CASE REPORT
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IDIOPATHIC GRANULOMATOUS MASTITIS: DIAGNOSIS AND FOLLOW-UP WITH MAGNETIC RESONANCE IMAGING

Mastite granulomatosa idiopática: diagnóstico e seguimento com ressonância magnética

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ABSTRACT

Granulomatous mastitis is a rare and benign condition of the breast that, in some cases, has an unknown etiology of benign inflammatory disease know as idiopathic mastitis. Your diagnosis is usually made by exclusion. Imaging tests have shown nonspecific findings that may suggest an inflammatory disease, a carcinoma, or no changes. A differential diagnosis should be made with other causes of mastitis, always alerting to the risk of inflammatory carcinoma. Imaging tests are more useful to rule out malignancy than to confirm idiopathic granulomatous mastitis. Because both imaging and physical examination can mimic a malignant lesion of the breast, the histopathological report is fundamental to establish the diagnosis. Its etiology remains unknown, so the treatment is controversial in the literature, with some authors recommending surgery, others immunosuppression, and, finally, some antibiotics. We report the case of a 21-year-old woman with a suspected lesion in the breast associated with papillary discharge. During the investigation, there was a 12 x 6 x 8.5 cm enhancement on magnetic resonance imaging associated with inflammatory signs on the skin and lymph nodes. Anatomopathological examination revealed a idiopathic granulomatous mastitis. The enhancement disappeared completely after conservative treatment with corticosteroids. Mammography and ultrasound may also demonstrate nonspecific changes, such as focal asymmetry, undefined mass, or distortion. Despite the limitations of the imaging studies, it has been demonstrated in this report that MRI can be used to monitor the clinical response to conservative treatment and follow-up by the risk of recurrence.

KEYWORDS: Diagnostic imaging; mastitis; granulomatous mastitis.

RESUMO

A mastite granulomatosa é uma condição rara e benigna da mama que, em alguns casos, possui etiologia desconhecida de doença inflamatória benigna, a mastite idiopática. Seu diagnóstico normalmente é feito por exclusão. Os exames de imagens têm demonstrado achados inespecíficos que podem sugerir uma doença inflamatória, um carcinoma ou não apresentar alterações. Deve ser realizado um diagnóstico diferencial com outras causas de mastites, sempre alertando para o risco de carcinoma inflamatório. Os exames de imagem servem mais para descartar uma malignidade do que para confirmar a mastite granulomatosa idiopática. Em função de tanto os exames de imagem como o exame físico poderem simular uma lesão maligna da mama, o laudo histopatológico é fundamental para estabelecer o diagnóstico. A sua etiologia permanece desconhecida, portanto, o tratamento é controverso na literatura, com alguns autores recomendando cirurgia, outros a imunossupressão e, por fim, alguns antibióticos. É apresentado o caso de uma paciente de 21 anos com uma lesão suspeita na mama associada à descarga papilar. Durante a investigação, houve um realce de 12 x 6 x 8,5 cm na ressonância magnética associado a sinais inflamatórios na pele e nos linfonodos. O exame anatomopatológico evidenciou um quadro de mastite granulomatosa idiopática. O realce desapareceu completamente após o tratamento conservador com corticoterapia. A mamografia e o ultrassom também podem demonstrar alterações inespecíficas, tais como assimetria focal, massa indefinida ou distorção. Apesar das limitações dos exames de imagem, demonstrou-se, neste relato, que a ressonância magnética pode ser utilizada para monitorar a resposta clínica ao tratamento conservador e o acompanhamento pelo risco de recorrência.

PALAVRAS-CHAVE: Diagnóstico por imagem; mastite; mastite granulomatosa.

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INTRODUCTION

There is a great spectrum of breast infection and inflammatory diseases. One of these inflammatory conditions is granulomatous mastitis, which is a rare benign breast condition. There are also some etiologies like tuberculosis, sarcoidosis, parasitic and mycotic infections. In some cases, there is an unknown etiology of an inflammatory disease called idiopathic granulomatosis mastitis (IGM). Kessler and Wolloch first described it as a specific entity in 1972. It occurs in non-lactating young women. The clinical presentation is a local inflammatory sign in the breast. Diagnosis of IGM can be difficult and frequently late. The radiographic evaluation is unspecific, with a variety of presentations. There are some controversies about the best treatment to it. We present the case of IGM with complete response in the magnetic resonance imaging (MRI) follow-up[1-4].

