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Extreme presentation of lower limb post-traumatic chronic osteomyelitis: a 56-year-long untreated and actively secreting fistula

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Summary

Untreated chronic osteomyelitis presentations are becoming less common, especially in industrialized countries, however the post-traumatic and post-surgical osteomyelitis are the most frequent causes of bone infection in the developed countries, accounting for 80% of bone infections. Around 10 to 30% of cases of the acute form become chronic. Moreover, chronic osteomyelitis and extensive bone defect still represent a challenging medical condition for orthopedic surgeons.

Key words: chronic osteomyelitis, lower limb amputation, multidrug resistant microorganism

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Introduction

Untreated chronic osteomyelitis presentations are becoming less common, especially in industrialized countries. However, post-traumatic and post-surgical osteomyelitis are the most frequent causes of bone infection in developed countries, accounting for 80% of bone infections [1]. Around 10 to 30% of cases of the acute form become chronic [2]. Moreover, chronic osteomyelitis and extensive bone defect still represent a challenging medical condition for orthopedic surgeons [3].

Description

A 78-year-old man came to the hospital complaining of pain and purulent discharge with a foul smell from his left leg, covered by an inappropriate dressing. After bandage removal, there was scar tissue along the full length of the antero-medial side of the tibia, fixed equinus foot deformity, and stiffness of knee and ankle joint. There was a draining sinus anteriorly just 7 cm below the anterior tibial apophysis and a second one 4 cm above the ankle joint (Fig. 1).
An X-ray exam of the left lower limb showed extreme bone destruction of the medial and distal third of the tibial diaphysis with proximal third sclerosis and huge geodic cavities, both femorotibial and tibiotarsal arthrosis with severe osteoporosis of the tarsus and forefoot (Fig. 2).

Figure 1. Scar tissue along the full length of the antero-medial side of the tibia (A) with a double draining sinus, 7 cm below the anterior tibial apophysis and 4 cm above the ankle joint (B), combined with a fixed equinus foot deformity and stiff knee flexion (C).

The combination of both clinical and radiographic information directed the clinicians to chronic osteomyelitis as consequence of a trauma at the age of 22, or a 56-year-long chronic osteomyelitis. The medical history revealed an exposed tibial diaphysis fracture caused by a car accident, which was inappropriately treated, back in the 1960s, with an external fixator and exposure focus suture; unfortunately no clinical documentation was available.

A first diagnosis of post-traumatic infected non-union was given in another hospital in 2012, where an attempt of distraction osteogenesis was tried together with a dangerous sinus coverage by skin graft, procedure completed one year later with fixator removal and cast application. There was clinical evidence of resolution of infection nor X-ray demonstration of bone healing; the patient never returned to the previous hospital and had been living with osteomyelitis for 10 additional years.

On physical examination, the extensive putrefied and suppurative wound of the left lower limb showed a double outbreak of bone exposition, full of smelly serum; several cultural samples of that fluid were taken. The left knee was stiff in 30° flexion with a simultaneous retraction of the thigh flexors, while the ankle stiffness was associated with equinus foot and algodystrophic features; the entire left lower limb was shortened by 12 cm compared to the contralateral one. Neurovascular and muscular status of the anterior compartment of the left leg was compromised, the anterior tibialis and dorsalis pedis were unidentified, and foot tactile sensation was reduced.

The vital parameters were stable and no fever was referred during the days before hospitalization. The Lower Extremity Functional Scale (LEFS) had a score of 31/80 [4].

The patient was first treated with daily dressing and empirical ceftriaxone antibiotic (2 gm IV daily).

Microbiological analyses of the wound pus revealed a high level of gentamicin and kanamycin resistant *Enterococcus faecalis* and colistin resistant *Pseudomonas aeruginosa* with selective meropenem sensitivity.

The laboratory investigation revealed elevated serum inflammatory markers with lymphocytosis.

