CASE REPORT

Chronic granulomatous mastitis with axillary lymphadenopathy in a nulliparous woman

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SUMMARY

Chronic Granulomatous Mastitis (CGM) is a rare disorder and this generally occurs in young women with a recent history of lactation. Development of this disease in nulliparous women are rare with an incidence of 10%. Axillary lymphadenopathy is seen in 15% of cases diagnosed with CGM. We present a case of CGM in a 23-year-old nulliparous woman presenting with a breast mass and multiple axillary lymphadenopathy. To the best of our knowledge there are no documented cases of both these rare clinical features occurring simultaneously. The use of oral steroids prevented surgery and effectively induced remission.

INTRODUCTION

Chronic Granulomatous Mastitis (CGM) is a benign condition which may mimic breast carcinoma. First described as a case series of five patients by Kessler and Wolloch in 1972, all patients were parous, of the childbearing age group and presented with a painless, firm to hard breast mass without axillary lymphadenopathy. The authors highlighted the importance of excluding a carcinoma and avoiding a mastectomy in their relatively young group of patients. Now, CGM is a recognised benign inflammatory breast disease which can be managed medically. We describe an atypical presentation of CGM with axillary lymphadenopathy in a young nulliparous lady.

CASE REPORT

A 23-year-old nulliparous, Malay nurse presented with complains of a right breast and axillary swelling of 3-month duration. The breast lump had increased in size prompting a clinical consultation. She had no nipple discharge, fever, cough, loss of appetite or weight. She had no past or family history of malignancy and tuberculosis.

On examination a 3x4cm mobile lump was palpable at the upper outer quadrant of the right breast with multiple enlarged axillary nodes. Ultrasound guided core needle biopsy of the breast lump and axillary node confirmed granulomatous inflammation in both locations. Histopathology reported tissue inflammation with neutrophils, plasma cells, lymphocytes and granuloma formation consisting of epithelioid macrophages, multinucleated giant cells and areas of necrosis. There were no fungal bodies or acid fast bacilli demonstrable. Blood investigations for autoimmune and infective screening taken was unremarkable except for an elevated C-reactive protein of 6.3. Following a trial of oral Azithromycin for one week, a repeat ultrasonogram demonstrated a reduction in the size of axillary lymph nodes with no change in size of the breast lump.

She was referred to the respiratory and infectious disease physicians to rule out Tuberculosis (TB) and an underlying systemic fungal infection. A repeat core needle biopsy was done for TB PCR, TB culture, fungal culture and histopathology. At the time of repeat biopsy, 20mls of frank pus was aspirated from a fluctuant swelling detected on clinical examination and sent for culture as well. Mantoux test was positive and anti-tuberculosis treatment was initiated. All other tests were negative and histopathology again reported granulomatous mastitis with features similar to the initial biopsy. Anti-tuberculosis treatment was stopped and she was started on oral prednisolone 30mg daily. At clinic review five days later there were no palpable breast lump and axillary lymph nodes were smaller. After two months of tapering dose of prednisolone, clinical and sonographic examinations revealed no breast lesion with only a 0.9x0.5cm impalpable axillary lymph node.

DISCUSSION

CGM is a chronic inflammatory disorder affecting the breast of unknown aetiology. Autoimmune, hormonal and infectious pathophysiologies have been proposed but not substantiated. Patients are young, parous women in the childbearing age group usually but not exclusively with a history of recent lactation. We describe here the atypical occurrence of this disease in a nulliparous woman. Previous studies have demonstrated CGM in nulliparous women with an incidence of 10%. These patients tend to be outliers either of the pubertal or geriatric populations.[1] Clinically it can mimic breast cancer presenting as a painless, firm to hard mass with overlying skin changes including erythema, ulceration, sinus tract formation and nipple retraction. It may also present as recurrent breast abscesses. Axillary lymphadenopathy is seen in 15% of cases.[2] Interestingly, axillary lymph node biopsy in our case also demonstrated features of granulomatous inflammation prompting multidisciplinary team involvement and more aggressive investigation for a tuberculous and autoimmune aetiology.

