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Xanthogranulomatous Osteomyelitis: Two Rare Cases Report

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Abstract

Background: Xanthogranulomatous osteomyelitis (XO) is a rare chronic inflammatory lesion that is histologically characterized by the presence of foamy histiocytes and plasma cells. Radiologic and gross examinations can mimic malignancy (9), so carful workup and definitive diagnosis should be made by histopathologic evaluation.

Case Report: There are two rare cases being reported here; the first one is a 21-year-old male who presented with chronic pain and swelling of upper leg for one year. He has history of healed traumatic fracture of fibula 7 years ago. Radiological examination revealed radio opaque lesions of upper fibula. The second case is a 30 years old female, presented with pain and swelling of wrist for six months with clinical suspicion of Ewing sarcoma. Radiological examination revealed highly suspicious radio opaque, lytic lesions of lower radius. Biopsy of both lesions were done. Microscopic examination showed marked chronic inflammatory cells infiltration mainly foamy histiocytes and lymphoid cells with many foreign body giant cells. No evidence of tuberculosis or malignancy.

Conclusion: As these lesions were clinically highly suspicious of malignancy or associated with other diseases, so proper diagnostic roles in xanthogranulomatous osteomyelitis should include histopathological examination in order to rule out any malignant conditions of the bone.

Key words*:* Xanthogranulomatous Osteomyelitis, Chronic Inflammatory, Lesion.

INTRODUCTION

Xanthogranulomatous osteomyelitis is a rare form of chronic inflammatory disease confirmed microscopically with characteristic features of foamy histiocytes, neutrophils, lymphocytes, activated plasma cells and many multinucleated giant cells (1-8). It mimic malignancy both by radiological and gross examination (9). Fibroblastic cells proliferation may exist and forming considerable fibrosis in addition to occasional clefts of cholesterol (1,10,13,15).

Many bones are prone for development of xanthoma, particularly upper and lower extremities as humerus, femur, tibia, radius and ulna are frequently affected bones (16-20), while calcaneus and vertebral bodies and spines were less frequent (1,6). Interesting fibula is less likely to be affected by xanthoma

(17). Most xanthomas are lytic lesions, but intraosseous xanthomas are osteolytic expansile lesions and often associated with hyperlipidemia (20-22).

CASE REPORT

Case No.1. A 21-year-old male presented to orthopedic doctor with painful swelling of upper leg, he has history of trauma and fracture of fibula 7 years ago. No history of tuberculosis, diabetes, and other disease. Radiological examination revealed lytic expansile mass of upper fibula (Fig.1).

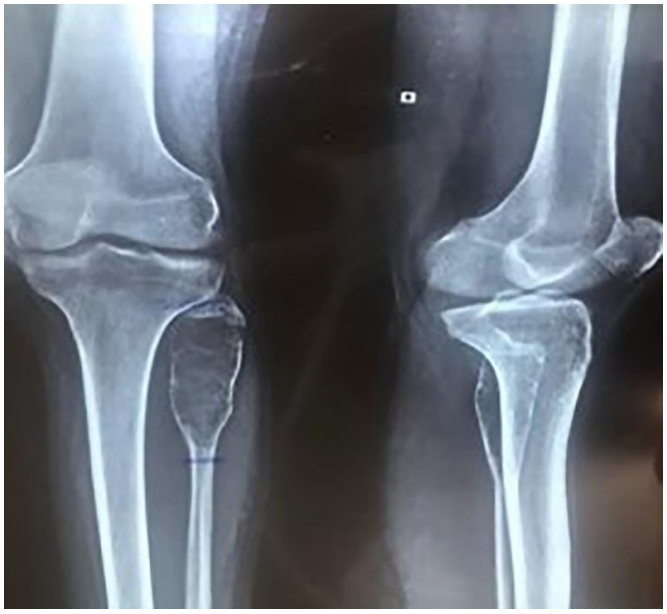

Fig.1: radiological image of left leg bones showed expansile lytic lesion of upper fibula.

Gross feature showed one piece of bone, covered by cartilage cup, measured 8x3 cm, several pieces were taken in three blocks. Microscopic features of bone lesion sections revealed abundant benign looking distorted osteoid tissue, showed marked chronic inflammatory cells infiltration mainly foamy histiocytes and lymphoid cells with many foreign body giant cells. No evidence of tuberculosis or malignancy (Fig.2).

