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Case Report

Isolated calcaneal tuberculous osteomyelitis in a 13-year female: a diagnostic dilemma - rare case report and review of literature

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ABSTRACT

Foot and ankle involvement in osteoarticular tuberculosis is uncommon and isolated calcaneal tubercular osteomyelitis of foot bones sparing nearby joints with an osteolytic defect is even more rare. Worsening of the disease can occur over a time involving nearby joints and small bones if diagnostic and therapeutic phase are delayed. We present a retrospective study of osteolytic variety of foot calcaneal bone tuberculosis. We present a case report of 13-year female with isolated osteolytic variety of calcaneal tuberculous osteomyelitis to highlight the clinical, radiological features and management challenges of the condition. Biopsy was done to confirm tissue diagnosis based on Gene Xpert, AFB staining, MGIT culture, and histopathology. Surgical intervention was done in form of debridement and sinus tract removal as a diagnostic and therapeutic purpose to prevent further involvement of a nearby joints and small bone with cystic destruction. Isolated calcaneal tubercular osteomyelitis cases are still found to be seen in our environment though considered a rare. When tuberculous pathology is limited to the isolated calcaneal bone sparing the other joints, the prognosis is better than in osteoarticular disease, as there is less deformity, and hence, less residual pain and disability.

Keywords: Calcaneal, Foot-ankle, Infection, Osteolytic, Tuberculosis

INTRODUCTION

Tuberculosis is transmitted primary through inhalation or ingestion of mycobacterium tuberculosis. The infecting organism is usually Mycobacterium tuberculosis, a thin rod like bacillus with round ends. Infestations by atypical mycobacteria like *M. bovis, M. kansasii, M. avium* complex, *M cheloniae* and *M. fortuitum* are also possible, and some have been reported after penetrating injuries. After exposure, the infection may be cleared by the host and lead to a primary infection or can later be reactivated from latent infection. Skeletal tuberculosis is always a secondary infection from a primary focus elsewhere. Tuberculosis commonly affects the pulmonary system but can affect virtually any organ system of body. Skeletal tuberculosis accounts for 10% to 35% of cases of tuberculosis and is a problem frequently encountered in the Asian and African nations. Skeletal tuberculosis, although not the commonest type of extra-pulmonary presentation, has thus achieved the potential to become a significant problem for the orthopaedic surgeon in general. India alone was postulated to have one fifth of the world population of Tuberculosis patients. Among the skeletal tuberculosis, spine is considered the most affect site but the foot and ankle are relatively infrequent sites, with the incidence in the literature varying from 3% to 6-7% to as high as 12%[1-4]

Nevertheless, osteo-articular tuberculosis is not an infrequent presentation to the orthopaedic surgeon in the developing world.[5] The paucibacillary nature of the discharging sinus makes the bacteriological confirmation more difficult and warrants the use of invasive procedures to establish the diagnosis.[6] The problem becomes complex when it occurs at rare sites, or has an unusual presentation, and the clinician is not aware of the disease.

Tuberculosis of the calcaneus can have varied presentation and can be mimicked by other heel pathologies. This coupled with a lack of awareness on the part of the surgeon often delays the diagnosis and can hence lead to an unsatisfactory clinical outcome and residual functional disability.[11]

To highlight these issues, we present a case of a isolated destructive bony lesion of the calcaneus, which was referred to us and came to OPD. Sometimes a mistake of delaying the correct diagnosis can be made by the treating physician in an attempts to definitively rule out malignancy and prove TB in the rare case presentation. This case draws attention to the 'diagnostic dilemma' and should help raise awareness of atypical presentations of skeletal TB such as foot ankle so that early management can help ensure a good prognosis and prevent unwanted residual deformity.

RESULTS

A 13-year-old female, presented to us in OPD with complaint of right heel pain, warm swelling, inability to weight bear and discharging sinus since 1.5 years. She had an alleged history of trivial trauma to the right heel 1.5 years ago following which she developed insidious onset of pain and warm swelling along with local induration and redness which resulted into a discharging sinus with straw coloured discharge aggravating since 8 months. It was associated with limping and intermittent fever.

She gave a history of multiple visits to the local quack where they performed local debridement and wound wash 6 months back, but it didn't resolve her symptoms. The patient also gave us a history of weight loss, loss of appetite along with evening rise of temperature over a period of 8 weeks. She had a positive family history of pulmonary tuberculosis (TB) contact and with no other co-morbidities. The patient was admitted in our tertiary care hospital for surgical debridement and further management.

