

XANTHOGRANULOMATOSIS - MASQUERADING MALIGNANCY OF THE BREAST RECONSTRUCTED WITH LICAP FLAP

Oncology

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ABSTRACT

Breast cancer is the most prevalent malignancy among women in India, accounting for 26.6% of cases according to GLOBOCAN 2022. As a result, the presentation of benign breast masses in elderly women poses a diagnostic challenge for oncologists. Xanthogranulomatous inflammation is a benign pathological condition considered to be a primary disease process of unknown etiology. Xanthogranulomatous mastitis affecting the breast is very rare. Accurate diagnosis is crucial to minimize morbidity by preventing the need for multimodal therapy and to enable cosmetically acceptable reconstruction after resection when indicated. In our case report, we are presenting an elderly women with a large breast mass who underwent total mastectomy with LICAP reconstruction

KEYWORDS

Mastitis, Granulomatous, LICAP, Mastectomy

INTRODUCTION:

Xanthogranulomatous inflammation is a benign pathological condition associated with rare form of chronic inflammation[1]. It involves various organ systems such as the gall bladder, kidney, pancreas, appendix, eyes, female genital tracts, colon, and urachus [2]. Breast involvement seen with this inflammation has been limited to only few rare cases of mastitis. We would like to report an unusual involvement of this inflammatory process presenting as a solid lump resembling a malignancy.

They are characterized by eosinophilic, granular, periodic acid-Schiff (PAS)-positive histiocytes in initial stages, followed by foamy macrophages, activated plasma cells, suppurative foci, and hemorrhages [3]. Eventually, these lesions and neoplasms become fibrotic. Recently, epithelial tumors have been identified in soft tissue and bone in six cases that presented with features of xanthogranulomatous inflammation. These cases involved young patients, and the tumors behaved like low-grade neoplasms[4]. Very few reports have been published in literature on xanthogranulomatous mastitis.

CASE PRESENTATION:

A 62- year old female presented to our institute outpatient department with a large mass in the right breast for 1 month duration. The mass was initially small and then increased to present size. She had no comorbidities. On examination entire right breast was replaced by the tumour and had intrinsic mobility(Figure-1).



Figure- 1: Clinical picture of a 62yrs old postmenopausal women with right breast lump

No palpable axillary nodes. Initial investigation with xray mammogram and ultrasound revealed a large well circumscribed equal density lesion of size 11 x 9 cm replacing the entire right breast. Internal echoes were noted with no internal vascularity (BIRADS – 3)(Figure-2).

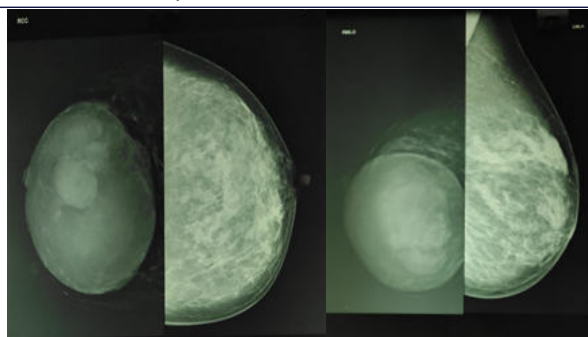


Figure- 2: Xray mammogram – CC & MLO view of both breast showing right breast lump

Trucut biopsy showed multinucleated giant cells and histiocytes suggestive of xanthogranulomatous mastitis and no evidence of malignancy. Immunohistochemistry performed was positive for GATA3, CD 68. After pre-operative work up and preparation patient underwent a Simple mastectomy with LICAP flap reconstruction(Figure-3).



Figure- 3: Intraoperative picture of total mastectomy with LICAP reconstruction

Gross morphology showed 17 x 12 x 6cm tumour weighing 631gm. Cut surface shows haemorrhagic fluid with thick cyst wall. Microscopy revealed cyst wall with dense chronic inflammatory infiltrate with collection of foamy histiocytes, granulomas and cholesterol clefts. Multinucleated giant cells with hemosiderin laden macrophages were also noted favouring xanthogranulomatous mastitis (Figure-4).

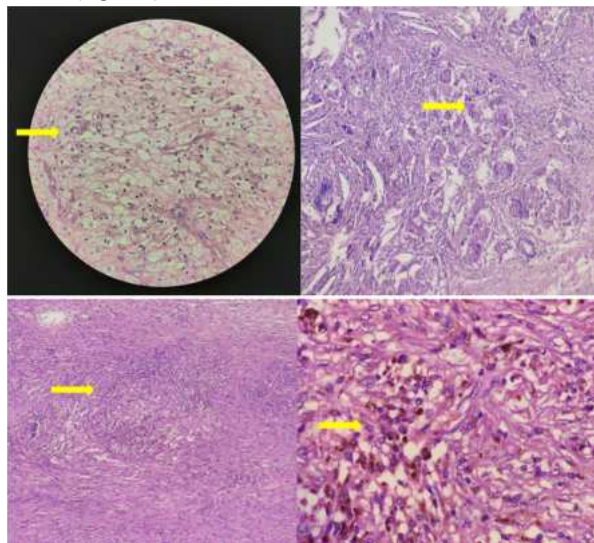


Figure- 4: a) Photomicrograph showing foamy histiocytes
b) H & E stain showing multinucleated giant cells
c) Photomicrograph showing granuloma
d) H & E stain depicting hemosiderin laden macrophages

DISCUSSION:

Xanthogranulomatous inflammation is a benign pathological condition considered to be a primary disease process of unknown etiology. Xanthogranulomatous mastitis affecting the breast is very rare, commonly affecting the kidney and gas bladder [5]. The exact etiology of the disease is unknown; organ obstruction, suppurative infections and hemorrhage trigger tissue damage within the involved organs, usually eliciting a microscopic response of the disease process. First case of mammary presentation of juvenile xanthogranuloma was described by Shin et al in 2005 [6].

