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RESEARCH ARTICLE

A RARE CASE REPORT OF XANTHOGRANULOMATOUS OSTEOMYELITIS OF RIGHT TIBIA

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ABSTRACT

Xanthogranulomatous Osteomyelitis is a specific type of chronic inflammatory process characterized by compilation of foamy macrophages admixed with mononuclear cells. Pathogenesis may be due to cell mediated immunity of delayed hypersensitivity type. We describe the case of a 25-year-old female presenting with pain in left leg and disease confirmation was made by histological examination as Xanthogranulomatous Osteomyelitis.

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INTRODUCTION

Xanthogranulomatous Osteomyelitis is an infrequent kind of inflammatory process which is characterized by composition of immune cell aggregation on histological studies (1). Delayed-type hypersensitivity response of cell-mediated immunity may be concerned in its pathogenesis (1). Gross and radiological examination can mimic malignancy, and differentiation should be confirmed by histopathological evaluation. Xanthogranulomatous osteomyelitis has been typically described by the presence of, PAS positive histiocytes which are granular and eosinophilic in the initial stages with a mixture of activated plasma cells and foamy macrophage and the presence of suppurative foci and hemorrhage (2). It has been found in various tissues like the kidney, fallopian tube, ovary, vagina, prostate, urinary bladder, testis, gallbladder, epididymis, colon and appendix. Very rarely, it can affect lungs, brain, or bone (3).

Case presentation

A 25-year-old woman presented to the out-patient department with a bony hard swelling on the extensor aspect of her right leg. The swelling was slowly progressive in nature. On local examination there was a small tender prominence of 1 × 1.5 cm located in the lower one-third of the right Tibia. Both anteroposterior and lateral radiographs revealed an expansile sclerotic destructive lesion involving the mid one-third of diaphysis of the right tibia, with an ill defined zone of transition. No matrix calcification or periosteal reaction is seen (Fig.1). MRI showed focal cortical thickening of tibia in the lower half of medial aspect with homogenously hypointense in signal, without nidus with normal surrounding soft tissues (Fig.2), rest of the investigations including serum uric test, urine examination, thyroid profile, liver, renal function tests and nerve conduction studies were normal. A primary bone tumour was clinically suspected. The bone site involved was curetted and thoroughly lavaged and bone wax was placed. On gross examination the tissue was sent for histological examination was powdery in consistency, whitish in color with few bony particles. Histologically the lesion showed necrotic bony tissue and a few fomy histiocytes, spindle cells and occasional osteoclastic gaint cells with no granuloma seen. Section from bone shows thick bony lamellae separated by fibrovascular tissue containing a few lymphocytes.

The tissue cultures did not show growth of any organisms. As Birbeck granules were not identified hence diagnosis of Langerhans cell histiocytosis was ruled out. The postoperative period was uneventful and pain gradually subsided, with healed wound and no discharge. Xanthogranulomatous inflammation has been related to bacterial infection in several organs such as gastrointestinal system, kidneys and gall bladder but remains undetermined for bone.

DISCUSSION

Xanthogranulomatous Osteomyelitis is encountered in different tissues i.e. the urinary bladder, gallbladder, fallopian tube, kidney, vagina, testis, ovary, epididymis, colon and appendix.

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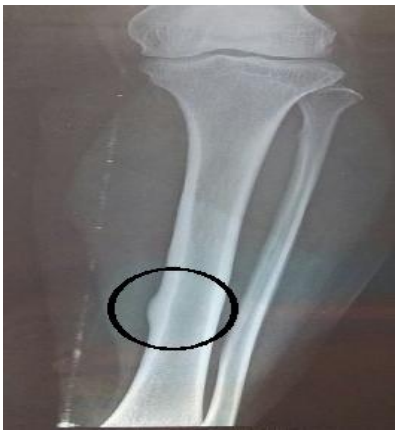

Figure 1 Radiograph of an antero-posterior view of the right leg displays an expansile sclerotic destructive lesion in the distal tibia.

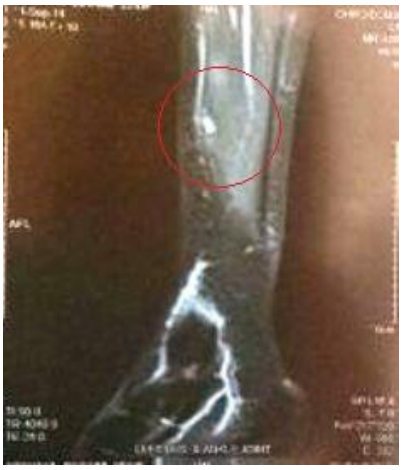

Figure 2 Coronal contrast enhanced T1-weighted MRI showed a showed focal cortical thickening of tibia in the lower half of the medial side of the distal tibia.

