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Saudi Orthopedic Association

Journal of Musculoskeletal Surgery and Research

Case Report

Calcaneum tuberculosis – A rare entity: Case report and review of literature

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Received: 21 December 2022
Accepted: 02 March 2023
EPub Ahead of Print: 17 March 2023
Published: 06 May 2023

DOI
10.25259/JMSR_158_2022

Quick Response Code:

ABSTRACT

Calcaneum tuberculosis (TB) is a rare entity, though it is the commonly involved tarsal bone. It does not present with the typical features of TB and can be confused with other pathologies affecting the heel. Very few case reports and case series studies are present to date for describing this rare entity. We report a 23-year-old female who presented to the orthopedic outpatient clinic with pain and swelling around the left heel and posterior aspect of the ankle for 3 months. Her radiograph showed a lytic lesion in the posterosuperior aspect of the calcaneum. Therefore, magnetic resonance imaging was done, which showed a lesion around the posterior-superior aspect of the calcaneum with the involvement of retrocalcaneal bursa. An incisional biopsy was performed, followed by *en bloc* excision of the lesion. The histopathological examination report suggested granuloma formation with caseous necrosis and the cartridge-based nucleic acid amplification test was also positive for mycobacterial TB infection. Proper clinical history, radiological examination, and biopsy are of utmost importance in these rare entities as they help in making the definitive diagnosis. Anti-Koch treatment is an effective treatment for TB, and the patient improved within 2 months of treatment both clinically (pain, swelling, and activity of daily routine) and radiologically (for bone formation and no recurrence). This report aimed to present a lytic lesion of the calcaneum that turned out to be TB and its management. The follow-up was 18 months post-surgery and showed no signs and symptoms of recurrence.

Keywords: Anti-Koch treatment, Calcaneum, Lytic lesion, Retro-calcaneal bursa, Tuberculosis

INTRODUCTION

In the era of modern medicine, infections like tuberculosis (TB) remain a major concern in developing countries like India. TB can be seen in the skeletal system, tissues, and visceral organs. The prevalence rate of TB in the Indian population is 193/100,000.[1] TB involving the skeletal system accounts for only 2% of all cases affected by TB,[2] whereas the involvement of foot and ankle joints accounts for approximately 10% of the skeletal involvement.[3] Of all the foot bones, the calcaneum is the most commonly involved, followed by the talus, distal end of the first metatarsal, cuneiform, navicular, and cuboid bone involved in TB of osteoarticular origin.[4-6]

Calcaneum TB is a rare entity. It does not present with typical features of TB and can be confused with other pathologies affecting the heel. A rare occurrence of TB of the foot and lack of

How to cite this article: Choudhary P, Saxena M, Chouhan J, Bharwani N, Bhati M. Calcaneum tuberculosis – A rare entity: Case report and review of literature. J Musculoskelet Surg Res, 2023;7:135-41.

awareness among orthopedic surgeons lead to difficulty and delay in diagnosis, which can further lead to patient disability in terms of function and poor clinical outcomes.[7]

Very few studies, including case reports and case studies, describe the presentation of foot and ankle TB and its management are present. We report a case with an unusual presentation of calcaneal TB without any other bony involvement. We also performed a literature review to study the involvement of calcaneum in foot and ankle TB, as well as the different treatment modalities.

In most cases of foot and ankle TB, medical management has a key role, whereas surgery is required in cases not responding to anti-Koch treatment (AKT) for tissue collection, deformity correction, pain relief, and reconstruction of the joint.[8]

CASE REPORT

A 23-year-old female presented to the orthopedic clinic complaining of pain and swelling around the left heel and posterior aspect of the ankle for 3 months. Her examination showed minimal swelling around the posterior aspect of the ankle with a normal overlying skin and tenderness on deep pressure. There were no local (hotness and sinus formation) or systemic features (weight loss and loss of appetite) of TB and no lymphadenopathy of inguinal lymph nodes.