Koo and Jung suggested that the cause of xanthogranulomatous mastitis can be obstruction and rupture of ectatic duct with foamy histiocyte aggregates. Immunohistochemistry shows these histiocytes positive for CD68 and negative for cytokeratin [7]. The diagnosis for this disease represents a challenge for the physician, as the breast sonography findings are non-specific. The typical appearance describes an irregular mass, heterogeneous, hypoechoic with posterior acoustic enhancement. On the other hand, mammography will describe a mass suspicious of malignancy, with heterogeneous composition, non-well defined borders that may even present spiculations. Kok KY et al described the insufficient diagnostic value that the radiologic studies have in order to differentiate this entity from breast cancer, because of the radiologic similarity that both entities present although breast ultrasound has demonstrated to be a useful resource in patients with Xanthogranulomatous mastitis accompanied by abscesses [8]. Xanthogranulomatous inflammation in breast can occur in cancer regression due to neoadjuvant chemotherapy, as mammary involvement in Erdheim-Chester disease and malakoplakia. Cancer regression in neoadjuvant chemotherapy is characterized by histiocytes, elastosis, inflammation, and fibrosis which is like findings in xanthogranulomatous mastitis and can be differentiated by chemotherapy history. Recently, it has been expressed that infectious processes sustained by Gram-positive and Gram-negative bacteria can affect the soft tissue and appear as xanthogranuloma [9].

Chronic inflammatory lesions are known to produce firm lesions which may mimic neoplastic lesion. In our case, definitive diagnosis could not be achieved in FNA smears due to the absence of adequate number of xanthoma cells. Hence a trucut biopsy was performed and Final diagnosis was made on histopathology examination. Xanthogranulomatous inflammation is characterized microscopically

by the presence of multinucleated giant cells, lipid-laden macrophages (xanthoma cells), and cholesterol crystals. It also shows accumulation of cells including lymphocytes, plasma cells, and some polymorphonuclear leukocytes. The most typical findings in xanthogranulomatous inflammation are interlacing of foamy macrophages and activated plasma cells [10].

Recommended treatment of solid lumps with diffuse involvement is usually surgical as this condition is not pre-malignant. A complete surgical excision negates the need for any further treatment. There is no evidence in the literature to support any recurrence in the native or other organs following the primary surgical treatment.

CONCLUSION:

Xanthogranulomatous mastitis is an extremely rare entity especially in elderly population. Clinical presentation is quite variable but recognizing the features of xanthogranulomatous inflammation may have a significant impact on the management of the patient. Imaging modalities and FNAC are inadequate for the diagnosis. Histopathological examination is essential for definitive diagnosis.

This case report brings to light the findings of a probable xanthogranulomatous tumor in breast tissue. Although most xanthogranulomatous reactions represent benign processes, more aggressive forms can present as seen in our case. Key to diagnosis is essential to reduce the morbidity by avoiding multi-modality therapy as in malignant cases and to perform a cosmetically acceptable reconstruction after resection when indicated. Due to the rarity of xanthogranulomatous tumors in the breast, prognosis and a standardized treatment including further surveillance and follow-up have yet to be established.

REFERENCES:

- Hussain T, Elahi B, Long E, Mahapatra T, McManus PL, Kneeshaw PJ, et al. Xanthogranulomatous inflammation involving latissimus dorsi donor site and implant breast reconstruction: Case report and literature review. *World J Surg Oncol* 2012;10:166.
- Kansakar PB, Rodrigues G, Khan SA: Xanthogranulomatous cholecystitis: a clinicopathological study from a tertiary care health institution. *Kathmandu Univ Med J (KUMJ)* 2008, 6:472-475.
- Cozzutto C, Carbone A: The xanthogranulomatous process. *Xanthogranulomatous inflammation. Pathol Res Pract*. 1988, 183:395-402.
- Fritchie KJ, Torres-Mora J, Inwards C, et al.: Xanthogranulomatous epithelial tumor: report of 6 cases of a novel, potentially deceptive lesion with a predilection for young women. *Mod Pathol*. 2020, 33:1889-95.
- Franco V, Aragona F, Genova G, Florena AM, Stella M, Campesi G, et al. Xanthogranulomatous cholecystitis. *Histopathological study and classification. Pathol Res Pract* 1990;186:383-90.
- Shin SJ, Scamman W, Gopalan A, Rosen PP. Mammary presentation of adult-type "juvenile" xanthogranuloma. *Am J Surg Pathol* 2005;29:827-31.
- Koo JS, Jung W: Xanthogranulomatous mastitis: clinicopathology and pathological implications. *Pathol Int* 2009, 59:234-240.
- Kok KY, Telisingshe PU. Granulomatous mastitis: presentation, treatment and outcome in 43 patients. *Surgeon*. 2010;8(4):197-201.
- Barnes PJ, Foyle A, Haché KA, Langley RG, Burrell S, Juskevicius R, et al. Erdheim-Chester disease of the breast: A case report and review of the literature. *Breast J* 2005;11:462-7.
- Bourm KS, Menias CO, Ali K, Alhalabi K, Elsayes KM. Spectrum of xanthogranulomatous processes in the abdomen and pelvis: A Pictorial review of infectious, inflammatory, and proliferative responses. *AJR Am J Roentgenol* 2017;208:475-84.