Case Report

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Isolated cystic tuberculosis of medial cuneiform: a case report

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ABSTRACT

Isolated tuberculosis of foot is a rare entity. Plenty of literature is available on osteoarticular tuberculosis but literature on tuberculosis of foot is limited. Additionally the condition mimics variety of other foot pathologies which can delay diagnosis and add to morbidity. We present a twelve year old boy who presented clinically with swelling, pain and discharging sinus of foot with a cystic lesion in medial cuneiform on radiography. Though histopathology and Ziehl Nielsen staining later on established the diagnosis, but initially it was mistaken as a case of mycotic osteomyelitis. Tuberculosis of foot, though rare, should always be one of the differential diagnoses in a patient presenting with bony lesion on radiography irrespective of absence of constitutional symptoms and negative tests for tuberculosis. If battery of tests fails to establish diagnosis, open biopsy with curettage and histopathology should be considered to work out the final diagnosis.

Keywords: Cuneiform, Cystic tubercular osteomyelitis, Mycotic osteomyelitis, Tuberculosis

INTRODUCTION

Skeletal tuberculosis accounts for 1 to 3% of extra pulmonary tuberculosis of which 10% involve foot and ankle. To ther words foot and ankle tuberculosis constitute 0.1 to 0.3% of extra pulmonary tuberculosis. Spine is the most common site and involvement of foot is rare. Exact incidence of involvement of different bones of foot is not known but calcaneum is the most common bone involved. The bones involved are usually the calcaneum, talus, first metatarsal, navicular and medial and intermediate cuneiforms. Because of rarity, diagnosis of tuberculosis of foot remains a dilemma especially when confined to a single bone without articular involvement. So there is always a chance of misdiagnosis and a delay in diagnosis which may add to morbidity. Tuberculosis of foot can mimic variety of

disease like chronic pyogenic osteomyelitis, mycotic osteomyelitis, Madura mycosis and even bone tumors. 3,6-9 In addition isolation of organism by Zhiel Nielson staining and culture of the discharge and pathological tissue is a rarity. 3, 4 All this makes diagnosis of this entity difficult. Presumptive diagnosis can be made on the basis of histopatholgy of the pathological tissue which reveals granulomatous inflammation with or without casseation.

CASE REPORT

A twelve year old boy presented to our Orthopaedic Out Patient Department with swelling, pain and discharging sinus of left foot for last six months. Swelling and pain had progressively increased over last six months to extent that patient had difficulty in walking without support. There was no history of fever, weight loss, loss of appetite, chronic cough, haemoptysis, genitourinary symptoms, and similar involvement of other parts of body or of the other family members. There was history of thorn injury to same foot two years back after which patient developed pus discharge which was managed by oral antimicrobials and antiseptic dressings and the lesion healed within two weeks following which patient had no symptoms for one and a half year till present array of symptoms appeared.

On examination there was swelling of dorsomedial aspect of foot with multiple sinuses surrounded by local erythema on medial aspect discharging straw coloured fluid (Figure 1).



Figure 1: Multiple discharging sinuses with surrounding erythema on medial aspect of foot.

Tenderness was elicited over mid foot region medially. No inguinal lymphadenopathy was seen.

Routine investigations were normal. ESR was 55 mm after one hour. Mantoux reaction was positive (18 millimetres at 72 hours). A lytic lesion of medial cuneiform containing sequestrum was noticed on radiograph with apparently normal tarso-metatarsal and inter-tarsal joints (Figure 2). Based on the past history of thorn injury a provisional diagnosis of fungal osteomyelitis was made.



Figure 2: Antero posterior and lateral radiographs of foot showing lytic lesion (arrows) containing sequestrum in medial cuneiform.

Chest radiograph appeared normal. Gram staining, Ziehl Nielsen staining, wet KOH mount, and culture of the discharge for fungi, pyogenic organisms and mycobacterium tuberculosis were negative.

Because of failure to isolate fungi, bacteria or acid fast bacilli from the sinus discharge surgical intervention was done by excising the sinus tract, debridement of unhealthy soft tissue and curettage of the medial cuneiform. Intraoperatively soft tissue surrounding medial cuneiform was unhealthy, there was breach in the medial cortex of medial cuneiform and the cavity inside it contained unhealthy granulation tissue. Adjacent joints were normal. All the excised tissue and curettage was sent for Gram staining, Ziehl Nielsen staining, KOH mount, culture for fungi, pyogenic organisms, and mycobacterium tuberculosis and for histopathological examination. Ziehl Nielsen staining of curetted material revealed acid fast bacilli (Figure 3) and culture on Lowenstein Jensen media grew mycobacterial colonies (Figure 4). Histopathology revealed granulomatous inflammation, epitheloid cells, multinucleate giant cells, and plenty of plasma cells with casseaous necrosis.

