

A Fascinating Experience with A Rare Case of Granulomatous Mastitis

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Abstract

We Report a case of Idiopathic granulomatous mastitis in a 30 year old pregnant woman who came to our hospital with complaints of painful lump in her right breast because the results of initial aspiration cytology was mammary abscess with focal mastitis, incision and drainage was performed when MRI breast showed intermediate concern for malignancy - Excision biopsy was done. However the final histopathological diagnosis was granulomatous mastitis with no evidence of malignancy.

A review of literature revealed that idiopathic granulomatous mastitis has a tendency to affect young woman with a history of child birth on oral contraceptive usage. Clinical diagnosis and imaging has often been difficult. Complete resection or corticosteroid therapy can be recommended as appropriate treatment. Since recurrence rate is as high as 50%, long term follow up is indicated in such patients.

Keywords: Idiopathic granulomatous mastitis; Corticosteroid therapy; Pregnancy.

Introduction

Idiopathic granulomatous mastitis (IGM) is a rare, chronic inflammatory disease of the breast

of unknown etiology. IGM most commonly affects young, healthy women and usually presents as an ill-defined breast lump simulating malignancy. Women often present with painful, unilateral, discrete breast masses that have a tendency to recur.¹ IGM is also considered a variant of periductal mastitis by some experts.² The pathologic and imaging diagnoses of IGM remain difficult. Histologically, IGM demonstrates noncaseating granulomas limited to the mammary lobules with or without associated microabscesses resulting from a local immune process.³ Possible etiologies include an autoimmune process, trauma, infection, oral contraceptive use, and prolactinemia.^{4,5} IGM may also be confused with other conditions besides malignancy, conditions such as tuberculosis, sarcoidosis, erythema nodosum, and Wegener granulomatosis. Thus, it is important to confirm evidence of IGM on histopathology.⁶ The ideal treatment of IGM also remains unclear. Studies have demonstrated moderate success with varying options including observation, steroids, and immunosuppressants.⁷ Often, surgical management is the last resort, although lesions may recur and result in poor aesthetic outcomes.⁶

Case report

A 30-year-old woman, G2P1 with history of 3 months of amenorrhea, presented with painful lump in the right breast for the past 4 months, she also had history of febrile episodes. There was no family

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history of breast carcinoma. She delivered her last child 4 years before presentation and had breastfed continuously for 2 years. On physical examination, the patient was febrile (38.68 C), an ill-defined hard tender lump occupying upper medial quadrant with no associated nipple discharge or retraction. Left breast being normal. There was no appreciable axillary lymphadenopathy. Clinical diagnosis of focal mastitis was made and treated conservatively with ciprofloxacin and metronidazole elsewhere. Patient was symptomatically feeling better but still clinically palpable lump was present at the upper medial quadrant. Patient was treated again with linezolid after a gap of 10 days. Patient was brought to Bangalore with tender lump in the upper medial quadrant with bilateral lower limb edema. Patient was re-investigated. A comprehensive laboratory and radiologic workup was subsequently undertaken. Significant laboratory findings included a leukocytosis of 14,100 cells/cumm with neutrophilia (84%), ESR-54 mm/hr and CRP-74mg/L (normal range: 0-6mg/L). Sonographic evaluation revealed ill-defined area of altered echogenicity seen in upper inner quadrant extending into subareolar region of the right breast with mild hypoechoic thickening of the ducts and focal increased vascularity-findings suggestive of focal mastitis. Fine needle aspiration cytology revealed suppurative lesion-mammary abscess and advised excision biopsy to rule out malignancy. She was started on antibiotics and repeat ultrasound of the breast after a week revealed right breast extensive abscess showing fluctuant turbid fluid from 11-70 clock position with right axillary lymphadenopathy. As a result the decision of incision and drainage of the abscess was made and performed. Pus culture and sensitivity yielded no growth. Following which she had recurrence of her symptoms. MRI of the bilateral breast was done which showed right breast cystic/heterogeneous intraglandular space occupying lesions in the medial quadrant with deep seated lesion close to the intermammary cleft and associated marked regional non-mass enhancement. Type II/III time-signal intensity curves around the space occupying lesion of the right breast on a background of low ADC values-suggesting intermediate concern for malignancy (Differential includes-chronic organizing mastitis) with no significant finding in the left breast. Evaluation for tuberculosis were negative. The decision for excision biopsy of the right breast lump was made and performed under general anesthesia. (Fig 1 & 2) shows the intra operative pictures. Histopathology revealed Right breast-Cystic neutrophilic granulomatous mastitis

