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Idiopathic Granulomatous Lobular Mastitis Masquerading as a Breast Tumor: A Case Report

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Abstract

Introduction: Idiopathic granulomatous lobular mastitis (IGLM) is an inflammatory disease of the breast with an obscure etiology. It occurs mainly in women of reproductive age, and the lesion mimics carcinoma of the breast both clinically and radiologically
Case Presentation: We present the case of a 29-year-old female who visited our hospital in Kancheepuram, Tamil Nadu, with a 4 × 3 cm lump in the upper outer quadrant of her left breast. The clinical and radiological findings were indicative of a malignant lesion; however, fine-needle aspiration cytology (FNAC) revealed features of granulomatous mastitis, and the subsequent histology of the excised lump confirmed the diagnosis of IGLM.
Conclusions: IGLM should be considered as one of the differential diagnoses when granulomas are encountered in breast FNAC and biopsy. A definitive diagnosis of IGLM can be made by identifying its characteristic histomorphology and ruling out other causes for granulomatous inflammation. An exact diagnosis is essential since the treatment for different granulomatous conditions of the breast varies.

Keywords: Granulomatous Mastitis, Mastitis, Fine Needle Aspiration Biopsy

1. Introduction

Idiopathic granulomatous lobular mastitis (IGLM), which was first reported in 1972 by Kessler and Walloch, is a rare chronic granulomatous inflammatory lesion of the breast lobules with an unknown etiology (1). It is commonly seen in women of reproductive age with a mean age at presentation of 34 years (1). Clinically these patients present with a painful, unilateral breast lump, but cases with bilateral presentations have also been reported (2). Some patients also have associated skin changes, lymphadenopathy, nipple discharge, ulcers, and draining sinuses, thus mimicking malignancy. Rare presentations in the male breast have also been reported (3). An exact diagnosis is key before initiating suitable treatment to avoid unnecessary radical surgeries. Along with a literature review, we present a case of IGLM that masqueraded as a breast tumor in a non-lactating young woman.

2. Case Presentation

A 29-year-old female presented to our hospital (Shri Sathya Sai Medical College and Research Institute, Kanchipuram, Tamil Nadu) with a painful breast lump measuring 4 × 3 cm in the left upper outer quadrant that had been for the past 6 months. The patient was neither pregnant nor lactating. There were no other relevant clinical findings. The mass was tender and firm in consistency with restricted mobility. Mammography showed a 3.5 × 3 cm mass with an ill-defined margin that was classified as BI-RADS category 3. Because of the clinical suspicion of breast cancer, fine-needle aspiration cytology (FNAC) was done.

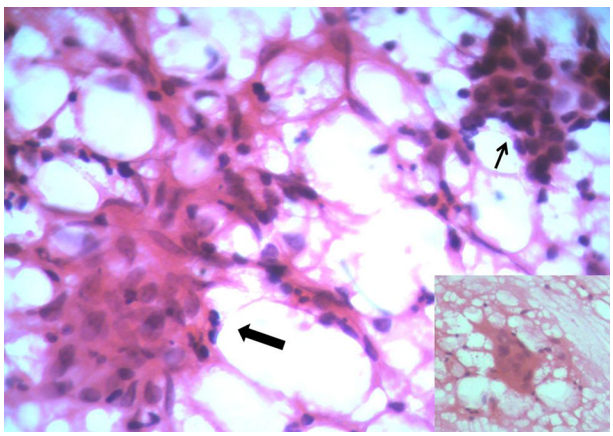
The FNAC smears showed a few cohesive clusters of ductal epithelial cells along with a focal collection of epithelioid histiocytes, scattered polymorphs, and lymphocytes in the background (Figure 1). The acid fast bacilli (AFB) smear was negative. Cytologically, a diagnosis of granulomatous mastitis was given, and the mass was subsequently surgically excised and sent for histopathological examination. Grossly, the mass was well circumscribed, firm to hard with a nodular surface. The cut surface color ranged from gray to tan with focal yellowish areas (Figure 2).