Hematological parameters showed lymphocytosis and raised erythrocyte sedimentation rate. A radiograph of the calcaneum showed a lytic lesion in the posterosuperior aspect of the calcaneum [Figure 1]. Magnetic resonance imaging of the left foot [Figure 2] was advised, which showed the presence of a lesion around the posterior-superior aspect of the calcaneum along with the involvement of retro-calcaneal bursa with the size of the lesion around 21 × 37 × 42 mm and altered signal intensity on both T1- and T2-weighted images suggestive of mainly infective pathology. Proper consent before the biopsy was taken from the patient and her relatives. In the present case, an incisional biopsy was performed, followed by *en bloc* excision.

Then, histopathological examination (HPE) report suggested granuloma formation with caseous necrosis and cartridge-based nucleic acid amplification test (CBNAAT) was also positive for mycobacterial TB infection.

The calcaneal TB in this patient belonged to "Stage-2" as per the Classification of Foot and Ankle TB by Martini *et al*.[9]

Stage Feature
1. No bony changes and localized osteoporosis
2. Cavity formation or bony destruction within the bone
3. Complete bony involvement without major joint involvement
4. Massive destruction

After that, an excisional biopsy was performed for the lesion affecting the calcaneum using a posterior-lateral approach around the ankle. Intraoperatively, lysis around the posterosuperior aspect of the calcaneum was seen, along with the involvement of retro-calcaneal bursa. Then, curettage of the calcaneum was done along with the retrocalcaneal bursa, and the wound was closed in layers. Next, a left below-the-knee plaster was applied for 4 weeks followed by partial weight-bearing after 4 weeks and then complete weight-bearing from the 8th week. The sample was sent for HPE, CBNAAT, and Ziehl-Nelson (ZN) staining. Superficial infection was seen postoperatively, which was managed with regular dressing under antibiotic (Tablet. Cefuroxime, 500 mg, bd and linezolid, 600 mg, bd orally for 2 weeks) cover. HPE showed caseating tubercular granuloma [Figure 3], the ZN stain showed the presence of tubercular bacilli and CBNAAT was positive.

Figure 1: Lateral radiograph of ankle suggestive of lesion around posterior-superior aspect of calcaneum.

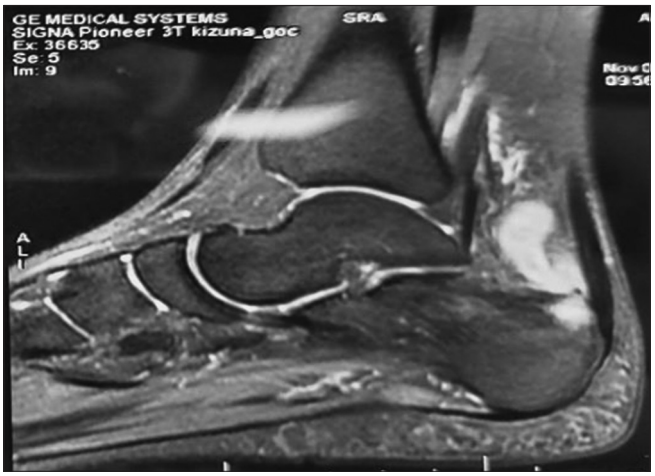

Figure 2: Sagittal section of magnetic resonance imaging ankle suggestive of bony and soft tissue involeument.

After these results, AKT was started (For the first 2 months, isoniazid [5 mg/kg], rifampicin [10 mg/kg], pyrazinamide [15 mg/kg], and ethambutol [25 mg/kg], for the next 10 months only isoniazid and rifampicin). The patient was called for regular follow-up, which consisted of radiological evaluation [Figure 4] and hematological investigations. Weight-bearing was allowed after 6 weeks. After 18 months of follow-up, it showed no signs of recurrence. Improvement in terms of clinical (pain, swelling, and activity of daily routine) and radiologically (for bone formation and no recurrence) was seen.

