



IDIOPATHIC GRANULOMATOUS MASTITIS - A CASE SERIES

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ABSTRACT Idiopathic granulomatous mastitis (IGM) is an uncommon benign inflammatory breast disease of poorly understood etiology. On evaluating clinically or radiologically, it can closely mimic two breast conditions frequently seen, i.e breast cancer or breast abscess. Some patients might undergo mastectomy due to high suspicion of breast cancer. Hence IGM should be considered while evaluating a painful breast mass. In this report we present 3 females who presented with painful breast lump in a tertiary care hospital over a period of 1 year with histopathological diagnosis of IGM and had remission with surgical management. **Conclusion** IGM is considered as a diagnosis of exclusion and can cause recurrence, many studies have reported the same and some have undergone remission with use of corticosteroids followed by surgical excision all three cases that we have studied here had disease localized to one quadrant of breast and had undergone remission by quadrantectomy and without postoperative use of steroids

KEYWORDS : Idiopathic granulomatous mastitis, Chronic granulomatous mastitis, mastitis, granulomas

INTRODUCTION

Idiopathic granulomatous mastitis (IGM) is chronic inflammatory condition of the breast (1) It is a rare presentation and is considered as a diagnosis of exclusion when specific infection, trauma, foreign body or evidence of sarcoidosis is ruled out. On histopathological evaluation granulomatous changes, non-caseating granulomas, around the breast lobules and ducts, with epithelioid histiocytes, multinucleated giant cells, plasma cells, lymphocytes, neutrophils, and eosinophils are observed. (2) There should be no evidence of specific infection, trauma, foreign body, or evidence of sarcoidosis to consider IGM as the diagnosis. (3,4). It has a poorly understood etiology. Possible risk factors include race (5), cigarette smoking, diabetes mellitus, obesity, breast trauma, infection, oral contraceptive pill (OCP) usage, lactation, autoimmune disease (6), hyperprolactinemia and immunologic response to milk leakage from the breast lobules (7). IGM usually affects women of childbearing age (8) It can mimic breast cancer clinically and by imaging-mammography. Some patients may even undergo mastectomy due to high suspicion of breast cancer. (9).

The present study included 3 patients with a histological diagnosis of IGM (granulomatous inflammation without caseous necrosis; giant cells; confined to breast lobules; no microorganism seen) treated between January 2023 and October 2023 at a tertiary care centre in Kerala.

Case 1 :

A 28-year-old female presented with 2 month history of painful lump in the right breast upper inner quadrant which was associated with nipple retraction. She had underwent incision and drainage of breast abscess recently followed by a course of empirically started antibiotics- Cefuroxime. On physical examination the patient was afebrile and there was a 6 x 3.5 cm tender lump in the upper inner quadrant of the right breast, nodular surface, well defined margins, firm in consistency, the skin overlying the mass showed linear scar of recent I & D, discharging sinus with purulent material in the medial end of the scar, the mass was fixed to the breast tissue and skin fixity was present, no pectoral or chest wall fixity. Axillary lymph nodes were not palpable. Systemic examination were within normal limits. Sonomammogram revealed well defined oval hypoechoic mass at 2 to 3 o'clock position which could represent fibroadenoma with cystic changes and infarction. FNAC from the lump was suggestive of granulomatous inflammatory changes. The patient failed to improve with antibiotics and subsequently underwent wide local excision of the breast lump. Per operatively, there was a firm mass with copious purulent discharge, involved lobules of the breast and intervening stroma were excised. Cultures and special stains for bacteria, mycobacteria and fungi done from this were negative. Histopathology, macroscopically the cut section appeared as focal grey white areas with granularity and haemorrhage, with areas of fibrosis. Microscopically the breast tissue showed adenosis and few dilated ducts, with some of the glands surrounded by dense

lymphoplasmacytic infiltrate. Intervening stroma showing dense mixed inflammatory infiltrate composed of lymphocytes, neutrophils, plasma cells, epithelioid macrophages and multinucleated giant cells with granuloma formation. She was continued with prolonged course of antibiotics. A decrease in the size of her breast mass was noted after 1 week, in follow up. The patient had an excellent clinical response with complete resolution of the breast mass on follow-up visit after one month.

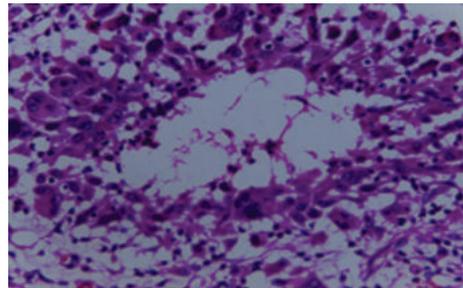


Figure- Epithelioid histiocytes And multinucleated giant cells along with lymphocytes, plasma cells forming attempted granuloma in the intervening stroma.

Case 2:

A 32 year-old female, presented with one month history of painful lump in the left breast upper inner quadrant. No history of nipple discharge, fever, loss of weight, loss of appetite, cough, evening rise of temperature. On physical examination the patient was afebrile and there was a 6 x 6 cm tender lump in the upper outer quadrant of the left breast, nodular surface, well defined margins, firm in consistency fixed to the breast tissue, skin over the swelling was normal. Axillary lymph nodes were not palpable. Systemic examination were within normal limits. Sonomammogram revealed well defined oval hypoechoic mass at 9 to 10 o'clock position, FNAC from the lump was suggestive of granulomatous inflammatory changes. The patient underwent wide local excision of the breast lump, and per operatively a firm mass 6 x 6 in the upper inner quadrant was identified with pultaceous material in the centre from which Cultures and special stains for bacteria, mycobacteria and fungi was sent and were negative. Histopathology, macroscopically the cut section appeared as a mass with multiple cystic spaces. Microscopically there were multiple granulomas composed of epithelioid histiocytes, multinucleated giant cells along with dense inflammatory infiltrates composed of lymphocytes, plasma cells and neutrophils. Patient was started on with a course of antibiotic pre and 1 week post procedure excision of the entire breast tissue in the upper inner quadrant, antibiotics given and on follow up the patient had improved clinically.

