

Case Report

A rare presentation of tubercular osteomyelitis of the foot

Marco Pes¹, Veronica Amorese¹, Andrea Baioni¹, Matthew Gavino Donadu^{2,3}, Paola Molicotti⁴, Fabio Milia¹, Carlo Doria¹

¹ Orthopaedic Department, University of Sassari, Italy

² Department Chemistry and Pharmacy, University of Sassari, Italy

³ Department of Biomedical Sciences, University of Sassari, Italy

⁴ S.C. Microbiologia, A.O.U. Sassari, Italy

Abstract

Tuberculosis is a communicable disease that is a major cause of ill health. It is one of the top ten causes of death worldwide and the leading cause of death from a single infectious agent. Its most common clinical presentation is pulmonary involvement. However, approximately 23–30% of tuberculosis patients have extrapulmonary symptoms. A rare (1%) clinical presentation of tuberculosis is foot and ankle infection. This is complicated by the fact that the diagnosis of osteoarticular tuberculosis is difficult. Our case was a 66-year-old multi-pathological pensioner, who, while working in the countryside, had an ankle sprain on the left foot, with a lacerated wound of about 2 cm diameter. The non-endemic area and the negative chest X-ray made the diagnosis extremely complex. However, a multidisciplinary approach with the radiologists and the infectious disease department led to clinical stabilization of the patient. Therefore, awareness and high index of suspicion of the disease is essential and referral to experts should be made if diagnosis is indeterminate despite extensive investigations. The knowledge allows early identification of the disease and prompt therapy in order to avoid long-standing untreated infections which typically cause bone destruction and loss of function. The knowledge is also mandatory for western physicians due to increasing international travel, immigration from less developed countries and increased use of immunosuppressive medications. We believe that this article can bring awareness around osteoarticular tuberculosis and help with improving outcome and eradication of the infection. Level of clinical evidence: 4.

Key words: foot; tuberculosis; osteomyelitis; extra-pulmonary tuberculosis.

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Introduction

Tuberculosis (TB), a communicable disease, is one of the top ten causes of death worldwide and the leading cause of death from a single infectious agent. About a quarter of the world's population is infected with *Mycobacterium tuberculosis* and thus at risk of developing the TB disease. Millions of people continue to be affected by TB each year. Globally, an estimated 10.0 million (range 9.0–11.1 million) people fell ill with TB in 2018 [1].

No bone is immune to involvement by TB, and the arthritis is monoarticular in 90% of cases. The most frequently involved joints are the weight-bearing joints such as hip, knee, shoulder or elbow [2]. Tuberculosis infections of the foot and ankle are very rare and comprise only 1% of all tuberculosis infections. Timing of diagnosis, patient compliance to therapy and rare presentation of symptoms are some difficulties that the clinicians can face. Although rare, musculoskeletal tuberculosis can be a problem. Its uncommon site, non-specific presenting symptoms and its ability to mimic

numerous disorders make it more difficult to formulate a definitive diagnosis, and this leads to therapeutic delays [3,4]. In addition, the diagnosis of osteoarticular tuberculosis (OT) is difficult to achieve. Therefore, awareness and high index of suspicion of the disease is essential and referral to expertise should be made if diagnosis is indeterminate despite extensive investigations [5]. The aim of this case report is to focus on awareness on this atypical presentation [6].

Case Report

A 66-year-old pensioner had an ankle sprain on the left foot and a lacerated wound of about 2 cm diameter while working in the countryside. The patient had a history of alcoholism, smoking (20 cigarettes/day until 40 years of age), hypertension, chronic obstructive pulmonary disease (COPD), and obesity in November 2015.

The patient went to the Accident and Emergency (A&E) department, where an X-ray imaging was done; the report was negative for fracture, and therefore the

patient was sent home with painkillers. The patient neglected himself for 7 months and in July 2016 he received the first specialist evaluation for pain in the foot and a Magnetic Resonance Imaging (MRI) scan (Figure 1) without contrast agent was made in August 2016. The MRI reported: “nodular mass of 49 × 25 × 19 mm between the IV and the V metatarsus”, suggesting the hypothesis of extra-articular ganglion or schwannoma.

After reviewing the imaging, we decided to perform excision of the mass. The patient was admitted to the hospital on 7 November 2016. During objective examination it was observed that the foot was swollen, with stretched and shiny skin, extremely painful and with a cutaneous fistula secreting a thick serohematic liquid. This liquid was sampled with two swabs; one of them tested positive for *Staphylococcus epidermidis*. The X-ray (Figure 2) of the foot showed osteoporosis

Figure 1. Image MRI Scan T2 with the presence of a nodular mass of 49×25×19mm between the IV and the V metatarsus.



“worthy of more investigations”; while the chest X-ray was negative.

On 21 November 2016, following the suspected diagnosis of osteomyelitis of the metatarsus, a surgical incision was made to remove the abscess. During the surgery, two additional swabs of the purulent liquid were taken at the surgical incision; both resulted in negative. The abscess broadened medially until the tendon and laterally to the bone; all the pus was removed and toilet of the wound and a surgical debridement with the position of a seton stitch were performed.

