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## CASE REPORT

### Idiopathic Granulomatous Mastitis - Report Of A Case With A Brief Review Of Literature

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#### ABSTRACT

Idiopathic granulomatous mastitis is a rare inflammatory disorder which is characterized by the presence of non-caseating granulomas within the breast lobules, in which no organisms are found. It is hypothesized that the disease is a hypersensitivity reaction to the lobular epithelium and its contents. Though it is frequently reported from western countries, very few cases have been described from India. It affects only parous women and clinically mimics inflammatory breast carcinoma and tubercular mastitis. FNAC alone is not sufficient for the diagnosis and this can only be confirmed by an open biopsy. We report here, a case of granulomatous lobular mastitis in a 25 year old lactating female patient.

**Key Words:** Granulomatous lobular mastitis, granuloma, Breast, non-caseating granuloma.

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#### Introduction

Idiopathic granulomatous mastitis is a rare lesion in which the granulomas are associated with micro abscesses which may respond to corticosteroid therapy [1],[2].The distinctive term of granulomatous lobular mastitis has been applied to this condition which is seen to arise in younger, parous women, where the inflammatory granulomatous process is centered on the structure of the breast lobule [3],[4]. The histological diagnosis of granulomatous mastitis is difficult because the features overlap with other granulomatous conditions, especially mycobacterium infections, including atypical mycobacterium infections. Definitive diagnosis depends on typical histology and negative microbiological profile [5],[6].We report here, a histological diagnosed case of granulomatous lobular mastitis.

#### Clinical Findings

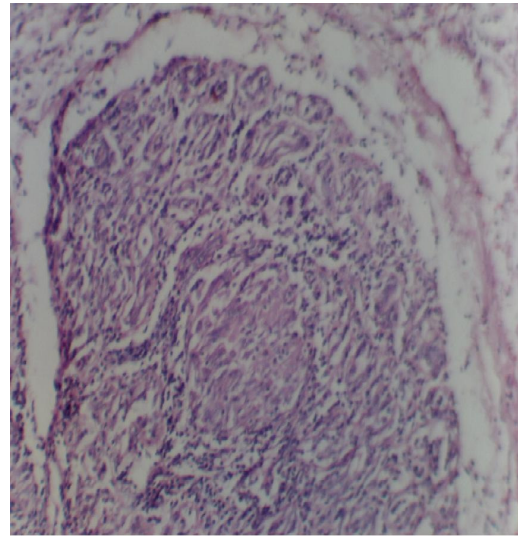
A 25 years old female lactating patient presented with multiple, firm, tender, nodular lesions in the right breast. FNAC revealed granulomatous mastitis and the stain for AFB was negative. Then biopsy was done to confirm the diagnosis and to rule out other granulomatous conditions.

#### Pathological Findings

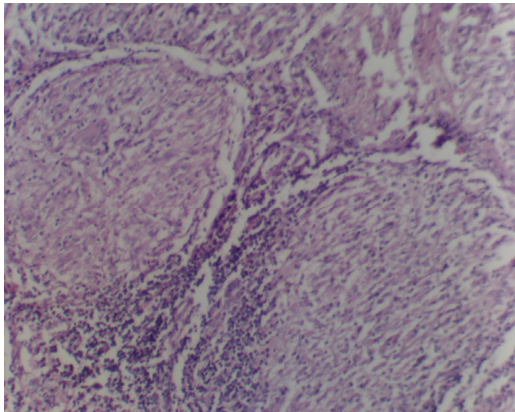
The gross specimen consisted of an irregular mass measuring (5x4) cm. The cut surface showed nodular areas with areas of necrosis [Table/Fig 1]. Histology showed multiple non-caseating granulomas with microabscess formation, which were confined to the breast lobules [Table/Fig 2], [Table/Fig 3], [Table/Fig 4] (Figure-2,3,4 HE-10x). The granulomas were composed of epithelioid histiocytes, giant cells including Langhans types, lymphomononuclear cells and neutrophils [Table/Fig 5] (Figure-5, HE-10x). Acid fast bacilli and fungal bodies could not be detected by special stains (Z-N and PAS). A diagnosis of granulomatous lobular mastitis was made.



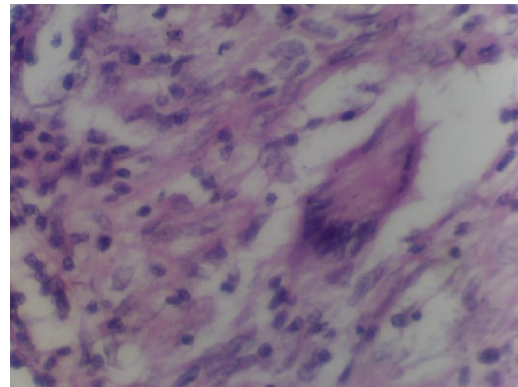
[Table/Fig 1]. Irregular nodular breast mass depicting minimal necrosis.



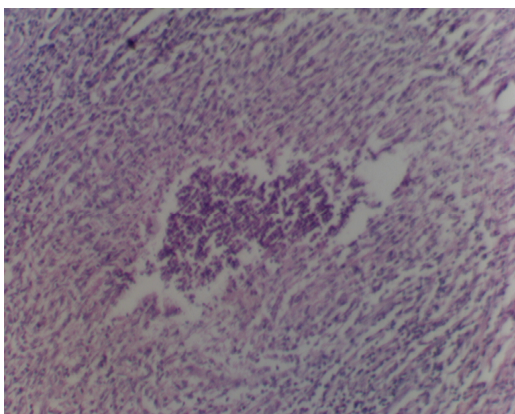
[Table/Fig 4]. Photomicrograph showing microabscess in the breast lobule.