A CT scan with contrast was performed to evaluate soft tissue involvement and lower limb vascularization, showing lack of opacification of the superficial femoral artery, with minimal rehabitation downstream in the distal section by superficial circulation collateral branches and deep femoral artery perforating branches, and filiform peroneal and anterior tibialis arteries (Fig. 3). In addition, there were found large areas of extensive

Figure 2. X-ray exam showing an extreme bone destruction of the medial and distal third of the tibial diaphysis with proximal third sclerosis (A), both femorotibial and tibiotarsal arthrosis with severe osteoporosis of the tarsus and forefoot (B).

osteolysis, probably consisting of huge geodic cavities, involving both the proximal third of the tibia and the distal femur.
It was clear that the patient suffered from severe diffused chronic osteomyelitis of the left tibia according to Cierny-Mader classification type IV class A, caused by multidrug resistant microorganisms; the vascular supply of the limb was residual [5].
After thorough analysis and planning, an above-knee amputation was established together with the patient. Early antibiotic treatment was made in order to minimize the risk of surgical site infection and to maximize the length of non-infected tissue.
The surgery was performed in spinal anesthesia and under tourniquet ischemia, drawing an anterior thigh flap whose tip reached the edge of the patella, splitting the muscle from the femur and elevating periosteum circumferentially 4 cm above the superior attachment of the knee capsule, then cutting the femur at the distal third by using an oscillating saw. Both the femoral artery and vein were clamped and tied with heavy suture, and the sciatic and saphenous nerves dissected and retracted about 5 cm. The elevated periosteal flap was used to cover the femoral medullary canal and, before skin suture, the tourniquet was released and all bleeding was controlled [6] (Fig. 4).
The surgery was successfully completed. A bone biopsy was taken from the drainage sinus to confirm the previous bacterial identification, and culture showed the presence of multidrug resistant *Enterococcus faecium* and *Pseudomonas aeruginosa*. Intravenous meropenem was administered for 2 weeks to avoid residual infections on the amputation stump. No post-operative complications, such as skin flap failure, wound dehiscence, seroma, or hematoma were seen.
As wound healing was completed, the patient was referred to an external prosthetic center for the completion of the rehabilitation pathway.

Discussion

As already mentioned, chronic osteomyelitis represents a challenging medical condition, especially concerning the choice of antibiotic choice and the surgical procedure.
The very first challenge is represented by the sampling procedure, which in case of a wound swab is burdened with a high risk of false positives, i.e. a high risk of contamination, and low specificity. Superficial samples or swabs from fistulas have low accuracy compared with bone biopsy culture and should not be used for pathogen identification. Monomicrobial identification from several samplings may be needed to start pre-surgical antibiotic therapy. Blood cultures have high sensitivity only in acute forms of osteomyelitis and are often useless for correct diagnosis of chronic bone infections [7-9].
Chronic osteomyelitis is caused by monomicrobial infection in 48 to 88% of cases in observational studies, while a polymicrobial etiology is more common in post-traumatic infection. Gram-positive microorganisms are the most commonly isolated (around 60%) in chronic osteomyelitis, with a predominance of *Staphylococcus aureus*. Gram-negative organisms, like *Pseudomonas* spp and Enterobacteriaceae, along with anaerobes, have been more frequently reported in cases with polymicrobial etiology. In recent years, antimicrobial resistant organisms, mainly extended-spectrum beta-lactamases (ESBL) producing organisms and multidrug resistant (MDR) bacteria, have been increasingly reported as causative agents of chronic osteomyelitis, posing new therapeutic challenges [10,11].
The management of antibiotic therapy is always a crucial and complicated point for the orthopedist. Indeed, data show that antibiotics will not be able to eradicate a bone infection after a certain duration due to biofilm [10,12]. The biofilm progressively turns into a mature form after 48-72 h from the infection and the adhesion to bone or hardware becomes irreversible. Therefore, antibiotic therapy alone is not sufficient and must always associated with surgical and chemical debridement using antiseptic washing solutions.
As a general principle, there is not a fixed duration of antibiotic therapy. The time of administration might change depending on

Figure 3. CT scan with contrast showing both a vascular impairment of the lower limb (A) and large areas of extensive osteolysis, probably consisting of huge geodic cavities (B,C), involging both the proximal third of the tibia and the distal femur (D-F).

the surgery performed, on the clinical and laboratory response and does not necessarily depend on the pathogen isolated with few exceptions in which case there is a need for a long-lasting treatment as for tuberculosis, Buruli ulcer, Q fever, nocardiosis, or brucellosis. According to some authors, the total duration of antibiotic treatment, if associated with surgery, should be limited to 6 weeks [13]. A paper in 1990 demonstrated an unacceptably high risk of treatment failure when antibiotics are administered for less than 4 weeks [14]. Trials including patients treated for longer periods with intravenous or oral antibiotics, 6 months or more, do not suggest any improvement compared with 6 weeks of therapy [15].

According to the literature in acute osteomyelitis, antibiotic therapy with no surgery for 6 to 12 weeks at the most, is sufficient for resolution in 90% cases. In chronic forms the same range duration is also recommended, using inflammatory markers as main indicators for drug suspension. Concerning the type of administration, in former times experts usually recommended intravenous (IV) therapy for 4 to 6 weeks, followed by an oral course of additional weeks or months [16,17].