During the 1^o admission the patient was afebrile. Antibiotic therapy was prescribed as follows: Amoxicillin and Clavulanic Acid 875 + 125mg, 1 cp 3/die from 7 November to 10 November; Ciprofloxacin 750 1 cp 2/die from 10 November to 5 December; and Rifampicin 600 1/die from 10 November to 21 November.

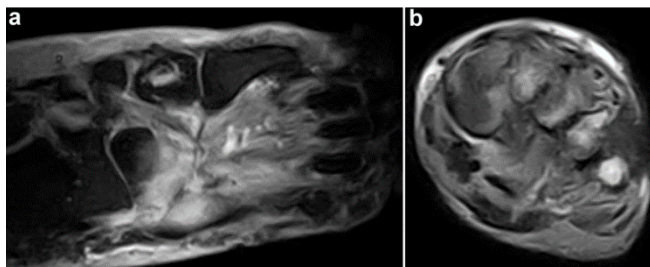
The patient was discharged on 28 November 2016 with the following indications: functional rest, leg elevation, walking with two crutches, out-patient medications and antibiotic therapy.

In March 2017 clinical conditions worsened on the left foot with a new cutaneous fistula on the previous surgical scar exuding a purulent and partially caseous secretion, increasing pain and functional deficit.

Figure 2. Lateral X-Ray of the foot showing osteoporosis among the metatarsus; worthy of more investigations in the opinion of the radiologist.



Figure 3. Erosion of the IV and the V metatarsus with pus and the presence of a fistula.



a. MRI STIR; b. MRI T2.

On the 29 March 2017 another MRI scan (Figure 3) was done, showing an erosion of the IV and V metatarsus with pus and a fistula. Therefore, we proposed a new admission for a second surgical approach.

The patient was admitted to the hospital for the second time on 27 April 2017. During the objective exam, it was observed that the foot was swollen, with dyskeratosis and hydropic swell of the skin, and there was a fistula over the previous surgery scar. Three new swabs were taken: the first was positive for *Kocuria kristinae*, *Corynebacterium amycolatum*, and multi-resistant *Corynebacterium pseudodiphth*, the second was positive for *Staphylococcus cohnii* spp, and the third was positive for *Mycobacterium tuberculosis*. These results were reported on 9 June 2017 and this delay was ascribed to the paucibacillary specimen and previous antibiotic treatments.

The patient underwent the second surgery on 15 May 2017 and a diamond cutaneous incision including the fistula was made. There was an emission of partially caseous purulent material at the origin of the fourth and fifth metatarsal, that had been completely exported. Therefore, a precise curettage of the bone was achieved and withdrawn for cultural and histological examinations for common bacteria and Koch's bacillus and positive findings were reported on 17 July 2017. This procedure was followed by an abundant wash out of the wound and the placement of a surgical drainage.

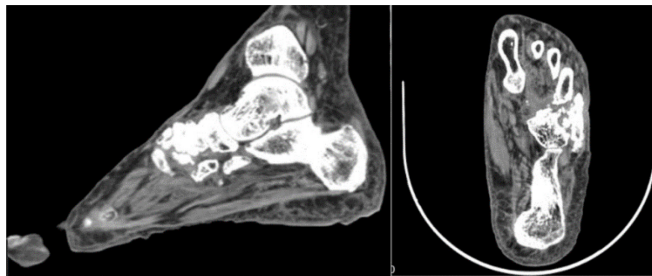
The antibiotic therapy which was prescribed following the indications of the Department of Infectious Diseases, University of Sassari, included: Daptomycin 750mg, 1 vial per day from 5 May 2017 to 1 June 2017, and Co-Trimoxazole 160 + 800mg 1 tablet 3 times per day from 2 June 2017 to 21 June 2017.

The patient was discharged on the 6 May 2017 and prescribed medications every two days for the treatment

Figure 5. Exterior condition of the foot at two years follow-up.



Figure 4. TC scan without contrast showing aspects of bones remodeling, compatible with tubercular osteomyelitis unchanged compared to the previous X-ray exams.



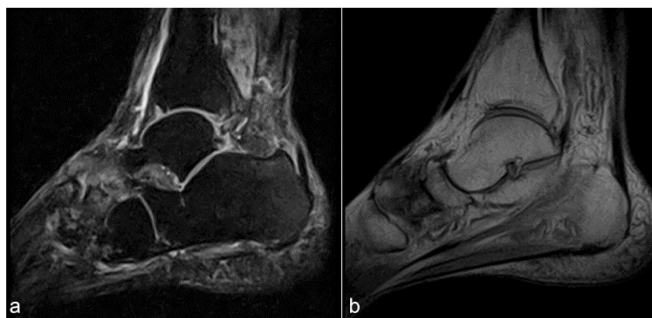
of the secreting fistula and periodic clinical follow-up at the Department of Infectious Diseases.

