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CASE REPORT

A case of recurrent idiopathic granulomatous mastitis

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ABSTRACT

We report a case of 46-year-old female patient who presented with a breast lump and was diagnosed as a case of idiopathic granulomatous mastitis. Patient was put on steroid therapy and her lesion completely disappeared, both clinically and radiologically. When we stopped the steroid therapy, the mass again recurred and patient had to undergo lumpectomy with no recurrence in follow-up of 1 year.

Key words: Granulomatous, idiopathic, mastitis

Introduction

Idiopathic granulomatous mastitis (IGM), also called as granular lobular mastitis, was first described in 1972 by Kessler and Wolloch [1]. The relationship of granulomatous mastitis to pregnancy or lactation has been strongly emphasized because this disease entity typically affects younger women, usually within 6 years of pregnancy [2,3]. Granulomatous mastitis generally manifests as a distinct, firm to hard mass that may involve any part of the breast but tends to spare the subareolar regions [2,3]. Radiologically, these lesions could be misdiagnosed as carcinoma. Awareness among the clinicians, radiologists and pathologists about this disease entity is required and multi-disciplinary approach is imperative to establish the diagnosis. We herein report a case of granular lobular mastitis to emphasize that this uncommon but not rare disease should be considered when usual therapy for breast lesion does not improve symptoms. Thus to prevent the morbidity of delayed diagnosis and misguided therapy, this entity should be considered in the differential diagnosis of any breast mass, especially in women of child bearing age.

Case Report

This was case report of a 46-year-old female patient who presented to our clinic with a history of painful right breast mass of few months duration. There was no family history of breast or endometrial cancer, nor any history of trauma. She was pregnant at the time of presentation (gravida 3, para 4) and her youngest third child was of 6 months age. Her first and second children were 18 and 10 years old. She had a positive history of oral contraceptive use for almost 10 years which was stopped a few months before her third pregnancy.

Clinical examination was remarkable for the presence of a palpable mass involving almost half of the right breast, extending from the right upper quadrant to the right nipple. The mass was painful with no evidence of any skin changes or axillary lymphadenopathy. The left breast was normal on examination. Routine blood investigations including hemogram, liver and renal function tests were unremarkable. Mammogram of the breast showed an irregular breast mass lesion with speculated irregular margins at the upper outer quadrant of the right breast, without any abnormality on the left side [Figure 1]. The right breast mass lesion had no evidence of micro-calcification and was classified on "breast imaging, reporting and data systems" as having a score of four. Breast ultrasound revealed the presence of an irregular hypoechoic mass lesion on the right breast at 9 O'clock position with tubular extensions and a striated echo-texture with normal appearing left breast. The magnetic resonance imaging (MRI) of the breast showed dysplastic enhancing right breast tissue with focal ductal areas which demonstrated contrast enhancement at the retro-areolar region, with right axillary lymph nodes having a benign radiological criterion [Figure 2]. Since all the radiologic investigations suggested malignancy, a Tru-cut biopsy [Figure 3] was performed which showed a non-classified granuloma inside breast lobules with inflammatory infiltration composed of epithelioid histiocytes, eosinophils, polymorphonuclear leukocytes and plasma cells. The presence of plasma cells was a feature suggestive of IGM which was later confirmed histologically after exclusion of other diseases, including but not limited to, tuberculosis, sarcodosis, parasitic infection and Wegener's granulomatosis.

The patient was treated initially with oral prednisolone 30 mg/day for 6 weeks with complete remission of the mass both clinically and radiologically and the drug was tapered until it was stopped. The patient was observed over a period of 2 months during which the mass started to reappear again until it reached to almost 2.0 cm. At this stage because of recent recurrence in short period of time, a wide local excision (breast conserving surgery) lumpectomy was done with negative margins. The histological diagnosis re-confirmed the initial diagnosis of IGM. The patient had 1 year of follow-up (4 visits/year almost every 3 months) with no clinical or radiological recurrence.

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CASE REPORT

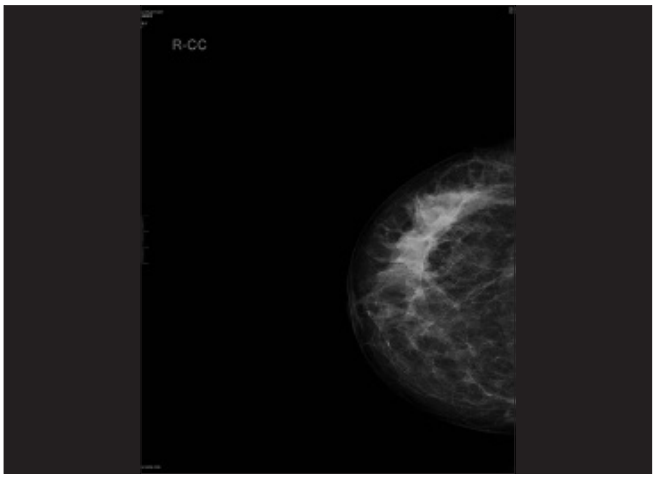

Figure 1 Mammogram showing irregular right breast mass lesion with speculated irregular margins

