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Granulomatous Lobular Mastitis : a Rare Chronic Inflammatory Disease of the Breast which Can Mimic Breast Carcinoma

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Abstract. Granulomatous lobular mastitis is a rare chronic inflammatory disease of the breast. The differential diagnosis with malign breast disease is often not easy. In most cases a surgical biopsy is needed for correct diagnosis. Idiopathic granulomatous mastitis is an exclusion diagnosis, based on the demonstration of a characteristic histological pattern, combined with the exclusion of other possible causes of granulomatous breast lesions. There is still no generally accepted optimal treatment. If surgery forms part of the treatment, a conservative approach seems to be adequate in most cases. Another option is a long-term steroid treatment. It is mandatory to exclude infectious causes of granulomatous mastitis before corticoid therapy is started.

Introduction

Idiopathic granulomatous mastitis (IGM) is a rare chronic inflammatory disease of the breast. Awareness of this condition is important because it can clinically mimic breast carcinoma.

Case report

A 57-year old woman presented with a painful inflammatory breast mass (5 centimetres), a discrete nipple retraction and no palpable axillary nodes. She had no particular medical history, never used oral contraceptives and breastfed two children.

Blood tests showed no sign of inflammation. Mammography and magnetic resonance (Fig. 1) of the breast revealed a diffuse lesion, very suggestive for breast cancer.

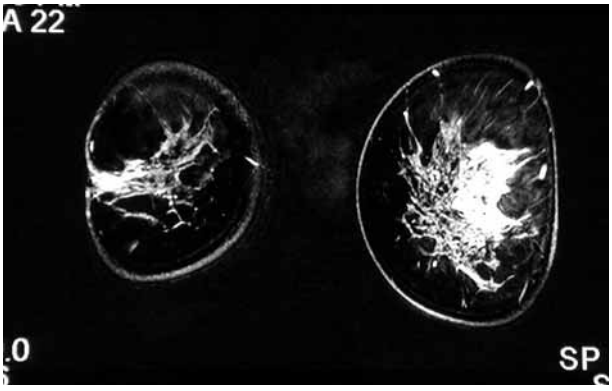

Fig. 1
Magnetic resonance of the breast showing a hyperintens lesion with irregular margins, suggestive for breast cancer.

Fine needle cytology (FNA) showed clusters of normal ductal cells. Open biopsy revealed well-formed noncaseating granulomas with Langhans and foreign body-type giant cells within fibrous background tissue. These exhibited a distribution mainly respecting the lobules (Fig. 2), but at some sites completely destroying these structures. A few micro-abscesses were found. Serological and haematological investigations were normal. No evidence of tuberculosis or sarcoidosis was found.

The diagnosis of idiopathic granulomatous mastitis was sustained.

A delayed wound healing was noted, but no recurrence of the disease occurred.

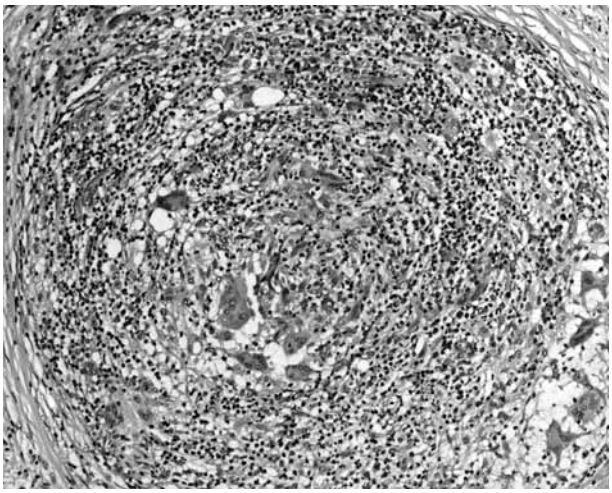

Fig. 2
Open biopsy revealing well-formed noncaseating granulomas with Langhans and foreign body-type giant cells. These exhibit a distribution mainly respecting the lobules.

Discussion

IGM is a rare chronic inflammatory disease of the breast, characterized by granulomas and abscess formation.

IGM mainly affects women between the ages of 17 and 42, is often associated with the use of oral contraceptives and breastfeeding, but no sufficient supporting data are available. The time between the last childbirth and first symptoms ranges from 2 months to 15 years. The patients present with a unilateral breast mass (0,5 to 9 cm). Regional lymfadenopathy may be present and the initial diagnosis is breast cancer in 50 % of the reports. IGM can also cause "peau d'orange" skin, ulceration and nipple inversion (1-2).

