

Case Report

Bilateral idiopathic granulomatous mastitis a rare benign inflammatory disease of breast: case report

Talib Hussain^{1*}, Subash Bhardwaj²

¹Department of Surgery, ²Department of Pathology, Government Medical College Jammu, Jammu and Kashmir, India

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*Correspondence:

Dr. Talib Hussain,

E-mail: Chouhanh190@gmail.com

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ABSTRACT

Idiopathic Granulomatous Mastitis (IGM) is an uncommon benign inflammatory disorder of breast. Peri-ductal mastitis is thought to be the initial inflicting factor for its development. IGM usually presents as painful swelling, abscess or sinus/fistula in breast. There are no specific findings on ultrasound or mammographic imaging, and moreover it looks BIRADS-IV on mammography. Histopathology provides the final diagnosis. Here authors report a case of bilateral IGM which was successfully treated with surgery.

Keywords: Fine needle aspiration cytology, Histopathological examination, Idiopathic granulomatous mastitis, Nipple areola complex

INTRODUCTION

Idiopathic granulomatous mastitis was first described by Kessler and Wolloch in 1972.¹ Women of childbearing age with a recent history of pregnancy and lactation are usually affected.² This condition is labelled as idiopathic granulomatous mastitis after excluding other granulomatous conditions like tuberculosis, sarcoidosis, Wegener's granulomatosis.

Various etiologic factors have been described for its development. The clinical and radiological findings are not specific for IGM.³ Histopathology plays an important role in the final diagnosis of IGM.⁴

Treatment involves initial conservative management with or without oral corticosteroids and may require wide surgical excision with or without lactiferous ducts excision / incision and drainage of abscess or even total mastectomy.⁵ So, the management of IGM cases need to be tailored according to the clinical presentations.

CASE REPORT

Married female 37 yrs old, a known case of hypertension with history of recurrent gastritis presented to hospital in May 2019 with complaints of swelling in right breast from last one month with history of occasional pain and low-grade fever. Last childbirth was 3 yrs. back. GPE was unremarkable. Local examination revealed single, 5×5 cm, hard, non-tender, mobile lump in upper inner quadrant of right breast. Nipple areola complex was normal. All baseline investigations were normal. Mammogram right breast revealed BIRAD-IV category (Figure 1).

FNAC showed neutrophils, macrophages, lymphocytes, histiocytes and multinucleated giant cells along with epithelioid cell granulomas suggestive of granulomatous mastitis. Wide local excision was done via peri-areolar incision on 31st May 2019. IOF revealed hard inflammatory lump of about 8×5 cm in size containing thick white pus. Histopathology showed chronic lymph nuclear inflammation, lymphoid follicles, epithelioid cell

granulomas and multinucleated giant cells. No caseous necrosis seen (Figure 2).

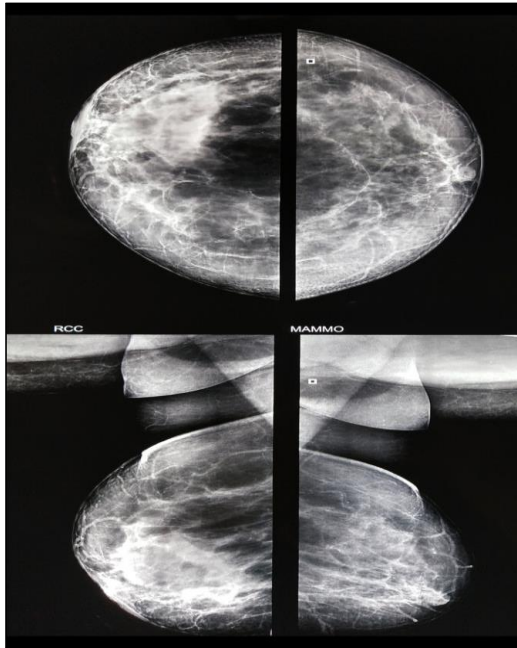


Figure 1: Mammogram showing irregular hyperdense lesion in supero-medial quadrant of right breast with thickening of overlying skin.

