Diagnostic Dilemma Of A Rapidly Progressive Ulcer Of The Breast: A Case Report

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Citation

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Abstract

Idiopathic granulomatous mastitis is a rare inflammatory breast disease of unknown etiology. It tends to occur in young females usually with a history of recent childbirth or oral contraceptive usage. The clinical presentation is varied and may mimic malignancy. It may be identified by presence of epithelioid histiocytes, lymphocytes, plasma cells, neutrophils and multinucleated Langhans-type giant cells usually without any caseation or necrosis.

INTRODUCTION

Idiopathic granulomatous mastitis was described as a specific entity in 1972 by Kessler and Wolloch. Despite many cases reported in the international literature, this pathology remains quite unknown.[1] It is a rare inflammatory breast disease of unknown aetiology and tends to occur in young female patients with a history of recent childbirth or oral contraceptive usage.[2] Histopathologically, it may be identified by presence of epithelioid histiocytes, lymphocytes, plasma cells, neutrophils and multinucleated Langhans-type giant cells usually without any caseation or necrosis.[3,4] It usually presents with one or more of clinical features like galactorrhea, inflammation, breast mass, tumorous indurations and ulcerations of the skin.[5]

CASE HISTORY

A 20-year-old female presented to us with complaints of a large ulcer in the region of left breast with sloughed-off areola and skin. The lesion had started as a swelling in the left breast below the nipple-areola complex about 2 month back which was small initially and then rapidly increased in size followed by sloughing off the nipple-areola complex with eventual progression to sloughing -off of the entire breast tissue. The patient had had full term normal delivery 2 months before. On clinical examination there was a large ulcer of about 12cm x 10cm size in the left pectoral region replacing much of the bulk of the left breast with slough and necrotic tissues. The floor gave appearance of a fungating mass. Local temperature was slightly increased, tender to touch and the ulcer was not fixed to underlying structures. No axillary or other lymph nodes were palpable. Imaging studies of chest and abdomen were normal. A provisional diagnosis of locally advanced breast malignancy was made. Biopsy showed congestion with chronic inflammatory infiltrate consisting of lymphocytes, plasma cells, histiocytes and Langhans-type of giant cells. Keeping idiopathic granulomatous mastitis as provisional diagnosis, the patient was treated conservatively.

Figure 1

Figure 1: Ulcer replacing left breast

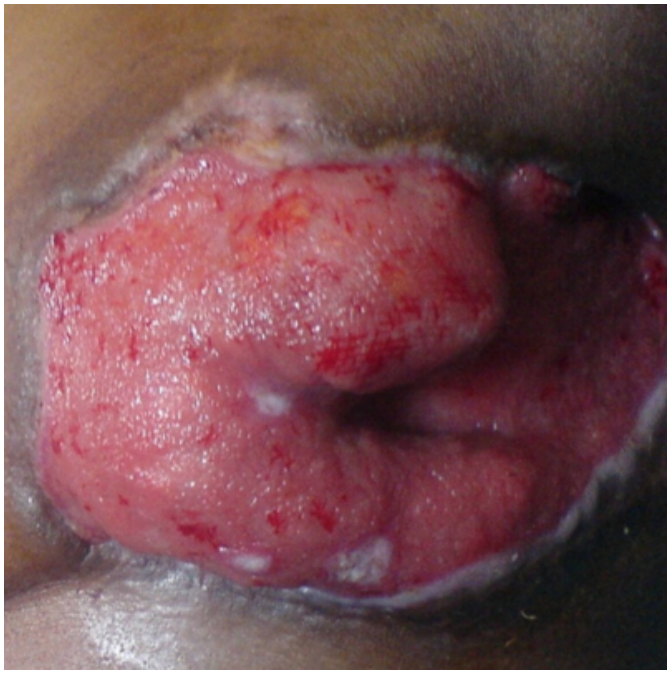
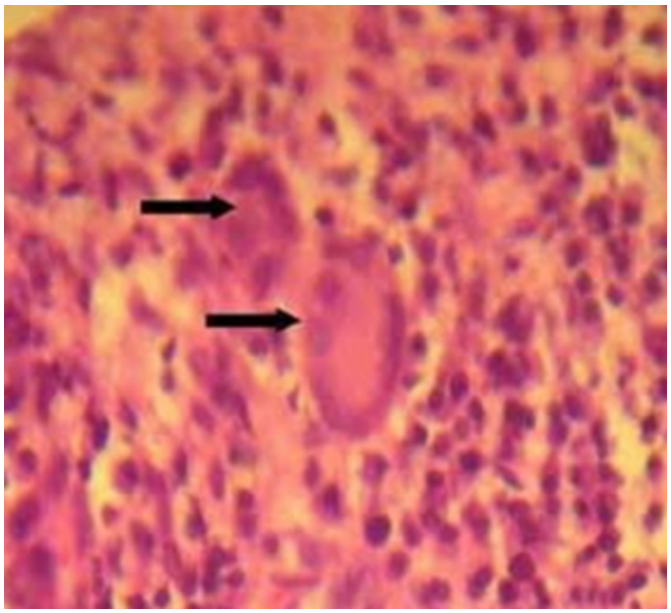