On local examination of right foot, a localized swelling was present over lateral aspect of heel about 3cm in maximum dimension below the lateral malleolus with discharging sinus containing pus on and off. The swelling was 3×2 cm in size, hard in consistency, non-mobile, non-compressible, and tender to touch. The margins of the sinus were macerated and had straw coloured pus discharge from it. With the probe the sinus length was examined and confirmed by inserting inside it. The skin overlying the swelling was normal and on palpation the area was warm with point tenderness. Further, the patient had a limping gait as she could not bear weight due to pain. The X-ray of right ankle was done which showed well demarcated lytic lesion over the lateral side of calcaneum with osteopenia around and a sequestrum could be seen (Figure 1). Computed tomography (CT) reconstruction was done in order to check for the cortex of the calcaneum and to outline the extent of the bone destruction (Figure 1). Magnetic resonance imaging (MRI) was also done and suggested of right calcaneum osteomyelitis (MRI demonstration of cortical fractures, early cavitations, and soft tissue fluid collection). The routine laboratory investigations revealed hemoglobin of 11.7 gm%, with a total leucocyte count of 15000 per cubic mm with 80% neutrophils, 26% lymphocytes and 3% eosinophils. The erythrocyte sedimentation rate (ESR) was 48mm after 1 hour and the C-reactive protein was 9.7 mg/l. The viral markers were normal. Routine radiograph of the chest was normal.

Figure 1: Preoperative (a) X-rays, (b) MRI and (c) CT scan of calcaneum showing expansile lytic lesion with cavitation.

Curettage of the lesion was performed after anesthetic clearance. A longitudinal L-shape incision was given over the swelling, the lesion was reached and a thorough curettage of the lesion done with complete excision of sinus tract (Figure 2). Whitish necrotic tissue was curetted out which was sent for Gram staining, Ziehl Neelsen staining, culture sensitivity and biopsy histopathology. Before closure, with 2 ampules of gentamicin powder wound wash was given. As the defect was small bone grafting was not done. A below knee plaster of Paris slab was applied post operatively. The intraop tissue sample were sent for Gene Xpert and yielded MTB on report. The histopathology report revealed presence of chronic inflammatory cells comprising chiefly of lymphocytes, plasma cells and epithelioid cells organized in granulomas

along with Langhans type giant cells. The histomorphology was suggestive of tubercular granulation tissue. The patient was then started on anti-tuberculosis drugs cat 1 4 FDC, but after 2 months it failed and she again developed discharging sinus from the old sinus.

Figure 2: Sinus over posterior aspect of lateral calcaneum with intraop pics of curetted lesion and residual cavity.

Samples were again sent for MGIT which was suggestive of MDR TB, and she was started on Bedaquiline based regime for MDR TB. She completed the entire course of treatment and after 18 months on follow up, the pre-operative symptoms of pain and difficulty in walking had subsided and the morbidity had reduced to a great extent with healed scar (Figure 3).

Figure 3: Follow up after 3 months showing healed wound with normal range of motion.

DISCUSSION

Osteoarticular tuberculosis of the foot is almost always secondary to a primary focus elsewhere in the body. The primary lesion does not manifest always in most of the cases.[10] The primary focus in the body, whether in the lungs or the gut, may itself be latent or active at the time patient presents; however, in a vast majority of cases with skeletal tuberculosis, this primary focus is not readily identifiable. In a previous report of tuberculosis of the foot bones, we found that only 19% of the cases had positive evidence of a primary focus elsewhere in the body.[6]

Tuberculosis of the bone, in general usually begins in the cancellous portion of the bones involved. Involvement of the foot is infrequent. The route of infection in these cases is either direct inoculation or via blood stream.[7,8]

There was a study of 224 cases of tuberculosis of the foot and ankle. At presentation, articular involvement was more frequently encountered, as only a few cases presented principally as osteomyelitis of the tarsal bones or small bones of the foot. Out of these, 51 cases were limited to the calcaneus and presented as tuberculous osteomyelitis; the ankle was involved in 55 cases, and the foot the most common site of presentation was the mid tarsal and subtalar joints. Of all the tuberculous infections in the foot and ankle, the osseous lesion was initially detected in the calcaneus in 22% of the cases, and the ankle joint was involved at presentation in approximately 25% of the feet.[9]

In the present series, these days the most common bone involved was the calcaneus, as has been noted in similar reports focused on the foot. The tuberosity of the calcaneus is most commonly involved in isolated osteomyelitis due to its vulnerability to trauma and since it is far away from the articular surfaces, it is perhaps often picked up prior to joint involvement. The possible reasons for this could range from calcaneus being the largest bone (making it possible to detect lesions still limited to it, in contrast to smaller bones where articular penetration into the joints is early), to its increased vulnerability to direct trauma.[9-14]

In spite of a high index of suspicion for tuberculous infection, bony involvement in isolation can lead to some confusion, and diagnostic delays are common. The possible contributing factors include lack of constitutional symptoms so often related to osteoarticular tuberculosis elsewhere and confusion of radiographic features with bone tumors.[9,10]

In our study, patient presented with painful swelling, discharging sinus, inability to weight bear over the calcaneum and intermittent fever which are similar to pyogenic osteomyelitis and this presentation warrants further investigation, particularly in regions where tuberculosis is endemic. Some differential diagnosis of calcaneal tuberculosis includes pyogenic or fungal infections, benign bone tumours or malignancies (e.g. Ewing's sarcoma).[9]

Sequestrum formation is another common feature of calcaneal tuberculosis. Most sequestrum resorb within 6 months of chemotherapy alone. Resolution of cavities takes a longer time and small, residual cavities are common and have no clinical significance. These cavities are probably filled with fibrous or fibro-osseous tissue and do not warrant surgical intervention.[10,11] Computed tomography and magnetic resonance imaging can show a sequestrum that is not readily apparent on radiographs, and can identify cortical breaks and show the extent of the bone destruction.

