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Diabetic Mastopathy: A Case Report

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Abstract

Diabetic mastopathy is a recently described and uncommon benign entity occurring in young women with prolonged and complicated type 1 diabetes mellitus. Its clinical and radiological signs are not specific and often mimic a breast carcinoma. However, the benign nature of this lesion is easily recognized on histological examination, visualizing dense keloid-like fibrosis, lymphocytic lobulitis and ductitis with lymphocytic perivascular inflammation, with or without epithelioid-like fibroblasts. Surgery can generally be avoided. The evolution of this entity is characterized by the risk of local growth, bilateralisation or recurrence after surgical treatment. Through this case we recall the clinical, radiological and outcome of this disease.

Subject Areas

Gynecology and Obstetrics

Keywords

Diabetes, Mastopathy, Diagnosis, Conservative Treatment

1. Introduction

Diabetic mastopathy is a rare and benign breast condition observed in patients suffering from long-standing type 1 diabetes. It represents less than 1% of benign breast lesions [1].

The pathogenesis of diabetic mastopathy, probably multifactorial, is not precisely known, but seems linked to an autoimmune process [2].

It takes on the appearance of a malignant lesion simulating breast cancer both clinically and radiologically. It is characterized by the proliferation of a dense and fibrous stroma of the breast tissue, lymphocytic lobulitis and the presence of dystrophic fibroblasts with an epithelioid appearance [1].

Clinicians and pathologists must recognize this benign condition since sur-

gery can make the condition worse. These lesions tend to recur more aggressively at the resection site. We present the case of a type 1 diabetic woman presenting with typical diabetic mastopathy and summarize the literature to discuss the diagnostic and management of this condition.

2. Case Report

The patient is a 45-year-old postmenopausal woman, with no family history of breast cancer, a medical history of celiac disease and type I diabetes under insulin therapy: 22 IU in morning and 20 IU at night. Her diabetes is poorly controlled, causing numerous episodes of metabolic decompensation.

She consulted for a lump in the right breast that had been present for 6 months.

Breast examination reveals two symmetrical breasts showing no abnormality, in particular no skin redness, peau d'orange appearance or nipple retraction.

On palpation of the right breast, we note a 4 × 2 cm nodule at the junction of the external quadrants, of hard consistency, painless, mobile in relation to the superficial and deep planes. There is no breast discharge or axillary lymphadenopathy. Examination of the left breast was unremarkable.

Breast ultrasound finds a poorly defined area, hypoechoic, with posterior attenuation and discreetly vascularized on color Doppler measuring 40 × 16 mm (Figure 1). The lesion is classified BIRADS 4B according to the ACR.

Breast biopsy reveals benign fibro-epithelial proliferation with absence of malignant tumor proliferation.

The tumor was excised.

The pathological exam of the tumor showed a firm and dystrophic breast tissue. Macroscopically, the biopsy fragment measures 4 × 3 × 2.5 cm made of very firm fibrous tissue. Histological examination showed that the lesion is composed of breast parenchyma with dense fibrosis surrounding the milk ducts and acini with the presence of a moderate inflammatory infiltrate, mainly lymphocytic without signs of malignancy (Figure 2). This aspect suggests diabetic mastopathy of the breast.

During her follow up, no signs of recurrence were noted.

Figure 1. Breast ultrasound showing a poorly defined hypoechoic area.

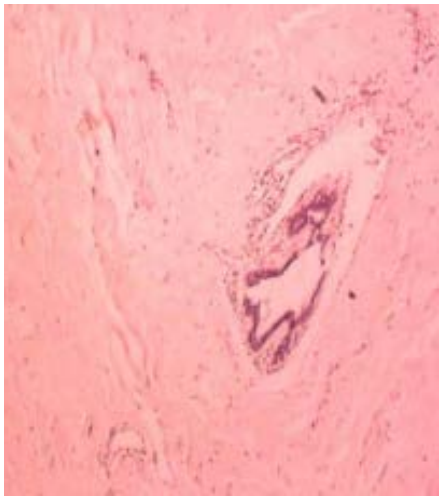

Figure 2. Lymphocytic infiltration around the mammary lobule.

