

Diagnosis of Idiopathic Granulomatous Mastitis in a Sudanese Woman

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Abstract

Idiopathic granulomatous mastitis (IGM) is a relatively rare specific chronic inflammatory process of unknown etiology, that diagnostically overlaps with common breast pathologies in Sudan, namely breast cancer (BC) and tuberculous mastitis (TBM). We report the case of a 34-year-old female who presented with a 1-month history of a painful lump in the lower outer quadrant of her left breast. A tru-cut biopsy showed features of granulomatous inflammation suggestive of IGM. Four months later, she presented with similar features and fine-needle aspiration cytology (FNAC) confirmed the presence of IGM and excluded the presence of both, BC and TBM. Histology once again confirmed the diagnosis of granulomatous mastitis with no evidence of breast cancer. Grocott's Methenamine Silver, Ziehl-Neelsen stain, and polymerase chain reaction were negative and accordingly the possibility of fungal infection and TBM were excluded. To our knowledge, this is the first case report of IGM in Sudan. FNAC helped in correct diagnosis of our case and importantly, conditions such as BC and TBM were both excluded as common mimickers of IGM. Although breast biopsy is the main golden approach in the diagnosis of IGM in addition to the usefulness of adjunct ancillary microbiological techniques, still further research is needed to establish whether FNAC can be a reliable tool in the diagnosis of IGM with the common practice of this diagnostic tool in Sudan.

Keywords: Breast cancer, idiopathic granulomatous mastitis, tuberculous mastitis

INTRODUCTION

Idiopathic granulomatous mastitis (IGM) is a rare chronic inflammatory disease of the breast that can clinically mimic breast carcinoma (BC) and tuberculous mastitis (TBM), as common breast lesions in Sudan. In >50% of reported cases, the initial diagnosis was considered malignant or suspicious for BC.^[1]

Since IGM was described for the first time as a distinct pathological entity of the breast in 1972,^[2] it represents both diagnostic and therapeutic dilemma for the clinicians worldwide; it is almost impossible to differentiate IGM from BC clinically and radiologically, and IGM is a diagnosis by exclusion, so it also needs to rule out other granulomatous lesions with cytological and histopathological overlap.^[3] This disease is characterized by chronic, non-caseating granulomatous lobulitis of unknown etiology.

Failure to recognize granulomatous mastitis, which may be consequently mistaken for breast carcinoma, has previously resulted in un-necessary mastectomies or open biopsies.^[4]

In Sudan, both BC and TBM represent common malignant and benign IGM mimickers, respectively, that necessitate reliable diagnostic tool to rule them out, as breast cancer is the most common cancer and represents one-third of investigated breast lesions at cytology clinic in Sudan.^[5-7] Our aim is to report a case of IGM, highlighting how fine-needle aspiration cytology (FNAC) is reliable, simple, and less invasive diagnostic modality, which can help in prompt diagnosis of IGM and differentiating it from BC and TBM as common pathological mimickers in Sudan.

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CASE REPORT

We report a case of 34-year-old mixed African female who presented with a 1-month history of a painful lump in the lower outer quadrant of her left breast. There was no discharge or overlying breast skin changes. She had two children and both were breastfed; her last child was born 6 years ago and breastfed for 2 years. The patient had no significant medical past history and denied previous use of oral contraceptives, estrogens, smoking, alcohol, or recent breast trauma. The patient consented for the publication of her case report.

She was febrile; her temperature was 38.5°C and had normal blood pressure. On physical examination, there was a mobile 3 cm in diameter left breast mass with obvious signs of inflammation that involving the overlying skin, while the ipsilateral axillary lymph nodes were not palpable. A tru-cut biopsy histopathological examination showed mixed inflammatory infiltrate centered around mammary lobules, composed of lymphocytes, plasma cells, scanty neutrophils, and aggregates of epithelioid histiocytes forming ill-defined granulomas [Figure 1A and Figure 1B] that revealed features of granulomatous inflammation. In addition Grocott's Methenamine Silver and Ziehl–Neelsen were all negative. The patient was discharged without antibiotics or steroids and advised to present 2 months for follow-up. She had an excellent clinical response with complete resolution of the breast mass 8 weeks later.

At the 16th week, she was admitted to the hospital again with recurrent hard breast mass; this time it appeared in the outer upper quadrant of the left breast, with nipple retraction and peau d' orange sign, but still the axillary lymph nodes were not palpable. Radiological examination performed which showed an irregular hypoechoic soft-tissue mass, widely speculated and measuring 6.3 cm × 4.4 cm in dimensions; the picture was similar to that of inflammatory breast

carcinoma, and it interpreted as suspicious left breast mass. FNAC was done and the aspirate revealed cellular smears with predominance of neutrophils, scattered necrotic tissue fragments and aggregates of cohesive epithelioid histiocytes or granulomas in a background of lymphocytes and Langhans giant cells [Figure 2]. This again confirms IGM even without provision of previous medical history. A polymerase chain reaction test for mycobacterium showed negative result; the chest X-ray was in normal limits and Montoux test for tuberculosis was insignificant.

Since that time, the patient had an excellent clinical response with complete resolution of the breast mass and there were no signs of recurrence. Now she is kept under regular follow-up and there is no recurrence of the disease in a period of 15 months.

Figure 3 depicts the comprehensive diagnostic procedures that were applied in our reported case.

