkable socio-economic burden [5]. Surgical treatment requires removal of dead bone pieces until bleeding bone margins are encountered (paprika sign) and reconstitution of the obliterated medullary canal [4]. Available treatment modalities involve debridement, flap coverage, filling the void with antibiotic beads, Papineau technique, free microvascular bone and soft tissue reconstruction and tibiofibular synostosis [5-7]. Debridement may result in large bone defect which may indicate skeletal fixation [8]. This is often achieved with a linear or ring external fixation- such as Ilizarov or Taylor Spatial Frame (TSF) [6,9] Treatment is often staged and multidisciplinary requiring, an orthopaedic surgeon, infectious disease specialist, plastic

surgeon and a radiologist. The first stage of surgery entails radical excision of all necrotic, infected tissue, and excision of sinus(es), while minimizing devascularization of bone and skin. Different techniques of managing the resulting bone void after debridement have been reported in literature with varying success rates. These include: masquelet technique, intercalary bone transport, Papineau technique and use of morphogenic proteins and tissue engineering techniques [9].

Masquelet Technique

The masquelet technique was first described in 1986 and is a two-stage procedure for bone loss management. The technique is relatively new and is indicated in bone loss due to trauma, resection of infected bone or tumour and has been shown to be effective in the treatment of tibial shaft non-unions after failure of previous treatment options [10]. The use of the Ilizarov technique is associated with an increased risk of fracture. Vascularized fibular autograft requires a microsurgical technique which is highly demanding and not widely available. The masquelet technique, on the other hand, relatively poses less technical demands and has been reported to offer satisfactory bone union after failed surgeries for Gustilo grade III open fractures. The procedure can be performed in the presence of a stabilizing implant. In addition, it is a cost-effective treatment method that does not require specialized training and sophisticated instruments [11-15]. The technique is preferred in the management of diaphyseal defects longer than 5cm and can be used to manage diaphyseal bone loss of up to 25cm in length [12]. The procedure entails placement of antibiotic impregnated Polymethyl Methacrylate (PMMA) cement spacer in the bone defect and temporizing for 6 weeks during which a pseudomembrane forms around the PMMA cement spacer. The cement is extracted at six weeks and the resulting void filled with autogenous bone graft which may be augmented with bone graft substitutes in large bone defects. The induced pseudo-membrane has biological and structural characteristics that promote bone union [3,9]. Moreover, it provides a vascular bed for graft incorporation into the adjoining bone. The pseudo-membrane further expresses growth factors such as transforming growth factor beta (TGF- β), bone morphogenic protein 2 (BMP-2), interleukin 6 (IL-6), interleukin 8 (IL-8), Von Willebrand factor (VWF) among others, that promote bone union. The membrane, additionally, provides an envelope to the graft that promotes graft revascularization. [10,13] However, the use of the masquelet technique on the tibia is more demanding compared to other long bones in the body due to the relatively poorer soft tissue envelop of the tibia [13]. Achieving favourable outcomes in the management of infected tibial diaphyseal non-union in a Lower-Middle Income Country could be uncommon due to resource constraints [13].

Methods

Case Summary

History: We report a case of a 16-year old female who was seen at the orthopaedic clinic with an 18-month history of recurrent discharging sinuses on the right leg following an initial closed fracture of the tibial diaphysis which was stabilized with plate and screws. The fracture failed to unite after a period of 12 months following which the plate and screws were removed and a stainless-steel tibial nail fixation of the fracture was performed. She had been out of school for 18 months. There was no history of immunosuppressive disorder such as diabetes, sickle cell disease, HIV or malnutrition.

Physical examination: General examination showed a well looking female with no fever, jaundice or pallor. The examination of the right leg showed multiple previous longitudinal hypertrophic surgical scars. There were three sinuses on the anteromedial right leg which were actively discharging malodorous greenish purulent fluid. There was shortening of the right leg by 5cm with stiffness of the right ankle joint and atrophy of the muscles of the leg. The initial x-ray showed an atrophic non-union of the right tibial diaphysis with an intramedullary nail in situ. The patient was counselled and prepared for a staged surgery and underwent three surgeries at separate sittings.