Case 3:

A 37-year-old female, presented with two month history of painful

lump in the left breast upper inner quadrant. No history of nipple discharge, fever, trauma to breast. On physical examination the patient was afebrile and there was a 8 x 8 cm tender lump in the upper outer quadrant of the left breast, nodular surface, well defined margins, firm in consistency fixed to the breast tissue, skin over the swelling was normal. Axillary lymph nodes were not palpable. Systemic examination were within normal limits. Sonomammogram reported as irregular shaped non-circumscribed heterogeneously hypoechoic wider than taller solid lesion at 11 to 12 o'clock position in periareolar region of left breast. FNAC from the lump was suggestive of suppurative inflammatory lesion possibly subareolar abscess. Core needle biopsy was done suggestive of granulomatous mastitis. The patient underwent wide local excision of the breast lump, and per operatively a firm mass 8 x 8 cm in the upper outer quadrant was identified. Cultures and special stains for bacteria, mycobacteria and fungi was sent and were negative. Histopathology, macroscopically the cut section appeared as a mass granular projection and areas of hemorrhage and cystic degeneration. Microscopically there were multiple collections of epithelioid histiocytes forming granuloma along with multinucleated giant cells in lobulocentric pattern, intervening area shows dense inflammatory infiltrate composed of neutrophils, lymphocytes, plasma cells and macrophages along with adipose tissue multiple dilated and congested blood vessels. Patient was started on with a course of antibiotic pre and 1 week post procedure, anti inflammatory drugs given and on follow up the patient had improved clinically.

The present study included 3 patients with a histo-logical diagnosis of IGM (granulomatous inflammation without caseous necrosis; giant cells; confined to breast lobules; no microorganism seen) treated between January 2022 and October 2023 at a tertiary care centre in Kerala. All three females were in reproductive age and parous, without a family history of breast disease or cancer. None had history of smoking and alcohol use all three had history of breast feeding within past 5 years and had presented with painful breast lump. One of them had nipple retraction and recurrence with sinus formation. In these patients we had found out clinically and radiologically that the lump was confined within a particular quadrant and none of them had axillary lymphadenopathy. FNAC was suggestive and biopsy reports were consistent with Idiopathic Granulomatous Mastitis following wide local excision of the mass, which were limited to a quadrant.

DISCUSSION

IGM is an idiopathic breast condition where there is granulomatous inflammation of the breast, IGM remains a diagnosis of exclusion. Factors like IgG-related disease(7), hyperprolactinemia(8) are discussed in relation to the pathogenetic basis of IGM which may explain the variation in response to treatment and recurrence pattern. The affected females were in reproductive age group, previously given birth and had lactation which suggest an association with hormone status for the disease and this may be a localized immune response to the secretion that has extravasated from the breast lobule.

According to the study by Memis et al(10) asymmetrically increased density in mammography was the pattern seen in majority of the studied cases. Mass-like appearance, tubular/nodular hypoechoic structures and focal decreased parenchymal echogenicity with acoustic shadowing are some of the other patterns observed. In the present study we had seen patterns mimicking fibroadenoma, well defined hypoechoic lesions and illdefined hypoechoic lesions in mammography. In the West treatment usually starts with antibiotics and steroids and in cases where not attaining remission surgical management is planned(11). Erozgen et al, from their study concluded that even though corticosteroids are seen to be beneficial in the treatment of IGM, surgical excision is considered to be the treatment of choice if indolent infection or fistula develops(12). Atak et al in their study also considers surgery as a mainstay management but suggests the use of corticosteroids preoperatively to minimize the size of a large lesion(13). In a study conducted in Bahrain among 29 cases which were surgically treated, excision, debridement and retroareolar ductal system removal, none of them had recurrence.

In the present study all three cases underwent excision of the involved breast tissue and lobules with intervening stroma, limited within a particular quadrant. In our study a good recovery was observed over 6 months follow up after quadrantectomy. According to the results of the present study and our review of the literature suggesting wide excision of the involved breast tissue for the patients with suspected IGM, excision can be considered as a diagnostic and therapeutic procedure ensuring that a significant breast volume is not lost and

avoiding inadvertent use of steroids and steroid induced complications.

Finally, as Luqman, et al (14) and Hasan, et al(15) have both reported cases of breast cancer occurring in association with IGM, we also suggest long-term follow-up and thorough investigation of patients diagnosed with IGM to avoid missed cancer diagnosis.

CONCLUSION

IGM is an under-recognized benign inflammatory breast disease, mainly affecting women of childbearing age. Patients commonly present with palpable mass and pain at any location of the breast. IGM may present within 5 years of the last breastfeeding period. Histopathological evaluation of the tissue is the gold standard to confirm the diagnosis. Corticosteroids and wide excision play major roles in management, an extensive or recurrent disease might need mastectomy to cure, but a beneficial result can be obtained by quadrantectomy when the lesion is limited to a particular quadrant and follow up is required in such patients. IGM should be hence be considered in the differential diagnosis while evaluating a painful breast mass.

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