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Case report

Osteoarticular tuberculous infection of the first tarsometatarsal joint - a case report

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ABSTRACT

Introduction: The prevalence of osteoarticular tuberculosis is increasing. Tuberculous infection in midfoot is rare. Establishing the diagnosis is difficult, leading to delay in management and leave many complications.
Case presentation: An 18-year-old man presents to our clinic with pain on his foot for the last 1 year. No other remarkable signs and symptoms. X-ray of his foot shown destruction of the 1st tarsometatarsal joint, later confirmed with MRI that shown synovitis and bone edema. Mantoux test and biopsy were done and established the diagnosis of tuberculous infection.
Clinical discussion: Intensive phase of anti-tuberculosis chemotherapy was given for 2 months, followed by continuation phase for 7 months. Surgical management of debridement and arthrodesis were performed as adjunctive treatment. At 10 months follow-up patient was pain free, fully weight-bearing and no signs of further destruction.
Conclusion: Osteoarticular tuberculosis is difficult to diagnose, a high index of suspicion is required to avoid delay treatment and complications. Anti-TB chemotherapy is still the treatment of choice, with surgical management is reserved for advance case.

1. Introduction

Tuberculosis (TB) is global major health problem and one the most concerned disease in the world [1]. People age between of 15 to 49 years old are affected the most, nearly three quarter of total cases [2]. The bacteria called *Mycobacterium Tuberculosis* causes this contagious disease. The transmission is believed to be airborne, and would happen if someone breathes the air containing thick saliva of the infected [2]. In the Western World, 1–2 % of all cases of tuberculosis accounts for musculoskeletal tuberculosis [3]. In young people with immunodeficiency, cases of TB infection in the bones and joints are increasing significantly and difficult to treat. The spine is most commonly affected, and involvement foot and ankle region are rare, even in the endemic countries [4]. Because the clinical and radiological findings are non-specific, the foot involvement is difficult to diagnose. This caused a delayed diagnosis and advanced disease with poor outcomes [5,6].

Amongst the endemic countries for TB infection, Indonesia is in the 9th of the highest numbers of TB cases, and ranks out of 20 countries with the highest estimated TB cases in HIV patients [7]. And in this paper, we will report the case of a tuberculous osteomyelitis of the first tarsometatarsal (TMT) joint of the right foot, without the incidence of immunodeficiency or malnourishment. To the best of our knowledge, this is the first paper from Indonesia reporting tuberculosis infection of the midfoot.

2. Case report

An 18-year-old man came to the outpatient clinic with a history of pain in the right foot for the past 1 year. The patient was an active and healthy student with no recent history of significant trauma, beside he likes to play football. The pain was dull and persistent, but he can still bear his daily activity. Aside from pain, there was swelling in the medial dorsal side of his right foot, he said it gets bigger after activity and gets smaller again during the night. No history of contacts with TB suspected patient and no respiratory symptoms, fevers, night sweats or weight loss, and no other past medical history. There is also no other specific past illness and medication from family history.

From physical examination we found tenderness over the base of the

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1st tarsometatarsal joint, with swelling of the dorsal lateral aspect of the right foot. Ankle movements within normal limit. There was warmth of the overlying skin, but no color change was seen. Other joints were also within normal range of motion, and systemic examination was normal, with no fever and unremarkable respiratory, cardiovascular and abdominal examinations. He had a normal CRP of 0.16, elevated ESR of 20, and negative HIV screening test.

Foot X-ray examination showed abnormal lucent lesion at the 1st tarsometatarsal joint of right foot, and destruction at the base of 1st metatarsal bone and also at the medial cuneiform bone. No sign of old fracture and other bones within normal findings (Fig. 1). Chest x-ray examination showed no abnormality or signs of TB infection. From MRI examination, prominent bone marrow edema was seen with destruction at the base of 1st metatarsal bone and also at the medial cuneiform bone. Degenerative changes were seen in the 1st tarsometatarsal joint and fluid collection at dorsal aspect of mid foot causing a prominent localized tenosynovitis around the anterior tibialis tendon. The general appearances were suggestive of either septic arthritis, osteomyelitis, or bony tumor (Fig. 2). The patient underwent Mantoux test at local clinic, and the result suggested TB infection. Tissue culture for pyogenic organisms was negative. Biopsy result was supported TB infection with Langhans giant cell and multiple granulomas. A diagnosis TB osteomyelitis was made and the four-drug anti-tubercular (anti-TB) chemotherapy was given (Rifampicin:R, Isoniazid:H, Pyrizinamide:Z, Ethambutol:E) by pediatrician, with doses adjusted to the patient's weight.