DISCUSSION

In developing nations like India, infections like TB continue to be a major concern in the public health sector, leading to morbidity and mortality. Lungs are considered the most common organ affected by TB, but it also involves extra-pulmonary sites. Spine involvement is commonly seen when

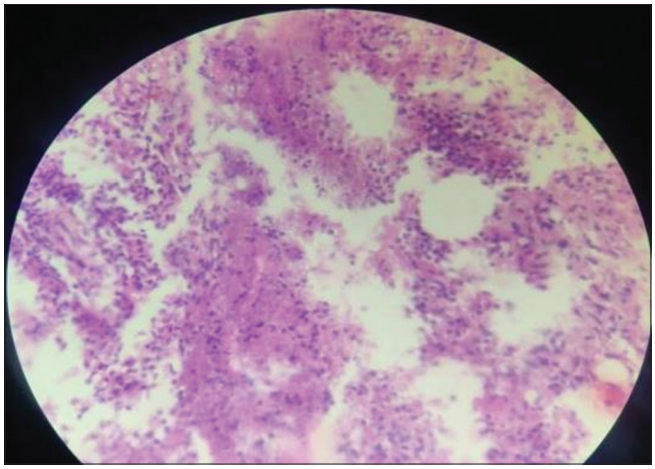

Figure 3: Histopathological examination showing caseating tubercular granuloma.

Figure 4: Lateral radiograph of ankle at 12th follow-up.

TB affects the skeletal system, whereas foot and ankle joint involvement is rare.[10] Prompt diagnosis of infections like TB affecting bone and joints is of utmost importance as it leads to massive destruction, which leads to a disability affecting daily routine activities.

TB affecting the skeletal system is a paucibacillary type and rare presentation around the foot leads to delayed diagnosis and treatment.[11] The disease usually starts from the cancellous part of the bone through direct inoculation or a hematogenous route.[12] The order of involvement in skeletal TB is the dorsal spine, pelvic bones, femur and hip joint, tibia and knee, and ribs, in that order. The elbow, ankle, and foot constitute 2% each and multiple-site involvement is merely 3%.[13,14]

Of all the bones of the foot, the calcaneum is the most commonly affected by TB, as seen in the literature, also supported by the study conducted by Kumar *et al*., which showed that out of 312 cases of foot TB, 88 (28.2%) cases had calcaneum involvement.[4] A possible explanation is the larger bone size and the fact that it is the commonly injured foot bone by trauma.[7,15] Tubercular infections of the foot can present with pain, swelling, discharging sinus, difficulty in weight bearing, heel-up signs, and symptoms like evening rise of temperature, night sweats, and weight and appetite loss.[16]

The Google Scholar and PubMed data were searched for TB around the foot and ankle by two authors (NB and MS) with keywords – "Tuberculosis," "Calcaneum," "Foot," and "ankle." We included the search results for the past 10 years (2013–2022) and included all type of studies. Out of the results searched, we found ten relevant studies with the involvement of calcaneum.

The literature review of the past 10 years shows that Male: Female is 3:1.[4,12,17-24] The disease duration varied from 1 to 24 months. The side involvement does not show much difference in involvement with Left: Right; 1.27:1. All the studies tabulated above show the involvement of the calcaneal body mainly and in few cases, show the involvement of the anterior process (three patients) and[12] tuberosity (seven patients).[19,20,22] The most common symptoms noted were pain with weight bearing and swelling.

In the present case, only painful swelling was seen around the foot and ankle, along with heel-up signs on prolonged walking.[25] Intraoperatively, sequestrum was found in the present case and similar findings have been reported in a study on foot TB by Dhillon and Nagi.[11] In contrast, a study by Mittal *et al*.[15] showed no sequestrum in calcaneum TB. Calcaneum TB can be confused with heel pathologies such as pyogenic and fungal infections, Haglund's deformity (posterosuperior tuberosity prominence as a result of overuse or footwear), and lytic lesions of bone.[17]

For differentiating TB from pyogenic infections affecting the foot and ankle, as a result of the absence of proteolytic

