Case Report

Xanthogranulomatous osteomyelitis of femur masquerading as neoplasm

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ABSTRACT

Xanthogranulomatous osteomyelitis (XO) is a very rare chronic granulomatous inflammation. It can mimic malignant bone tumors on its clinical presentation, gross features, and radiological imaging. However, histopathological examination can differentiate it from malignancy. We describe the case of a 20-year-old male presented with fever and pain in the right knee joint for 4 months. Plain radiography and magnetic resonance imaging of the right knee joint revealed osteolytic lesion in the lower metaphyseal region of femur. With this clinical presentation and radiological imaging, a diagnosis of primary bone tumor was made. However, XO was confirmed by histopathological examination. This case highlights the rare occurrence and also it can mimic as bone tumor.

Key words: Bone tumor, femur, osteomyelitis, xanthogranulomatous inflammation

INTRODUCTION

Xanthogranulomatous reaction is a rare form of chronic inflammation, histologically consisting of collections of foamy histiocytes admixed with polymorphonuclear leukocytes, lymphocytes, and activated plasma cells.[1,2] This entity has been described in various organs; however, rarely, it can occur in organs such as lung, brain, and bone.[1,3] The presence of xanthogranulomatous inflammation in the bone is known as xanthogranulomatous osteomyelitis (XO). It can present mass-like lesion extending to adjacent structures and can mimic infiltrative carcinoma.[4] To the best of our knowledge, only 10 cases involving bone have been reported so far in literature.[1-3,5-9] We report the 11th case of XO with a brief review of literature.

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CASE REPORT

A 20-year-old male presented with fever, pain, and swelling in the right knee joint for 4 months. Pain aggravated on walking with no history of night pain/trauma. On local examination, tenderness was present in the lower end of right femur with no joint line tenderness and normal range of movements of knee. Systemic examination was normal. Complete blood count was within normal range. Serum parathormone and alkaline phosphatase were also normal. X-ray of right knee joint showed suspicious lytic lesion with sclerotic margin in the lower metaphyseal region of the femur [Figure 1a]. Magnetic resonance imaging of the right knee joint revealed hyperintense lesion in the lower metaphyseal region of the femur with well-defined sharp margin and minimal hyperintensity in the adjacent part of the femur [Figure 1b]. A clinical suspicion of primary bone tumor was entertained. The involved bone was curetted and specimen was sent for histopathological examination and also for culture.

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Grossly, the specimen was consisting of two gray brown bony tissue bits. Microscopically, there were sheets of foamy macrophages, exuberant lymphoplasmacytic inflammation along with dead bone [Figure 1c]. There was no evidence of malignancy or granuloma. Periodic acid–Schiff stain was positive for foamy macrophages [Figure 1d]. Ziehl–Neelsen stain for tissue of acid-fast bacilli was negative. Microbiological culture of the tissue sent at the time of surgery failed to reveal the growth of organisms after 48 h of aerobic incubation. The postoperative period was uneventful.

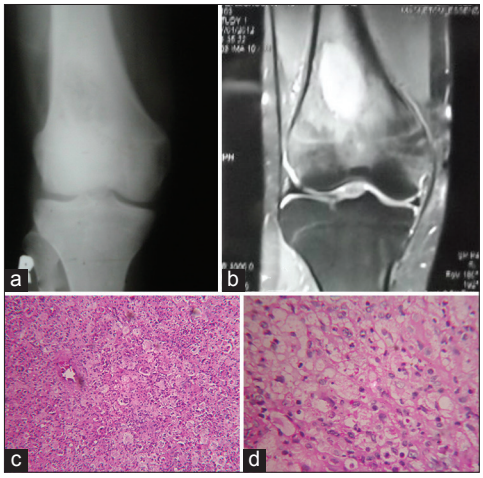

Figure 1: (a) X-ray of right knee joint showing suspicious lytic lesion with sclerotic margin in the lower metaphyseal region of femur. (b) Magnetic resonance imaging of the right knee joint revealing hyperintense lesion in the lower metaphyseal region of femur with well-defined sharp margin and minimal hyperintensity in the adjacent part of femur. (c) Photomicrograph showing sheets of foamy macrophages (H and E, ×100). (d) Photomicrograph showing foamy macrophages and chronic inflammatory cell infiltrate (Periodic acid–Schiff, ×400)

DISCUSSION

The first two cases of this entity were described by Cozzutto in 1984.[1] The previously reported cases of XO are summarized in Table 1. In most of the previously reported case reports, it was clinically as well as radiologically mimicking malignancy. Hence, histopathological conformation is mandatory for proper diagnosis and management of patients.

