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Case Report Open Access

# Primary Xanthoma of Tibia in Normolipidemic Patient: A Case Report

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**Abstract**

Primary xanthoma of bone is an exceedingly rare disorder with unsettled histiogenesis that may pose a challenge due to its wide range of differential diagnosis. Due to its aggressive radiographic appearance other primary bone tumors and metastatic lesions need to be ruled out. Herein, we present a case of primary intraosseous xanthoma of right tibia that was misdiagnosed as giant cell tumor on radiology in a women with no aberrant lipid metabolism or evidence of pre-existing bone lesion.

**Keywords:** Xanthoma; Bone tumor; Giant cell tumor

## Introduction

Primary intraosseous xanthoma is a rare benign lesion with unsettled histiogenesis characterized by abundant foamy histiocytes (xanthoma cells), giant cells and mono nuclear macrophages. This lesion may pose a diagnostic challenge to radiologist, pathologist and clinician. This lesion has a wide range of differential diagnosis as giant cells, sheets of foamy macrophages and cholesterol deposits are often seen as secondary or degenerative findings in many neoplastic and non-neoplastic bone lesions. Xanthoma of bone is solitary and mostly flat bones are frequent sites of involvement. Herein we present a case of 30 year old female with no aberrant lipid metabolism or evidence of pre-existing bone lesion [1]. Xanthoma of bone is an exceedingly rare harmless primary bone tumor. Intraosseous xanthomas are lytic, expansile lesions composed of lipid-laden histiocytes, regularly seen in patients with hyperlipidemic conditions. Xanthomas are histologically distinguished 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. Xanthoma is an abnormal deposition of cholesterol or triglycerides, which can observed in any part of the body and in many kind of disease states. These lesions contain abundant foamy histiocytes, and commonly occur in superficial soft tissue, such as skin, subcutaneous fat, or tendon sheaths. Primary Intraosseous Xanthoma (PIX) is very limited and, as such, its predictors, such as age, gender, and race, are not well known [2,3]. They have been reported in the appendicular skeleton, including the tibia, calcaneus, and humerus. Although several cases of PIX have been reported, they usually appear in the setting of hyperlipidemia, especially primary or familial. To our knowledge, no case of PIX of the proximal femur in a normolipidemic patient has been reported [4].

## Case Report

A 30 year old female sought medical attention at our primary health care center with complaint of right leg pain for the last three months.

On examination diffuse swelling and tenderness was noted in right leg with no signs of local inflammation. The range of motion was not limited. Patient's lipid profile and other laboratory investigations were in normal limits [5,6]. X-ray showed a well circumscribed, lobulated, expansile osteolytic lesion with thick sclerotic margin and internal septations in diaphysis of right tibia. The case was diagnosed radiologically as giant cell tumor of bone (Figure 1).

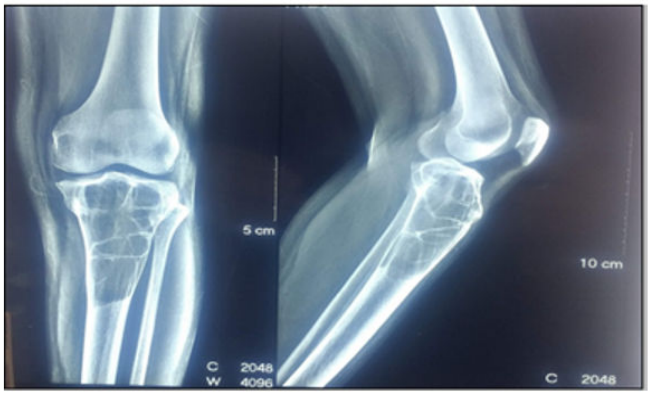


**Figure 1:** Plain x-ray of right lower leg showing expansile osteolytic lesion in proximal part of righttibia.

Based on radiological findings bone curettage was done and the specimen was sent for histological evaluation. On gross examination multiple yellow, friable, irregular bony fragments and soft tissue pieces measuring 9 × 5 × 3 cm in aggregate were seen. Representative sections were first processed but subsequently the specimen was submitted entirely [7,8]. Microscopic examination from bone and soft tissue revealed a lesion composed predominantly of abundant cholesterol clefts, hemosiderin laden macrophages. Foamy histiocytes (xanthoma cells) with central to eccentric nuclei and pink granular cytoplasm admixed with few multinucleated giant cells in a background of mixed inflammatory cells and fibrous tissue and at

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places foreign body giant cell reaction was noted. There was no significant pleomorphism and mitoses noted (Figures 2 and 3).

