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Granulomatous Mastitis – Diagnosis and Treatment Modality – Case Report and Review of Literature

Dr Preethi Elumalai[1], Dr Anil Malleshi Betigeri[2], Dr Sagarika Sarkar[3], Dr Anantharaman D[4]

[1](Tutor, Department of Pathology, East Point College of Medical Sciences and Research Centre, Bengaluru)
[2](Associate Professor, Department of Pathology, East Point College of Medical Sciences and Research Centre, Bengaluru)
[3](Assistant Professor, Department of Pathology, East Point College of Medical Sciences and Research Centre, Bengaluru)
[4](Professor, Department of Surgery, East Point College of Medical Sciences and Research Centre, Bengaluru)
Corresponding author: Dr Preethi Elumalai

Abstract: *Granulomatous mastitis (GM) is an uncommon chronic inflammatory disease of breast of undetermined etiology. Despite its low incidence, it has gained importance since it mimics breast carcinoma. There is still no definite treatment modality for this condition. Treatment with corticosteroids and/or wide excision is most frequent mode of therapy reported in the literature. We report a case of granulomatous mastitis in a 38-years-old female, who had presented with swelling in her left breast for 2 months. She had a past history of incision and drainage for the same complaint 3 months back. Presently, she was clinically diagnosed as a case of antibioma and underwent lumpectomy. Since it is a rare disease with chances of recurrence, ensuring appropriate diagnosis and optimum treatment is essential.*

Keywords : *Idiopathic granulomatous mastitis, Breast abscess, Inflammatory mastitis, Lobulocentric granulomas of breast.*

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I. Introduction

Kessler and Wolloch first described Idiopathic granulomatous mastitis in 1972.[1] Idiopathic granulomatous mastitis may present as mass like lesion. It occurs in the age group of 17 to 52 years. It usually occurs in parous individuals. Any association with breast feeding hasn't been established. This disease manifests as a firm to hard lump and can involve any quadrant of breast. In mammogram, granulomatous mastitis presents as multiple clustered hypo and hyperechoic masses.[2] Histologically, it is characterized by granulomatous reaction confined to lobules. Early detection with proper treatment is essential to avoid recurrence and unnecessary radical surgery which poses a major trauma to patients.

In this article, we report a case of granulomatous mastitis in a 38- year- old female who was initially clinically diagnosed as a case of antibioma. She presented with a history of lump in the breast for 3 months. There was no improvement following antibiotic therapy or even after incision and drainage. Eventually she underwent lumpectomy.

II. CASE PRESENTATION

A 38- year- old non-lactating female had complaints of swelling in her left breast for 3 months. She had a history of fever on and off. There was pus discharge from the swelling since last 3 weeks. She had been treated with antibiotics for 1 month in a nearby hospital following which her swelling was reduced minimally but did not resolve completely. This was followed by incision and drainage (I&D). However, after 3 weeks the patient again noticed swelling and pus discharge from the lesion. She didn't have any history pertaining to tuberculosis or autoimmune disorders.

On examination of left breast, a swelling measuring 8x7 cm size was noted in the upper inner quadrant extending to sub areolar region and lower inner quadrant. There was pus discharge from the swelling. Incision scar was evident in the upper inner quadrant. Nipple and areola appeared normal. There was no lymphadenopathy. Pus aspiration was done thrice and the samples were sent for microbiological studies. Culture reports were negative. The patient then underwent lumpectomy and the specimen was sent for histopathological examination.

We received an irregular fibrofatty mass measuring 8x8x3 cm, cut section of which showed necrotic and fibrotic areas. Fibrinopurulent exudates were noted at few foci. Multiple sections were taken for histopathological examination. Microscopy revealed numerous granulomas with dense inflammatory cell infiltrate composed of lymphocytes, plasma cells with multinucleated giant cells. Granulomatous reaction was lobulocentric and was seen extending into adjacent breast parenchyma. No caseation necrosis or malignant cells were seen. Stain for AFB and fungus were negative.

