CASE REPORT

Xanthogranulomatous Mastitis: Report of a Rare Case and Review of Literature

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ABSTRACT

Background: Xanthogranulomatous inflammation (XGI) is a rare form of chronic inflammation characterized by the presence of lipid-laden macrophages, multinucleated giant cells, and cholesterol crystals. It is an uncommon finding in the breast. Our literature search revealed twenty-four documented cases in the English literature.

Case report: An 18-year-old female presented with a mass in the left upper lateral quadrant of the breast with ulceration for a 1-month duration. The mass was initially small about 2cm in its widest diameter and continued to increase in size to 5 x 3 cm. The overlying skin is smooth and not attached to the underlying structures. It is firm with mild tenderness and ill-defined margins. Two months later she developed a left breast ulcer that measured 2 x 1 cm on the surface of the lump. It has a sloping edge with a necrotic floor. There was no associated axillary lymphadenopathy. Fine needle aspiration cytology suggested an inflammatory process. Tissue biopsy histology shows abundant foam cells, multinucleated giant cells of foreign body type as well as numerous lymphoplasma cells infiltrating the breast tissue with geographic areas of necrosis in keeping with xanthogranulomatous mastitis.

Conclusion: Xanthogranulomatous mastitis is a very rare entity, it is self-limiting in most cases. The diagnosis of XGM can be made by excluding other diseases that elicit inflammation in the breast. The lesion can mimic granular cell tumor, histiocytoid carcinoma, invasive carcinoma NST, and lipid-rich breast cancer; therefore, clinicians should always consider these as differential diagnoses to avoid unnecessary surgery.

Keywords: Xanthogranuloma, Mastitis, Rare

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Introduction

Xanthogranulomatous inflammation (XGI) is a rare chronic inflammation characterized by the presence of lipid-laden macrophages, multinucleated giant cells and cholesterol crystals.[1] The lesion is a stable or regressing inflammatory histiocytic lesion that usually occurs during childhood or adolescence.[2]

Xanthogranulomatous inflammation has been reported almost across all organ systems and spans a vast clinicopathological spectrum.

Xanthogranulomatous pyelonephritis (XGP) and xanthogranulomatous cholecystitis (XGC) are the most common because of their higher incidence and the high propensity to be symptomatic.

There are two types of XGI, the juvenile and adult type.

Most of the juvenile lesions are found on the head and neck region where it undergoes spontaneous regression within months to years, however, the adult forms constitute 15 to 30% of XG. It appears mostly as a solitary lesion and it tends to persist.[3,4]

Xanthogranolomatous mastitis (XGM) is a very rare entity that was first described by Shin et al in 2005, Hwang et al reported bilateral xanthogranulomatous mastitis in 2007.[3,4]

The presence of the lesion on the breast is extremely rare with a large gap in the diagnosis and treatment of the disease. It may masquerade as invasive carcinoma NST leading to gratuitous surgery. This prompted the need for this case report.

We present an 18-year-old female who was clinically diagnosed of benign left breast neoplasm and has had lumpectomy to rule out cancer and the histopathology, confirmed XGM.

Case Presentation:

An 18-year-old female presented with an abnormal mass on the left upper lateral quadrant of the breast with ulceration of 1-month duration. The mass was initially small about 2 cm in its widest diameter and continued to increase in size to 5x3 cm. The overlying skin was smooth and not attached to the underlying structures. It is firm with mild tenderness and ill-defined margins. Two months later, she developed left breast ulcer that measured 2 x 1 cm on the surface of the lump. It has a sloping edge with necrotic floor. There was no axillary lymphadenopathy seen. Fine needle aspiration cytology was requested and the result shows foamy macrophages along with background lymphocytes and plasma cells suggestive of an inflammatory process. Tissue biopsy histology shows abundant foam cells, multinucleated giant cells of foreign body type as well as numerous lymphoplasma cells infiltrating the breast tissue with geographic areas of necrosis in keeping with xanthogranulomatous mastitis (Figure 1). Ziehl-Neelsen stain done was negative. Verbal consent was obtained from the patient before the report of the case.