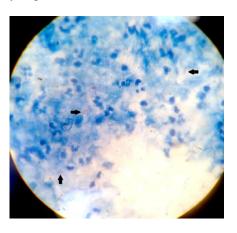


Figure 3: Ziehl Nielsen staining of curetted specimen showing acid fast bacilli (arrow).



Figure 4: Culture on Lowenstein Jensen medium showing dry and buff coloured colonies of mycobacterium tuberculosis.

On the basis of histopathology and isolation of mycobacteria, diagnosis of tuberculosis of medial cuneiform was made and antitubercular therapy was started and foot splinted with below knee posterior slab. Weight bearing was restricted. At 6 weeks follow up, splint was removed and active range of motion of ankle started. Partial weight bearing was started at 12 weeks and progressively increased as tolerated.

Antitubercular therapy was given for 12 months, four drugs namely isoniazid, rifampin, pyrazinamide, ethambutol for two months followed by isoniazid and rifampin for ten months. Final follow up at one and a half year revealed healed sinuses, painless non tender weight bearing foot with normal range of motion. Radiograph revealed small cystic cavities in medial cuneiform with surrounding sclerosis and mineralization of surrounding bones (Figure 5).



Figure 5: Follow up radiograph at one and a half year, cystic lesion in medial cuneiform has decreased in size with surrounding subtle sclerosis (arrows).

DISCUSSION

Isolated tubercular involvement of foot bones with an osteolytic defect is a rare entity.^{3,10} Dhillon MS et al studied 92 cases of foot tuberculosis over 20 years, 23 were of osteolytic variety out of which 2 had lesion in the cuneiform.³ Mittal et al has classified tubercular lesions radiologically into five type namely, cystic, rheumatoid, subperiosteal, kissing and spina ventosa.¹⁰ Cystic lesions are relatively uncommon, characterized by pure interosseous lesions with or without soft tissue component.^{3,10} As they have not yet invaded the joint prognosis is good if early treatment is instituted. In our case there was a cystic lesion of medial cuneiform, which had breached the medial cortex (nonarticular surface) but not the articular surface. There was soft tissue involvement with a discharging sinus.

Because involvement of cuneiform is rare and it may mimic other pathologies such as fungal osteomyelitis, madura mycosis, chronic pyogenic osteomyelitis, bone tumors (aneurysmal bone cyst, giant cell tumor, telangiectatic osteosarcoma and angiosarcoma) diagnosis may be delayed which may add to morbidity and deteriorate prognosis especially if it involves the adjacent joint. In our case diagnosis of mycotic osteomylitis was initially made based on history of vegetative trauma but failure to isolate fungi made us to reconsider our diagnosis.

Other factors which contribute to diagnostic delay is lack of constitutional symptoms so often related to tuberculosis elsewhere, relative normal picture on laboratory investigations, failure to isolate organism in Ziehl Nielsen stain and cultures. 3,4,6,8 ESR is almost always elevated in case of tuberculosis. 2,12,13 In our case the only diagnostic clue was positive mantoux test and raised ESR both of which are not diagnostic. It was histopathlogy of the tissue specimen, Ziehl Nielsen staining and isolation of mycobacteria on Lowenstein Jensen media which finally lead to diagnosis.

Prolong drug therapy (for a minimum of 12 months) proves effective for eliminating or sterilizing the persistent bacilli, which are small populations of metabolically inactive microorganisms. 14 Treatment should not be delayed waiting for culture results because experience suggests that delay in treatment may result in less than optimal outcome. 14 However, in various studies, the duration of therapy has varied widely: 6 months in sacral TB, 12-18 months in various spinal sites, 12-18 months in TB of craniovertebral junction, 14-18 months in sternoclavicular joint involvement, 12-20 months in TB affecting the talus, 12 months in tuberculosis of metacarpals and phalanges.¹⁴ Short course antitubercular therapy has high recurrence in osteoarticular tuberculosis, so should be given for a minimum of 12 months.^[5] We followed the same protocol. Aim of treatment is to eradicate the organism and to obtain a supple, pain free weight bearing functional foot.

With treatment radiological changes do take place but cavities may persist for years and are of no clinical significance. In our case at final follow up of one and a half year radiographs showed decrease in size of cavities, surrounding sclerosis and improved mineralisation with a painless, supple weight bearing foot.

It is concluded isolated cystic tuberculosis of medial cuneiform is a rare entity which may pose a diagnostic challenge for an orthopaedic surgeon. Whenever diagnosis is doubtful, one can go for open biopsy to confirm it by histopathology, Ziel Nielson staining and culture for mycobacterium tuberculosis and institute early anti tubercular therapy for a minimum of 12 months for better prognosis and to prevent recurrence.

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