Xanthogranulomatous osteomyelitis

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ABSTRACT

Xanthogranulomatous inflammation in bone is a very rare entity. We came across this rare case of xanthogranulomatous osteomyelitis. We present a rare case of 50-year-old male who presented to the hospital with the chief complaint of pain and swelling in left ankle for 5 months and fever for 2 days. Total leukocyte count and differential leucocyte count was normal. Renal function tests and liver function tests were normal. Montoux test was negative. Chest X-ray was normal. Pus culture was negative for acid-fast bacilli but showed heavy growth of Pseudomonas. X-ray imaging revealed soft tissue mass, periosteal reaction and cortical disruption in the lower end of left tibia. Sensitivity to Amikacin and Imipenem was found on pus culture. So patient was started with these antibiotics. However, pain was not relieved. So arthrodesis was done, and curetted material was sent to histopathology lab where it was diagnosed xanthogranulomatous osteomyelitis. The patient was followed-up for 1 month. He did not have any complaints. All the previously existing complaints subsided. Xanthogranulomatous inflammation can be seen in bones also, though rarely reported.

Key words: Arthodesis, xanthogranulomatous osteomyelitis, tibia

Introduction

Xanthogranulomatous inflammation has been described in various organs such as kidney, gallbladder, colon, pancreas, and salivary gland.[1] However, xanthogranulomatous osteomyelitis has not been much described in the literature. Organs such as lung, brain, and bone are rarely affected. Bone involvement is accompanied by systemic and regional clinical presentations such as pain, fever, and leukocytosis.[2] Hence far seven cases of xanthogranulomatous osteomyelitis have been reported in the world to the best of our knowledge,[3-8] We report the eighth case of xanthogranulomatous osteomyelitis.

Case Report

The patient provided consent to report this case.

A 50-year-old male was admitted to the hospital with the chief complaint of pain and swelling in left ankle for 5 months and fever for 2 days. Patient was diagnosed to be suffering from tuberculosis 4 years back for which he had completed full course of antituberculous treatment (isoniazid, rifampicin, ethambutol, and pyrazinamide daily for 2 months, followed by 4 months of isoniazid and rifampicin).

Physical examination showed tenderness and swelling in the ankle with a pus point on swelling.
Laboratory investigation showed raised erythrocyte sedimentation rate. Total leucocyte count and differential leucocyte count was normal. Renal function tests and liver function tests were normal. Montoux test was negative. Chest X-ray was negative.

X-ray imaging revealed soft tissue mass, periosteal reaction and cortical disruption in the lower end of left tibia [Figure 1].

On microbiological examination, pus culture was negative for acid fast bacilli but showed heavy growth of *Pseudomonas*. Sensitivity to Amikacin and Imipenem was found. Hence, the patient was started on these antibiotics. However, the pain was not relieved, and arthodesis was done. Curated material was sent to histopathology lab.

Grossly, the specimen received was soft and gray-brown. Microscopically, mixed inflammatory infiltrate comprised of sheets of foamy histiocytes, lymphocytes, plasma cells, and few polymorphs was seen infiltrating the bony trabeculae. Stain for acid-fast bacilli was negative. Thus, a histopathological diagnosis of xanthogranulomatous osteomyelitis was made [Figure 2a-d].

**Discussion**

Thus, xanthogranulomatous osteomyelitis is a rare entity. Delayed-type hypersensitivity reaction of cell-mediated immunity may be implicated in its pathogenesis. Grossly, it is a mass-like lesion extending to enclosing tissues, which can mimic the infiltrative cancer.[2]

Microscopically, it shows the presence of features of osteomyelitis (admixture of inflammatory cells including neutrophils, lymphocytes, plasma cells) with abundant macrophages.[3]

Although association between bacterial infection and the xanthogranulomatous inflammation has been determined in several organs such as kidneys, but it is still to be proven for bone.[3]

The first two cases of xanthogranulomatous osteomyelitis were diagnosed by Cozzutto in 5-year and 14-year-old males in fist rib and tibia respectively.[3] Borjian et al. described a case of xanthogranulomatous osteomyelitis involving two bones independently, one in the right humerus and other in left fibula, simultaneously.[9]

The key features of previously diagnosed cases described in the literature are discussed in Table 1.

Thus, majority of the patients presented with fibrile illness, pain, and swelling in the bones.

Laboratory investigations revealed the neutrophilic leucocytosis and culturable micro-organism in few patients. X-ray revealed osteolytic lesions in most patients.

Microscopically, xanthogranulomatous osteomyelitis has to be differentiated from Langerhan’s cell histiocytosis, Erdheim-Chester disease, and lipid storage disorders. Langerhan’s cell histiocytosis commonly affects mid-shaft portions of long bones and often has infiltration of eosinophils. Erdheim-Chester disease is a multifocal disorder with frequent involvement of extraskeletal tissues. Histology shows presence of cholesterol clefts along with foamy histiocytes. In the case of Gaucher’s disease and Niemann Pick disease, foam cell transformation is usually seen within the bone marrow, and a clinical history plays an important role in differentiating storage disorders from xanthogranulomatous osteomyelitis.[3]

**Figure 2:** (a) Bony trabeculae (H and E, ×100). (b) Chronic inflammatory infiltrate (H and E, ×400). (c) Xanthogranulomatous change (H and E, ×100). (d) Xanthogranulomatous change (H and E, ×400)
Thus, we conclude that xanthogranulomatous osteomyelitis is a rare diagnosis which can be treated easily. Hence, care has to take to differentiate it from other benign and malignant lesions of bone.

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Table 1: Salient features of reported cases of xanthogranulomatous osteomyelitis

| Xanthogranulomatous osteomyelitis | Vankalakunti et al.[4] | Cennimo et al.[5] | Borjian et al.[6] | Kamat et al.[7] | Seung et al.[8] |
|----------------------------------|------------------------|-------------------|------------------|-----------------|-----------------|
| Age/sex                          | 50 years/female         | 41 years/male     | 14 years/male    | 13 years/male   | 59 years/male   |
| Site                             | Ulna                   | Index finger and wrist | Humerus, fibula | Ankle           | Wrist           |
| Presentation                     | Pain and swelling for 2 years | Pain and swelling for 1-year. history of TB | History of trauma. Pain for 8 weeks | Pain and swelling in ankle | Pain, swelling of wrist (2 months), history of MDS |
| Significant lab findings         | No microorganism on culture | Positive culture of Mycobacterium marinum | Neutrophilic leucocytosis, raised ALP, raised ESR. S. aureus present | Raised ESR. S. aureus was isolated | Leucocytopenia, thrombocytopenia |
| Radiological investigation       | X-ray: Destructive lytic lesion | X-ray: Soft tissue swelling | X-ray: Periosteal reaction | X-ray: Periosteal reaction, bone marrow infiltration. | X-ray: Submetaphyseal lytic lesion in the distal tibia with a sclerotic margin around it |
| Procedure done                   | Curetted out, cancellous iliac crest graft interposed | Synovectomy and minocycline, clarithromycin, and ethambutol | MRI: Synovial enhancement | MRI: Signal abnormalities | Excisional biopsy |

CT: Computed tomography, MRI: Magnetic resonance imaging, MDS: Myelodisproliferative syndrome, ALP: Alkaline phosphatase, ESR: Erythrocyte sedimentation rate, S. aureus: Staphylococcus aureus