CASE REPORT

A 21-year-old healthy female sought a breast surgeon complaining of a three-month history of cyclic mastalgia and nipple discharge in her left breast. Her menarche was at 13 years-old, with regular menstrual cycles. At the time of admission, she was taking oral contraceptives and denied smoking, alcohol abuse or any preceding breast trauma. Her uncle and maternal grandmother have a history of breast cancer, and her maternal grandfather of hypertension and diabetes. During the physical examination, the patient had breast hyperemia in the inner quadrants of the left breast and a thickened area of approximately 10 cm. The axillary lymph node was extended.

The ultrasound results demonstrated inflammatory signs in the skin, subcutaneous tissue and breast parenchyma on the left side with ductal ectasia (BI- RADS® 2), as seen in Figure 1.

After the dynamic MRI contrast agent, we found an intense enhancement measuring 12 x 6 x 8.5 cm with regional distribution in the inner quadrants, from the nipple to the pectoralis major muscle. The MRI also showed skin edema and lymph node alterations, suggesting an inflammatory process (Figures 2 and 3).

The investigation continued with core biopsy (Figure 1B) and histopathology, which presented a granulomatosis mastitis with giant cells. The cultures were negative for fungi and bacteria (Figures 4, 5 and 6).

The treatment was conservative with corticosteroid therapy for two months. She had complete clinical response afterwards.

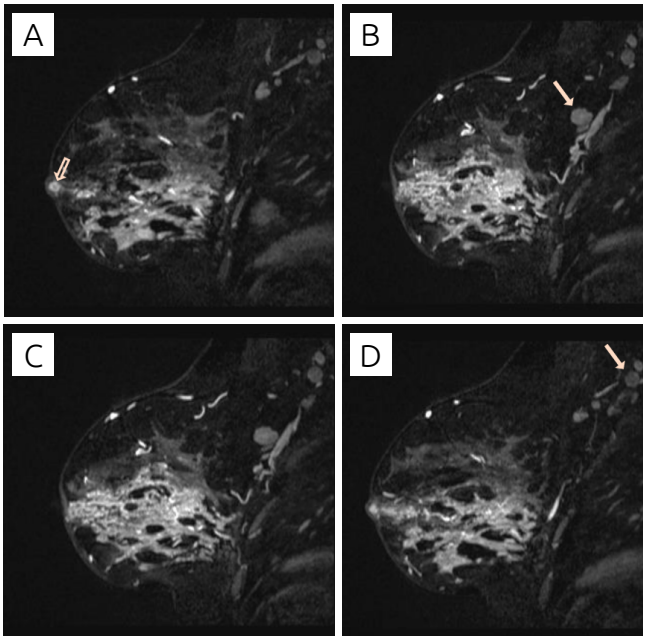

Figure 2. Axial magnetic resonance imaging (A) T1; (B) T2 STIR, skin edema and thickening in lateral quadrants; (C) and (D) subtraction, multiregional enhancement. Ductal ectasia.

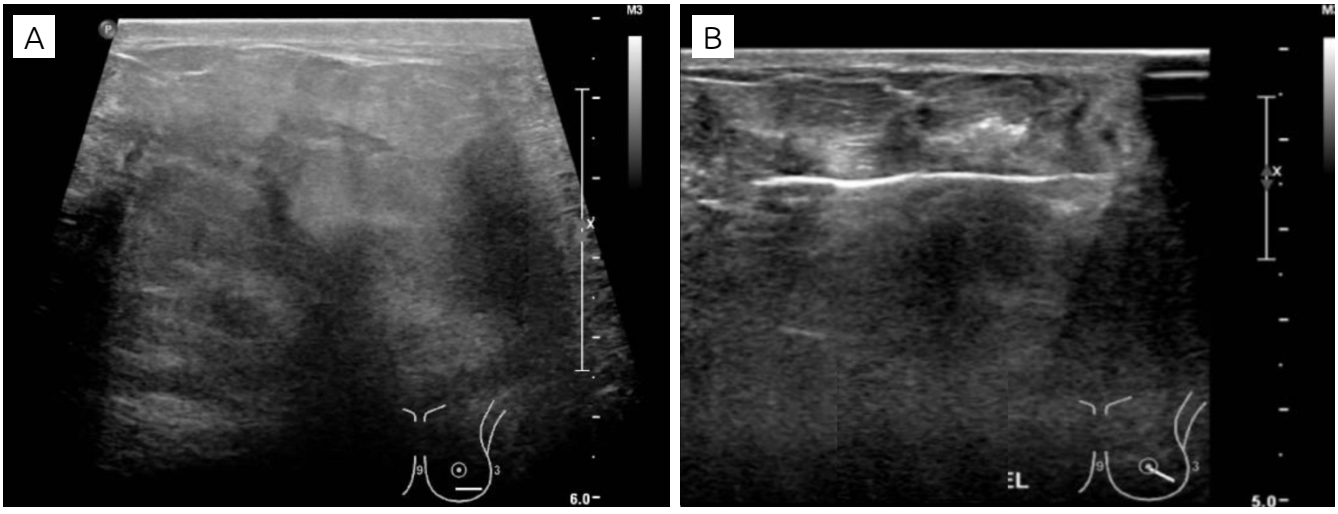

Figure 1. Ultrasound (A) with skin edema and enhancement with parenchymal hyperechogenicity; (B) core biopsy.