The aetiology remains unknown. There has been consideration of a possible infectious aetiology. Differential diagnosis includes Mycobacterium tuberculosis, blastomycosis, cryptococcosis, histoplasmosis, actinomycosis and filarial infection.

Some believe that damage to ductal epithelium produced by local trauma, local chemical irritant or infection, could allow luminal secretion to escape into the lobular connective tissue, thereby causing a granulomatous response (1,3). However, serologic tests for ANA and RF autoimmune antibodies, which are evidence of an autoimmune phenomenon, are usually found to be negative. Some authors suggest a possible relationship between smoking and IGM. However, more clear evidence is needed (4).

The diagnosis of IGM is based on the demonstration of a characteristic histological pattern, combined with the exclusion of other possible causes of granulomatous breast disease.

Radiological and cytological findings are in many cases unable to resolve the differential diagnosis of inflammatory process and malignancy. The imaging findings of granulomatous mastitis are not well documented and often mimic breast carcinoma (multiple small masses or a large, focal asymmetric density). Ultrasound usually reveals a large inhomogeneous, hypoechogenicity with internal tubular lesions. Doppler examination reveals increased vascularity of the lesions and surrounding tissue (4-5).

ENGIN *et al* came to the conclusion that neither mammography, nor Doppler sonography played a significant role in the differential diagnosis of granulomatosis versus carcinoma and stated that all cases require histopathological confirmation (6).

Magnetic resonance images could be useful in order to indicate active lesions and to locate the extent of the lesions, but it is impossible to differentiate an active inflammatory process from a neoplastic process since the technique focuses on morphology rather than on vascular physiology. A "Dynamic contrast-enhanced magnetic resonance mammography" focuses on the dynamic rather than morphological attributes of the lesions and indirectly reveals their vascular nature and could potentially be used to discriminate between benign and malignant processes, but the difference between carcinoma and inflammatory process remains difficult (7).

The cytological features of IGM can be difficult to distinguish from those of carcinoma. The relatively rare occurrence of this lesion and his cytological features make it a potentially diagnostic pitfall for the pathologist. FNA may reveal scattered aggregates of epithelioid cells mixed with multinucleated Langhans-type giant cells, neutrophils, lymphocytes and stromal cells (4,8). False positive and negative results are described in literature.

An adequate tissue sampling biopsy is very often needed for a firm diagnosis. Core biopsy can be very useful but may not always differentiate between IGM and other granulomatous disease of the breast. If granulomatous inflammation of the lobular units, necessary for the diagnosis of IGM, is present on core-biopsy then a definitive diagnosis can be established. The histological findings include non-caseating granulomatous inflammation, centred on breast lobules, composed of epithelioid cells, polymorphonuclear leucocytes and multinucleated Langhans type giant cells. The most important histological feature is the fact that the inflammatory process has a predominantly lobular distribution. Abscess or micro-abscess formation can occur. Special stains for bacteria and fungi are negative (1,9).

Little is known about the optimal therapy. The natural history of idiopathic granulomatous mastitis is often that of a self-limiting condition and 50 % of the cases recover uneventfully. The usual therapy is wide local excision of the breast mass. Postoperative wound infection, fistulas, chronic suppuration and recurrence are often seen, necessitating repeated surgical interventions. Limited excision alone has a strong tendency for persistence or recurrence (3-4).

The effect of a short course of high-dose prednisone (60mg/day during three weeks) can be quite impressive (reducing size of the lesion and enhancing complete lesion after surgery). Some authors suggest that in patients with proved granulomatous mastitis a trial of corticosteroids should be given before excision, allowing more conservative surgery. Recurrence can be treated successfully with another course of corticosteroid therapy. In patients with a high prolactin level who have recurrence, medical treatment to control prolactin level may be useful. In cases of delayed wound healing a short therapy of high-dose steroids can be efficient (2-3).

The doses of the corticosteroids should be gradually tapered to avoid an exacerbation of the disease process. Both complete resection or corticosteroid therapy can be recommended as treatment options, but we have to be

aware that there is insufficient data to support the effectiveness of prednisone in treating granulomatous lobular mastitis and that the possible disadvantages of long-term steroid therapy must be considered (3-4).

Long-term follow-up is indicated since more than 30 % of the patients with IGM experience recurrence (10).

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