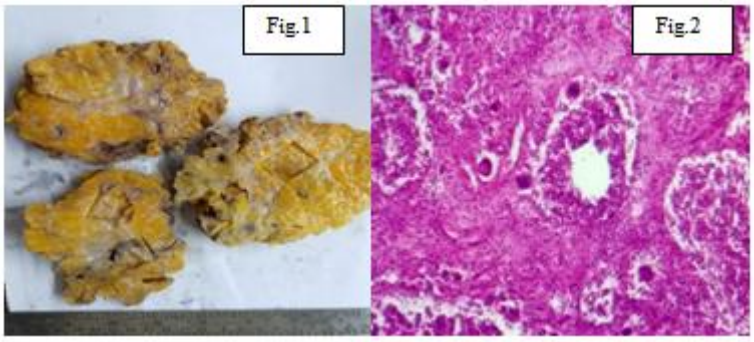

Fig.1: Gross image of fibrofatty mass with necrotic and fibrotic areas.
Fig.2: Histopathology (H&E,x400): Lobulocentric granulomas with dense inflammation and multinucleate giant cells.

III. Discussion

There are many inflammatory and reactive lesions of breast that include - fat necrosis, breast infarct, plasma cell mastitis, granulomatous lobular mastitis, sarcoidosis, inflammatory pseudotumor, vasculitis, scleroderma, dermatomyositis, paraffinoma, silicone mastitis and diabetic mastopathy. There are few inflammatory lesions that are exclusively seen in pregnancy namely puerperal mastitis, mammary infarction and galactocele.

Idiopathic granulomatous mastitis occurs mostly in young women. Since the etiology of granulomatous mastitis is unclear, various hypotheses have been put forward. One of the basic pathogenic mechanisms resulting in granulomatous mastitis is believed to be from the extravasated secretions from lobules which elicit a local immune response. Another study states that any local trauma or infection which disrupts the ductal lining can produce immune response. Granulomatous mastitis is a disease with heterogeneity and variable clinical features.[3] It occurs in age group of 17 to 52. Clinically it presents as a firm to hard mass exclusively sparing subareola. It is often accompanied with tenderness.

On gross examination, the cut surface of Idiopathic granulomatous mastitis shows grey tan areas with vague nodular formations. Sometimes abscess formation can be identified. Histologically, there is a lobulocentric granulomatous inflammation.[4] There is dense lymphoplasmacytic infiltration with multinucleate giant cells. When lesions become confluent, fibrosis and necrosis are often evident. There are no demonstrable microorganisms. Foci showing dilated ducts can also be noted. Differential diagnoses include tuberculosis especially in tropical countries, followed by sarcoidosis and Cat scratch disease. Multiple sections are to be taken to rule out malignancy.

There are various recommended treatment modalities available for granulomatous mastitis. It includes medical management and surgical management. Since the etiology of Idiopathic granulomatous mastitis is not specific, treating it poses a challenge. There is no standard treatment.

TABLE:1 Recommended treatment modalities

MEDICAL TREATMENT	SURGICAL TREATMENT
Appropriate antibiotics	Incisional biopsy with drainage
Corticosteroids	Open biopsy with steroid therapy
Methotrexate therapy	Wide local excision(WLE) ,Mastectomy (rarely)[5]

In a study of 24 patients by Bani-Hani et al, wide local excision is the main stay of treatment in order to avoid unnecessary of removal of breast.[6] Lai et al stated that 50% of cases with Idiopathic granulomatous mastitis had spontaneous resolution without any treatment.[7] In a study of 541 patients, 112 patients were treated with steroids, however 22 patients had recurrence and steroid induced Diabetes mellitus was observed in 5 patients. There was no response in 4 patients. Mass was decreased in size in 2 patients. Later steroid was changed to methotrexate and brought satisfactory results. Only for 2 patients, mastectomy was done due to

recurrence. Thus methotrexate is the effective treatment by preventing the complications.[8] Sakurai et al reported that treatment of mass lesions with steroids give complete resolution.[9]