Due to persistent pain and marked degenerative changes, surgical option was discussed with the patient as a next step of therapy. A surgical management was then performed and conducted by first author in a tertiary referral hospital. A longitudinal approach between 1st and 2nd metatarsal was made, which facilitated the exposure of the 1st metatarsal and medial cuneiform bones. Intraoperatively, the caseous mass was found within the 1st tarsometatarsal joint. The lesion was completely debrided, leaving a significant gap in the joint. The gap was filled with autologous cancellous bone graft and fixed with 2.7-footft plate. Non-weightbearing activity was instructed for 6 weeks, with partial weight bearing for the next 2 weeks. At 8 weeks patient was allowed to fully weight bear. The wound is healed uneventfully and no complications were seen.

Intensive phase (HRZE) of anti-TB chemotherapy was continued for 2 months, followed by continuation phase (Isoniazid and Rifampicin) for 7 months. At 10 months follow-up, the patient was pain free, fully weight bearing on affected foot, and no deformity is seen. X-ray examination at 10 months showed no further bony destruction (Fig. 3). The work has been reported in line with the Updating consensus surgical case report (SCARE) 2020 criteria [8].

3. Discussion

Extrapulmonary tuberculosis is more often seen in patients with immunocompromised disease or malnourishment. In immunocompromised patients, Tuberculous osteomyelitis occur as a result of

