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Case Report

Idiopathic Granulomatous Mastitis: A Case Report

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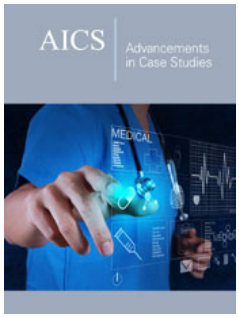

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Abstract

Idiopathic granulomatous mastitis is a rare disorder of unknown etiology with non-specific findings. It mostly affects young premenopausal women often after the lactation period. The incidence of this disease is not well known and its pathogenesis is not fully understood. It should be considered in the differential diagnosis of breast carcinoma, infectious mastitis and systemic diseases. We report a case of an idiopathic granulomatous mastitis in a 36-year-old breastfeeding woman who presented with a history of left painful breast lump, erythema and swelling for the last two weeks.

Keywords: Idiopathic granulomatous mastitis; Nonpuerperal mastitis; Granulomatous lobular mastitis

Introduction

Idiopathic granulomatous mastitis (IGM) is a rare, benign, inflammatory breast disorder first described in 1972 by Kessler and Wolloch [1]. It predominantly affects women of reproductive age, particularly those with a recent history of lactation. Clinically, IGM presents as a unilateral painful breast mass, often accompanied by erythema, breast edema, and sometimes axillary lymphadenopathy. The etiology of IGM remains unclear, though it is characterized histologically by lobulocentric granulomatous inflammation [2]. Differential diagnoses include breast malignancy and other granulomatous diseases such as tuberculosis and sarcoidosis. Given its rarity and the potential for misdiagnosis, IGM poses a significant diagnostic challenge. This case report aims to contribute to the existing literature by detailing a recent case of IGM, highlighting the clinical presentation, diagnostic process, and management strategies employed.

Case Report

We report the case of a 36-year-old woman, G1P1A0 who presented to the gynecology department for left mastodynia, breast volume increase and erythema with fever. The patient had no personal or family history of breast or ovary neoplasms. She was breastfeeding for the last three months. The initial suspected diagnosis was infectious mastitis and the patient was treated for 10 days with antibiotics and paracetamol with no improvement. She was then addressed to the radiology department for mammography and ultrasound exam.

On physical examination, the left breast was painful on palpation with diffuse skin erythema. The patient had no breast fistula nor nipple discharge. A mass was found at the upper quadrants' union of the left breast.

Mammogram revealed breasts of scattered densities with scattered areas of fibroglandular tissue (BIRADS B). A dense irregular mass with indistinct margins measuring 39 x 34mm was found in the left breast's upper quadrants union with skin thickening and nipple retraction. Focal asymmetries were found on the upper outer quadrant and the union of the outer quadrants of the left breast. No microcalcifications nor architectural distortion were found.

Ultrasonography revealed skin thickening and diffuse edema. A large heterogeneous hypoechoic mass with angular margins, tubular extensions and internal vascularization on color Doppler was found in the retroareolar region. Two hypoechoic microlobulated masses

were found on the lower outer quadrant and the upper outer quadrant of the left breast. The surrounding tissue was hyperhemic.

Breast MRI showed a large heterogeneously enhancing retroareolar mass with increased signal intensity throughout the left breast at T2-weighted imaging. Kinetic curves demonstrated a type II morphology. Two irregular ill-circumscribed masses were found in the lower outer quadrant of the left breast. The patient underwent ultrasound-guided percutaneous core needle biopsy. The histological exam revealed granulomatous mastitis (Figures 1-3).