Study	Patients' Demographics	Chief Complaints	Treatment	Follow-up
Gillot and Ray[17] (2013)	66-year-male	Presented with complaint of pain and swelling around the right ankle for 2 years with symptomatic relief from steroid injection Local examination-Fluctuant swelling around the ankle (cold abscess, discharging sinus) with normal range of motion Radiograph-Suggestive of calcaneal involvement MRI-T1 and T2 wt. images suggestive of calcaneum body destruction Biopsy – suggestive of granulomatous TB	AKT for 18 months (6+12 months) and immobilized with an air cast boot	18 months, on final follow-up radiograph suggestive of new bone formation on calcaneum
Hayat *et al*.[18] (2014)	66-year-male	Presented with complaints of pain and swelling around left heel for 10 months with no relief from NSAIDs Local examination-Swelling, redness and hypersensitivity around the posterior aspect of calcaneum with no discharging sinus Radiograph – posterior aspect of calcaneum showing diffuse involvement (3.6×2.6 cm) with 2×1 cm patchy lucency in the posterosuperior corner CT scan shows destruction of the posterosuperior aspect of the calcaneum Biopsy – shows granulomatous inflammation suggestive of TB	Surgery (Debridement +curettage and cementing) and AKT for 9 months	4 months, no recurrence.
Chater *et al*.[19] (2014)	7-year-male	Presented with complaints of pain around the right foot for 6 months Local examination – swelling around right foot and lateral aspect of the ankle Radiograph – shows a lytic lesion of the calcaneum Biopsy – shows caseous necrosis with granuloma of epithelioid type	AKT for 9 months	18 months, no recurrence
Agarwal and Jain[12] (2015)	Ten patients had (one calcaneal body, three anterior process and body, three diffuse, and three tuberosity involvement), with seven males and three females, six left side and three right side, and one with bilateral involvement. (Mean age – 9.8 years)	Presented with complaints of pain and swelling around heel and lateral malleolus with the duration of symptoms from 1 month to 12 months Radiograph – seven patients had a single lytic lesion in the calcaneum, three patients had multiple lytic lesions.	AKT for 12 months, foot orthosis for 6 weeks, partial weight-bearing in the next 6 weeks, and full weight-bearing in the 10th week	Mean follow-up 17 months. Re-mineralization of bone was seen in all patients after 3 months
Tiwari *et al*.[20] (2017)	Nine patients (five calcaneal body, two anterior process, and two tuberosity involvement), seven males and two females, with five left and four right side involvement. Mean age – 21.33 years	Presented with complaints of pain, swelling and discharging sinus with a duration of symptoms ranging from 1 month to 14 months The diagnosis was made based on CBNAAT (1), ZN-staining (2) and Biopsy (6)	AKT for 12–18 months is given in all five patients. Below knee cast was given for 6 weeks, followed by partial weight bearing after 6 weeks and then after full weight bearing as per tolerance	The mean follow-up was 26.1 months. No recurrence

Study	Patients' Demographics	Chief Complaints	Treatment	Follow-up
Kumar *et al*.[4] (2019)	19-year-male	Presented with complaints of pain and swelling around the distal area of the right leg for 2 months Local examination - swelling, tenderness, induration and discharging sinus around lateral malleolus The radiograph and CT scan show a lytic lesion in the lateral malleoli with a cortical breach in the posteroinferior aspect MRI shows an ill-defined lytic lesion on the plantar aspect of the calcaneum without cortical breach Biopsy – showed caseous necrosis with epithelioid granuloma and Langerhans-type giant cells PCR – positive for mycobacterial TB	AKT for 18 months	36 months, Radiograph and CT-scan showed no recurrence.
Yadav *et al*.[21] (2020)	21-year-male	Presented with complaints of pain and swelling around the right heel with difficulty walking for 5 months Local examination – swelling and deep tenderness present around the right calcaneum (Past history of pulmonary TB) Radiograph – well-defined lytic lesion in calcaneum on Harris-axial view MRI – T1 and T2 wt. images showed a well-defined lytic lesion in the calcaneum Biopsy – showed caseous necrosis with granuloma ZN-Stain and Gene-xpert – were positive	AKT for 12 months and below knee cast for 4 weeks, with partial weight-bearing in the next 4 weeks, followed by full-weight bearing on the 12th week	18 months, the radiograph showed healed lesion.
Ramanathan *et al*.[22] (2020)	Six patients (four calcaneal body and two Posterior tuberosity) involvement, with three females and three males Mean age – 49.1 years	Presented with complaints of pain and swelling around foot and ankle Local examination – shows swelling, tenderness, discharging sinus and toe-tip gait Biopsy and Gene-Xpert were used for diagnosis. Radiograph, CT scan, and MRI show a well-defined osteolytic lesion in the calcaneal body	AKT for 12–18 months in all 4 patients with additional surgery in 1 out of 4 cases	Mean follow-up 14 months, no recurrence.
Bikoroti *et al*.[23] (2021)	9-year-male	Presented with complaints of pain and swelling around left heel extending to left lower limb for 4 months with constitutional symptoms Local examination – shows swelling and discharging sinus with no tenderness The right hip also elicits tenderness and mild shortening CT scan showed a lytic lesion in the calcaneum Biopsy – showed acute, chronic inflammatory cells with multi-nucleated giant cells Auramine-rhodamine stain was positive	AKT for 12 months	21 months follow-up, healed sinus, weight gain, and limb length discrepancy of around 1 cm