The patient was afebrile during the outpatient course in July 2017 with the persistence of two secreting branches of the fistula, characterized by absence of leaks of purulent material when squeezed. The skin did not appear warm on palpation but the patient continued to be in pain. When the histological findings were received, the Department of Infectious Diseases decided to begin *Mycobacterium tuberculosis* therapy in August 2017. The patient was prescribed Isoniazid (300 mg per day) and Rifabutin (150 mg per day). One branch of the fistula was still present in September 2017 and it leaked some caseous material when squeezed.

On November 2017, during the objective examination the skin was normochromic and any secretions were not highlighted as long as there was any swelling. X-ray examination did not show any difference from the previous findings.

The patient reported pain during deambulation and functional deficit on the tibio-tarsic joint in March 2018; this led to a new MRI examination which indicated, a decrease of the inflammatory process compared to the previous MRI, and severe aspects of

Figure 6. Imaging at 2 years follow-up evidencing the chronic state of the infection with relevant damage of the articulation, but without new evident outbreak.



a. MRI STIR; b. MRI T2.

erosion of the tarsus and meta-tarsal joints with elements of bone liquefaction ascribable to tubercular osteomyelitis.

A CT exam of the left foot was performed in May 2018 (Figure 4) and it showed aspects of bones remodeling, compatible with tubercular osteomyelitis. This was similar to the previous X-ray reports.

Meanwhile the patient was followed in the outpatient clinic. The inflammatory indices and white blood cell (WBC) were checked and these values were trending towards a steady low, in response to the antibiotic therapy.

During the rest of 2018 the patient was followed regularly by the Orthopaedic outpatient clinic and the Infectious Disease outpatient clinic. Antibiotics therapy and the medications of the foot were continued. In December 2018 we started to notice a progressive improvement in the local status and a complete closure of the precedent fistula. The infectious disease department decided to end the antibiotics therapy in February 2019 and declared the patient clinically stabilized.

Objective examination at two-year follow-up indicated that the foot was not swollen, not warm on palpation, and the skin was not stretched. There was a minimal residual pain during deambulation and a slightly limited range of motion (Figure 5). The MRI executed in January 2021 underlined the chronic state of the infection with relevant damage of articulation, but without new sign of active infection (Figure 6).

Discussion

The manifestation of extra-pulmonary tuberculosis of the bone is observed in only 1-3% of all tuberculosis patients and tends to occur in the major weight-bearing joints such as the hip and knee, due to a haematogenous dissemination from elsewhere in the body [3]. Tuberculosis infections of the foot and ankle are very rare and happens in only 1% of the cases. Therefore, diagnostic and therapeutic delays are typical due the uncommon site, lack of awareness, and its ability to mimic other disorders clinically and on radiographs [7]. Skeletal TB is even more rare without evident pulmonary involvements, as was observed in our patient [8]. There have been significant discussions about the role of trauma in the development of OT and these have focused on two perspectives. Firstly, trauma probably draws attention to a mild focus or it may activate a latent focus already present [9]. Secondly, in some cases a penetrating trauma can be related directly as the cause of OT. In fact, previous reviews of skeletal tuberculosis reported higher incidences of foot

involvement in people from endemic areas where it is not uncommon to walk around barefoot [3,10]. A recent review of the incidence of OT in western countries showed the increasing incidence in that area over the last decade, with the majority of the cases among local citizens [11]. The incidence of tuberculosis in our region Sardinia, Italy, is 1 in 100,000. Even if the incidence is low, physicians need to be aware of this disease and be prepared for atypical presentation, such as our case, especially when there are multiple failed antibiotic therapies [7].

Furthermore, since the disease is paucibacillary, a positive acid-fast bacilli culture is rare, thus delaying the diagnosis. Diagnosis is usually confirmed by obtaining granulomatous tissue on biopsy [8]. Positive histological yields can be obtained in 64–90% of all patients. Therefore, biopsy is essential in diagnosing tuberculosis [11]. Even in the absence of positive results, it is recommended to start empirical anti-tuberculosis treatment in order to limit the potentially permanent destruction of affected skeletal segments in patients with high clinical suspicion [11]. The importance of a prompt diagnosis is underlined by the fact that in the early stages, when the disease is limited to the bone, medical treatment leads to excellent healing and limited residual disabilities. On the other hand, spread from a periarticular bony focus, in the midfoot to the interconnected joints, leads to a stiff foot and serious residual deformities [8].

In conclusion the diagnosis of osteoarticular tuberculosis is challenging for every physician, especially when presenting without pulmonary involvement. Therefore, tuberculosis should always be considered in the differential diagnosis when diagnosing patients who present with nonspecific symptoms, and history of pain and swelling without systemic illness. This knowledge allows an early identification of the disease and a prompt therapy in order to avoid long-standing untreated infections which typically cause bone destruction and loss of function. The knowledge is also mandatory for western physicians who treat OT due to increasing international travel, immigration from less developed countries and increased use of immunosuppressive medications. Histological examination combined with the culture of the sample is crucial. We believe that this article can bring awareness around osteoarticular tuberculosis and help with improving outcome and eradication of the infection.

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Corresponding author

Marco Pes, MD
Resident, Orthopaedic Department, University of Sassari
Viale San Pietro 43, 07100, Sassari, Italy.
Tel: +39 079 208133
Email: marco92pes@gmail.com

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