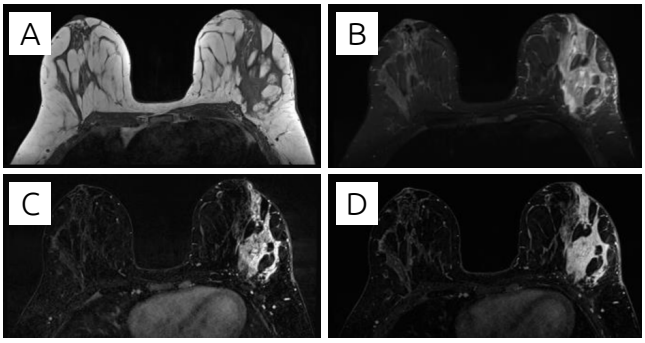

Figure 3. Magnetic resonance imaging enhancement.

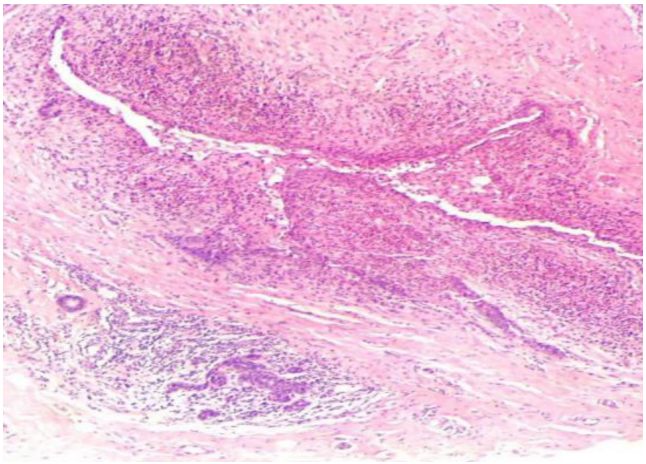

Figure 4. Lobules with inflammation.

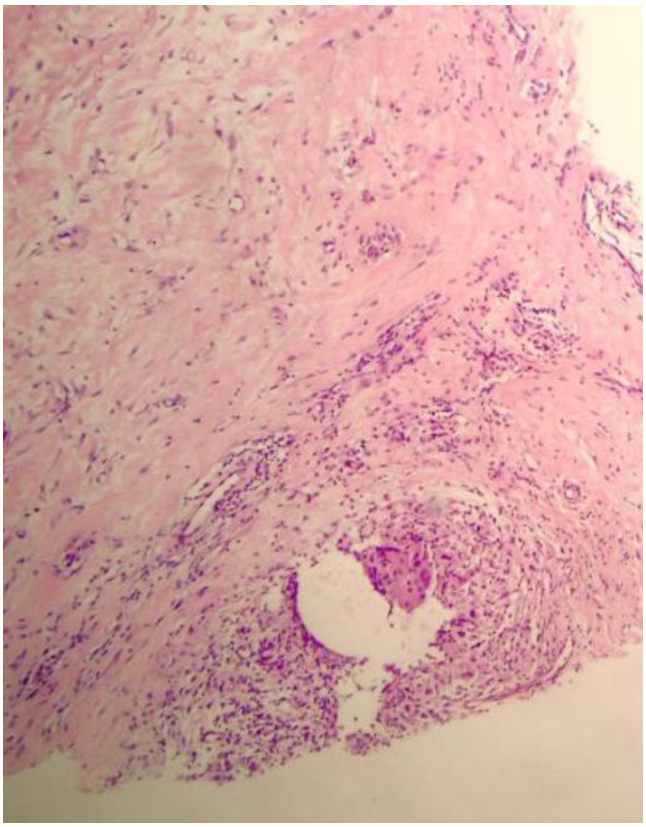

Figure 5. Granuloma with giant cells.

Subsequently, new imaging exams were ordered for follow-up. The new ultrasound showed preserved subcutaneous tissue and skin, and the MRI no longer had enhancements on the left breast (Figures 7 and 8).