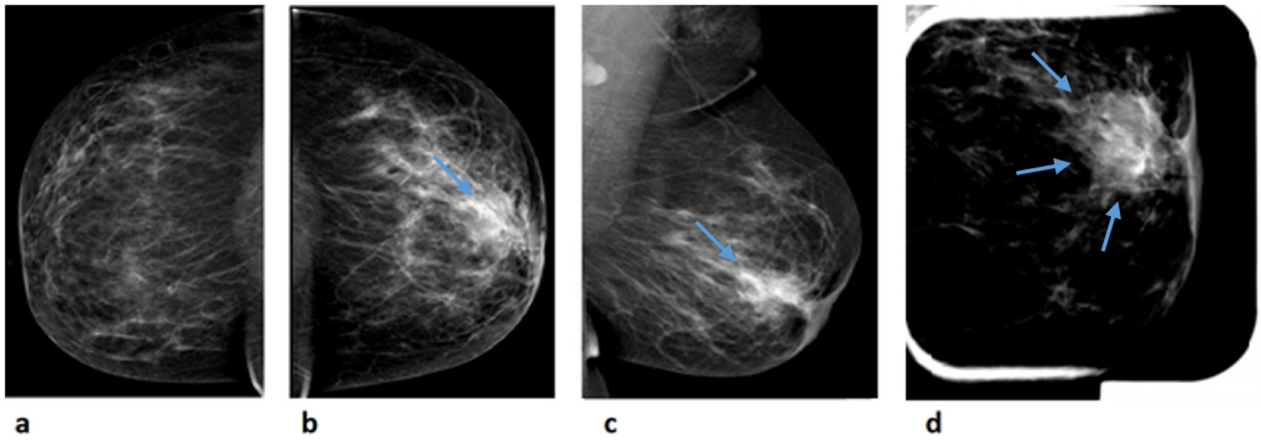

Figure 1: Right breast cranio-caudal CC (a), left breast cranio-caudal CC (b), left breast medio-lateral oblique MLO (c) and spot compression view (d) show increased density of the left breast, a retroareolar irregular dense mass with indistinct margins (arrows) and skin thickening.

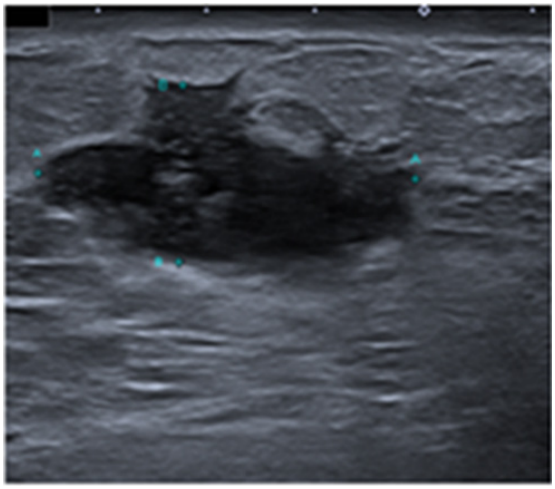

Figure 2: Left breast ultrasound obtained in the retroareolar region showing a heterogeneous hypoechoic mass with angular margins.

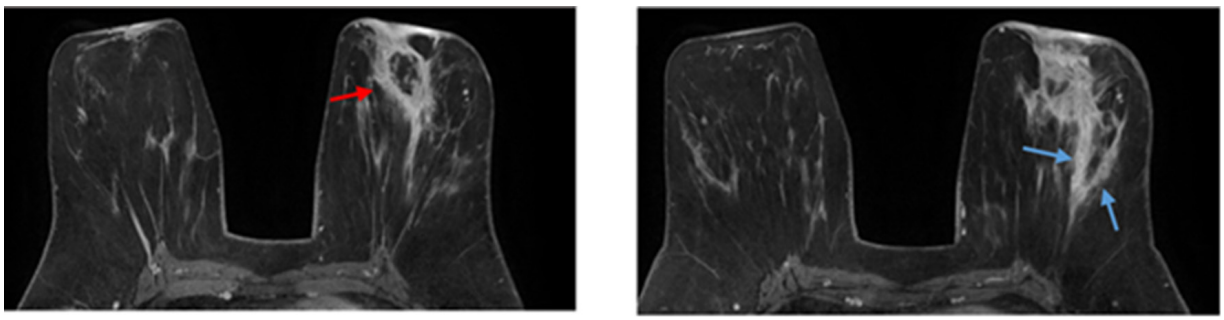

Figure 3: Gadolinium-enhanced T1-weighted fat-saturated images (f,g) show large heterogeneous enhanced retroareolar mass (red arrow) with skin thickening. Linear non-mass enhancement is shown in the outer quadrants' union (blue arrows).