In a retrospective study of 46 patients over 12 years, complete excision was done in 18 patients, 25 patients were treated with steroids and concluded that treatment depends upon severity and extent of the disease.[10] In an article by Gulten kayak et al, 24 patients of Idiopathic granulomatous mastitis were followed up. Wide local excision was performed in 15 patients, of which one patient recurred after 2 years in another quadrant. Incisional biopsy with abscess drainage was done in 9 patients. It was concluded that better treatment is wide local excision with steroid therapy unless there is diffuse involvement or any complication of abscess or fistula formation.[11]

In a study of 13 patients by Mohammed Esmail et al, they favoured non- surgical management. Hence, they recommended repeated aspiration with antibiotics.[12] In a study of seven patients by Sanint et al, all patients were treated with glucocorticoids. 2 patients had recurrence and underwent surgical drainage.[13]

TABLE:2 Management of Idiopathic granulomatous mastitis from literatures

S.No.	Study name	Total cases	Medical therapy	Surgical therapy	No. of cases recurred	Recommended treatment of choice
1	Bani-Hani et al 2004	24		WLE -23 Mastectomy -1	4(>1 year)	WLE
2	Lai et al 2005	9	Close follow up -8	Lobectomy -1	4	Close regular surveillance
3	Sami, Davut, sule 2011	541	Steroids -112 Methotrexate given to 13 unsuccessful steroid treated patients Methotrexate with steroids - 3	Mastectomy - 2	22, Steroid induced DM -5, No response-4 14 methotrexate treated patients - Satisfactory	Methotrexate is satisfactory
4	Sakuari et al 2011	8	Steroid -5 Steroid with antibiotics -3	- 1 out of 3 patients	No	Steroid therapy is effective
5	Ebru et al 2013	46	Steroids - 25	Complete excision- 18 Excision with steroids - 3	8	Depends upon severity and extent of disease
6	Gulten et al 2014	24		WLE – 15 I&D - 9	1	WLE For complicated IGM – WLE with close follow up
7	Md esmail et al 2014	13		Repeated aspiration with antibiotic therapy		Recurrent drainage
8	Sanint et al 2015	7	Glucocorticoids-7	2 recurred cases with surgical drainage	2	Glucocorticoids is the treatment of choice
9	Present case	1	Antibiotics	Lumpectomy		

Depending upon the size of the mass and the immune status, patient has to be treated with steroids or methotrexate. If the mass becomes extensive wide local excision is to be done. Mastectomy has to be avoided. Proper and timely workup has to be started to rule out other causes and to limit the size of the mass. In our case, microbiological studies and autoimmune workup was done which was found to be negative. Patient recurred with lump which had increased in size with pus discharge even after course of antibiotics and incision and drainage. Finally, Wide local excision of the mass was done.

IV. Conclusion

Though idiopathic granulomatous mastitis is a rare disease, it has to be diagnosed and treated with utmost care as it has increased chances of recurrence. There should be proper approach in the management of idiopathic granulomatous mastitis depending upon the size, extent of the disease and patient compliance.