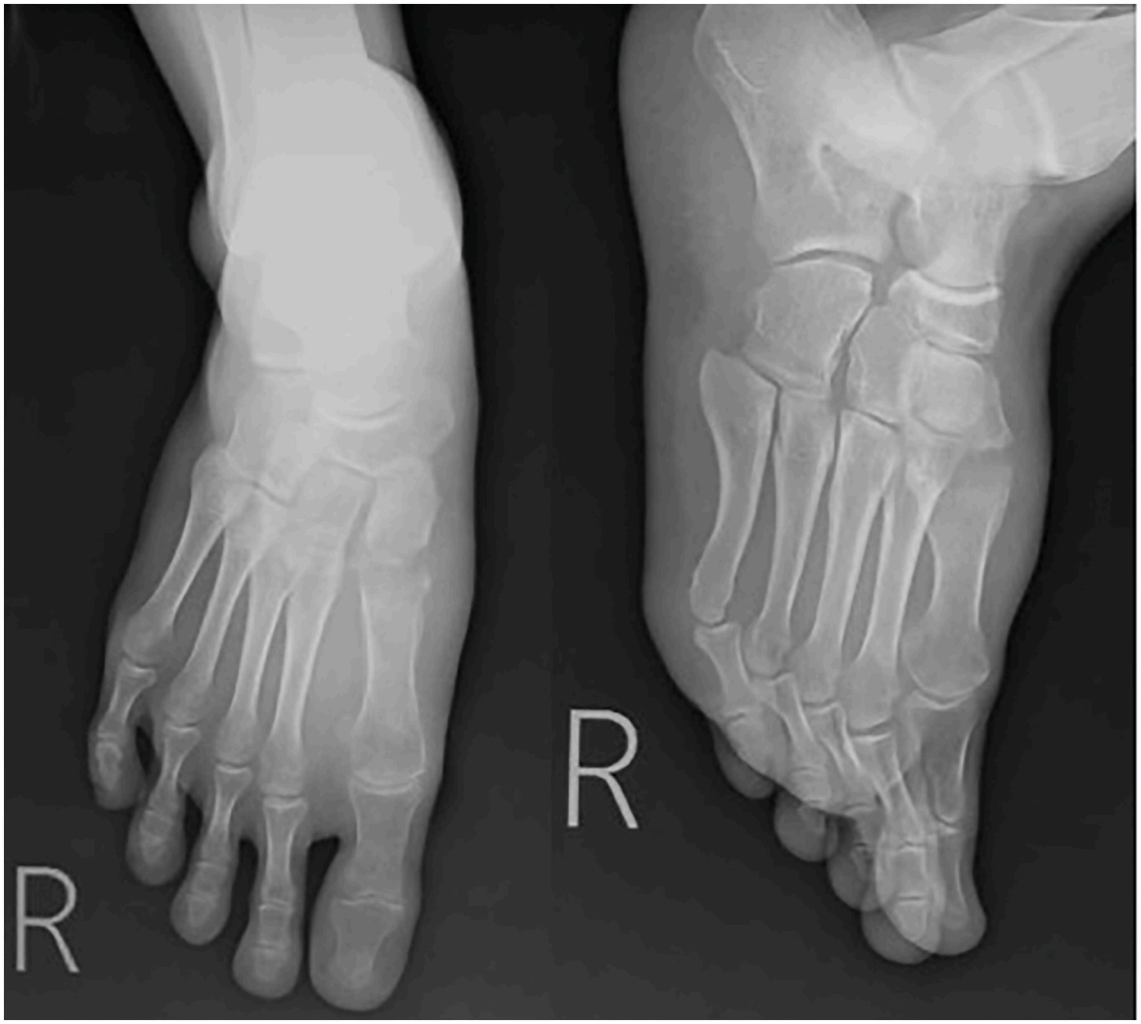

Fig. 1. Right foot x-ray taken on 1st admission to the clinic, destruction at the base of 1st metatarsal bone is noted.

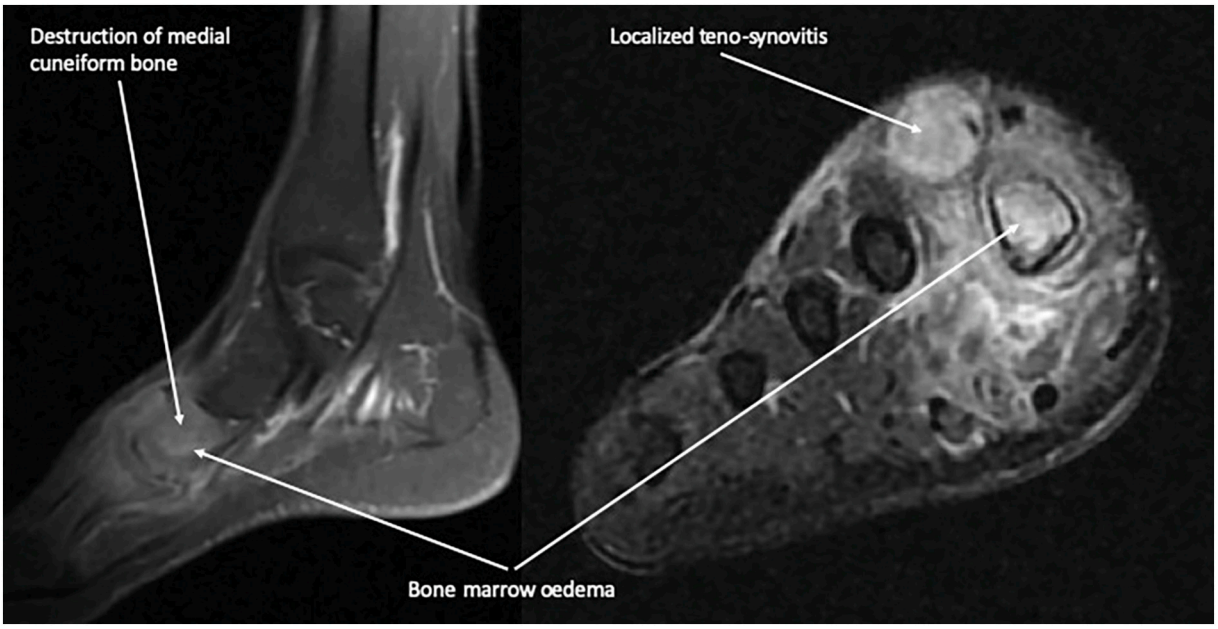

Fig. 2. Degenerative changes are seen in MRI of the 1st TMT joint. Joint destruction, bone marrow edema, and joint effusion extending to dorsal aspect of mid foot causing a prominent localized teno-synovitis are shown.