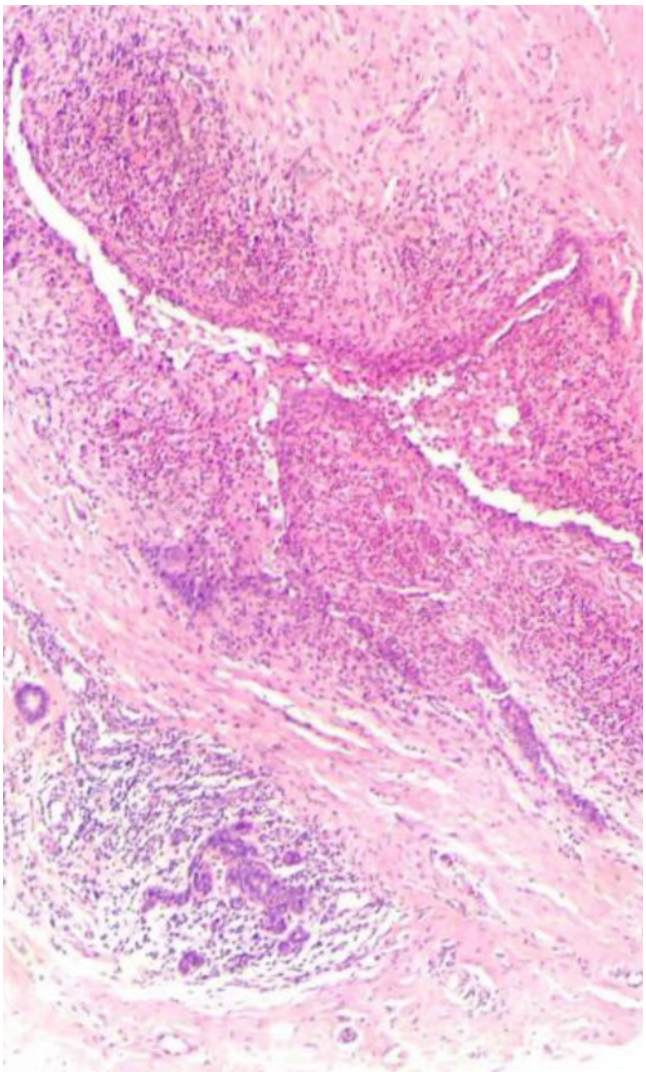

Figure 6. Obstructed duct with inflammation.

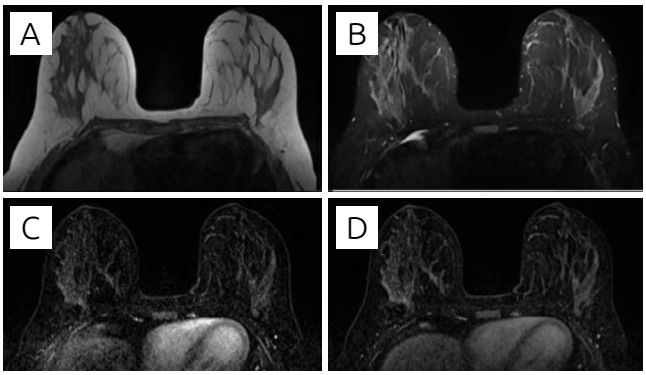

Figure 7. After treatment, axial magnetic resonance imaging (A) T1; (B) T2 STIR; (C) and (D) subtraction, inflammation resolution.

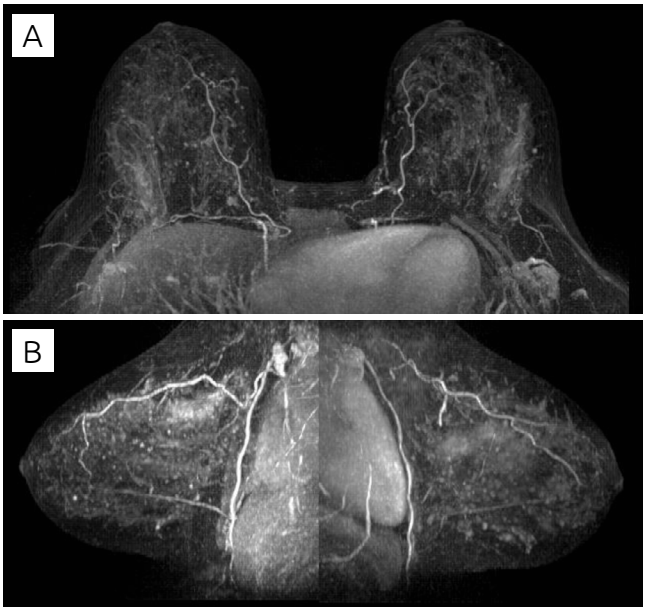

Figure 8. After treatment, MIP reconstruction without suspect enhancements.

