Bilateral Tibial Xanthoma in a Normolipidemic Patient—Report of a Rare Case with Review of Literature

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ABSTRACT

Xanthoma of bone is a rare benign bone disorder. It radiologically presents as a lytic lesion, often with cortical expansion or disruption, mimics primary bone tumors and metastatic lesions. Histopathological study gives the definitive diagnosis. Xanthoma of bilateral tibia in a normolipidemic patient is rare and not yet reported in literature. Here with we present one such rare case, with review of literature.

Keywords: Xanthoma; Normolipidic; Tibia; Lytic Lesion

1. Introduction

Xanthoma of bone is an exceedingly rare benign primary bone tumor. Intraosseous xanthomas are lytic, expansile lesions composed of lipid-laden histiocytes, [1] often seen in patients with hyperlipidemic conditions [2,3]. Xanthomas are histologically characterized by mononuclear macrophage-like cells, abundant foam cells, and multinucleated giant cells. Occasionally, spindle cells are present, which has led investigators to include this lesion as a subset of benign fibrous histiocytoma (BFH) of bone [4]. One 45 years old lady who presented to our institution was discovered to have an intraosseous xanthoma in both tibia in the absence of hyperlipidemia. The lesion showed abundant giant cells, mononuclear cells, foam cells and cholesterol crystals. We present this rare case of intraosseous xanthoma, along with review of literature.

2. Case Report

A 45 year old housewife presented with swelling in upper 1/3rd of both lower limbs, pain & inability to walk on left leg following a slip & fall. On examination there was a diffuse swelling and tenderness in left leg. Patient was asymptomatic before fall and hence duration of the swelling could not be determined. On investigating, x-ray of left leg revealed fracture of both bones of left lower limb and an osteolytic lesion in the proximal part of left tibia (Figure 1). X-ray of right leg showed an osteolytic lesion in the upper 1/3rd of tibia (Figure 2). MRI-T1, T2 & FATSAT of Lt leg showed multiseptate lesion involving epiphysseo-metaphyseal region with extension upto proximal diaphysis with enhancing signal in the lesion at

Figure 1. X-ray of left leg showing fracture of both bones and an osteolytic lesion in proximal tibia.

Figure 2. X-ray of right lower leg showing osteolytic lesion in proximal part of tibia.
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fracture site, suggesting Haemorrhage. MRI-T1, T2 & FATSAT of right Leg showed multiseptate lesion of epi-
physeal region with extension up to proximal diaphysis on right side. No obvious extracortical soft
 tissue extension was seen, normal joint space & articular surface was maintained. The patient was submitted for
Biochemical investigations and the results showed, Serum calcium –9.5 mg/dl (8.5 - 10.5), Serum phosphorus
–4 mg/dl (3.4 - 5), Alkaline phosphatase –65 IU/L & Parathyroid hormone levels –24 pg/dl (10 - 55) were
within normal limit. No abnormality was noted in remaining other systems. Following this, curetting biopsy of left
Tibial lesion with bone graft and nailing was done and specimen was sent for histopathological opinion. The
bony and soft tissue bits were gray to yellow in colour, and histopathologically showed bundles of hyalinized
collagen fibers traversed by small & large blood vessels lined by endothelial cells, non specific granulation tissue
at the periphery. Cholesterol clefts & giant cells at focal areas. No cyst was identified in multiple sections studied.
To rule out haemangioma venous doppler of left & right Lower limb was done which was normal. This was fol-
lowed by Fine needle aspiration of right tibial lesion which yielded 3 cc of straw coloured fluid and this fluid
showed few cells consisting of foamy macrophages and occasional lymphocytes, neutrophils in a proteinaceous
background. Following this, curettage biopsy and bone graft of the right tibial lesion was done and the tissue was
sent for histopathological opinion. Biopsy constituted of multiple bony and soft tissue bits and these showed
sheets of foamy histiocytes interspersed with occasional inflammatory cells, in a fibrocollagenous stroma. There
were abundant cholesterol clefts with giant cells around them and Cholesterol clefts between bony spicules were
seen (Figures 3). Special stains were done, and Vangij-
esson’s stain showed delicate collagen in the interstitium,
whereas Reticulin stain was negative, no blood vessels or individual cell reticulin was seen. Schultz reaction on frozen
section was done which was weakly positive, showing light blue green cholesterol crystals. Polarizer on frozen
sections showed focal birefringent cholesterol crystals as needles & block brightness. Histologically benign fibrous
histiocytoma and non ossifying fibroma were considered as differential diagnosis. The benign fibrous histiocytoma
shows spindle cells arranged in storiform pattern which was lacking in our case, where as non ossifying fibroma
shows cellular fibrous tissue arranged in storiform pattern, scattered osteoclasts, foamy & hemosiderin laden
macrophages and sometimes bizarre nuclei, and all these features were lacking in our case. And thus depending on
the above findings and radiological and biochemical findings the final diagnosis was rendered as primary tuberous
xanthoma of both tibial bones.

Figure 3. Abundant cholesterol clefts with giant cells around them. Sheets of foamy macrophages interspersed with occasional inflammatory cells. There are also bony spicules between cholesterol clefts.

Seeing the histology of the samples received, the patient was submitted for lipid profile and the family members
were also advised lipid profile study. The lipid levels of the patient were within the normal limit, and the son of
the patient who was the only relative available refused to cooperate.

3. Discussion—Review of Literature

Xanthoma is a tumour like collection of foamy histiocytes. They may occur in all five subtypes of essential hyper-
lipidemia, in disease states associated with secondary hyperlipidemia (e.g., primary biliary cirrhosis, diabetes
mellitus) and occasionally in the normolipidemic state. Usually xanthomas occur in the skin and subcutis, but
occasionally they involve deep soft tissue such as tendon or synovium and bone. Cutaneous xanthomas are design-
ated into 5 types, 1) Eruptive, 2) Tuberous, 3) Tendinous xanthomas, 4) xanthelasma, 5) Plane xanthoma [5].

Xanthoma of bone is an exceedingly rare tumor (<70 cases reported in the appendicular and axial skeleton till
was also followed in our case. Grafting of the lesion is the treatment of choice, which has been reported [31, 32]. Surgical Curettage & bone

In view of the rare intraosseous location with bilaterality in normolipimic individual and with the histology corresponding to a tuberous xanthoma, our case is unique in the sense that our case is first with symmetrical localisation involving tibia and hence this is being presented.

REFERENCES


