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Case Report & Review of the Literature

Lymphocytic Mastopathy – A Rare Entity and A Diagnostic Challenge

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ABSTRACT

Lymphocytic mastopathy is a benign rare entity, commonly associated with long standing diabetes. It has also rarely been reported on patients with autoimmune diseases, particularly Sjogren's syndrome. There are very few cases reported, with the literature suggesting an incidence of <1% of benign breast diseases. Despite being considered a benign disease, lymphocytic mastopathy presents similarly to breast cancer on imaging, posing a diagnostic challenge. This report describes a case of a postmenopausal woman with significant autoimmune history that presented with a right-sided breast lump found to have lymphocytic mastopathy on histology.

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Case Report

A 70 year-old female with right-sided breast lump was referred for investigation with presumably periductal mastitis that failed antibiotic treatment. The patient noticed a progressive right breast swelling over a 2-week period, associated with erythema and pain. This was mostly localized to right upper outer quadrant, adjacent to nipple. She denied nipple discharge but noticed a slight inversion of the same. Other symptoms included lethargy, malaise and subjective fevers. The patient was treated as chronic subacute mastitis with oral antibiotics (cephalosporin for 5 days) and referred to the Breast Surgeon for further assessment. Despite her trial of antibiotic, there was minimal improvement of her symptoms, on her 1-week follow up appointment. The patient was having biennial mammographic screening, had no previous recalls or history of breast lesions. There was no known family history of breast or ovarian cancer. Her comorbidities consisted of long standing Systemic Lupus Erythematosus (SLE) and Sjogren's syndrome on long-term corticoid, immunomodulator and immunosuppressant therapy. In addition to this, she also had bronchiectasis, asthma, obstructive sleep apnea and esophagitis. She denied smoking or alcohol use.

On physical examination an 11 o'clock 5cm mass was palpable, with irregular borders and overlying erythematous changes to skin but no peau d'orange features. The mass was painful on palpation, hard and not adherent to adjacent planes. There was no palpable lymphadenopathy. An ultrasound organized during an acute admission showed an irregular hypoechoic mass measuring 15x10mm at 11 o'clock, with infiltrative borders and minimal peripheral vascularity (Figure 1). No lymphadenopathy was appreciated. A Breast Surgeon performed a repeat US, mammogram and a subsequent core biopsy for further investigation. On the mammogram, a non-specific density with architectural distortion was appreciated, corresponding to a 5cm hypoechoic irregular mass found on US. The lesion was classified as a BIRADS 4a. A core biopsy was performed, and histology showed occasional irregularly dilated ducts, surrounded by prominent plasma cell population without features of malignancy.

A second course of oral cephalosporin was given for further 2 weeks, and the patient was planned for review in rooms in 1 month. She however, represented the following week with persistent symptoms. Given the patient's imaging results and failure of conservative management, decision was made to perform an excisional biopsy for further diagnostic elucidation and symptom control. The procedure went as expected, and on her post-operative follow up the pathology report

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was reviewed. The specimen measured 45x38x25mm and on microscopic examination multiple dilated ducts with periductal fibrosis and intense lymphoplasmacytic inflammation was identified. No features of atypical hyperplasia, in-situ or invasive malignancy were seen. The pathological diagnosis of lymphocytic mastopathy was made. She was reviewed 3-months post procedure she has had no signs local recurrence. She's currently 1-year excision and remains disease free.

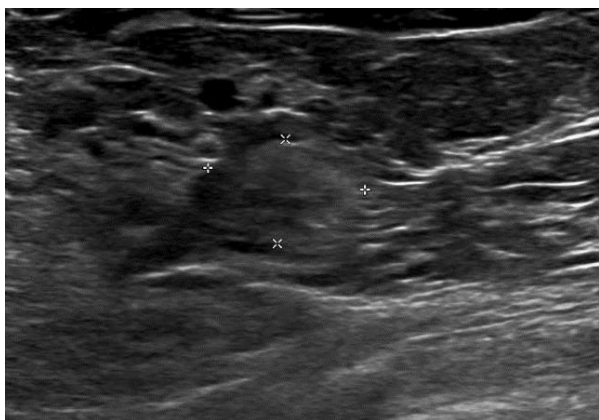


Figure 1: Ultrasound findings – hypoechoic irregular lesion at 11 o'clock.

