



Original Article

Idiopathic Granulomatous Mastitis: In Search of a Therapeutic Template

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Abstract

Background: Granulomatous mastitis also known as granulomatous inflammatory lesions of the breast can be divided into idiopathic granulomatous mastitis and granulomatous mastitis occurring as a rare secondary complication of a great variety of other conditions. Treatment is radically different for idiopathic granulomatous mastitis and other granulomatous lesions of the breast, the precise diagnosis is therefore very important.

Objectives: The purpose of this study was to find the clinicopathological features of idiopathic granulomatous mastitis, as well as the authors' experience with surgical treatment of these patients.

Methods and Materials: A hospital based retrospective study done with twenty diagnosed patients of idiopathic granulomatous mastitis enrolled from department of surgery, Ad-din women's medical college hospital, Dhaka in the period between July 2012- July 2015.

Results: Mean age of presentation was 38.1 years. All patients had a history of childbirth and breastfeeding. Sixteen (80%) patients were premenopausal. The main clinical feature was a mass in the breast in all patients and clinical findings suggesting an infection accompanied the mass in seven patients (35%). Surgery was the definitive procedure in all patients. Wide local excision done in all patients. None of the patients received steroids. Median follow-up time was 12 months. During follow-up, two patients (10%) presented with recurrence. All patients with recurrence had a mass in their breasts on the same side of previous disease.

Conclusion: Increased recognition of this disease will improve its understanding and management. Long-term follow-up is necessary.

Key words: Breast, granulomatous mastitis, surgery

Introduction

Idiopathic granulomatous mastitis (IGM) is an uncommon, benign, chronic inflammatory condition of unclear etiology, usually seen during pregnancy or lactation period¹. It is also known as granular lobular mastitis and usually poses a diagnostic and therapeutic

dilemma². It is a relatively recent reported condition that was first described in 1972 by Kessler and Wollock³. IGM is a rare inflammatory disease of the breast, but it is important because of two reasons. First clinical and radiological findings of IGM mimicks breast carcinoma and the differential diagnosis can

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only be confirmed histopathologically. Second, it is difficult to treat IGM especially if presented with fistula and abscess formation. The primary presentation is a firm breast mass, frequently associated with local pain and eventually skin ulcerations, abscesses and fistula develop^{2,4}. The diagnosis is made only after excluding breast cancer and other infective and non-infective causes of granulomatous inflammation, such as tuberculous, parasitic and fungal infections, sarcoidosis, Wegener's granulomatosis, giant cell arteritis, polyarteritis nodosum, and foreign-body reactions. The optimal treatment of IGM is still controversial. Although surgical removal is accepted as the mainstay of treatment, antibiotics, steroids, and even chemotherapeutic agents were used to treat these patients⁴.

Materials and methods

A hospital based retrospective study done with twenty patients of idiopathic granulomatous mastitis managed over July 2012- July 2015 enrolled from department of surgery, Ad-din women's medical college hospital. This study was approved by our institutional review board. Pathologic diagnosis of IGM and ruling out the other possible causes of granuloma formation were considered as inclusion criteria. Definitive diagnosis was based on excisional biopsy from the affected breast. We consider open biopsy to be the safest method to rule out malignancy. The histological diagnosis of granulomatous mastitis was made when noncaseating, nonvasculitic granulomatous inflammatory reaction, composed of epithelioid cells, lymphocytes, and foreign body type of giant cells, centered mainly on breast lobules. Microabscess formation was noted in some of the cases. Special stains (Gram, Ziehl-Neelsen, and periodic acid-Schiff) for microorganisms were negative. In this study, surgical excision and antibiotics were the primary treatment modalities. Patients with inflammatory findings, but without abscess formation were initially received antibiotic regimen of Cefixime and flucloxacillin for at least one week. In case of no regression and negative culture (with probability of fastidious nature of certain microbes), the treatment would be changed to Clindamycin. Incisional biopsy was obtained from patients with abscess formation. If there was fistula formation to the skin, affected skin was excised too. Wide local excision to obtain disease-free margins was performed in patients with an isolated mass. No patient received corticosteroids or methotrexate in this study. Treatment methods and related complications were evaluated. Patients were followed every 6 months with a thorough clinical examination. Bilateral breast ultrasonography and, if required, mammography were performed yearly.

Recurrences detected during follow-up and their treatments were recorded as well.

Results:

Table 1.

Distribution of granulomatous mastitis by age ranges.

Age ranges(years)	Number of cases	Percentage (%)
19-29	3	15
30-39	13	65
40-49	2	10
50-59	2	10
Total	20	100

Mean age was 38.1 years with extreme values of 19 years and 56 year. None of the patients presented a documented history of autoimmune disease. The time to presentation of idiopathic granulomatous mastitis in relation to the patient's most recent birth ranged from 8 days to 15 years. The timescale was less than five years in 12 cases, which equates to 60% of cases. All patients had a history of childbirth and breastfeeding and one patient was pregnant with her second child at the time of diagnosis. None of the patients in our series had received hormone replacement therapy. A family history of breast cancer was found in one patient. None of the patients were smokers.

The mean duration of symptoms of granulomatous mastitis in our series was 5 months with extreme values of 2 weeks and 2 years. The main clinical feature was a mass in the breast in all of the patients. The mean size of the lesion was 5.5 cm with extreme values of 2 cm and 18 cm in diameter. The predominant area involved was the upper outer quadrant. Axillary lymphadenopathies were present in five patients (25% of cases). Twelve cases involved the left breast (60%), while eight involved the right breast (40%).