Surgical Procedure

The first stage of surgery took place in November 2020 and involved the use of the anterior approach to the tibial diaphysis. During the surgery, about 50mls of pus was evacuated, the intramedullary nail was removed. There were cerclage wires on the bone which were extracted as well. Extensive debridement was performed that entailed excision of fibrous tissue, sinuses and necrotic sclerotic bone until punctate bleeding from bone edges was encountered. There was an 8cm bone defect post-debridement. The medullary canal was reamed to remove all infected and necrotic tissue and the canal and the wound thoroughly irrigated with 9 litres of normal saline. During the procedure, tissue biopsy for histopathology, pus for bacteriological culture and sensitivity, were undertaken but the results were unremarkable. The leg was pulled to length and

the tibia stabilized with a Taylor Spatial Frame (TSF). We used gentamicin-impregnated PMMA cement spacer (Stryker PMMA cement, SimplexR HV with Gentamicin, Germany) to fill the resulting bone void. The incision was closed in layers with vicryl 1 interrupted sutures to subcuticular tissue and nylon 2 interrupted sutures to skin. Following surgery, the patient received intravenous cefuroxime for 7 days and oral clindamycin for 5 weeks making a total of 6 weeks antibiotic treatment. She commenced weight bearing as tolerated from post-operative day 1, with the aid of a pair of axillary crutches.

At the second stage of surgery, the PMMA cement spacer was extracted without disrupting the induced membrane and the resulting void filled with cortico cancellous ipsilateral iliac crest bone graft with retention of the TSF. The autograft was augmented with tri-calcium phosphate crystals [14]. The second surgery was performed 6 weeks after the first procedure, after which, the patient received intravenous Cefuroxime for 72 hours and then oral clindamycin for 14 days. The patient continued weight bearing as tolerated from post-operative day 1, with the aid of a pair of axillary crutches. At the third stage of surgery, the TSF was exchanged for a titanium interlocking, intramedullary hollow nail and additional ipsilateral cancellous iliac crest bone grafting undertaken to further stimulate bone union. Post-operatively, the pin sites were dressed weekly with Povidone iodine-soaked gauze pieces. The patient underwent isometric and range of motion exercises of the leg, knee and ankle. Following 6 weeks of antibiotics, the levels of Full Blood Count (FBC), C-reactive protein (CRP) and ESR normalized - (CRP- was less than 10mg/L, ESR was less than 20mmfall/hr). The intra-operative appearance of the tibial non-union, the extracted tibial nail and sequestra and the bone void after debridement are shown in figure 1. The extracted PMMA cement at 6 weeks, the immediate post-operative appearance of the affected leg and the TSF are depicted in figure 2. The appearance of the leg and the post-operative radiographs at 6 weeks are shown in figure 3. The state of the affected leg and the post-operative x-rays at 12 weeks following the second surgery and after removal of the TSF, have been depicted in figure 4. Figure 5 shows the appearance of the leg at one-year follow-up, with healed sinuses.



Figure 1: First surgery: Intra-operative appearance of the tibial non-union (left), the extracted tibial nail and sequestra (middle), the bone void after debridement (right).



Figure 2: Second surgery: Extracted PMMA cement at 6 weeks (left); immediate post-operative appearance of the right leg, with retention of the TSF (right).

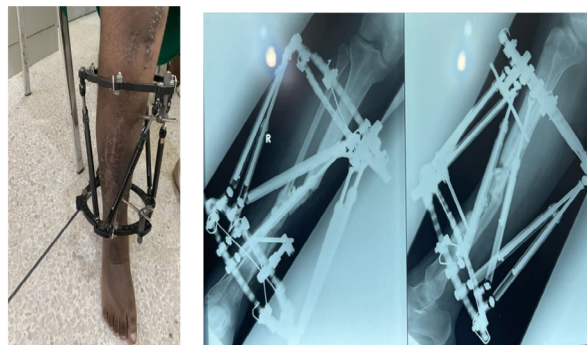


Figure 3: Appearance of the right leg at 6 weeks (left) and a post-operative x-ray.



Figure 4: Appearance of the right leg (left) and post-operative x-ray (right) at 12 weeks following the second surgery.



Figure 5: Appearance of the leg at one-year follow-up, showing healed sinuses (left), the x-ray appearance of the tibia (right) at 12 weeks after the third surgery (right).

Clinical Outcome Assessment at 1 year

The outcome of treatment was assessed using the association for the Study and Application of Methods of Ilizarov (ASAMI) instrument to evaluate bone union and limb function [15].

ASAMI score for bone union

- i. Union, no infection, deformity <7 degrees, limb length discrepancy <2.5 cm: Excellent
- ii. Union + any 2 of the absence of infection, deformity <7 degrees and limb length discrepancy <2.5 cm: Good
- iii. Union + any one of the absences of infection, deformity <7 degrees, limb length discrepancy <2.5 cm: Fair
- iv. Non-union, re-fracture or union + infection + deformity >7 degrees + Limb length discrepancy >2.5 cm

ASAMI score for limb function

- i. Active, no limp, minimum stiffness (loss of <15 degrees knee extension, <15 degrees ankle dorsiflexion, no RSD, insignificant pain: Excellent

- ii. Active, with 1 or 2 of the following: limp, stiffness, RSD, significant pain: Good
- iii. Active, with 3 or all of the following: limp, stiffness, RSD, significant pain: Fair
- iv. Inactive (unemployment or inability to perform daily activities because of injury): Poor
- v. Amputation: Failure.