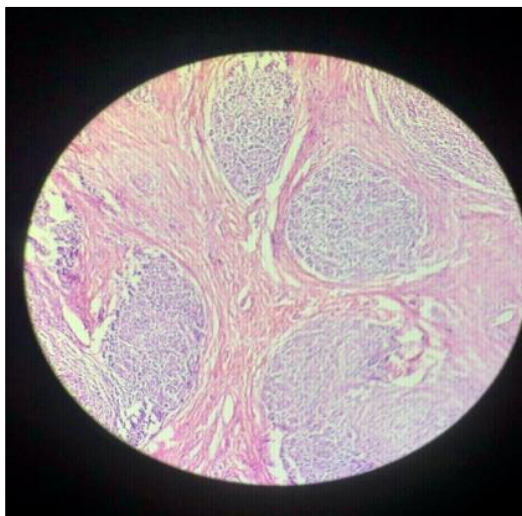


Figure 2: HPE showing chronic inflammatory cells, multinucleated giant cells and non-caseating epithelioid cell granulomas.

Ziehl-Neelson staining didn't show any acid-fast bacilli and culture of pus showed no organism.

Two months later patient developed another swelling in same breast in retro areolar region. Mammography showed chronic infection with abscess formation in retro areolar region. Wide local excision along with radical duct excision was done via peri-areolar incision (Figure 3).

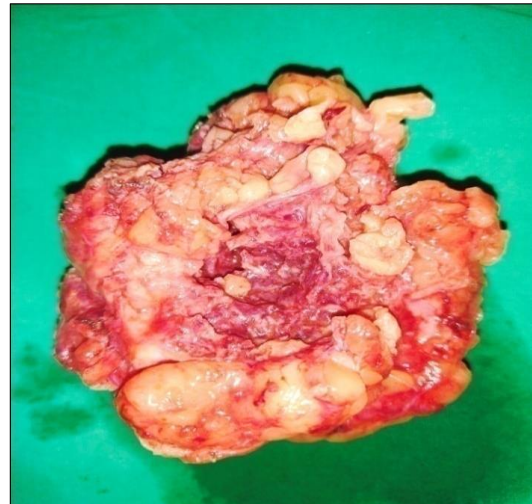


Figure 3: Excised specimen from right breast showing hard inflammatory mass with a central cavity which contained thick white pus.

The HPE showed acute on chronic mastitis along with foci of adenosis, large histiocytes and occasional multinucleated giant cells. Culture of pus showed no organism.

One and half month later patient developed tender swelling in upper inner quadrant of contralateral (i.e. left) breast with erythema of overlying skin. NAC was normal. Ultrasound breast showed multiple hypoechoic areas in retro areolar region and upper inner quadrant. Excision of inflammatory mass with radical duct excision was done (Figure 4 and 5).

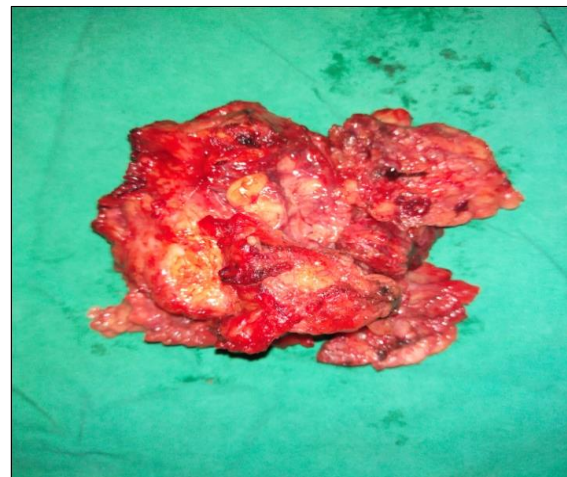


Figure 4: Excised specimen from left breast showing hard inflammatory mass with a central cavity which contained thick white pus.

HPE showed large dilated ducts surrounded by heavy chronic inflammation rich in eosinophils, plasma cells, lymphocytes, epithelioid cells and multinucleated giant cells. No caseation necrosis/neoplasia seen (Figure 6).