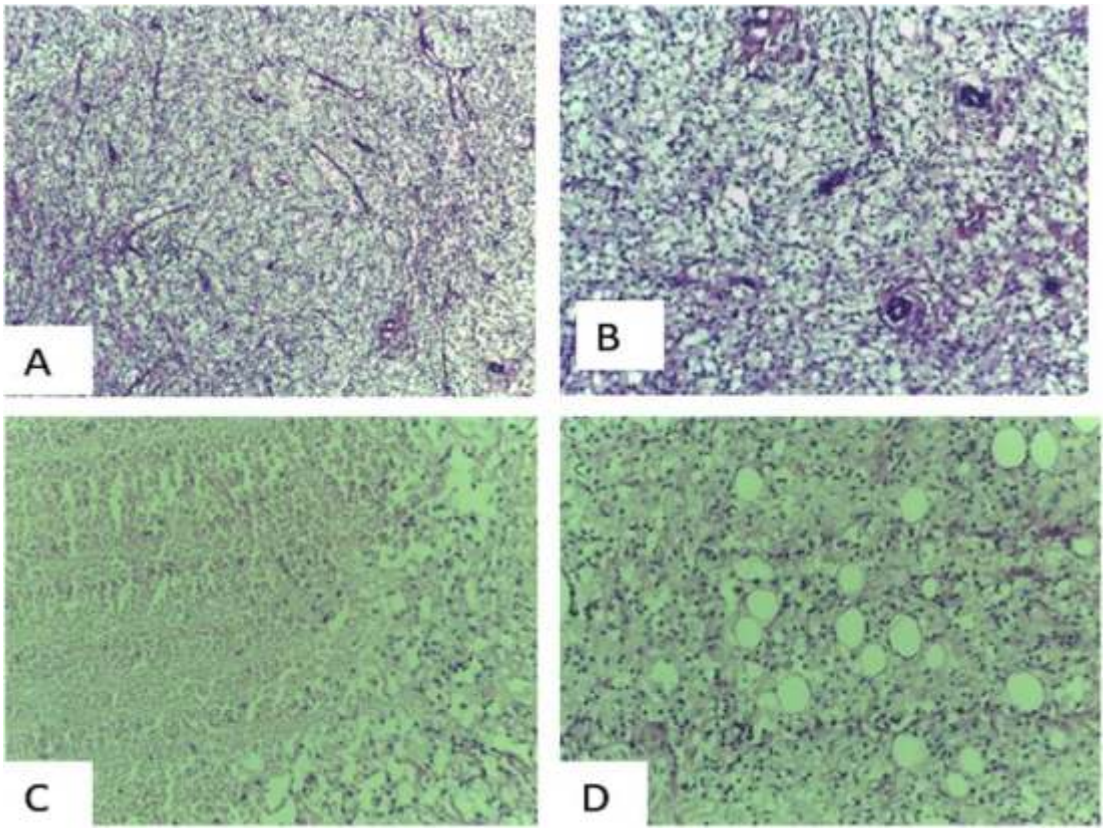

Figure 1: Photomicrograph of xanthogranulomatous mastitis showing sheets of foamy macrophages (a,b), areas of necro-inflammation (c) and (d) adipose tissue infiltrated by histiocytes

Table 1: Literature review of XGM

S/NO.	Authors/ Year	Age(Yrs)	Sides of breast lump	Size (cm)	Clinical symptoms	Radiological findings	Diagnosis	Past medical history	Other relevant findings
1	Hwang et al 2007	60	Bilateral	3.0		BI-RADS 4b	XGM		
2	Koo et al 2009 (case series)	46	Rt	1.2		BI-RADS3	XGM		
		44	Lt	1.5		BI-RADS3	XGM		
		46	Lt	1.0		BI-RADS3	XGM	Breast cancer surgery	
		47	Lt	1.2		BI-RADS3	XGM		
		37	Rt	1.5		BI-RADS3	XGM		
		46	Rt	1.5		BI-RADS3	XGM		
		53	Lt	2.0		BI-RAD2	XGM	Breast cancer surgery	
		70	Bilateral	3.0	Palpable lumps	BI-RADS4b			
		37	Lt	0.8		BI-RADS 4a			
		55	Lt	0.5		BI-RADS 4a			
		36	Rt	1.5		BI-RADS 4a			
		36	Lt	1.8		BI-RADS 4a			
		26	Rt	4.8	Palpable lumps	BI-RADS 3			
		44	Lt	1.2		BI-RADS 5			
		67	Rt	1.3		BI-RADS 3			
		42	Lt	0.5		BI-RADS 3			
2	Hussain et al 2012	56	Lt	10	Palpable lumps		XGM		
3	Jyoti et at 2014	19							
4	Dinets et al 2016	30	Lt	4 x 5					

5	Bananikar et al 2017	42	Lt	4	Palpable lumps	BI-RADS3	XGM with intraductal papilloma
6	Oliveira et al 2017	47	Bilateral	6.8	Palpable mass	BI-RADS 4c	XGM
7	Leong et al 2018	41	Lt				
8	Zahid et al 2021	92	Rt	5	Palpable lumps	BI-RADS 4	XGM

Discussion:

Xanthogranulomatous inflammation is a rare chronic inflammation characterized by the presence of lipid-laden macrophages, multinucleated giant cells and cholesterol crystals.[1]

Although xanthogranulomatous inflammation has been reported almost across all organ systems and spans a vast clinicopathological spectrum.

