

## Case Report

# Navicular and cuneiform tuberculosis: a rare localization of tarsal tuberculosis

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**Abstract:** Navicular bone and cuneiform were infrequent locations of tarsal tuberculosis. The present study reported an unusual case of navicular and cuneiform tuberculosis who was successfully treated with Streptomycin-loaded bone cement. A 71-year-old Chinese male patient was admitted to our hospital with complaints of swelling and pain at the dorsal aspect of the right foot for 6 months. The patient presented obvious swelling and distension, skin redness and fever as well as an ellipse skin lesion (1.5×1.5 cm) at the dorsal aspect of the right foot. Radiological examination displayed a reduction in right tarsal bone density, trabecular thinning and indistinct joint space but no sequestrum; right lower extremity vascular ultrasound showed atherosclerosis of the lower extremity and a formation of plaque at the right femoral artery; and magnetic resonance imaging (MRI) revealed edematous marrow in the right navicular bone, medial, middle and lateral cuneiform bones. Swear and culture of blood and wound secretions were normal. Debridement of focal lesion and Streptomycin-loaded bone cement implantation were performed. Postoperatively acid-fast staining of lesion sample revealed *acid-fast bacilli*. The patient received two weeks of anti-tuberculous chemotherapy and obtained a good clinical outcome. In conclusion, the diagnosis of tarsal tuberculosis should be strongly considered the presence of acid-fast bacillus in the lesion smear or culture and a suggestive MRI image. Streptomycin-loaded bone cement implantation following the surgical debridement of focal lesion was an effective treatment for tarsal tuberculosis.

**Keywords:** Tarsal tuberculosis, navicular, cuneiform, *tubercle bacillus*, streptomycin-loaded bone cement

### Introduction

Tuberculosis caused by *Mycobacterium* infection still remains a major health problem in both developed and developing countries [1, 2]. It was responsible for 9.6 million new cases and 1.5 million deaths in 2014 [3]. Pulmonary tuberculosis is the most common presentation for this disease, whereas bone tuberculosis represents less than 3% of all tuberculosis [4, 5]. Besides, foot tuberculosis is quite rare and accounts for only 5-10% of osteoarticular tuberculosis patients [6]. Furthermore, the infrequent occurrence and the nonspecific signs and symptoms usually lead to a delayed diagnosis of foot tuberculosis [5]. Herein, the present study reported a case of an elder male patient with navicular and cuneiform tuberculosis which were rare locations of tarsal tuberculosis.

### Case presentation

A 71-year-old Chinese male patient was admitted to our hospital on September 15th, 2015

with 6 months history of swelling and pain at the dorsal aspect of the right foot. The patient initially presented red swelling and warmth at the dorsal aspect of the right foot without evident inducements, accompanied by walking pain; however, no mass or skin ulceration was reported. After fluid therapy with antibiotics such as Penicillins and Cephalosporins, the red swelling and fever at the dorsal aspect of the right foot became progressively lessened but thereafter these symptoms were on and off and recurrently attacked. The patient received anti-infective therapy in various local hospitals, but the symptoms didn't improve completely. In addition, the red swelling, pain and fever at the dorsal aspect of the right foot were worse two months ago, and a local liquid mass (1.5×1.5 cm) was witnessed. Then the incision and drainage were performed and draw out about 5 mL of red purulent secretions, with the wound opened and unstitched. Within these days, the patient had recurrent hyperpyrexia (body temperature of 38.5°C-41.0°C) and mild weight

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**Figure 1.** Clinical examination showed an ellipse but not deep skin lesion (1.5×1.5 cm) at the dorsal aspect of the right foot, with a small quantity of necrotic tissues at the base and a little turbid exudate.

loss but normal appetite and defecation, and he had a clear consciousness and remained in good spirits.