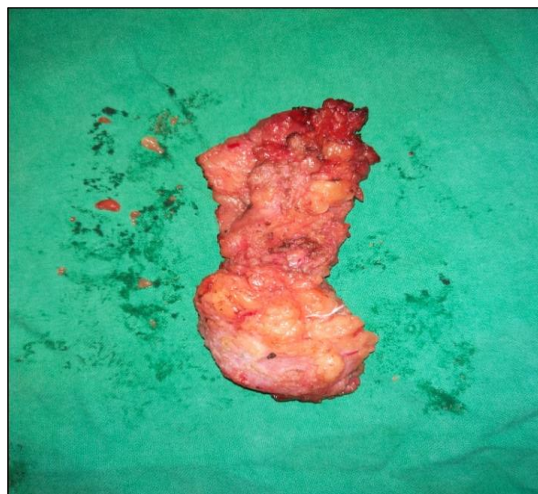


Figure 5: Excised lactiferous ducts from left breast.

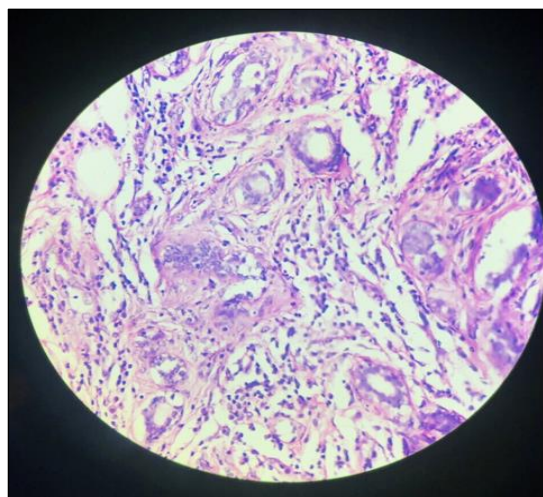


Figure 6: HPE of excised left breast lump showing inflammatory cells and multinucleated giant cells.

No Acid-Fast Bacilli detected in pus and no growth of organism detected after 48 hrs. of incubation. She was not put on steroids in view of history of recurrent attacks of gastritis. Patient is on follow up and doing well with invisible incision scars and symmetrical bilateral breasts despite of undergoing two surgeries on right breast.

DISCUSSION

The term granulomatous mastitis was introduced by Vessiere et al, and later described by Kessler and Wolloch in 1972.⁶ The mechanism of development of IGM is postulated to be initiated with ductal epithelial damage followed by extravasation of glandular secretions in to the connective tissue of the lobule resulting in local inflammatory lesions.⁷

The exact etiology is not known. There can be association with use of oral contraceptive pills, autoimmune diseases, pregnancy, breast feeding, breast trauma,

hyperprolactinemia, alpha-1 antitrypsin deficiency, infection with histoplasma and corynebacterium kropsenstedtii.⁸

IGM usually presents as progressive painful, firm, tender, ill-defined lump in any quadrant of breast and is unilateral. Bilateral involvement may be seen in 25% of patients⁹. It is rarely accompanied by axillary lymphadenopathy. IGM can cause skin thickness, sinus/fistula and abscess formation.¹⁰

IGM is a diagnosis of exclusion. The differential diagnosis includes Tuberculosis, sarcoidosis, foreign body granulomas, Wegener's granulomatosis, breast abscess and breast cancer.¹¹

Radiologic imaging findings are not specific for IGM. Ultrasound findings include large lobulated irregular hypoechoic mass with tubular extensions or distortion of the parenchyma with acoustic shadowing.¹² Mammographic findings include ill-defined mass, asymmetrically increased diffuse or focal density with parenchymal distortion or microcalcifications. MRI findings include round, smooth contoured mass-like lesions.¹³ However, MRI cannot differentiate between IGM and other granulomatous disorders.

Histopathology remains the most effective method of diagnosis. Findings include non-caseating granulomas, multinucleated langerhans giant cells, neutrophils, eosinophils, macrophages, epithelioid cells, fat necrosis and microabscess formation.¹⁴

There is no universally accepted treatment for IGM.¹⁵ Majority of patients undergo a combination of medical and surgical approaches. Surgery may be combined with corticosteroids and other immunosuppressants like methotrexate and azathioprine. Antibiotics have no role in the management of true cases of IGM.¹⁶ Hyperprolactinemia if present, should be evaluated and treated to prevent recurrence. The use of steroids was first proposed by Dehetrogh et al.¹⁷ Steroids are started at a dose of 1 mg/kg/day and tapered slowly according to clinical response, usually over a period of four weeks. Response usually occur within weeks of treatment, but patients may require treatment for several months. Surgical options include incision and drainage of abscess, wide local excision with or without lactiferous ducts excision and mastectomy. Limited excision alone has little benefit because of strong tendency for recurrence. Complete lactiferous ducts excision should be considered along with wide local excision in patients who have completed their families to prevent future development of peri-ductal mastitis and subsequent granulomatous mastitis.