[Table/Fig 2]. Photomicrograph showing multiple granuloma



[Table/Fig 5]. Photomicrograph depicting Langhans giant cells within mixed inflammatory background



[Table/Fig 3]. Photomicrograph showing microabscess in the center of granuloma.

## Discussion

Idiopathic granulomatous mastitis was first described as a specific entity by Kessler and Wolloch in 1972 [1]. It was further elaborated by Cohen [7] in 1977. Granulomatous mastitis is an uncommon and curious condition of unknown aetiology (6). However, its association with the use of oral contraceptive pills, autoimmune disorders, hyperprolactinaemia, alpha-1 antitrypsin deficiency and *Corynebacterium* spp has been proposed [8],[9],[10],[11],[12]. Studies show that women

who are affected by IGM, belong to third decade of life [8],[9],[13]. All the cases described by Kessler and Wolloch [1972] and Fletcher 1982, occurred within 6 years of pregnancy. Our case presented during the lactation period. Studies show conflicting data associating the role of oral contraceptives in patients diagnose as IGM range from 0 to 33 % [8],[14], [15]

It occurs in young parous women and presents as a firm tender lump that may be mistaken for carcinoma [1],[6]. In a study, all cases with histopathologically proven Idiopathic granulomatous mastitis initially showed breast masses which were suspected of having breast carcinoma [16]. So, detailed histopathological examination of the cases which are suspected as carcinoma is mandatory.

Histopathologically, IGM may mimic tubercular mastitis showing well defined granuloma, caseous type secretions, epithelioid cells and langhans giant cells and can result in a mistaken diagnosis of tuberculosis. Treating tuberculosis with steroids would aggravate the infection, whereas unnecessary antitubercular drugs may cause numerous side effects. So, the differential diagnosis of tubercular mastitis needs to be considered [18],[19]. The only diagnostic proof of tubercular mastitis is the demonstration of tubercle bacilli in a microscopic smear or culture or by PCR for mycobacterium tuberculosis. The sensitivity of PCR in AFB smear negative cases as low as 50% have been reported in some series [20]. A case study by KB Sriram and D. Moffat highlight the difficulty in differentiating culture negative tuberculosis from granulomatous mastitis and the importance of a high index of clinical suspicion [21]. The presence of caseous necrosis, langhans giant cells and granuloma favour the diagnosis of TB mastitis, whereas IGM represents a lobular distribution of mixed chronic inflammatory processes which are composed of lymphocytes, plasma cells, giant cells, the presence of neutrophil infiltration and lack of caseation. Additionally, micro abscess formation and squamous metaplasia of the lobular and ductular epithelium may occur [1],[27].

Farhan Abbas and Anwal in their study, found that fat necrosis was the most predominant feature. Fat damage was the main cause of the formation of granuloma and giant cells which were surrounded by lymphocytes, plasma cells and neutrophils. [22]

Because the GM is essentially a diagnosis of exclusion, other differential diagnoses are rare specific causes of granulomatous inflammation, including fungal infections or non infectious causes, sarcoidosis, Wegner's granuloma, granulomatous angiopanniculitis of the breast, fat necrosis, foreign body granuloma, plasma cell mastitis, cholesterol granuloma and milk granuloma. Combination of any of the above mentioned conditions is also a possibility, which must be taken into consideration [18],[19]. In this case, the diagnosis was done clinically and histologically. This case appears to be a distinct disease entity, as neither organisms nor foreign body materials have been identified.

Several reports describe the association of granulomatous lobular mastitis with erythema nodosum and polyarthritis, thus strengthening the argument of an autoimmune cause and therefore resemble to granulomatous thyroiditis, or granulomatous orchitis [23],[24]. GM mastitis may occur due to exogenous hormones (oral contraceptives) or endogenous hormones (prolactins). Oral contraceptives induce hyperplasia in the lobular ductule, thus leading to the obstructive desquamation of the ductules, distention of the ductules and perilobular inflammatory reactions [25]. Prolactins lead to postlactational granulomatous mastitis and this is associated with pregnancy.

Fletcher et al. (1982) suggested that the finding of polymorphs in some of the ductular lumina might indicate a primary damage to the epithelium by some unknown agent, resulting in a leakage of contents and a subsequent granulomatous response in the surrounding stroma. In the recently reported cases, immunostaining showed that the lesions contained predominantly stromal T lymphocytes which favoured the possibility of a local immune response [26]. This condition may

respond to steroids and be associated with a high incidence of postoperative wound infections [6]. In this patient, the lesion resolved with steroid treatment. Granulomatous mastitis can recur in up to 50% of the cases, usually within 6 weeks to 11 months after stopping treatment [11]. In refractory cases or in those with persistent collection, immunosuppressants like methotrexate have been utilized along with surgical excision.

## Conclusion

IGM is rare and benign inflammatory process which is commonly mistaken for malignancy and other disease entities which is why it is often treated incorrectly. Correct diagnosis requires the exclusion of infectious aetiologies, other causes of granulomatous mastitis and malignancy combined with definitive histopathological confirmation. One must not accept granulomatous mastitis as tuberculosis, even in the area where tuberculosis is common, until and unless there is a clear history of tuberculosis and the involvement of other organs like the lymphnodes, in order to avoid the pitfall of prolonged treatment and side effect.

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