DISCUSSION

IGM is considered a peculiar pathological entity for which no etiological factors have been identified. IGM may reveal similar presentation to BC and TBM. It is worth mentioning that breast cancer in Sudanese women represents almost 30% of the total FNAC performed in Sudan.^[5-7] Other causes of GM, such as fungal infections, foreign-body reaction, sarcoidosis, and Wegener's granulomatosis, are rare but still should be excluded.^[8] Different diagnostic modalities are in use to grasp the final diagnosis of IGM including; chest X-ray, ultrasonography, magnetic resonance imaging, cytology, histopathology, microbiological studies, and molecular tests.^[9] The treatment of patients with IGM varies significantly as it can last up to 12 months before complete healing occurs. Different approaches of management were used for instance, conservative/supportive care, oral prednisone or methotrexate, or surgical excision.^[10] In this case report, the

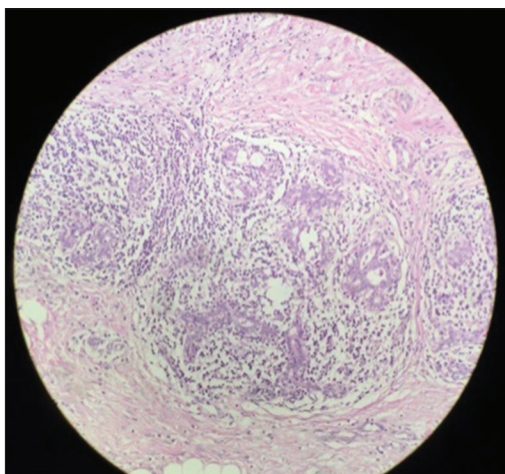


Figure 1A: Histopathological examination showing mixed inflammatory infiltrate centered around mammary lobules, composed of neutrophils, lymphocytes, plasma cells, and aggregates of histiocytes forming ill-defined granulomas (H and E, ×10)

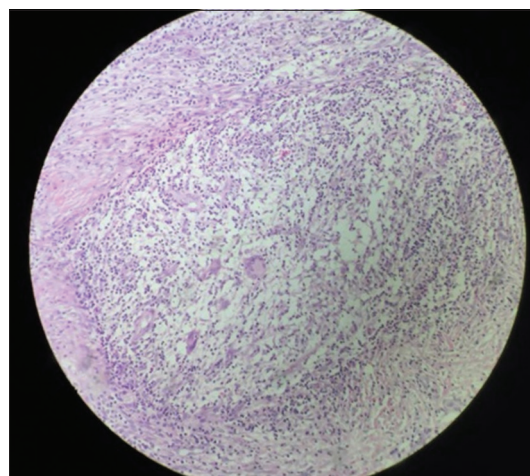


Figure 1B : Histopathological examination showing granulomatous reaction with scattered lymphocytes, plasma cells, multi-nucleated Langerhans giant cells and epithelioid histiocytes cells

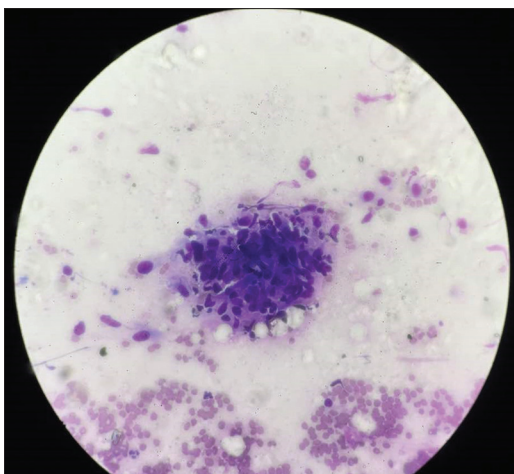


Figure 2: Fine-needle aspiration cytology smear showing a background of chronic inflammatory cells mainly lymphocytes and cohesive aggregates of epithelioid histiocytes (Diff Quick, ×40)

disease recurred after 4 months but healed with supportive treatment. Interestingly, IGM was reported in 13 males can also be associated with Bacillus Calmette–Guerin treatment for bladder cancer.^[11,12]

To our knowledge, this is the first case report of IGM in Sudanese patient. Importantly, we have used FNAC, which showed similar outcome to histology. As well FNAC has excluded BC and TBM as common mimickers of IGM. It worth mentioning that the absence of background caseous necrosis is an important cytological findings in exclusion of TBM. Further research is needed to establish whether FNAC can be used as diagnostic tool for IGM. This due to the fact that the usefulness of FNAC in granulomatous mastitis has been debated, while some authors confirming the useful role of FNAC, others have concluded that the various causes of granulomatous inflammation cannot be confidently differentiated by FNAC.^[4,13]

CONCLUSION

This the first case report of IGM in a Sudanese patient. FNAC is useful in establishing the correct diagnosis and comparable to histology as the gold standard in the diagnosis of IGM. Further microbiological and molecular assays are necessary in order to exclude TBM and other tropical diseases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initial will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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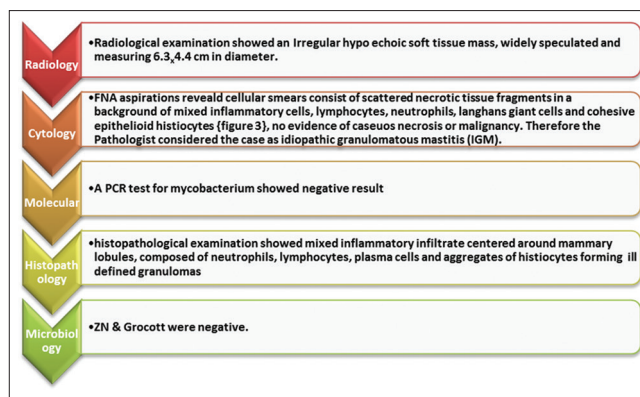


Figure 3: Idiopathic granulomatous mastitis comprehensive diagnostic procedures

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Conflicts of interest

There are no conflicts of interest.

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