Figure:1 After incision and drainage of pus

Table 2.

The clinical signs revealing idiopathic granulomatous mastitis.

Clinical signs	Number of cases	Percentage
Breast mass	20	100%
Inflammatory plaque	4	20%
Abscess	3	15%
Skin induration	2	10%
Breast discharge	1	5%

Excisional biopsy was the initial surgical intervention to obtain a histopathological diagnosis in 17 patients (85%). Incisional biopsy was performed in 3 (15%) patients.

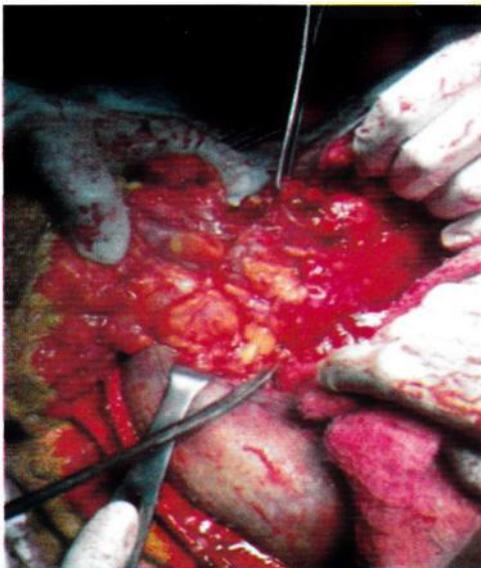


Figure 2: Per operative view yellowish white mass

Median follow-up time was 12 months (range,6-36). During follow-up, two patients (10%) presented with recurrence. All patients with recurrence had a mass in their breasts on the same side of previous IGM. These two patients were treated with further wide local excisions and did not show any signs of further recurrence.

Discussion

The current patient series represents one of the largest series (20 patients) in Bangladesh, reported from a single center until today, although the data were collected retrospectively. In addition, patients were followed for a long period of time years (median one year), to delineate the clinical course of the disease after initial treatment.

IGM represents 24% of all histopathologically defined inflammations in the breast⁵. Today, widely accepted mechanism states that autoimmune response to

secretions originating from damaged ducts may have a role in IGM. Accumulation of secretions in mammary ducts leads to ductal ectasia and, eventually, to the rupture of the involved ducts. Secretions spreading to the stromal tissue after ductal rupture result in a chronic inflammation⁵. Multiple breast-feedings and prolonged lactation periods may be the reason for dilatation and rupture of the ducts and acini. In addition, Al-Khaffaf et al. compared demographic characteristics of IGM patients to patients with periductal mastitis and normal controls and reported that IGM patients are younger and have given birth recently supporting the above mentioned hypothesis⁶.

The patient's age at the time of diagnosis varies depending on the series, ranging from 17 to 83 years⁶. The mean age for developing the disease was 38.1 years old in our series, with extreme values ranging from 19 to 66 years. In our series, four women were postmenopausal.

IGM presents most commonly with a painful, firm, tender, ill defined mass in the breast and unilateral⁷. In consequence of granulomatous inflammation, IGM can cause skin thickness, sinus and abscess formation, axillary lymphadenopathy and nipple retraction which may be clinically mistaken for breast carcinoma⁷. All patients were admitted with painful mass to our institution, 20% of these accompanied with skin changes. The lesions were located in any location but there were the tendency the subareolar. Bilateral involvement is reported very rare⁸, there was no patients in our series with bilateral and diffuse involvement. Enlarged axillary lymph nodes were present in 5 (25%) patients, but all of these enlarged nodes were established reactive and not suspicious for malignancy.

Since physical examination and imaging modalities fail to diagnose and differential diagnose, histopathologic diagnosis must be performed. Histopathologic diagnosis can be achieved with fine needle aspiration cytology (FNAC), core, incisional or excisional biopsy⁹. In our patients, the histopathologic diagnosis was obtained from excisional and incisional biopsies, we have not preferred FNAC.

The optimal treatment of IGM remains controversial. Surgical excision still seems to be the best treatment. Wide local excision can be appropriate treatment also provide exact diagnosis and treatment. After wide local excision, if possible, further therapy is not needed. Different recurrence rates (range 5.5%-50%) are reported after wide local excision. There were two (10%) recurrence of 20 patients that wide local excision was performed in our (range 5.5% - 50%)

are reported after wide local excision. There were two (10%) recurrence of 20 patients that wide local excision was performed in our patients. As a matter of fact, complicated IGM with abscess, fistula or diffuse involvement poses the problem. There is no ideal treatment for complicated IGM. In these patients, wide local excision cannot be possible. Here there is a chance of recurrence.

We hesitate to recommend corticosteroids in complicated cases for several reasons. First, there is no consensus about timing, duration and dose of corticosteroid administration. Second, according to the study of Sakurai et al., complete resolution of mass lesions with corticosteroid was achieved from 4 to 10 months¹⁰. No patients in our study receive corticosteroid as it takes long time of treatment.

Conclusion

Surgeons, radiologists, and pathologists should be more familiar with IGM due to its clinical significance. Differentiating IGM from breast infections and malignancy is the most critical part of the management. Multiple assessments including clinical, radiologic, and histopathologic examinations are required for an accurate diagnosis. Long-term follow-up preferably by the same doctors will be a logical solution for the management of such a difficult disease.

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