Microscopy showed granulomatous inflammation centered around the lobules, which were composed of epithelioid histiocytes and Langhans giant cells, admixed with neutrophils, lymphocytes, and plasma cells. A clear space rimmed by a thin band of neutrophils was seen at the center of the granulomas (Figures 3 and 4). No asteroid bodies or Schaumann bodies were identified. Lactational change was not seen in the surrounding lobules. The

Table 1. Summary of IGLM Cases in Literature

Study Group	NO OF CASES	Sex	(Mean) Age	Clinical Presentation	Other Findings
Gurleyik et al. (1)	19	F	34	Large irregular painful mass in 15 cases, 3 cases had ulcerative skin lesion, 2 had axillary lymphadenopathy.	Eight patients had history of contraceptive pill usage.
Altintoprak Fet al. (2)	26	F	37.5	Pain, swelling, and inflammation on the affected breast, along with superficial erosion or open fistulae on the breast skin	Three patient had history of oral contraceptive use and six had history of smoking
Reddy KM et al. (3)	Case report	M		Swelling and pain	Nil
Bakaris et al. (4)	Case report	F	27	Breast mass, nipple discharge and nipple retraction	History of breast cancer in the family
Ergin AB et al. (5)	Case report	F	40	Painful swelling 4.6 × 1.8 × 4.6 , erythema, nipple discharge	Nil
Akahane et al. (6)	9	F	36	2.4 - 10.0 cm palpable lumps, skin thickening, or axillary lymphadenopathy	Seven patients were diagnosed within 5 years of their most recent pregnancy
Lai et al. (7)	9	F	45.7	Breast mass	Recurrence in 4 patients
Lin et al. (8)	Case report	F	39	9 × 6 cm breast lump with localized redness	Associated prolactinemia
Our case	Case report	F	29	4 × 3 cm painful mass, firm in consistency with restricted mobility	Nil

Figure 1. The cytology Smear Shows a Cluster of Epithelioid Cells (Thick Arrow), Benign Cohesive Ductal Cells (Thin Arrow), and a Few Lymphocytes



The inset shows a giant cell (H and E, 10 ×).

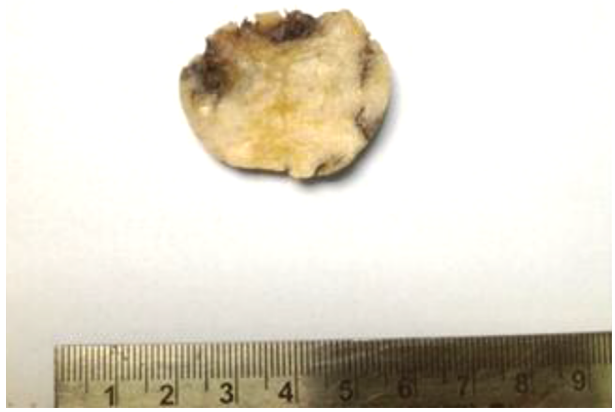


Figure 2. Cut Surface of the Breast Lump Showing Gray-to-Tan to Focal Yellowish Areas

sections were stained using a Ziehl–Neelsen method and Gomori's methenamine silver (GMS) and viewed with a polarizing microscope to rule out mycobacterial, fungal, and foreign body etiology, respectively. Based on the above findings, the diagnosis of idiopathic granulomatous lobular mastitis was given. The patient was later treated with corticosteroids, and there was good clinical recovery from the disease. The patient follow up has been uneventful to date.

3. Discussion

The cause of IGLM is unknown. Though several hypotheses have been suggested, the one that is most widely accepted is damage to the mammary ducts by various factors, including trauma, local irritants, and infections that allow luminal material to leak into the stroma thus evoking a granulomatous inflammation (9). The association of IGLM with pregnancy, lactation, the use of oral contra-

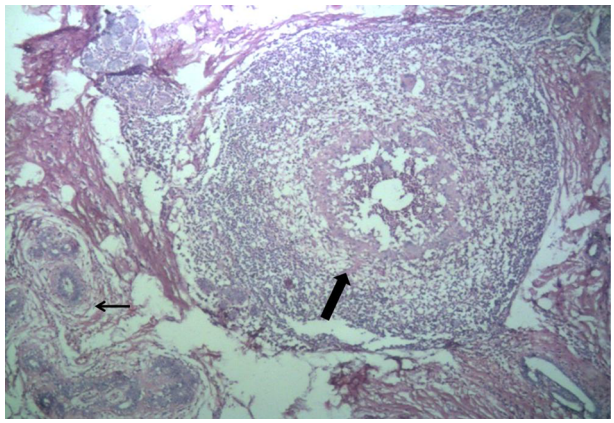


Figure 3. Photomicrograph Showing a Well-Formed Granuloma (Thick Arrow) Centered Around a Lobule With Adjacent Breast Acini (Thin Arrow) (H and E, 4 ×).