CONCLUSION

IGM is an uncommon benign inflammatory disease of breast, which may resemble breast cancer both clinically and radiologically. Diagnosis is confirmed on histology.

Treatment is tailored as per clinical presentations. Due to the increased risk of recurrence, this rare disease can trouble both patient and treating surgeon. So, radical duct excision should be considered in patients who have completed their families to prevent future recurrence.

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REFERENCES

1. Kessler E, Wolloch Y. Granulomatous mastitis: a lesion clinically simulating carcinoma. *Am J Clin Pathol.* 1972 Dec 1;58(6):642-6.
2. 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 Mar 1;15(1):119-23.
3. Patel RA, Strickland P, Sankara IR, Pinkston G, Many W, Rodriguez M. Idiopathic granulomatous mastitis: case reports and review of literature. *J General Int Med.* 2010 Mar 1;25(3):270-3.
4. Akcan A, Akyıldız H, Deneme MA, Akgun H, Arıtas Y. Granulomatous lobular mastitis: a complex diagnostic and therapeutic problem. *World J Surg.* 2006 Aug 1;30(8):1403-9.
5. Asoglu O, Ozmen V, Karanlık H, Tunaci M, Cabioglu N, Igci A, et al. Feasibility of surgical management in patients with granulomatous mastitis. *Breast J.* 2005 Mar;11(2):108-14.
6. Veyssiere C, Vives M, Smadja A. Difficultés diagnostiques de la tuberculose mammaire. Le problème de la mastite granulomateuse. *Lille Chir.* 1967;22:104-9.
7. Akcan A, Akyıldız H, Deneme MA, Akgun H, Arıtas Y. Granulomatous lobular mastitis: a complex diagnostic and therapeutic problem. *World J Surg.* 2006 Aug 1;30(8):1403-9.
8. Taylor GB, Paviour SD, Musaad S, Jones WO, Holland DJ. A clinicopathological review of 34 cases of inflammatory breast disease showing an association between corynebacteria infection and granulomatous mastitis. *Pathol.* 2003 Jan 1;35(2):109-19.
9. Diesing D, Axt-Fliedner R, Hornung D, Weiss JM, Diedrich K, Friedrich M. Granulomatous mastitis. *Arch Gynecol Obstet.* 2004;269(4):233-6.
10. Brown KL, Tang PH. Postlactational tumoral granulomatous mastitis: a localized immune phenomenon. *Am J Surg.* 1979 Aug 1;138(2):326-9.
11. Katz U, Molad Y, Ablin J, Ben-David D, Paran D, Gutman M, et al. Chronic idiopathic granulomatous mastitis. *Ann N Y Acad Sci.* 2007;1108:603-8.
12. Anik Y. Radiological findings of idiopathic granulomatous mastitis. *Turkiye Klinikleri J Med Sci.* 2010;30(1):286-92.
13. Dursun M, Yilmaz S, Salmalıoglu A, Yavuz E, Igci A, Acunas G, et al. Multimodality imaging features of idiopathic granulomatous mastitis: Outcome of 12 years of experience. *Radiol Med.* 2012;117:529-38.
14. Baslaim MM, Khayat HA, Al-Amoudi SA. Idiopathic granulomatous mastitis: a heterogeneous disease with variable clinical presentation. *World J Surg.* 2007 Aug 1;31(8):1677-81.
15. Al-Khaffaf B, Knox F, Bundred NJ. Idiopathic granulomatous mastitis: a 25-year experience. *J Am Coll Surg.* 2008 Feb 1;206(2):269-73.
16. Wilson JP, Massoll N, Marshall J, Foss RM, Copeland EM, Grobmyer SR. Idiopathic granulomatous mastitis: In search of a therapeutic paradigm. *Am Surg.* 2007;73:798-02.
17. DeHertogh DA, Rosssof AH, Harris AA, Economou SG. Prednisone management of granulomatous mastitis. *N Engl J Med.* 1980 Oct 2;303(14):799-800.

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