Literature Review

Lymphocytic mastopathy is a rare entity commonly associated with autoimmune disorders and diabetes, commonly seen in perimenopausal women [1-4]. Literature review shows that it is responsible for less than 1% of benign breast lesions [1]. Defined as an ultimately benign condition, its radiological features are notorious due to its mimicking nature to malignant breast disease. Historically this disease has been broadly associated with long-standing diabetic patients, being previously reported on both type one and two regardless of their insulin dependency status [5-7]. The pathogenesis of lymphocytic mastopathy is still unclear, but there is strong evidence linking a local autoimmune response to the breast tissue, particularly related to Sjogren's syndrome and Hashimoto thyroiditis [5, 8]. The current case report support the relationship between autoimmune disorder and lymphocytic mastopathy once the patient does not have a diagnosis of diabetes. Pathological features consist of predominant lymphocytic infiltrates, made almost exclusively by B cell lineage [9]. Clinical presentation varies from a single palpable lump, painful or not, multicentric with and/or bilateral breast involvement. Lymphocytic mastopathy lesions tend to reoccur, some authors finding recurrence in nearly a third of patients [8]. So far, that has not appeared to be our patients case given she does not have disease reoccurrence 1-year post wide local excision.

Imaging assessment for this pathology has always been challenging, due to its contradicting features when compared to its lack of malignant pathological findings. Worrying radiological characteristics suggestive of malignancy are usually present, particularly on ultrasound (US). Irregular, hypoechoic masses with increased acoustic shadow are the main seen features [2, 8, 10]. Mammograms usually do not offer any additional information, with dense parenchymal mass not distorting the normal breast tissue but with presence of microcalcifications commonly reported [1]. Interestingly a recently published article found that nearly

half of the diabetic patients who had diabetic mastopathy had negative mammogram findings, favoring US as the investigation of choice in this setting. A retrospective review from 2015 compared the sonographic features of diabetic mastopathy versus of breast cancer, and suspicious sonographic signs for malignancy were equally seen in both groups [11, 12]. This corroborates with the diagnostic challenge encountered in this patient pool.

Invariably these patients will undergo further diagnostic procedures such as fine needle aspirations and core biopsies, and eventually those will lead to wide local excisions. Even though core biopsy samples could be conclusive for lymphocytic mastopathy, the presence of a suspicious breast lesion on a postmenopausal woman virtually warrants surgical excision. This is easily applicable day-to-day when dealing with newly diagnosed breast mass on a previously disease-free (from the breast perspective) patient. This disease has a tendency to reoccur, varying from single to multiple foci, from unilateral to bilateral [5, 14]. It is still however not possible to predict how it will progress. There have been reports in the literature of recurrence rates of up to 63% [5]. A paper from 2000 did a short 5 case retrospective study and found that 4 of them had disease recurrence within their 6 year follow up span. Even though that was a small sample, it reinforces the prevalent recurrence rate in this pathology.

Some authors however have been questioning the necessity of surgical management in patients with previously known lymphocytic mastopathy, with regular radiological surveillance being favored. Most recent publications show that there is no evidence to support this disease as being pre-cancerous [5, 8, 9, 13]. The literature suggests that annual routine imaging follow-up in this group would be beneficial [13]. There has been one report of such a severe case of multicentric bilateral diabetic mastopathy that the patient opted for nipple sparing bilateral mastectomies with immediate direct-to-implant reconstruction [4]. In a systematic review performed by the same authors, there have been rare reports of mastectomies from severe disease. This shows that even though a rare and benign entity, lymphocytic mastopathy has a potential to carry significant burden of disease in some cases.

Conclusion

Lymphocytic mastopathy is a diagnosis to be considered in patients with long-standing diabetes or those with a significant history of autoimmune disease. It is shown to be a diagnostic challenge as it presents similarly to breast cancer on imaging, with negative biopsy samples. It has a tendency to reoccur and should be followed up regularly by a breast surgeon due to its potential for deforming disease.

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