One year following the last (third) surgery, the patient scores excellent on the ASAMI scale for bone union and for limb function. She fully bears weight on the right lower limb, there is no limb length discrepancy and no recurrent discharge. There is absence of pain, absence of abnormal movement at the fracture site and no Reflex Sympathetic Dystrophy (RSD). The limb is well aligned and there is absence of local signs of infection pain, redness or sinus discharge with improved right ankle mobility. The patient will be followed-up for signs of recurrent infection for at least 2 years.

Discussion

The patient of this report presented to us with an 18-month history of recurrent discharging sinuses on the right leg, she had been out of school for the duration of the disease. Additionally, the infection followed open reduction and internal fixation of a tibial shaft fracture and the patient had had multiple unsuccessful procedures prior to presentation to our clinic. There were multiple scars on the affected leg with leg shortening and ankle stiffness. The socioeconomic cost of infected non-union, has been highlighted by a number of authors.

Flores and others observed in their study that infected non-union remarkably undermines patients' quality of life, personal finances and hospital systems incurring indirect cost from reduced productivity and missed work [16,17]. Furthermore, in the treatment of the disease, we performed the surgical procedures in a staged manner beginning with radical debridement which resulted in an 8cm bone defect. The void could not be completely filled with autograft, necessitating commercial tri-calcium phosphate augmentation. The multistage operative treatment of infected non-union and the supplementation of autograft with commercial bone graft in large bone defects, 8cm or more, are corroborated by several studies. A study by McNally et al concluded that the multistage approach reduces infection recurrence rates [18]. Ozpolat and others, similarly, reported supplementation of autograft with commercial bone graft in large bone defects, recommending that tri-calcium phosphate is safe and effective autograft expander in the masquelet technique [14].

In the case of our study, following debridement, the tibia was stabilised with TSF which allowed the patient to bear weight with a pair of axillary crutches, as tolerated, on day 1 after surgery, and the TSF was retained on extracting the PMMA cement spacer at the second stage of surgery. Similarly, Durand and others in their study observed that an external fixator is the most common fixation method for infected non-union after debridement and reported that the masquelet procedure can be performed without removing a stable previous implant and that retaining the initial fixation device reduces operative time [13].

In our study, the extraction of PMMA cement and bone grafting was performed when there was no evidence of persistent infection. The cement was extracted without disrupting the induced membrane and the void filled with corticocancellous iliac crest bone autograft while the TSF was maintained [9]. In the use of the induced membrane technique, bone grafting is performed after resolution of infection. Furthermore, some authors have recommended the use of wound drain in the induced membrane to prevent partial graft loss due to haematoma, and progressive weight bearing over 6-7 months after bone graft placement. However, we did not believe that suction drainage was mandatory and did not use one in the case of our report [19]. The traditional masquelet technique is a two-stage surgery, however, in our case we performed a third surgery to exchange the TSF for an intramedullary nail upon resolution of the infection, at the request of the patient, who found the TSF bulky to bear [19]. In addition, the bacteriological culture of tissue and pus did not identify the offending organism(s) in our case. If it did, it could have guided our antibiotic treatment more effectively. In the patient of this case report the masquelet technique has proven to be safe and effective and the patient has excellent ASAMI score at 1 year. She fully bears weight on the right lower limb, there is no pain at the fracture site, no limb length discrepancy and no recurrent discharge. The safety and effectiveness of the masquelet technique has been reported by a number of authors. In their case series, Ozpolat et al. and Pesciallo et al. observed absence of pain at the fracture site at 3 months follow-up and concluded that the masquelet technique is an effective method of treatment of infected non-union with low recurrence rate [10,19,20].

Conclusion

Infected tibial shaft non-union may run a protracted course with reduced productivity and missed work. It may be refractory to multiple surgical procedures and result in limb deformity. Operative treatment of the disease with the masquelet technique could be safe, effective and applicable in a limited-resource setting. Additionally, the masquelet technique improves bone union and limb function, and tri-calcium phosphate augmentation of autograft in the management of post-debridement large bone defects, increases graft volume and could promote bone union.

Finally, staged surgical treatment of infected tibial diaphyseal non-union involving thorough debridement, maximises treatment success.

Lesson from the article

Treatment of infected non-union of the tibial diaphysis with the masquelet technique could be safe, effective and applicable in a limited-resource setting.

Clinical Message

Surgery improves function in patients with infected non-union of the tibial diaphysis.

Declaration of patient consent

Informed consent of the parents of the patient was sought and obtained for the images and clinical information to be reported. The parents of the patient understand that their names nor the name of their child will not be published and measures will be taken to conceal their identity.

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