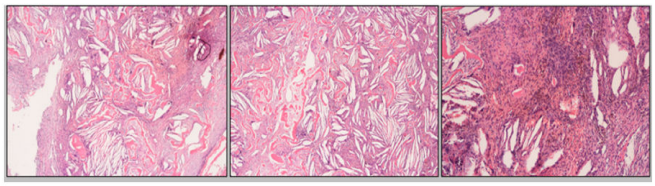

**Figure 2:** Photomicrograph hematoxylin and eosim of soft tissue showing abundant cholesterol clefts with sheets of foamy and hemosiderin leden macrophages and occasional gaint cells and inflammatory infiltrate.

Immunohistochemically CD68 was positive showing histiocytic origin. Other co-exisiting bone lesions could not be found after total processing of the specimen. The final diagnosis of prmary xanthoma of bone was rendered [9].

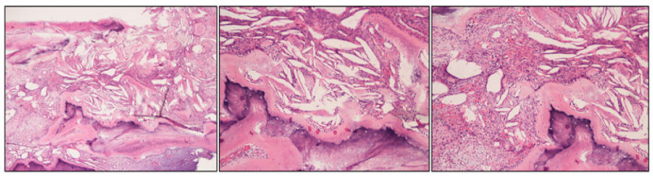

**Figure 3:** Photomicrograph hematoxylin and eosin of bone showing abundant cholesterol clefts with sheets of foamy hemosiderin laden macrophages and occasional gaintcells and inflammatory on filtrate.

## Results and Discussion

Xanthoma is a reactive proliferation and localized collection of lipid laden histiocytes. The most common site is skin and subcutaneous tissue but it also involves tendons and synovium. Intraosseous xanthomas are exceedingly rare bone lesion, particularly in normo lipidemic patients and occurs due to abnormal deposition of cholesterol in bone. Xanthoma of bone usually presents in patients above 20 years of age with male predominance. The main presenting complaint is pain [10]. It is always solitary. Xanthoma has been described in various intraosseous locations: Skull, mandible, femur, hand, radius, ulna, ribs, humerus, pelvis, sacrum and spine. The flat bones (skull, pelvis, ribs) are frequently involved. Normolipidemic xanthomas are either idiopathic or occur due to abnormal protein, lymphoproliferative disorders or they are post traumatic. Radiologically it appears as osteolytic lesion with cortical expansion or disruption and has a rim of sclerosis and it can mimic as metastatic bone deposits and primary bone tumors. Final diagnosis depend on histopathology [11]. Histologically intraosseous xanthomas are characterized by foamy and occasional non foamy macrophages. The lesion also has multinucleated giant cells occasionally Touton type, cholesterol clefts and fibrosis. Spindle cells are not a prominent feature of these lesions. Surgical excision of the lesion is elective treatment [12].

The histogenesis and classification of fibrohistiocytic lesions involving bone and soft tissue containing an admixture of fibrous tissue, foam cells and giant cells are confusing and include several overlapping entities including metaphyseal fibrous defect, non-ossifying fibroma, fibrous cortical defect, fibroxanthoma, xanthoma and benign fibrous histocytoma of bone. The differential diagnoses include both neoplastic and non neoplastic conditions which include bone involvement in sinus histiocytosis (Rosai-Dorfman disease), Erdheim Chester disease (a multisystemic granulomatosis), giant cell tumor of bone, eosinophilic granuloma, malignant fibrous histiocytoma, Clear cell metastasis and in children non-ossifying fibroma and fibrous cortical defect. More important is the differential diagnosis with metastatic clear cell carcinoma. In our case, systemic clinical examination, immunoreactivity to CD68 (histiocytic marker) helped exclude this condition and establish diagnosis of primary xanthoma of bone. The prognosis is satisfactory [13,14].

## Conclusion

This is a rare case of bone lesion with unsettled histiogenesis which can be misdiagnosed as other lesions. Histopathology plays a key role in diagnosis. It is very important to evaluate the entire specimen carefully to rule out primary bone tumors with xanthomatous degeneration and to confirm the diagnosis of primary xanthoma of bone as the clinicoradiologic features of skeletal xanthomas mimic bone tumors.

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