Study	Patients' Demographics	Chief Complaints	Treatment	Follow-up
Kadu *et al*.[24] (2016)	8-year-female	Presented with complaints of pain with dull aching character, unable to bear weight on the left heel for 4 months Local examination – shows swelling around the calcaneum with tenderness elicited by deep pressure with no constitutional signs and symptoms Radiograph – shows a small punctate lytic lesion in the metaphysis of the calcaneum Biopsy and ZN-staining were used for diagnosis	AKT was given for 12 months A below-knee cast was given for 6 weeks, followed by partial weight bearing after 6 weeks and at the 10th week, full weight bearing started	24 months follow-up with no recurrence and healed lesion

MRI: Magnetic resonance imaging, *AKT: Anti-Koch Treatment, ZN: Ziehl-Nelson, CBNAAT: Cartridge-based nucleic acid amplification test, CT: Computerized tomography

enzymes, there is a preservation of joint space in TB compared to pyogenic infections. Second, the Phemister triad (peripheral bony erosion, decreased joint space and juxta-articular osteoporosis), is not commonly seen in calcaneal TB.[10,26]

Dhillon *et al*.[7] study on foot TB showed that out of 92 cases, the cystic form was seen in 24 cases. In contrast, a study by Mittal *et al*.[15] showed the presence of the cystic form in only 15 cases out of 44. Articular involvement was seen as a common finding whenever a delay in diagnosis was present. While treating lytic lesions of bone, other infections and tumors are considered a differential diagnosis, so it is important to do a detailed clinical and radiological evaluation followed by a biopsy of the lesions for a definitive diagnosis.

The rare occurrence of TB in the foot and ankle and lack of associated symptoms such as evening temperature rise, appetite and weight loss, night sweats, and radiographic appearance make a definitive diagnosis difficult.[11,27,28] TB is of a paucibacillary nature when bone and joints are involved; therefore, more than one diagnostic modality is required for diagnosis.

To date, many diagnostic tests are available. Still, none of them is highly sensitive and specific for diagnosing skeletal TB, as stated by Dhillon *et al*.[7] Therefore, the present case report confirmed the diagnosis from all the available tests.

The improvement in anti-TB drugs showed clinical improvement, and 12–18 months of AKT is given for TB of osteoarticular origin. However, in the presentation where TB is seen in some unfamiliar locations, AKT can be continued for a few months under strict supervision.[29,30] 12 months of AKT is recommended. With the short duration of treatment, recurrence is commonly due to the paucibacillary nature of skeletal TB, as some of the organisms may remain dormant, leading to resistance to AKT.[31]

With improvement in AKT, the need for a surgical procedure is beneficial for diagnosis when in doubt, lesions not responding to medical treatment or cases with deformities, arthrodesis, and neurological involvement in the tubercular spine.[31]

CONCLUSION

AKT plays a crucial role in managing TB affecting the foot and ankle. Therefore, surgery is usually reserved for cases with unclear diagnosis, pain, or when not responding to medical treatment and deformity correction.

AUTHORS CONTRIBUTIONS

NB convicted the idea for the case report and PC and JC wrote the case report. Finally, the manuscript was reviewed by NB, MS, and MB. All authors have critically reviewed and approved the final draft and are responsible for the manuscript's content and similarity index.

DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

FINANCIAL SUPPORT AND SPONSORSHIP

This study did not receive any specific grant from the public, commercial, or not-for-profit funding agencies.

CONFLICTS OF INTEREST

There are no conflicting relationships or activities.

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