Diagnosis is by exclusion of other granulomatous diseases including tuberculosis, fungal infections, foreign body reaction and sarcoidosis.[3] Imaging studies are not specific

This article was accepted: 19 August 2019
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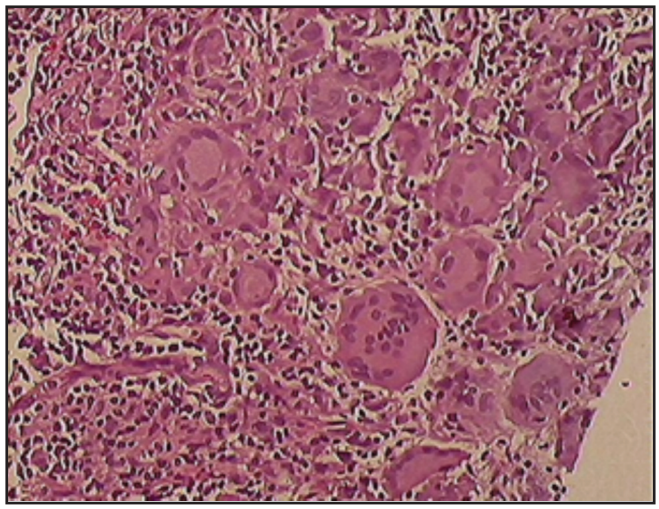

Fig. 1: Multinucleated giant cells (HPEX100).

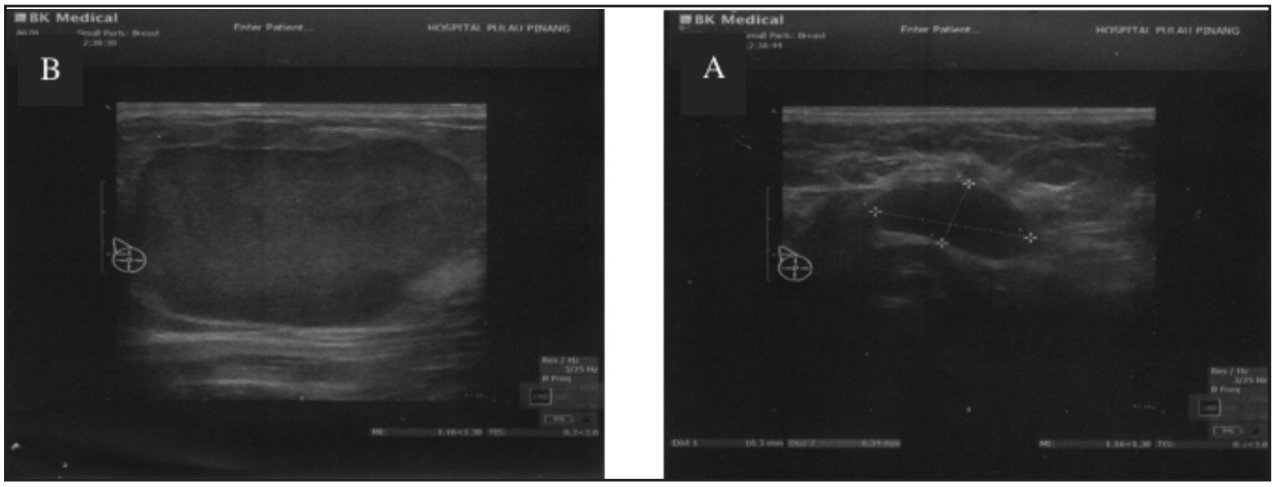

Fig. 2: Initial Sonographic Images of Breast(A) and Axillary(B) lesions

and often report a breast lesion of suspicious morphology. Mammogram, if performed, generally reports a suspicious lesion of at least BIRADS 4A. Ultrasound demonstrates an irregular hypoechoic lesion with skin thickening and subcutaneous edema.[4] Demonstration of non-caseating granulomas with epithelioid histiocytes, multinucleated giant cells centred on a breast lobule and abundant plasma cells, lymphocytes and neutrophils is diagnostic.[5]

Medical treatment with oral steroids can attain remission in over 60% of cases, highlighted by its successful use in this case. For disease relapses while on steroids or resistant cases, addition of methotrexate while gradually tapering the dose of oral steroids is usually successful.[4]

Surgery is indicated in cases with co-existing abscess formation which needs drainage and treatment with antibiotics prior to initiation of steroids or methotrexate. Surgery is able to eradicate most of the disease and reduce recurrence rates but this has to be weighed against the risks of infection, persistent sinus and aesthetic concerns of the patient. It should be the last resort in patients who fail medical therapy.

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