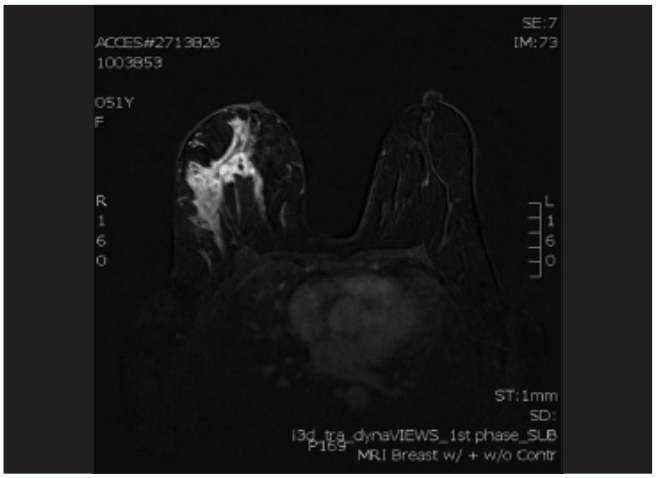

Figure 2 Magnetic resonance imaging showing dysplastic right breast tissue with focal ductal areas demonstrating contrast enhancement at the retro-areolar region

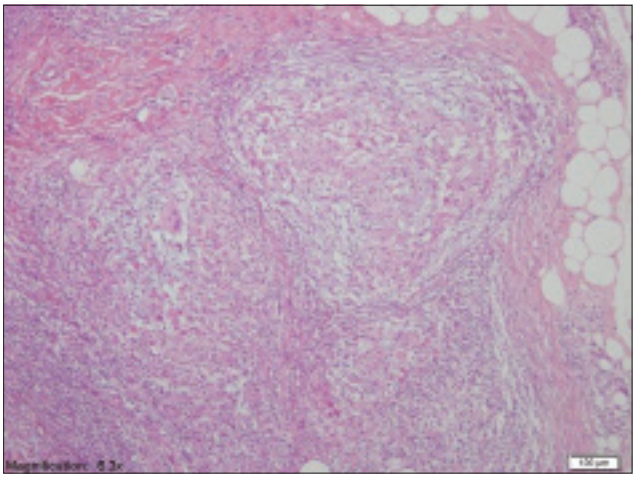

Figure 3 Histopathology showing non-classified granuloma inside breast lobules with inflammatory infiltration composed of epithelioid histiocytes, eosinophils, polymorphonuclear leukocytes and plasma cells

Discussion

The term "granular lobular mastitis" was coined by Going et. al. [2] and later described in detail by Kessler and Wolloch [1], based on typical histopathological features of a lobule-centered distribution [2]. It is a rare, benign and chronic inflammatory condition of uncertain etiology. An association with pregnancy and lactation has been noted and recently re-emphasized without any certainty. IGM usually presents primarily as a firm, tender breast mass with bilaterality in 25% of patients [3]. It may often present as a long standing breast mass, tumor indurations, skin ulcerations, local breast pain and tenderness, galactorrhea, abscesses and fistulae [4,5]. The usually defined clinical presentation is with unspecific findings of an expanding breast mass with surrounding local inflammatory signs [6,7]. As with most patients with IGM the initial presentation leads to the use of antibiotics. Some authors have suggested that IGM is a self-limiting condition [3], usually subsiding within a period of 2-24 months [4] but a chronic presentation could last for several years [8].

IGM is seen more frequently in third decade of life but the age of presentation can widely vary (range: 11-83 years) [9]. Associations have been suggested with breast feeding, use of contraceptives, alpha-1 antitrypsin deficiency, autoimmune disorders and hyperprolactinemia [6,7]. In our patient, IGM was probably associated with chronic use of oral contraceptives. IGM is usually a diagnostic and therapeutic dilemma, which initially can mimic other conditions and is a diagnosis of exclusion. Diagnostic tools such as ultrasound, mammogram, Tru-cut biopsy, fine-needle aspiration biopsy and contrast-enhanced MRI can be helpful in diagnosis in some patients.

Diagnosis is made by pathologic examination after eliminating infectious and autoimmune causes of granulomatous inflammation, such as malignancy, fungal, bacterial and parasitic infections, autoimmune conditions and other granulomatous diseases such as tuberculosis and sarcoidosis. A Tru-cut biopsy is recommended to clarify the diagnosis with characteristic histopathologic changes which include chronic granulomatous lobulitis without caseating necrosis and micro-abscesses in the presence of giant cells, leucocytes, epithelioid cells and macrophages [10].

There is no unanimity regarding the ideal management of IGM. A conventional non-operative management is usually suggested for IGM patients with minor symptoms [6]. Treatment is usually recommended for patients with drug-induced hyperprolactinemia and these patients are usually shifted to a prolactin-sparing agents. For patients with more severe symptoms, not responding to conservative management, oral prednisolone is recommended [11]. Patients not responding to oral steroids may be treated with immunosuppressive drugs as an alternative [12,13]. Finally, in persistent or recurrent cases, surgical management with wide surgical excision and/or mastectomy should be considered [14].

CASE REPORT

Conclusion

Based on the above case it can be concluded that IGM is a diagnosis of exclusion, after all the possible differential diagnosis of breast mass, including but not limited to malignancy, fungal, bacterial and parasitic infections, autoimmune conditions and other granulomatous diseases such as tuberculosis and sarcoidosis are excluded. A high index of suspicion is required to prevent the morbidity associated with delayed diagnosis and misguided therapy.

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Authors' Contribution

All authors contributed to the manuscript, have read and approved the final version.

Consent

The authors certify that a written informed consent was obtained from the parents of the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

Competing Interests

The authors declare that they have no competing interests.

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