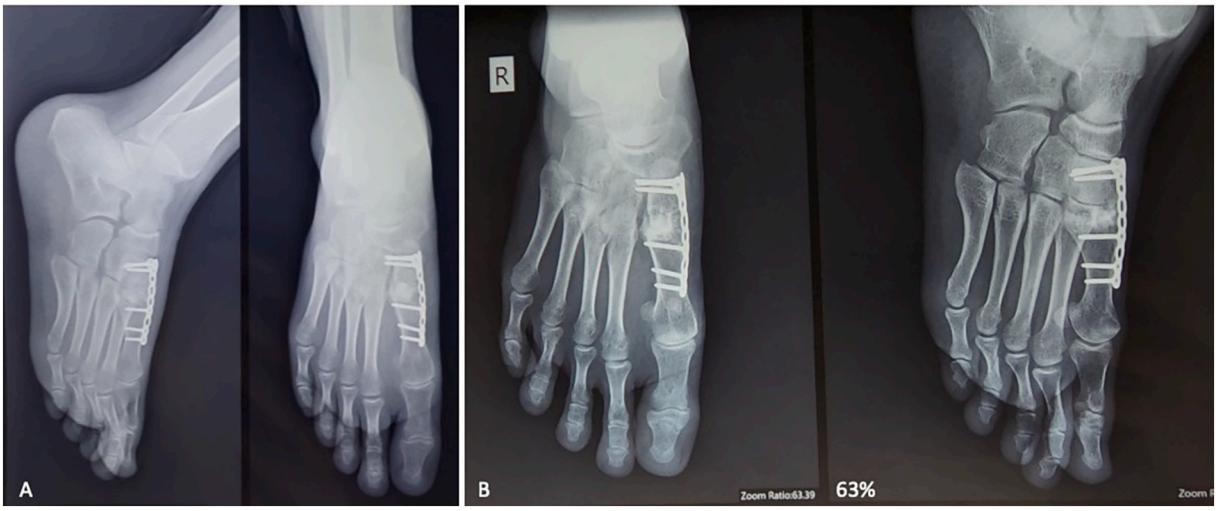

Fig. 3. Comparison between immediate post-op x-ray (A) and follow-up x-ray at 10 months shows no further bony destruction.

lymphatogenous or hematogenous spread from a primary focus of infection in the lung [9,10]. In our patient, neither immunocompromised disease nor malnourishment were detected. Tuberculous osteomyelitis has a low prevalence (1–3 %); a high index of suspicion is required to accurately diagnose the patient. Inadequate knowledge and experience of tuberculous osteomyelitis, and lack of consideration in the differential diagnoses, become the challenge to accurately diagnose this disease [11]. Even though the cause of TB infections in osteoarticular is still controversial, some theories stated that repetitive trauma is one of the cause, it encourages the local aggregation of macrophages that carry the bacilli that survived the primary infection [12]. The classic presentation of TB infection, such as localized pain, together with fever and weight loss, is rarely seen in tuberculous osteomyelitis. Blood tests may show a raised ESR. Pulmonary involvement is only about one third of patients with tuberculous osteomyelitis, making chest x-ray screening less useful [13,14].

Tuberculous osteomyelitis of the foot is uncommon and almost always a secondary infection, with primary focus elsewhere in the body. In most of the cases of a healthy patient, the primary lesion is not always identifiable, as seen in the current study. There are 4 basic types of infection, and in the foot, the infection occurs as one of the types [4,5]. The first and most common is as a granulomatous focus adjacent to a joint and will spread to involve the adjacent joint if not treated properly. This has disastrous effects in the midfoot area and the potential for widespread disease exists. Because of the interconnection of the joints, and also communicate with the Lisfranc and subtalar joint. The patient in current study is included in this type. The second type, which is more common in children, is that of a central granuloma. Phalanges and metatarsals are the most affected site with similar lesions in the hands and the contralateral foot, that is important to look for. The two other types are primary hematogenous synovitis, and tenosynovitis or bursal tuberculosis. Even though it is possible, synovitis is very rare, and tenosynovitis usually is a sequel to established infection in the neighboring joint or bone.