In the case reported by, Mittal et al who observed lack of sequestrum in the cystic lesions of the calcaneus.[13] They however noted sequestrate in the smaller bones like the cuboid. We found that coke like sequestrates were of no

consequence and got resorbed over time, with adequate anti tubercular therapy (ATT) .

In our case study, radiologically sequestrum was not seen in the cystic lesion of calcaneum but intraoperatively a small chips of sequestrum could be detected in minute amount deep inside cystic lesion which was believed to be resorbed by the time we operated. Intraoperatively it was seen that osteolytic cystic lesion was confined to calcaneal bone sparing the articular surface and nearby joints which was evident for us to diagnose osteomyelitis of isolated calcaneum.

Also Mittal et al described five radiographic types with the cystic variety being most common (15 of 44 cases).[13] In contrast to that, our case study revealed isolated cystic involvement of calcaneal bone.

Since the isolated osteomyelitis is usually seen only in the early stages of the disease process, early diagnosis and appropriate therapy remains a goal to get good long-term results. Neither the concomitant extra skeletal lesions or evidence of primary pulmonary tuberculosis are always seen nor does the culture or smear give positive results in majority of the cases due to the paucibacillary nature of the biopsy material. Thus a high index of suspicion to be kept always.

In many cases, the suspicion of bone tumor was entertained by the treating physician and the biopsy was done keeping that in mind; the true nature of the lesion was revealed only after tissue biopsy showed features suggestive of tuberculosis. As in our case study we did a debridement and biopsy of proper tissue from deep cystic lesion was sent for sampling to confirm the diagnosis.

Anti-tubercular chemotherapy is the main treatment modality and a minimum of 14 months of chemotherapy is necessary to prevent recurrence. Debridement or resection, with or without arthrodesis, should be reserved for those not responsive to chemotherapy or for those with deformity or painful joints. During debridement, sinus tract should beidentified and complete excision is mandatory leaving no residual. This may hasten healing, and curettage of juxta-articular cavities that threaten to invade an adjacent joint may improve the overall prognosis by minimizing chances of joint involvement.[10]

The outlook of patients suffering from tuberculosis has improved dramatically after the introduction of modern multi drug chemotherapy; however, the best regime for osteoarticular disease still remains a matter of debate.[15] We recommend a minimum of 12 months of chemotherapy due to the higher incidence of recurrences with shorter courses.[5] This prolonged treatment in osteoarticular tuberculosis is justified by the fact that it is a paucibacillary infection with many organisms being in a dormant state, making them resistant to chemotherapy.[5]

Resolution of cavities typically takes a long time and small residual cavities may be visible even years after treatment, and are of no great clinical significance.[10]

If however, the imaging (radiographs/CT/MRI) done at six to seven months of adequate chemotherapy shows evidence of deterioration of the lesion, one should suspect a "non-responding lesion," which may be caused by drug resistant disease, immune compromised state, or a nontuberculous pathology. Tissue diagnosis (with or without debridement) becomes mandatory in these patients.[9] Clinical and radiologic features, along with histopathologic evidence of granulomatous pathology should be sufficient to initiate therapy.[12]

CONCLUSION

There is a high incidence of tuberculous disease in the developing world. India being a endemic region, a high suspicion index, careful clinical examination, X-ray and modern imaging modalities is mandatory which help to establish the diagnosis of osseous, articular, or synovial disease at an early state. Isolated calcaneal osteomyelitis though considered a rare, when presented in the early stages without joint involvement, it should be evaluated in detail and confirm with biopsy for early diagnosis and therapy which is imperative for good long term results. If diagnosed late threatens to cause joint involvement and gives compromised results. Multi-drug therapy can heal most of the early cases with near complete resolution of disease. Operative intervention may be indicated for non-responsive cases, uncertain diagnosis, or to save a joint threatened by a juxta-articular focus. When the disease has healed, surgery may be helpful to arthrodese a painful joint or correct an unacceptable deformity. Under the influence of modern antitubercular drugs the healed status can last life-long. Nearly 2% of cases however may develop reactivation of disease or recurrence of infection in any part of body. This may occur as late as 10 to 25 years after healing of disease.

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