with adjacent breast showing fibrocystic disease. Postoperatively she was treated with antibiotics (augmentin) for a week and discharged. She started noticing hardness in the right breast after 10 days (Fig 3), hence decision taken to start oral corticosteroids (wysolone 10 mg tid for a week and tapering of the dosage was done thereafter). Patient responded very well and improved symptomatically with corticosteroid. After the corticosteroid therapy breast felt soft and no evidence of lump in the breast.



Fig. 1: Picture showing inflamed mammary tissue extensively with arrows showing prominent blood vessels.

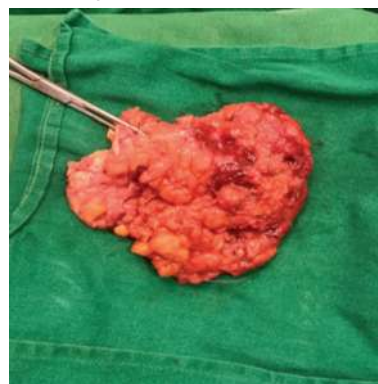


Fig. 2: Picture showing multiple cavities filled with greenish pus.



Fig. 3: Picture showing serous discharge (tissue fluid) from the sutured site.

Discussion

Kessler and Wolloch first described IGM in 1972 as a rare benign chronic inflammatory disease of the breast, characterized by presence of noncaseating granulomas confined to the breast lobules.⁸⁻¹² The exact pathogenesis of IGM is unknown, although it is thought to be immunologically mediated.⁸⁻¹⁴ IGM affects young healthy parous women and is often associated with pregnancy and lactation.⁸⁻¹² IGM is diagnosed after exclusion of infection, malignancy, foreign body reaction and other autoimmune disorders.⁸⁻¹² The absence of caseating necrosis and a predominantly neutrophilic background with multiple cavities on histopathology were important clues favoring diagnosis of IGM.¹³ IGM often has a chronic, relapsing course without an established standard treatment approach.⁸⁻¹² The principle treatment options include antibiotics, surgery, corticosteroid agents, or a combination of them.^{9,15-17} Some investigators suggest that systemic corticosteroid therapy may be the initial treatment of choice in IGM, whereas surgical resection should be reserved for recalcitrant lesions.^{12,15,18} The thought process behind this approach is that systemic corticosteroids usually melt the breast mass, allowing for a more conservative surgical resection should the disease persist.^{14,16,18} Nonsteroidal immunosuppressive drugs including azathioprine and methotrexate have been used successfully in managing IGM.^{9,12,14,18} Immunosuppressive therapy should be continued until a complete remission is reached, as recurrence rates as high as 50% have been reported.¹⁷ A recent prospective cohort study of 49 women with IGM found that the treatment period required to achieve a full remission varied from 3 to 18 months.¹⁵ Alternatively, some investigators propose that IGM should be treated with wide local excision at the onset of disease, citing a lesser chance of recurrence with surgical therapy.¹⁵⁻¹⁶ However, review of the literature finds that relapses can still occur despite surgical resection, and it may require repeated, potentially deformative surgeries to cure the condition.^{11,15} Prompt diagnosis and medical treatment of this rare condition is important, as it may prevent patients from undergoing potentially disfiguring surgery. We advocate for an initial trial of corticosteroid therapy, as it proved to be very successful in our patient.

Conclusion

After treating the patient with anti-biotics and surgical excision, the author noticed recurrence of symptoms and administered corticosteroid 10 mg

TID for 12 days with a tapering dose of 10 mg BD for 12 days and 10mg OD for 12 days. On assessment there was disappearance of tumor clinically. Hence the author recommends administration of corticosteroid to achieve symptomatic relief.

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