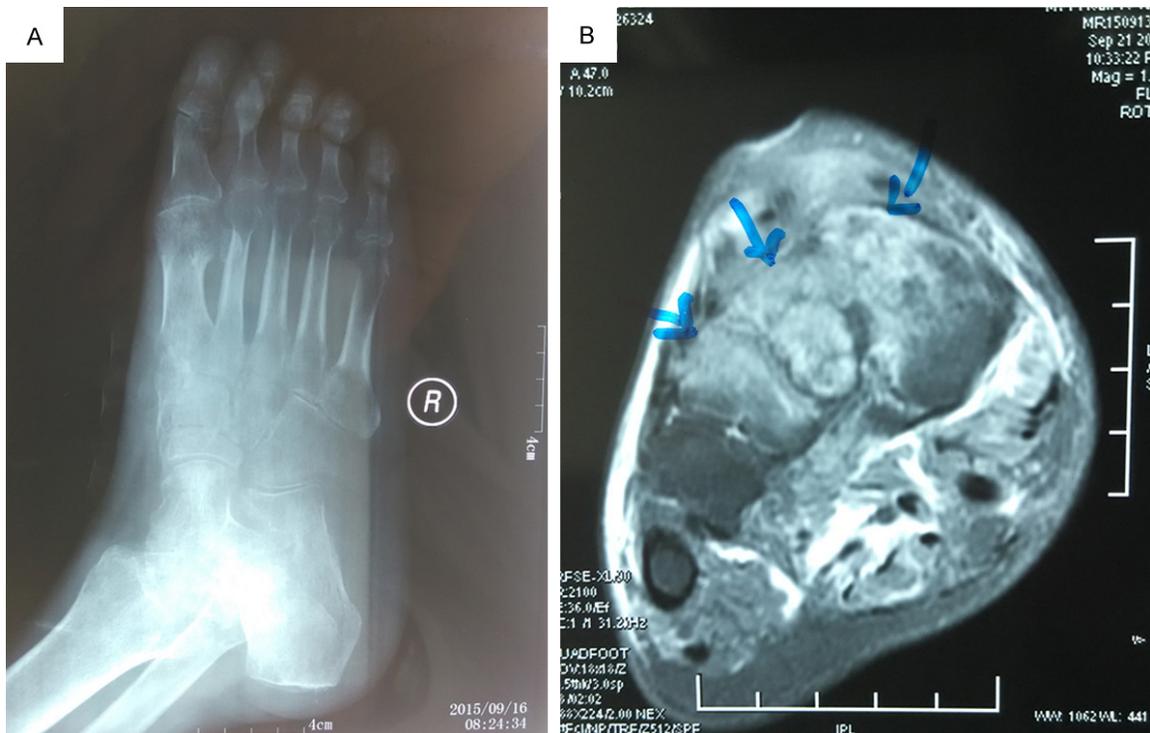
On admission, the patient was found to have obvious swelling and distension, skin redness and fever as well as an ellipse skin lesion (1.5×1.5 cm) at the dorsal aspect of the right foot. The lesion was not deep, with a little turbid exudate but no obviously abnormal flavor (**Figure 1**), and there were a small quantity of necrotic tissues at the base of the lesion. All toes of the right foot had good activities, and cutaneous sensation and blood circulation were both normal at the distal limbs. The patient was diagnosed with infection at the dorsal aspect of the right foot, and erysipelas and diabetic foot were ruled out.

Laboratory examinations revealed the following levels: erythrocyte sedimentation rate (ESR) of 36 mm/h, C-reactive protein (CRP) of 109.55 mg/L, red blood cell (RBC) of  $3.15 \times 10^{12}/L$ , white blood cells (WBC) of  $3.9 \times 10^9/L$  (82.2% neutrophils, 11.7% lymphocytes and 6.1% monocytes), platelets (PLT) of  $169 \times 10^9/L$  and hemoglobin (HGB) of 100 g/L. In addition, blood and wound secretion samples were collected and cultured, but no *acid-fast bacillus*, *tubercle bacillus* or other bacteria were detected. Computerized tomography (CT) of the chest demonstrated multiple lesser tubercle and cavity in the two lungs, signs of chronic bronchitis and fungal infection, a small quantity of pleural effusion and thickening and adhesion of pleura on the right. Preoperative bronchoscopy demonstrated chronic inflammatory changes in tracheal and bronchial mucous