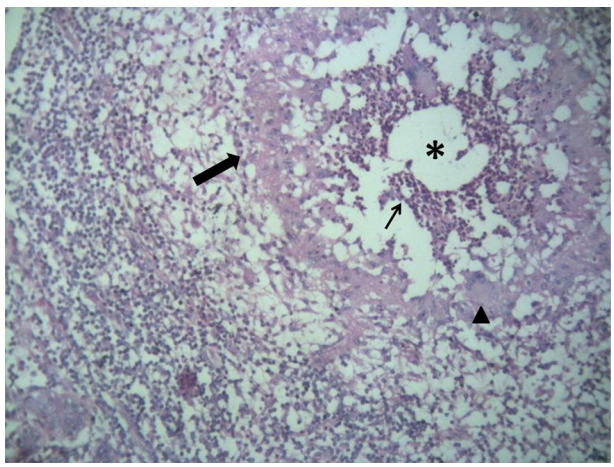


Figure 4. Photomicrograph Showing a Granuloma With a Central Clear Space (Asterisk) Rimmed by Neutrophils (Thin Arrow), a Band of Epithelioid Cells (Thick Arrow), and a Langhans Giant Cell (Arrow Head) (H and E, 10 ×).

ceptive pills (OCP), trauma, and autoimmune diseases has been documented. Additionally, recent studies have highlighted the involvement of hyperprolactinemia and galactorrhea with IGLM (10, 11). There are increased mammary acinar secretions with these conditions, which may lead to distension, rupture of the acini, and extravasation of the luminal contents causing a granulomatous response. Nevertheless, our case was not associated with any of the aforementioned risk factors. The age of presentation in our case was also considerably lower than reported elsewhere.

Hovanessian Larsen et al. (12) proposed an autoimmune etiology due to the response of IGLM to corticosteroids; however, they were ultimately unable to prove the existence of an autoimmune origin. Altintoprak et al. (2) evaluated the anti-nuclear antibody (ANA) and extractable nuclear antigen (ENA) levels in patients with IGLM by indirect immunofluorescence (IIF). Their results also failed to support an autoimmune basis for IGLM.

According to Akahane et al. (6) the most common presenting symptoms of IGLM are mastalgia and breast mass. They also reported that 58.3% of the cases in their study had diffuse multiple lesions. Features like breast lumps, pain, diffuse involvement of the breast, and lymph node enlargement are of concern to clinicians because of high suspicion for inflammatory carcinoma of the breast. Verfaillie et al. (9) reported that neither mammography nor Doppler sonography helps in the diagnosis of ILGM, and it can only be confirmed by histopathology.

ILGM is histologically characterized by granulomas that are non-caseating and centered around lobules. There is no definite evidence for an infectious etiology. A characteristic clear space rimmed by neutrophils is typically seen at the center of the granuloma. This clear space is due to the lipid material from the degenerated cells dissolved during tissue processing (13). The final diagnosis is made by identifying ILGM's characteristic histological pattern and excluding other causes of granulomas, such as tuberculosis, sarcoidosis, Wegener's granulomatosis, a fungal infection, or a foreign body (4).

In Kok et al.'s (14) study, only 17% of the IGLM patients were diagnosed by FNAC alone, while Akahane et al. (6) were unable to diagnose any cases by FNAC alone. This highlights the limitations of FNAC in diagnosing IGLM due to the lack of features characteristically seen in histopathology. In our case, the FNAC diagnosis was granulomatous mastitis; however, a final diagnosis of IGLM could only be made by histopathology after ruling out other possible causes of granulomas.

The definitive treatment of this condition is still unclear although corticosteroids remain the mainstay of treatment in patients with IGLM due to their anti-inflammatory action. Patients with minimal symptoms are better managed conservatively by close, regular surveillance without surgery (7). Nevertheless, Gurleyik et al. (1) reported that none of their patients treated with corticosteroids had a complete clinical recovery from the disease. They recommended that local excision of the remaining lesion after steroid therapy would give a better result and decrease the chances of recurrence. Unnecessary mastectomies should not be done for this condition as mastectomy is strictly reserved for cases with multiple recurrence and persistent lesions. In cases with multiple recurrence, secondary causes including prolactinemia due to pituitary adenoma or drugs like risperidone should be ruled out (8). A diagnosis of IGLM should be made cautiously since erroneously diagnosed cases, if treated

with steroids, will aggravate granulomatous disease of an infectious etiology. Similarly, cases of IGLM, if misdiagnosed as infectious granulomatous conditions, will never respond to antibiotics.