More recently, this trend has switched to IV treatment during the first 2 weeks. Reasons for limiting IV therapy are: better patient comfort, high costs, and prevention of catheter-related complications. A few retrospective studies suggest that regimens with an early switch to oral antibiotics are as effective as prolonged parenteral regimens [13,18].

In a review comparing IV to oral antibiotics in patients with osteomyelitis, there was no statistically significant difference in the remission rate at 12 months follow-up, but the rate of adverse events was significantly higher with IV administration [19]. In a retrospective study among 49 episodes of implant-free chronic osteomyelitis, 20% recurred. In addition, the authors concluded that 1 week of IV therapy had the same rates of remission as 2-3 weeks or more than 3 weeks [20]. Many classes of oral antibiotics might be considered as a valid option for treatment: metronidazole, quinolones, linezolid, and clindamycin.

Furthermore, another orthopedic challenge is represented by the surgical strategy to allow a complete removal of the infected bone and biofilm to both promote healthy bone and soft tissue healing and guarantee an adequate vascular supply and exposure to subsequent antibiotics. Moreover, in case of post-traumatic chronic osteomyelitis, there is often associated fracture non-union, with limb length discrepancy and deformity, so that eventual limb salvage procedure has to manage all these aspects at the same time, trying to impact as little possible the patient's quality of life. The infectious problems are