Figure 2

Figure 2: Histological slide showing giant cells



DISCUSSION

Granulomatous mastitis is a rare disease, which predominantly occurs in premenopausal women shortly after their last childbirth. Its aetiology is unclear, however, the disease has been shown to be correlated with breast-feeding and the use of oral contraceptives. An autoimmune aspect has also been considered.[5] A localized immune response to extravasated secretions from lobules may be considered to play a role, since many patients have previously given birth or were lactating at the time of the initial symptoms. In a recently reported case, immunohistochemical staining showed that the lesion contained predominantly stromal T lymphocytes. It is possible that damage to the ductal epithelium produced by local trauma, a local chemical irritant, or viral infection caused a localized immune response, and induced lymphocyte and macrophage migration. However, systemic immune abnormalities such as formation of autoantibodies or antigen-antibody complexes have not been reported so far.[2]

It usually presents with the clinical symptoms of breast mass, galactorrhea, inflammation, tumorous indurations and ulcerations of the skin. Breast mass is the most common presentation. The lesions are usually unilateral and known to occur in every quadrant region except for the subareolar region. Most of the patients are relatively young parous women below 50 years of age. About one third of the patients are found to have a previous history of use of oral contraceptives. The period between the most recent delivery and the initial complaint of a breast mass ranges from a month to 8 years. The reports suggest that most of the patients suffer from hormonal perturbation.[1,2]

Since the clinical manifestations simulate mammary carcinoma, this condition has sometimes been misdiagnosed as a malignancy and treated as such. Idiopathic granulomatous mastitis should be differentiated from other chronic inflammatory breast diseases such as mammary duct ectasia (plasma cell mastitis, subareolar granuloma and periductal mastitis), Wegener's granuloma, sarcoidosis, tuberculosis and histoplasmosis.[2] Puerperal mastitis is a condition thought to originate from reflux of bacteria into the milk containing breast by the nursing infant. It is especially prevalent during periods of increased engorgement such as weaning and so requires special consideration in the above scenario.

Histolopathologically, the condition is characterized by chronic lobulitis with granulomatous inflammation. Findings consist of a non-caseating granulomatous inflammation, centred on breast lobules, composed of epithelioid cells and multinucleated Langhans-type and foreign body type giant cells along with lymphocytes, plasma cells, macrophages and polymorphonuclear cells as well as abscesses.[1,2,3,5,6,7] In mammography and sonography, nodular opacities and hypoechoic nodules are found.[5] Absence of caseation necrosis helps in differentiating the entity from tuberculosis though confirmation is only after AFB staining or other specific tests for mycobacteria.[6]

Therapeutic options include a course of oral corticosteroids, non-steroidal anti-inflammatory drugs or colchicines to reduce the size of the lesion followed by conservative surgery. Local excision alone may lead to recurrence.[1,5] Oral steroids alone may be offered.[2] Methotrexate has also been found to be successful.[5] Complete remissions have been reported even with only expectant management without any specific medications.[7]About 38% of patients have been reported to experience recurrence, so long-term follow-up is indicated.[2]

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