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Xanthogranulomatous osteomyelitis: A case report

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Abstract

Xanthogranulomatous Osteomyelitis is a rare form a chronic osteomyelitis. Most cases presents with severe chronic inflammation with pain, fever and leukocytosis. Radiological findings in all described cases revealed a lytic lesion, which was most often diagnosed clinically as a bone tumor.It is characterized by composition of immune cell aggregation on histological studies. Delayed-type hypersensitivity reaction of cell-mediated immunity may be implicated in its pathogenesis. [6]

Keywords

Xanthogranulomatous Osteomyelitis, Inflammatory, Femur

1. Introduction

The term XanthogranulomatousOsteomyelitis (XO) was first coined by Cozzutto in 1984 characterised by prominent foamy macrophage collection on histopathology.Xanthogranulomatous Osteomyelitis is a very rare form of chronic osteomyelitis with only 5 reported cases in literature till date. Xanthogranulomatous inflammation has been reported kidney, pancreas, gall bladder etc but very rarely in bone. We report a case of 20 yrs old female who presented with lytic lesion at Proximal end of femur and was clinically diagnosed as a primary bone tumor for which currettage and bone grafting was done. Histopathological examination revealed sheets of foamy histiocytes with scanty plasma cells and lymphocytes and was diagnosed as xanthogranulomatous osteomyelitis. Patient was doing well at six months of follow up. This case is presented on accounts of its extreme rarity.

2. Case Report

We report a case of a a 20 years old female presented with complaints of pain in left hip for 1 year. There was no fever. Radiological examination revealed a lytic lesion at proximal left femur and was clinically diagnosed as primary or secondary bone tumor. [Fig 1] Curettage and bone grafting was performed and curetted material was sent for histopathological examination.

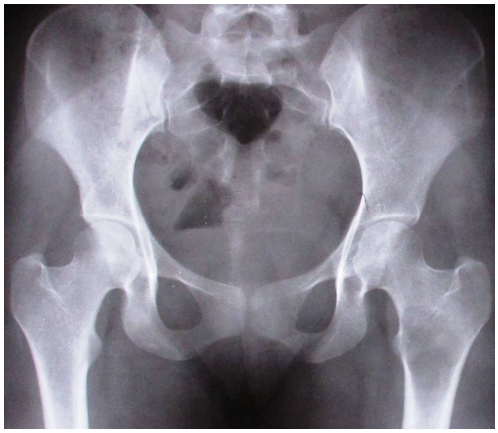

Fig 1. *X-ray of femur showing osteolytic lesion at proximal end.*

Grossly multiple greyish brown soft tissue bits were received along with scanty bony spicules aggregate measuring 3 X 3 X 1 cms. The entire tissue was processed for examination.

Microscopically sections showed bony spicules intervened by marrow spaces showing dense collection of foamy histiocytes having round vesicular nuclei and abundant cytoplasm. Surrounding fibroconnective tissue showed chronic inflammatory infiltrate including scanty plasma cells and lymphocytes. [Fig-2] PAS &Mucicarmine stains done are

negative favouringpseudogaucher cells. So a final diagnosis of chronic osteomyelitis compatible with Xanthogranulomatous Osteomyelitis was rendered. Patient was doing well at 6 months of follow up with no reoccurrence.

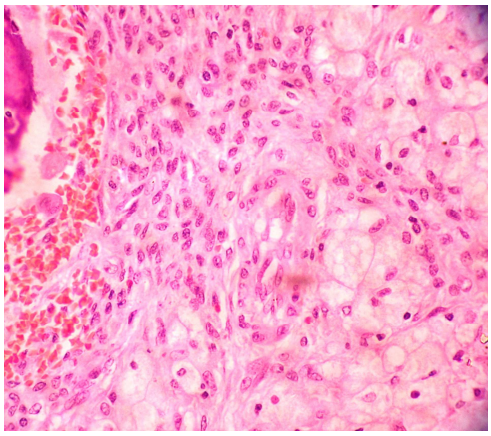

Fig 2. Light Microscopy showing sheets of foamy histiocytes with scanty lymphocytes and sclerotic bony spicules (H & E, 40X)

3. Discussion

Xanthogranulomatous Osteomyelitis is a very rare form of chronic osteomyelitis with only 8 cases reported is literature till date.[1,2,3,4,5,6 and 7]

Cases of xanthogranulomatous inflammation have been reported in organs like Gall bladder, Kidney,Urinary bladder, Fallopian tube, Ovary etc. [8,9]Very rarely it involves lung, brain, prostate or bone.

To the best of our knowledge only 8 cases have been reported in literature till date out of which in 4 cases the disease was seen in young individuals (5, 13,14 and 14 years) with three cases were reported in aolder patients (41, 50 and 59 years) .

Clinically all cases presented with complaints of pain and swelling of involved bone. The disease mostly involves males, with male: female ratio of 6: 1. Site of involvement of these cases were first rib, tibial epiphysis, ulna, distal end of right tibia, femur and fibula.

Radiologically most cases presented with lytic lesion of involved bone with a radiological diagnosis of primary or secondary bone tumor.(Table 1 Case Reports of xanthogranulomatous Osteomyelitis)

However microscopically differential diagnoses are Langerhan's cell Histiocytosis, and lipid storage diseases. In Langerhan's cell Histiocytosishistiocytes are admixed with eosinophils and show nuclear grooving [10].Histiocytes in Lipid storage diseases show characteristic fibrillary appearance and are intensely PAS positive.

This case herein reviews the clinical and histopathological features of this rare entity. Besides the rarity, Xanthogranulomatous Osteomyelitis should always be kept in mind when a lytic bony lesion shows dense histiocytic infiltrate on histopathology.

Table 1. *Case Reports of xanthogranulomatous Osteomyelitis)*

Case reports	Number of patients	Bone involved	Age	Sex	X ray Finding
Cozzutto C. [1]	Two patients	1rst Rib	5 year	male	-
		Proximal metaphysic of tibia	14 year	male	-
Vankalakunti M., Saikia U.N., Mathew M. et al. [2]	One	Diaphysis of ulna	50 year	female	Expansile lytic destructive lesion involving the mid one-third of diaphysis of the right ulna, with an ill-defined zone of transition
Kamatetal [3]	One case	Right tibia	13 year	male	Submetaphyseal lytic lesion in the distal tibia with a sclerotic margin around it
Kashanietal [4]	One case	femur	-	-	-
Cennimo et al.	One Case	index finger and wrist	41 year	male	-
Borjian A., Rezaei F., Eshaghi M.A. et al. [5]	One case	Metaphysis of the humerus and the medulla, metaphysis, and diaphysis of the fibula	14 year	Male	Mixed density, periosteal reaction, and cortical disruption with soft tissue swelling
Lee etal [6]	One Case	Distal tibia	59	male	Bulging mass-like lesion located at the distal ulna with a round radiolucent lesion of the distal ulna, suggesting an osteolytic mass

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