3.1. Conclusion

IGLM should be considered as one of the differential diagnoses when granulomas are encountered in fine-needle breast aspirates, but the final diagnosis should only be made by histopathology with the aid of special techniques and by ruling out other possible causes. A correct diagnosis is of prime importance for the appropriate management of these lesions.

Footnote

Authors' Contribution: Thulasi Raman R, Drafted the manuscript; D Manimaran, diagnosed the lesion.

References

1. Gurleyik G, Aktekin A, Aker F, Karagulle H, Saglamc A. Medical and surgical treatment of idiopathic granulomatous lobular mastitis: a benign inflammatory disease mimicking invasive carcinoma. *J Breast Cancer.* 2012;**15**(1):119–23. doi: 10.4048/jbc.2012.15.1.119. [PubMed: 22493638].
2. Altintoprak F, Karakece E, Kivilcim T, Dikicier E, Cakmak G, Celebi F, et al. Idiopathic Granulomatous Mastitis: An Autoimmune Disease?. *Sci World Journal.* 2013;**2013**:1–5. doi: 10.1155/2013/148727.
3. Reddy KM, Meyer CE, Nakdjevani A, Shrotria S. Idiopathic granulomatous mastitis in the male breast. *Breast J.* 2005;**11**(1):73. doi: 10.1111/j.1075-122X.2005.21404.x. [PubMed: 15647084].
4. Bakaris S, Yuksel M, Ciragil P, Guven MA, Ezberci F, Bulbuloglu E. Granulomatous mastitis including breast tuberculosis and idiopathic lobular granulomatous mastitis. *Can J Surg.* 2006;**49**(6):427–30. [PubMed: 17234073].
5. Ergin AB, Cristofanilli M, Daw H, Tahan G, Gong Y. Recurrent granulomatous mastitis mimicking inflammatory breast cancer. *Case Reports.* 2011;**2011**(jan20 1):bcr0720103156. doi: 10.1136/bcr.07.2010.3156.
6. Akahane K, Tsunoda N, Kato M, Noda S, Shimoyama Y, Ishigakis S, et al. Therapeutic strategy for granulomatous lobular mastitis: a clinicopathological study of 12 patients. *Nagoya J Med Sci.* 2013;**75**(3-4):193–200. [PubMed: 24640175].
7. Lai EC, Chan WC, Ma TK, Tang AP, Poon CS, Leong HT. The role of conservative treatment in idiopathic granulomatous mastitis. *Breast J.* 2005;**11**(6):454–6. doi: 10.1111/j.1075-122X.2005.00127.x. [PubMed: 16297091].
8. Lin C-H, Hsu C-W, Tsao T-Y, Chou J. Idiopathic granulomatous mastitis associated with risperidone-induced hyperprolactinemia. *Diagnostic Pathology.* 2012;**7**(1):2. doi: 10.1186/1746-1596-7-2.
9. Verfaillie G, Breucq C, Sacre R, Bourgain C, Lamote J. Granulomatous lobular mastitis: a rare chronic inflammatory disease of the breast which can mimic breast carcinoma. *Acta Chir Belg.* 2006;**106**(2):222–4. [PubMed: 16761483].
10. Rowe PH. Granulomatous mastitis associated with a pituitary prolactinoma. *Br J Clin Pract.* 1984;**38**(1):32–4. [PubMed: 6538433].
11. Cserni G, Szajki K. Granulomatous Lobular Mastitis Following Drug-Induced Galactorrhea and Blunt Trauma. *Breast J.* 1999;**5**(6):398–403. [PubMed: 11348321].
12. Hovanessian Larsen LJ, Peyvandi B, Klipfel N, Grant E, Iyengar G. Granulomatous lobular mastitis: imaging, diagnosis, and treatment. *AJR Am J Roentgenol.* 2009;**193**(2):574–81. doi: 10.2214/AJR.08.1528. [PubMed: 19620458].
13. Rosen AC, Dexter F. Lessons from evidence-based operating room management in balancing the needs for efficient, effective and ethical healthcare. *Am J Bioeth.* 2009;**9**(4):43-4. doi: 10.1080/15265160802716894. [PubMed: 19326313].
14. Kok KY, Telisinghe PU. Granulomatous mastitis: presentation, treatment and outcome in 43 patients. *Surgeon.* 2010;**8**(4):197–201. doi: 10.1016/j.surge.2010.02.002. [PubMed: 20569938].