Very rarely, it can affect lungs, brain, prostate, or bone. In 1984, Cozzutto *et al* first described 2 cases of involvement of the first rib in a 5-year-old boy and the proximal metaphysis of the tibia in a 14-year-old boy (1). Cennimo *et al* reported a case that manifested as a xanthogranulomatous reaction with culture results positive for *Mycobacterium marinum* in the index finger and wrist of a 41-year-old sushi chef in 2009 (2). In 2007, Vankalakunti *et al* reported a case of 50-year-old woman presenting Xanthogranulomatous osteomyelitis in form of osteolytic lesion in diaphysis of ulna (3). Another case of Xanthogranulomatous osteomyelitis, which presented as swelling in the right distal tibia which is osteolytic in nature with sclerotic border, in a 13-year-old boy was reported by Kamat *et al*. in 2011(4). Recently, a case of Xanthogranulomatous osteomyelitis invading 2 separate bones, the metaphysis of the right humerus and the medulla, metaphysis, and diaphysis of the left fibula, with mixed densities in a 14-year-old boy was published by Borjian *et al*. in 2011(5). Lee *et al* presented a case report of xanthogranulomatous osteomyelitis of the distal ulna mimicking a malignant neoplasm, with osteolytic lesion in 2013 (6). Only 7 cases of Xanthogranulomatous osteomyelitis in literature have been reported till date. This is the eighth case of Xanthogranulomatous osteomyelitis in bone that we are presenting in distal tibia. Based on our findings we found that Xanthogranulomatous osteomyelitis can present with variable densities such as osteolytic, sclerotic (as in our case) and mixed densities radiologically as summarized in the table below. Xanthogranulomatous osteomyelitis resembles malignancy, as it forms mass lesions in gallbladder, prostrate and kidney. Histologically these lesions revealed chronic inflammatory in nature with foam cells which were derived from monocytes or macrophages as they were positive for KP1, HAM56, CD11b and CD68 (7). It is a granulomatous disorder characterized by accumulations of macrophage foam cells and T cells. Pathogenesis may be due to cell mediated immunity of delayed hypersensitivity type (7). The relationship of Xanthogranulomatous osteomyelitis and trauma developing at the same site is hypothetical. The other causes of similar clinical presentation can be xanthoma, malakoplakia, metastatic renal cell carcinoma, Langerhans-cell histiocytosis, fibrohisticotytic tumor, chronic recurrent multifocal osteomyelitis, infiltrative storage disorder and erdheim-chester disease (8-9). Histopathological examination plays a vital role in diagnosis of xanthogranulomatous osteomyelitis along with magnetic resonance imaging and computed tomography.

Table 1 Summary of appearance of case reports based on site, year and Radiodensities.

Authors & Year	Site	Radio Densities
Cozzutto C.1984 (1)	1st rib, proximal metaphysis of tibia of different patients.	----
Vankalakunti M. *et al.*, 2007 (3)	Diaphysis of ulna.	Osteolytic
Cennimo DJ. *et al.*, 2009 (2)	Index finger and wrist.	----
Kamat G. *et al.*, 2011 (4)	Distal tibia.	Osteolytic
Borjian A. *et al.*, 2012 (5)	Metaphysis of the humerus and the medulla, metaphysis, and diaphysis of the fibula of same patient.	Mixed Density
Lee S.H. *et al.*, 2013 (6)	Distal ulna.	Osteolytic

CONCLUSION

This case is primarily presented due to its rarity and is the eighth case of Xanthogranulomatous osteomyelitis which is being reported in the literature to the best of our knowledge. The importance lies in the differential diagnoses of Xanthogranulomatous osteomyelitis which resembles tumor either benign or malignant based on gross and radiological examination and found that Xanthogranulomatous osteomyelitis can present with variable densities such as osteolytic, sclerotic (as in our case) and mixed densities radiologically. Histopathological examination plays a vital role in diagnosis of Xanthogranulomatous osteomyelitis and hence helps in appropriate treatment plan of the case.

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