Fig. 3: Approach to management of Granulomatous mastitis

Presence of microscopic granulomas on core needle biopsy of breast lump

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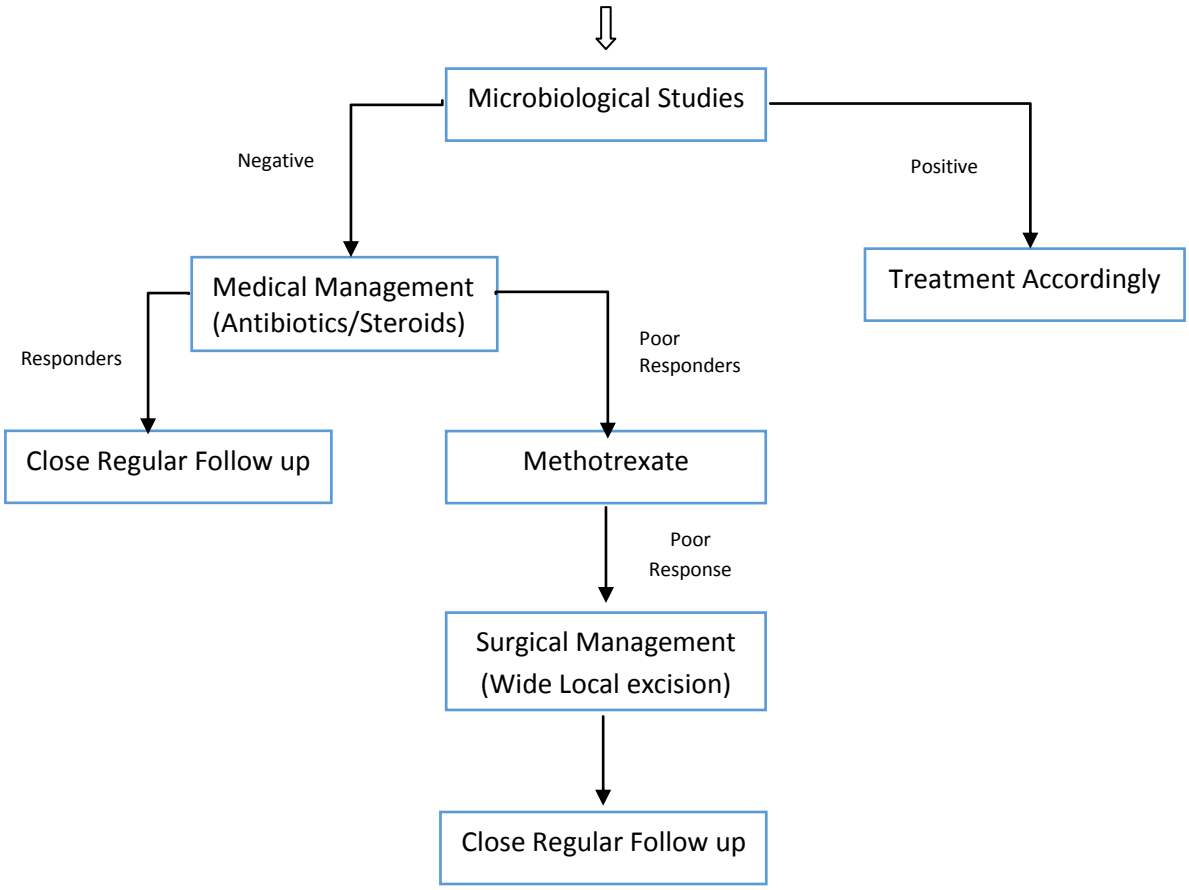
Differentials include

1. Tuberculosis of breast (granulomas, caseating necrosis)
2. Actinomycosis (yellow sulphur granules with filamentous structure and necrosis)
3. Coccidioidomycosis (granulomatous inflammation, large thick-walled spherules)
4. Histoplasmosis (granulomas with intracellular organisms)
5. Sarcoidosis (Non caseating granulomas, Asteroid bodies)
6. Foreign body reaction to silicone (Foamy histiocytes, giant cells, refractile structures)
7. Idiopathic granulomatous mastitis (Lobulocentric granulomas of unknown etiology)

Histopathological diagnosis of Idiopathic granulomatous mastitis



Thus, identifying and treating granulomatous mastitis poses a big challenge. Counselling and reassurance of the patient plays a vital role. Increasing awareness about this condition is necessary for patient follow- up. Early detection, appropriate histopathological diagnosis and standardising the treatment modality of granulomatous mastitis helps in limiting the spread and extent of the disease thereby preventing unnecessary mastectomies.

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