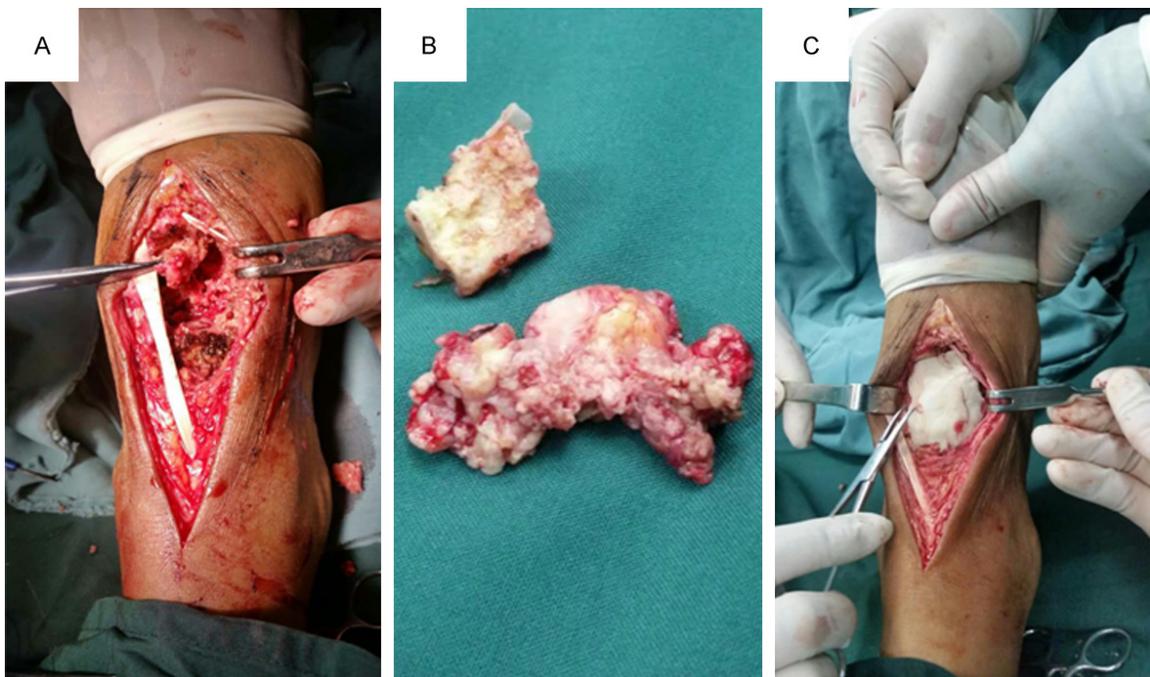
membranes, and abundant lymphocyte infiltration was observed by the biopsy of orificial mucous membrane at the basal segment of inferior lobe of right lung. The X-radiological examination of the right foot displayed a reduction in the tarsal bone density, trabecular thinning and indistinct joint space but with no sequestrum (**Figure 2A**); vascular ultrasound of the right lower extremity showed atherosclerosis of the right lower extremity and a formation of plaque at the right femoral artery; and magnetic resonance imaging (MRI) of the right foot revealed edematous marrow in the navicular bone, medial, middle and lateral cuneiform bones (**Figure 2B**), and sheeted long T1 and T2 signal shadow suggesting inflammatory changes.

On September 25th, the surgical debridement of the focal lesion was performed and antibiotic (Streptomycin)-loaded bone cement (1 g Streptomycin in 40 g bone cement) were implanted at the dorsal aspect of the right foot (**Figure 3A-C**) as described by Masri and his colleague [7]. During the modeling and implantation of the bone cement, the maintenance of medial longitudinal arch should be prudently considered. The lesion sample was also collected by the surgical biopsy. Postoperatively following ultrasonic washing and centrifugation, lesion sample was used to make smear and culture, and *acid-fast bacillus* was observed by microscopy (**Figure 4A**); however, no bacillus was observed on common smear (**Figure 4B**) or any cultures. Postoperative histological examination showed epithelioid granulomatous lesions and coagulation necrosis, as well as visible Langerhans cells, compatible with tuberculosis. Acid fast stain examination indicated a positive result of tubercle bacillus infection at the talus. After the surgery, ESR and CRP decreased to 26 mm/h and 43.28 mg/L, respectively, and routine blood tests revealed RBC of  $3.84 \times 10^{12}/L$ , WBC of  $4.2 \times 10^9/L$  (59.80% neutrophils, 23.60% lymphocytes and 16.6% monocytes), PLT of  $211 \times 10^9/L$  and HGB of 115 g/L. Then the patient was given general nutritional support, common antibiotics for infection prevention and quadruple antituberculosis drugs (streptomycin, isoniazid, pyrazinamide and ethambutol). Two weeks later, he was discharge from hospital after suture removal, with good wound healing.