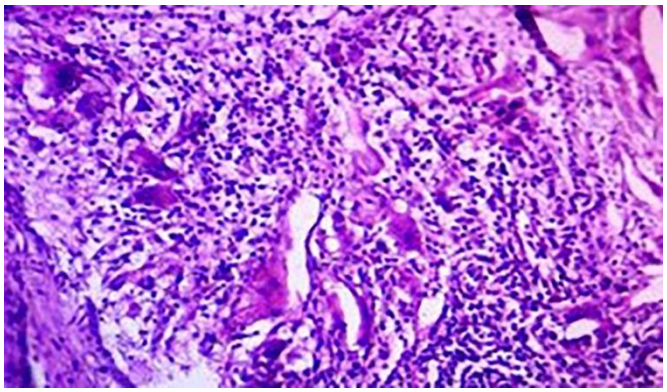

Fig.2: Histological section revealed marked chronic inflammatory cells infiltration mainly foamy histiocytes and lymphoid cells with many foreign body giant cells.

Case No.2. A 30 years old female presented to orthopedic doctor with painful swelling of left wrist for six months leg, without history of trauma or fracture. Radiological examination revealed lytic lesion of lower end of radius, with suspicion of Ewing sarcoma. No clinical evidence of any associated disease.

DISCUSSION

Xanthogranulomatous osteomyelitis is a rare disease firstly described and applied as a term in 1984 by C Cozzutto (22). Histologically, the lesion is composed from histiocytic and plasma cells infiltration in bone lesions (5). Till now little number of cases has been reported worldwide (Table.1). Only **20** cases have been recorded till writing this paper. Any age can be affected and most of them were ranging 5 years to 65 years (tabl.1). Both sexed were affected. Most of them were presented with painful swelling lesions preceded by traumatic fracture (18-20), while few were associated with other lesion as Crohn's disease (3), Xanthogranulomatous pyelonephritis (21) and genetic disorder (7). Some of cases mimic neoplastic lesions and give high suspicion of malignancy (10,13,15), while other was resembling tuberculosis (4). Most of reported cases were presented as unifocal lesions (1,5,15,18,22), while only 4 cases were multifocal (9,19,17,21). Long bones as tibia, femur and fibula were most frequently affected while rib and spine were rarely affected (1,12, 18).

In the presented two cases, both were presented with painful bone swelling. The first one was 21 years old with history of trauma accident 7 years ago, X ray revealed expansile mass of upper fibula, highly suspicious of bone tumor. So surgical resection was done with safe margin. Other case was 30 years female presented with painful swelling of the wrist, x ray revealed lytic lesion of radius, with high clinical suspicion of malignant tumor (Ewing tumor), curate biopsy was done. Both cases were not associated with any systemic disease. They were confirmed histologically as Xanthogranulomatous osteomyelitis. So the importance of this disease is enrolled in its clinical suspicion of malignancy that necessitates a special care and carful approach for diagnosis.

Conclusion

Owing to its rarity and clinical suspicion of malignancy, we present these two cases of Xanthogranulomatous osteomyelitis for documentation and making attention of surgical doctors and pathologists to take in consideration this disease entity for planning for proper management and to avoid misdiagnosis and unnecessary major surgical resection.

Table.1. The reported cases of Xanthogranulomatous osteomyelitis worldwide in sequence of dating.

Ref. N0.	D	S	N c s	A (	C f	E i	A d	L
21 .	1	1 d e	2	N	P	N b	, a f	N a
20	1	N	1	6	F p	T	T	S
19	2	F	1	5	P	U	-	S
18	2	N	1	1	P	T	-	S

					s			
16	2	M	1	1	P f	H a	-	N a
15.	2	M	1	5	P S	U	-	S
14	2	M	2	N	P	F	-	S
13	2	M	1	2	P	T	-	s
12	2	F	1	6	P	F	-	S
11	2	M	1	3	P	M ta c	-	N a
10	2	F	1	2	P	F a	-	N a
9	2	M	1	2	P	F	tr	S
8	2	F	1	3	P	T	-	S
7	2	F	1	5	P f	h	A s	N a
6	2	m	1	3	P	f	tr	S
5	2	M	1	1	P	T	T	
4	2	F	1	5	P	F	-	S
3	2	M	1	2	P	P	-	S
2	2	F	1	2	P	f b	-	S
1	2	F	1	6	S c c o	v	-	S

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