Discussion

Idiopathic granulomatous mastitis (IGM), also known as nonpuerperal mastitis or granulomatous lobular mastitis, is a rare benign chronic inflammatory breast disease that was first described by Kessler and Wolloch in 1972 [1]. It is a rare condition of unknown etiology. It usually affects young premenopausal women with history of breastfeeding for more than one year. It has been also reported to be associated with hyperprolactinemia, oral contraceptive use, α1 -antitrypsin deficiency, diabetes, trauma, auto-immune diseases and smoking [2].

IGM is characterized by the formation of non-necrotizing granulomas and sterile micro-abscesses. It should be distinguished from specific granulomatous mastitis (SGM) which is a manifestation of an underlying systemic disease like Wegener granulomatosis, sarcoidosis, or a chronic granulomatous infection such as tuberculosis, histoplasmosis, syphilis and foreign body reaction. When no underlying etiology is detected, granulomatous mastitis is called "idiopathic".

Patients may present with a tender breast lump, skin inflammation, ulceration, breast fistulas and axillary lymphadenopathies. The disease has a characteristic clinical course, with frequent recurrences and sometimes an eventual burn out.

Clinical and imaging diagnosis of IGM is challenging as it often mimics inflammatory breast carcinoma, infectious mastitis, diabetic mastopathy and systemic granulomatous disease.

Mammographic findings are non-specific. The most frequently encountered findings are asymmetrical density and non-circumscribed dense masses. Additional findings include parenchymal edema, skin thickening and axillary lymphadenopathies. Microcalcifications are rare in IGM and should raise the concern of an underlying breast carcinoma [3].

On ultrasound, fluid collections and irregular non-circumscribed masses with tubular extension are the most frequent findings. Mass lesions usually demonstrate lobular or indistinct margins. They are usually parallel to the skin. Posterior acoustic features vary greatly. Posterior acoustic enhancement and acoustic shadowing are both described. Fluid collections are found at an advanced stage of the disease [4]. Additional ultrasound findings include skin thickening, hyperechoic fat lobules, parenchymal distortion and inflammatory axillary lymph nodes.

The magnetic resonance imaging is nonspecific. The most encountered images are heterogeneously enhancing masses and "rim-enhancing" lesions. The breast stroma is usually hyperintese on T2-weighted images. Linear, segmental and regional non-mass enhancement can also be found. Small lesions with rim enhancement are found (30-80%) representing sterile micro-abscesses. Other findings include axillary lymphadenopathies, skin thickening and skin or nipple retraction [5].

The treatment is controversial and depends on the case presentation. Corticosteroid therapy is the treatment of choice. Methotrexate can be used in cases of steroid-resistant IGM and in patients who develop steroid associated side effects like glucose-intolerance or Cushing syndrome. Surgery is generally reserved for cases of refractory or recurrent IGM [6].

Conclusion

Idiopathic granulomatous mastitis (IGM) remains a challenging diagnosis due to its rarity and the non-specific nature of its clinical, ultrasonographic, and radiological findings. In this case, the presentation of a unilateral painful breast mass with erythema in a breastfeeding woman initially suggested an infectious etiology. However, the lack of response to antibiotic therapy and the imaging features, including a dense irregular mass with skin thickening and nipple retraction, prompted further investigation.

Advanced imaging modalities such as ultrasound and MRI revealed characteristic features of IGM, including heterogeneous hypoechoic masses with angular margins and tubular extensions, as well as hyperintense stroma on T2-weighted MRI. The definitive diagnosis of IGM was confirmed through histopathological examination, which revealed lobulocentric granulomatous inflammation.

This case underscores the importance of considering IGM in the differential diagnosis of breast masses, particularly in women of reproductive age with a history of recent lactation. Early recognition and appropriate management are essential to avoid unnecessary interventions and to provide optimal care.

Given the benign nature of IGM, conservative management with close monitoring is often sufficient, although more aggressive treatments may be warranted in refractory cases. This report contributes to the growing body of literature on IGM, highlighting the need for increased awareness and a multidisciplinary approach in managing this uncommon but significant condition.

Patient Consent Statement

The patient has provided written consent with regards to publication of her case.

Declaration of Interest

The authors have no conflicts of interest to declare.

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