XO has to be differentiated from the following conditions to arrive at the correct diagnosis: Langerhans cell histiocytosis (LCH), Erdheim-Chester disease, xanthoma, storage disease, malakoplakia, and metastatic renal cell carcinoma.[1-3]

LCH microscopically consists of Langerhans cells having reniform nuclei and eosinophils infiltration. X-ray and computed tomography scan typically reveal circumscribed lytic bony lesions without surrounding sclerosis.[1,10,11]

Erdheim–Chester disease is a multifocal disorder with frequent involvement of extraskeletal tissues. Histology shows foamy histiocytes, cholesterol clefts, and fibrosis without neutrophilic infiltrations.[1]

Xanthoma can occur in bone, secondary to hyperlipidemia. However, it may not show suppurative inflammation.[1]

Malakoplakia is described in bone, kidney, and other organs, and more common in immunosuppressed individuals.

Table 1: Summary of previously reported cases and present case of xanthogranulomatous osteomyelitis

Case reports	Age (years)/sex	Site	Radiological features	Clinical and radiological diagnosis
Cozzutto[1]	5/male	First rib	X-ray: Irregular osteolytic lesion	Chronic osteomyelitis Ewing's sarcoma
Cozzutto[1]	14/male	Tibia	X-ray: Irregular, mottled radiolucency	Chronic infection
Vankalakunti *et al.*[3]	50/female	Ulna	X-ray: Ill-defined osteolytic lesion	Bone tumor
Cennimo *et al.*[5]	41/male	Index finger	X-ray: Soft-tissue swelling MRI: Abscess collection and synovial enhancement	*Mycobacterium* infection
Borjian *et al.*[2]	14/male	Humerus, fibula	X-ray: Periosteal reaction and cortical disruption CT: Periosteal reaction and bone marrow infiltration MRI: Signal abnormalities	Malignancy Osteomyelitis
Kamat *et al.*[6]	13/male	Tibia	X-ray: Submetaphyseal lytic lesion with sclerotic margin	Brodie's abscess
Lee *et al.*[7]	59/male	Wrist	X-ray: Osteolytic lesion CT: Osteolytic lesion MRI: soft-tissue mass with lobulated margins PET-CT: Intense uptake	Bone tumor Metastasis
Rathi *et al.*[8]	50/male	Tibia	X-ray: Soft-tissue mass, periosteal reaction	*Mycobacterium tuberculosis*
Wang *et al.*[9]	45/male	Rib	X-ray: Osteolytic lesion CT: Osteolytic lesion	Bone tumor
Wang *et al.*[9]	46/male	Rib	X-ray: Osteolytic lesion CT: Osteolytic lesion	Bone tumor
Our case report	20/male	Femur	X-ray: Suspicious lytic lesion with sclerotic margin MRI: Hyperintense lesion with well-defined sharp margin	Bone tumor

MRI: Magnetic resonance imaging, CT: Computed tomography, PET: Positron emission tomography

Histologically, it shows large foamy macrophages and Michaelis–Gutmann bodies, which was not seen in our case.[12]

In case of storage diseases, clinical background, foamy macrophages within the bone marrow, and absence of suppurative component can differentiate the XO.[1]

Metastatic renal cell carcinoma can be easily differentiated by history and histologic tumor patterns.[1]

CONCLUSION

We present this case primarily due to its rarity, curability, and importantly, it can mimic primary or secondary tumor on clinical, radiological, and gross examination. Hence, radiologists must be aware of the possibility of the XO, and all the cases require histopathological confirmation for proper management. Currently, histopathological examination is most specific for the diagnosis of XO.

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Conflicts of interest
There are no conflicts of interest.

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