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**Figure 2.** The X-radiograph of right foot displayed reduced tarsal bone density, trabecular thinning and indistinct joint space (A); magnetic resonance imaging (MRI) scan revealed edematous marrow in the navicular bone, medial, middle and lateral cuneiform bones (arrow, B).

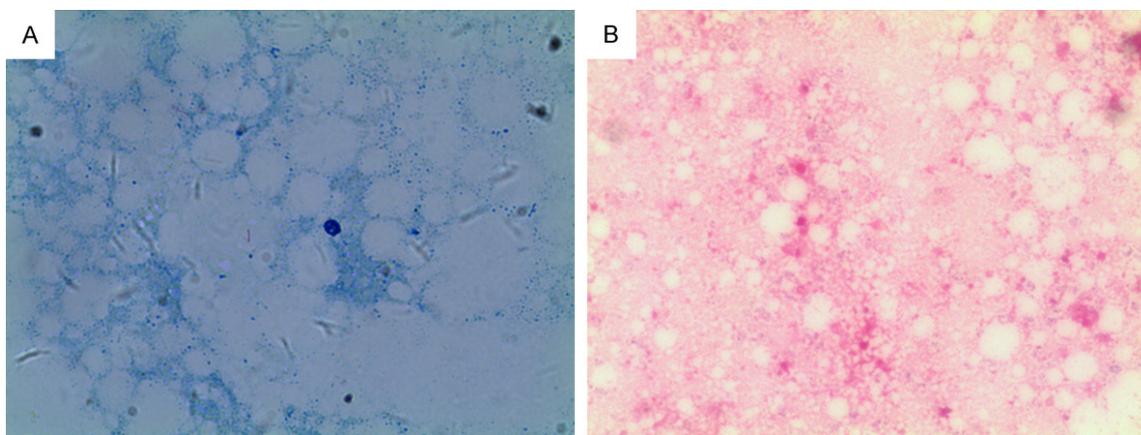


**Figure 3.** The procedure of debridement (A, B) and antibiotic-loaded bone cement implantation (C) performed at the dorsal aspect of the right foot.

The clinical characteristics and treatments of tuberculosis at the talonavicular joint published

between 2003 and 2015 are summarized in **Table 1**. The demographics of the cases in the

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**Figure 4.** Postoperatively the acid-fast staining smear of the lesion sample demonstrated *acid-fast bacillus* by microscopy (A) but no bacillus was observed on common smear (B).

literature review consisted of 4 male and 3 female patients, aged 7 to 74 years. The common symptoms were swelling (6 of 7), pain (5 of 7) and restricted range of motion (4 of 7). Most patients had no abnormalities in pulmonary tuberculosis (6/7). Pulmonary tuberculosis was seldom presented in cases with tuberculosis of the talonavicular joint (1 of 7). Among all of these reports included here, histological examination showed six cases with granulomata or accompanied with caseous necrosis in soft tissues. But the presence of *Mycobacterium tuberculosis* was very rare in the cultures of bone biopsy (1 of 7). All the patients improved well after at least 6 months of treatments, and no antibiotics-loaded bone cement was used in these cases.