DISCUSSION

Large varieties of benign inflammatory and infectious breast diseases are considered mastitis; thus, it is difficult to make a differential diagnosis. Clinicians should be alert to suspected inflammatory carcinoma. Breast mastitis can be lactation and occur during breastfeeding, or nonlactational, which, however, can be an infectious disease, such as tuberculosis, sarcoidosis, cat scratch, Cryptococci and other rare infections or inflammatory diseases like periareolar mastitis, fibrocystic change, or granulomatous mastitis[3,5].

Kessler and Wolloch first reported the IGM in 1972 like a lesion simulating breast carcinoma in five case reports[6]. This is a rare and benign condition, affecting young women, younger than 50 years of age. It is rare and there is a higher racial predilection for Asian and Hispanic women. Some authors believe that it is associated with a recent pregnancy and lactation[1]. Its incidence is unknown. Jayia et al. reported only 17 women at a breast cancer center during a 14-year period, and 2,500 women attended it[2,7-9].

A lump that can mimic carcinoma and can be painful or painless, with single or multiple masses with inflammatory signs like hyperthermia and redness are the main representations. It is usually unilateral and can involve any quadrant, but tends to spare the sub-areolar regions. Kamal et al. showed a predominance of right breast affection as seen in previous reports. In some cases, breast abscess, fibroses and fat necrosis can occur. Reactive axillary lymphadenopathy may also be presented, as in our case report[1,5,8,10,11].

Histology is characterized by a non-caseating granulomatous inflammatory response of the breast lobules in the absence of specific underlying causes. Pathological evaluations demonstrate granulomas, multinucleated giant cells, lymphocytes, plasma cells, micro-abscesses, and fat necrosis[12].

The pathophysiology is not completely understood, but there are some theories. One of them is that a local injury to the ductal epithelium produced by a trauma, infection or irritation can cause an immune response. Other speculated causes are chemical agents, such as smoking, using drugs, trauma and use of contraceptive pills. Oran et al. demonstrated that only one third in their records were smokers and 22% had a history of oral contraceptive use, therefore there is a lack of association[1,3,8].

Diagnosis is done by exclusion of cancer or other infectious diseases. We need to figure out if there is a cause for these alterations. Imaging methods are applied to rule out malignancy rather than to confirm the diagnosis. Unspecific findings may suggest IGM or breast radiologic findings can be normal. The mammogram can show a focal asymmetrical density, ill-defined mass or architectural distortion, and the ultrasound can show a hypoechoic lesion with a posterior shadow and tubular configuration, parenchyma heterogeneity. The MRI can include a parenchymal asymmetry or distortion with different contrast enhancement patterns (diffuse, heterogeneous and ring-shaped enhancement). In our case, the MRI showed a regional enhancement, and because there were other signs in the skin and lymph nodes, an inflammatory process was suggested[1,4,10]. Although radiology findings have a limited value, our report showed that they can be used to monitor the clinical response in a conservative treatment[13].

Because both clinical and radiological presentation can mimic a breast carcinoma, the final diagnosis has to be histopathological; therefore, our patient took a core biopsy. These pathological findings showed that a surgical excision for the diagnosis is not necessary. Other diagnostic forms are performed with fine-needle-aspiration biopsy (FNA) or surgical excisional or incisional biopsy.

The optimal treatment is controversial. In some cases, surgical excision, close observation, immunosuppressant therapy (steroids, azathioprine and methotrexate) and antibiotics are the mainstay of treatment.

Nowadays, surgical treatment has become less eminent and directed to specific cases. Kaviani et al. suggest an algorithm for the management of IGM. They mention that surgical treatments should be restricted to biopsies, by draining the abscess and excising fistulae[14]. They believe that surgical intervention can cause physiological distress, scars, disease exacerbation, fistula formation, and other surgical complications[9,14].

A retrospective study in Phoenix demonstrates that clinical observation can be an effective strategy for this nonmalignant disease, thus avoiding the costs and side effects of surgical and medical treatment[9].

The best dose and duration of medical treatment have not been established. We used the clinical remission to

decide when we should start to decrease the corticosteroid dose until to clinical presentation remission, and finally to its gradual stop. Our patient did not show side effects of the steroid therapy, such as glucose intolerance and Cushingoid features[1,8].

A prospective case series demonstrated a successful treatment in the majority (90%) of women by using only the prednisone therapy[11]. The prognosis is good; nevertheless, local recurrence is frequent[5].

CONCLUSION

We demonstrated the possibility of monitoring granulomatosis mastitis after steroid treatment with MRI.

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