In contrast, the rarity of Xanthogranolomatous mastitis (XGM) is most likely explained by a combination of truly low occurrence and under-detection due to its frequent asymptomatic and self-limited course in the majority of cases. Generally, XG inflammation occurred at birth in 1/5th of patients and 2/3rd of the patients develop the disease by 6 months of age while 20-40% of patients have the lesion after the age of 20 years 3 with a mean age of 44 years. [4]

The pathogenesis of XGM is unknown, but risk factors such as duct obstruction complicated by infection with low-virulence organisms, defective lipid transport, autoimmune disorders and allergic reactions have been reported.[3,4]

The clinical presentation of XGM is variable, however, most patients present with breast lumps which can mimic benign or malignant breast tumours.[4]

The index patient presented with a lump on the left upper lateral quadrant with mild tenderness which later ulcerated.

There was no history of trauma, lactation, or breast surgery prior to presentation which is similar to some reported cases. Koo et al (2009) in a case series reported some patients with a positive history of prior breast surgery while others had no such history.

Although the lesion is an inflammatory process, mimickers include granular cell tumor, histiocytoid carcinoma and lipid-rich breast cancer. Immunohistochemistry may be required to differentiate these lesions.

A comprehensive search through multiple search engines (Pubmed, Embase, Medline, Google scholar, Research-gate) from 1990 to February 2022, 24 cases were reported in the English literature as summarized (Table1).

xanthogranulomatous mastitis can affect both the young and the elderly, our review showed a median age of 30 years (age range 19 to 92).

Most of the cases present as a palpable lump, and the association with duct ectasia is a common finding. Some patients had a prior history of breast surgery.

Radiologically majority of cases show BIRADs 3 to 5 mammographic features. The finding of associated malignancy is not uncommon

Conclusion:

Xanthogranolomatous mastitis is a very rare entity. Although in most cases it is self-limiting, the

Diagnosis of xanthogranulomatous mastitis can be made by excluding other diseases that elicit xanthogranulomatous inflammation in breast.

The lesion can mimick granular cell tumor, histiocytoid carcinoma and lipid-rich breast cancer, therefore clinicians should always consider these as differential diagnoses.

The disease is associated with obstruction and rapture of ectatic ducts; therefore, clinicians should always consider this as a differential diagnosis to avoid unnecessary surgery.

References:

1. Koo JS, Jung W. Xanthogranulomatous Mastitis: Clinicopathology and Pathological Implications. Pathol. Int 2009; 59:234-240.
2. Bamanikar S A, Chandanwale S S, Pathak P, Gambhir A, Sheth J. A Rare Case of Xanthogranulomatous Mastitis with Intraductal papilloma. Med J DY PatilVidyapeeth 2018; 11:348-51.
3. Tasadooq H, Bilal E, Ervive L, Tapan M, Penelope LM, Peter JK. Xanthogranulomatous inflammation involving latissimus dorsi donor site and implant breast reconstruction: case report and literature review. World J SurgOncol 2012; 10:166.
4. J R. Goldblum, A L. Folpe, Sharon W. Weiss. Enzinger and Weiss's Soft Tissue Tumors 7th ed. Philadelphia: Elsevier 2020.
5. Hwang SH, Son EJ, Oh KK, Kim EK, Jung J, Jung WH. Bilateral Xanthogranuloma of the Breast. J Ultrasound Med 2007; 26:535-537.
6. Zabetian S, Friedman J B, Mchuargue C. A case of idiopathic granulomatous mastitis associated with erythema nodosum, arthritis and cough. JAAD case report 2016;2(2):125-127.
7. Nayak S, Acharjya b, Devi B,Patra M K. Juvinilexanthogranuloma in adult. Indian J.Dermatol 2009; 54(5):83-85.
8. Sikandra B, Qureshi M A, Avesi L, Sheraz N, Mirza T. Histopathological spectrum of breast disease in a multi-ethnic population of Karachi, Pakistan. RMJ.2015;40(1):51-58
9. Adam E G, Kemp J D, Holcomb K Z, Sperling LC. Xanthogranulomatous reaction to a raptured galactocele. J. Cutan.Pathol.2010;37(9): 973-976.
10. Li J, McGregor P H. Idiopathic granulomatous mastitis associated with hyperprolactinemia. Breast J 2017;23
11. Kapoor H, Zhang Y, Qasem S A, Owen W, Szabunio M M. Xantho-granulomatous mastitis preceded by cysts on ultrasound: Two cases with review of literature. Clinical Imaging 2021; 78:64–68.
12. Pluguez-Turull CW, Nanyes JE, Quintero CJ, et al. Idiopathic granulomatous mastitis manifestations at multimodality imaging and pitfalls. Radiographics 2018; 38:330–356.
13. de Oliveira I J, Viana C R, Sabino S M P d-S, Kerr L M, Vieira R A d-C. Xanthogranulomatous Mastitis Mimicking Locally Advanced Breast Cancer. The Breast Journal 2016; 1–3.

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