### Discussion

Foot tuberculosis is quite a rare kind of osteoarticular tuberculosis, with a prevalence rate of 5-10% [6]. Tarsal joints and calcaneal bone are the most two common lesion sites, but navicular and cuneiform tuberculosis are seldom seen [1]. Unlike spinal tuberculosis and major joint tuberculosis (hip and knee) [8], there may be delays in the diagnosis and therapy of tuberculosis at these small bones, more detected in advanced stages [9]. The main reason is that the symptoms of foot tuberculosis are nonspecific and often insidious. Besides, bone tuberculosis has a long treatment period lasting for 6 to 18 months [10]; hence, navicular and cuneiform tuberculosis are not easily cured and have a recurrence rate up to 26.4% [11]. In this study, we reported an elder female patient

with navicular and cuneiform tuberculosis who was successfully treated with Streptomycin-loaded bone cement and quadruple antituberculosis drugs in spite of the delayed diagnosis and treatment.

The review of several reports on tuberculosis of the talonavicular joint published between 2003 and 2015 revealed that the most common symptoms were swelling and pain, which were often accompanied with fever and diaphoresis [1]. The patient in this study complained of a walking tarsal pain, swelling and abscess for several months, but he did not present some tubercular toxic symptoms, mainly characterized by an irregular low-grade fever (body temperature between 37.4°C and 38°C) postmeridiam lasting for several weeks, poor appetite and night-sweat [12]. This might be the reason for the diagnostic errors, with progression to bones destruction and deformities, as the patient had recurrent hyperpyrexia (38.5°C-41.0°C) but normal appetite and defecation, and simple anti-infective therapy were of low usefulness [13, 14]. Besides, although body temperature was suspected by X-ray and chest CT and further supported by MRI scan, sequestrum was not observed. The abnormal changes in the chest by CT were also contrary to most cases in the review of several reports that patients had normal X-ray scanning [1, 5, 15-18]. Consequently, tarsal tuberculosis was not always associated with pulmonary tuberculosis.

Histopathologic and microbiologic examinations are of great value because they allow for

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**Table 1.** Summary of cases with tuberculosis of the talonavicular joint illustrating symptoms diagnosis and final diagnosis method

Study	Year	Gender	Age	Affected side	Symptoms	Final diagnosis method	Chest X-ray	Bone cement	Outcome (treatment period)
[19]	2004	Male	29	Left	Valgus deformity of the hindfoot, moderate swelling at hindfoot and ankle, decreased ROM of the ankle and subtalar joints	Culture of <i>Mycobacterium tuberculosis</i> , tissue histology showing granulomata with caseous necrosis	A small right pleural effusion and a bulky right hilum	No	Improving mobility (6 months)
[15]	2005	Male	7	Left	Warmth, tenderness, redness and limited painful ROM	Histological examination of biopsy sample showing granulomata and caseous necrosis	Normal	No	No pain, no significant limp, a good ROM (6 months)
[16]	2009	Female	11	Right	A 2-3 cm ulcerative lesion, localized warmth and slight swelling	Histological examination of biopsies showing chronic caseating granulomatosis	Normal	No	Pain-free, no limp on walking (9 months)
[5]	2014	Male	14	Left	Swelling and pain, positive history of loss of appetite	Histological examination of the biopsy specimen showing granuloma and central caseating necrosis, acid fast stain and PCR examination positive for <i>Koch's bacillus</i>	Normal	No	No pain while walking, able to perform daily activities without restrictions (20 months)
[17]	2014	Male	45	Right	Fluctuant swelling, slight localized tenderness on deep palpation, restricted ROM, slightly painful	Ziehl-Neelsen (Z-N) staining of the fluid confirming the presence of <i>acid fast bacilli</i> (AFB)	Normal	Bone grafting	Full and painless motion at the ankle and subtalar joint (20 months)
[18]	2015	Female	74	Right	Swelling, restricted subtalar motion	Soft-tissue biopsy showing a focus of granulomatous inflammation within the soft tissue, bone biopsy of the talonavicular joint revealing <i>Mycobacterium tuberculosis</i> infection	Normal	No	Normal gait with no residual foot pain or swelling (18 months)
[1]	2015	Female	68	Right	Swelling, firm and painful to touch	Histopathological examination revealing an epithelioid and giant cell granulomatous inflammatory process with caseous necrosis	Normal	No	Decreased pain and swelling of foot and improvement in gait (14 months)

ROM, range of motion.