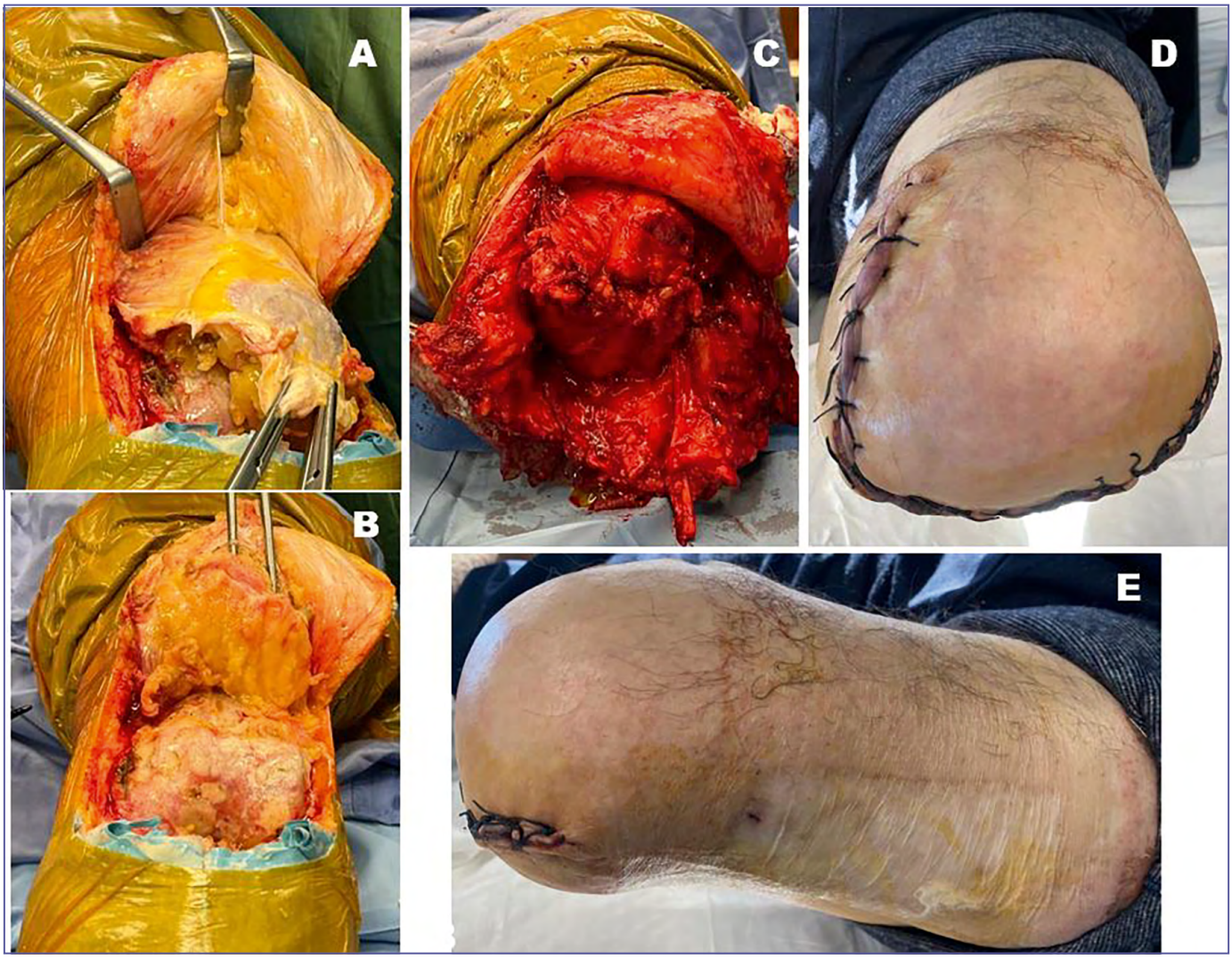

Figure 4. Surgical procedure by drawing an anterior thigh flap whose tip reached the edge of the patella (A), elevating periosteum circumferentially 4 cm above the superior attachment of the knee capsule (B), then amputating the femur above the knee and using the periosteal flap to cover the femoral medullary canal (C), before skin suture (D,E).

more complicated to manage in case of open tibia fractures because of the poor tibial healing due to the decreased soft-tissue coverage and naturally diminished vascularity of the region [21]. Even if the recent microsurgical orthoplastic solution has completely changed the modern surgical approach to "huge bone loss", clear guidelines on salvage vs amputation choice are not available [22]. Furthermore, amputation still represents a secure solution to the infection thanks to complete extremity eradication [23-25].

A recent systematic review focused on chronic/late onset fracture-related infection stated that recurrence of disease occurs in 6-9% of patients, leading to amputation of the affected limb in 3-5% of cases [26].

Recently, the AO Foundation and the European Bone and Joint Infection Society proposed a therapeutical algorithm for the management of fracture-related infection (FRI), starting from a consensus definition for FRI based on the presence of four confirmatory criteria: fistula, sinus or wound breakdown; purulent drainage from the wound or presence of pus during surgery; phenotypically indistinguishable pathogens identified by culture from at least two separate deep tissue/implant specimens; presence of microorganisms in deep tissue taken during an operative intervention, as confirmed by histopathological examination. Suggestive radiological signs of infection are bone lysis, failure of progression of bone healing (i.e. non-union), sequestration, and periosteal bone formation [27-29].

The appropriate surgical and pharmacological treatment should be chosen with a multidisciplinary approach, discuss-

ing the optimization plan and therapeutical options with the patient. The spectrum of surgical techniques in case of FRI includes DAIR (debridement, antibiotics, and implant retention), implant replacement, distraction osteogenesis, bone grafting with or without spacers, and amputation [30-32]. Surgical debridement is critical to the success of treatment in post-traumatic and post-interventional osteomyelitis [24].

However, despite guidelines, some cases of chronic osteomyelitis still escape the management of clinicians and evolve to irreversible and potentially life-threatening stages, as in the case report described of a 56-year-long fracture-related osteomyelitis. This should prompt reflection on how the length of the treatment path of osteomyelitis can often be exhausting and lifelong for the patient, up to the point of giving up the course of care to regain his own autonomy away from the hospital. The psychological impact of such a long "therapeutical journey", extricated from infection recurrences and residual dysmorphisms, is hardly taken in primary consideration in care decisions, because the irresolution of infectious disease is a shame for medical progress. But, according to the authors, sometimes the hardest choice for a clinician, i.e. limb amputation, can fit well to a specific patient, respecting both his/her wishes and mental balance.

Conclusions

Taking into consideration the patient's age and cultural background, amputation was an option considered for discussion; the neuromuscular and vascular status for most parts of the leg encouraged the decision, and the extreme pharmaco-resistance of the isolated microorganisms supported the choice. The aim of the amputation was removal of infected tissue, optimizing postoperative pain control and providing a residual limb that allows for prosthetic fit, in order to improve patient quality of care and life.

Conflict of interest statement

The authors declare no conflict of interest.

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Author contributions

MP (first author): mainly wrote this manuscript and performed the acquisition and interpretation of data; FRT, GP, AP: mainly performed the conception and design of study. All Authors read and approved the final manuscript.

Ethical consideration

This study was conducted ethically, with all study procedures being performed in accordance with the requirements of the World Medical Association's Declaration of Helsinki. Written informed consent was obtained from each patient for study participation and data publication.

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