3. Discussion

Diabetic mastopathy was first described in 1984 by Soler *et al.* as a condition simply attributed to long-term insulin-dependent diabetes [1]. According to Hunfeld and Bassler [3], it represents less than 1% of all benign breast lesions; this rate rises to 13% if we limit ourselves only to type I diabetic patients. Since then, multiple studies have shown its association with other autoimmune diseases, such as Hashimoto's thyroiditis and Sjögren's syndrome, suggesting an autoimmune origin [4] [5].

It is recognized as a rare benign clinicopathological entity with a few hundred cases reported in the English literature [6]. The disease particularly affects young and middle-aged women (34 to 47 years), with a small number of cases reported in men [7] [8].

The pathogenesis of lymphocytic mastopathy is not yet fully understood.

In diabetic patients, two hypotheses have been put forward to explain the pathogenesis of the disease:

- Hyperglycemia would cause an abnormal accumulation and a reduction in the breakdown of connective tissue in the extracellular matrix [2] [9] [10];
- Hunfeld and all [3] [7] [8] raise the hypothesis of an immune reaction, secondary to excess glycosylation, involving B lymphocyte cells and macrophages; the presence of cytokines would accentuate the phenomenon of collagenization.

The main clinical manifestation of diabetic mastopathy is a breast nodule, single or multiple, unilateral, or bilateral, of recent appearance, of irregular contours, of hard, even stony consistency, painless, of rapid growth, mobile in relation to both superficial and deep planes and variable seat. It is not associated with nipple discharge or inflammatory skin appearance [11].

Mammography findings are usually nonspecific and inconclusive because they cannot provide a detailed illustration of the dense, glandular breast parenchyma [12].

On the other hand, Logan *et al.* conducted a retrospective study to describe possible ultrasound findings of the disease and found that breast lesions of diabetic mastopathy typically manifest as hypoechoic masses with mild to marked acoustic shadowing [13].

Breast carcinoma therefore remains the main differential diagnosis of diabetic mastopathy, and a pathological sample is necessary to differentiate the two entities and reach a definitive diagnosis [14]. Biopsy is therefore the test of choice which allows the diagnosis.

The histological study highlights three types of elementary, non-specific lesions, variously associated:

- Lymphocytic lobulitis: mononuclear inflammatory infiltrate, dense, associating small lymphocytes, especially B lymphocytes, plasma cells and histiocytes, mainly in the mammary lobules, but also and less markedly perivascular and sometimes periductal.
- Fibrosis of the pallial tissue, homogeneous and dense.
- Inconstantly, stromal epithelioid cells distributed homogeneously.

Immunohistochemistry has no diagnostic indication [10].

Management of fibrous mastopathy follows the approach recommended for other benign breast lesions. Conservative treatment is usually initiated. The progression can be marked by a high rate of recurrence of lesions ranging from 20% according to Hunfeld and Bassler [15] to 100% according to Lammie [3]. Due to the high rate of recurrence, these lesions should not lead to repeated excisions. Simple monitoring is recommended, including an annual clinical examination and a mammogram every two years. The risk of cancer in these patients is like that of the general population, as reported by Kudva *et al.* and, therefore, the role of surgery remains very minimal. Surgical intervention is only preferred in cases where malignancy cannot be excluded or in cases of anxiety [15].

4. Conclusion

Diabetic mastopathy is a rare and benign condition occurring mainly in young type I diabetic patients with multiple degenerative complications. It clinically simulates breast cancer. Mammography is not very informative, and the definitive diagnosis is based on histological examination. Its evolution is dominated by the risk of recurrence. After exclusion of a neoplastic pathology by microbiopsy, its management is limited to simple monitoring.

Conflicts of Interest

The authors declare no conflicts of interest.

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