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the isolation and identification of microbes and the study of the sensitivity to antimicrobials [6]. Preoperatively microbiological cultures of the blood and wound secretions were negative for *acid-fast bacillus*, *tubercle bacillus* or other bacteria. Postoperatively, with regard to the lesion samples which were collected by surgical biopsy, ultrasonic washing and centrifugation were first performed then followed by high speed centrifugation, and finally the concentrated solution were obtained to increase the detection rate. Histological examination showing granulomata and coagulation necrosis was consistent with previous cases [1, 5, 15-19]. As expected, *acid-fast bacillus* was detected by smear under microscopy, in line with previous study [20] that reported tubercle was absent in many samples as *tubercle bacillus* located at the periphery of bone lesion. Despite tarsal tuberculosis symptoms similar to arthritis or osteomyelitis [21, 22], [the absence of bacteria on common smear or any cultures also confirmed no evidence of osteomyelitis.

Antibiotic therapy remains the most treatment for bone tuberculosis [23], and surgical treatments such as arthrodesis [24], resection arthroplasty [25], or prosthetic joint replacement [26] are also required in serious cases. However, high concentrations or long term of systemic antibiotic administration can be associated with toxicity [27] or potential drug-resistant tuberculosis and often achieve inadequate concentrations at the site of infection [10]. Antibiotic loaded bone cement has been commonly used for antibiotic delivery during total joint arthroplasty for the prevention or treatment of periprosthetic joint infection [28]. There were also several studies have reported treating serious cases of musculoskeletal tuberculosis with antibiotic-loaded bone cement, not only providing high local drug concentrations to control the infection but also sterilizing the joint [7, 29, 30]. In this present report, therapy was based on the surgical debridement of focal lesion and Streptomycin-loaded bone cement implantation, which has been described in a previous report that Streptomycin-loaded bone-cement (7 g streptomycin in 40 g bone cement) beads were used in the treatment of tuberculous bursitis and osteomyelitis of the greater trochanter in a 76-year-

old woman [7]. The antibiotics-loaded bone cement was implanted to fill up a large bone defect after the debridement of focal lesion and to achieve local effective antimicrobial concentration and the stabilization of the joint as well as consequent early recovery of weight-bearing. Because there were several joint surfaces between the tarsal bones (such as navicular and cuneiform bone) with the surrounding talus and proximal metatarsal, it was difficult to obtain a suitable bone covered with extensiveness articular cartilage from the total body for bone transplantation. The mismatching articular surface would cause the pain, unwell weight-bearing or osteoarthritis, whereas the adaptability in joint matching would be developed during the drying and hardening of the bone cement. Besides, the application of antibiotics-loaded bone cement avoided poor resistance infection, bone graft reinfection, bone resorption and the pain, high surgery cost and prolonged recovery caused by bone transplantation. Furthermore, during the modeling and implantation of the bone cement, we emphasized the maintenance of medial longitudinal arch and the recovery of the anatomic foot contours, which would conduce to postoperative walking and the recovery of weight-bearing function. Finally, the patient had good wound healing with two weeks of quadruple antituberculous chemotherapy.

There are a few learning points from this case. The diagnosis of tarsal tuberculosis should be strongly considered the presence of *acid-fast bacillus* in the lesion smear or culture and a suspected MRI image. Tarsal tuberculosis was not usually accompanied with pulmonary tuberculosis. Differential diagnosis of tarsal tuberculosis with other musculoskeletal disorders such as chronic osteomyelitis and metastasis, rheumatoid arthritis and septic arthritis should be considered [31]. Streptomycin-loaded bone cement implantation following surgical debridement of focal lesion might be an effective treatment for tarsal tuberculosis and should be started immediately to improve tissue healing and functional rehabilitation. However, the case study nature of the present report and the short-term evaluation of curative effect may be considered the limitations of the present report, and further tracking of more cases is needed.

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### Disclosure of conflict of interest

None.

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