Osteoarticular tuberculosis often presents a diagnostic challenge. A wide range of non-specific signs and clinical manifestations, such as pain, limitation of the range of motion, stiffness, and edema, make the early diagnosis extremely difficult [1]. Skeletal TB may mimic other chronic and acute diseases like osteomyelitis, septic arthritis, or pigmented villonodular synovitis (PVNS) [15]. Same with previously reported cases, we found non-specific signs and clinical manifestations in our patient. In x-ray examination, midfoot joint space narrowing might be difficult to quantify due to overlapping of the bones. The combination of peripheral osseous erosions, juxtaarticular osteoporosis, and joint space narrowing, which is the characteristic of tuberculous arthritis, can be seen more significantly in other larger joints, like knee, hip, and ankle joint [16]. As seen in this patient, even though the infection is happened in the midfoot, these features are appeared in x-ray examination. This so called "Phemister's Triad" is the result of granulation tissue formation in an inflamed synovium. Demineralization happens because this pannus erodes cartilage and bone. Since *Mycobacterium Tuberculosis* does not produce proteolytic enzymes, and the joint space will be preserved for a considerable time [16,17]. Non-specific findings are also commonly found in MRI examination. These findings may be consistent with osteomyelitis, avascular necrosis (AVN), bony tumors, inflammation, or neuropathic joint diseases [6].

There is lack of literature regarding diagnostic tools in TB of small joints, including the midfoot joints. Many authors have reported that in any tubercular osteomyelitis, bone sclerosis or PVNS may be seen in the early course of disease [18–20]. Ruling out other conditions is required, i.e. pyogenic infection, sclerosing osteomyelitis, and hyperostosis. Diagnostic confirmation by culture, PCR, and biopsy is essential. Laboratory tests to detect inflammation are commonly negative. The Mantoux test and Ziehl-Neelsen stain of synovial fluid may still in controversy and sometimes provide false-negative and false-positive results [21]. When TB is suspected, patient's history, clinical manifestations and radiological findings are still of diagnostic importance. Definitive diagnostic confirmation can be provided by histopathologic evaluation. As in our case, a prior biopsy and histopathology evaluation showed Langhans giant cell and multiple granulomas that confirms osteoarticular TB infection.

Conservative treatment including anti-tubercular (anti-TB) chemotherapy and splintage is the treatment of choice and usually sufficient to treat tuberculous osteomyelitis in early stage, although surgery is often required in late presentation or advance destruction of the bone [22]. In the early stages of this disease, isolated osteomyelitis can be seen, and to prevent possible sequelae, early treatment is required [4,6]. As soon as the diagnosis is established, quadruple anti-tuberculous therapy should be given. Daily treatment of Rifampicin, isoniazid, pyrazinamide and ethambutol are given for the first 2 months, with Rifampicin and isoniazid are continued for a further 4 months. When patients present late with extensive destruction of the bone, surgical debridement and excision of primary focus are required. And if arthritis and deformity are proofed from the examination, arthrodesis is often required [17]. As seen in our case, extensive destruction of the bone and severe arthritis of the 1st TMT joint are already taken place, so surgical management such as debridement, synovectomy, and arthrodesis with bonegrafting were performed as adjunctive treatment to the anti-TB chemotherapy.

4. Conclusion

The TB infection in foot and ankle area is rare, often leading to a delay in diagnosis and management. Laboratory findings and radiological features are usually non-spesific, makes a high index of suspicion is required, especially for surgeon who works in endemic areas. To prevent debilitating complications in any suspicion of TB cases, a biopsy should be performed to point out the accurate diagnosis and initiate treatment as soon as possible. Conservative treatment with anti-TB chemotherapy is still the first line of treatment, with surgical management is reserved for late and advance presentation of the tuberculous osteomyelitis.

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Consent

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Credit authorship contribution statement

Andi Praja Wira Yudha Luthfi: Writing the article, Data collection, surgeon in charge, analysis and patient follow-up.

Sigit Wedhanto: Responsibility for the research activity planning and execution, including mentorship external to the core team.

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Guarantor

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Declaration of competing interest

None declared.

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