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Case Report

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Case Report : Lymphocytic Lobulitis With Fibrocystic Disease Of The Breast

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Abstract :Lymphocytic lobulitis , is a fibroinflammatory benign condition of the breast which is associated with type 1 diabetes mellitus. This benign condition is uncommon and may be mistaken for inflammatory carcinoma of the breast. We report the case of a 61 year old female patient who presented with a lump in the right breast with a history of Type 2 diabetes mellitus. The lump was associated with discharge and recent onset pain. On examination a vague lump which was non mobile, involving the right breast was palpable. A single firm mobile right axillary lymph node was palpable. Peau d'orange or orange peel appearance which occurs due to blockage of sub dermal lymphatics by tumour infiltrates, was noted over the skin. Clinically the features were suggestive of inflammatory carcinoma. Mammogram suggested an inflammatory carcinoma. Ultrasound of the breast was suggestive of right duct ectasia and diffusely thickened breast with a Breast Imaging Radiology and Data System (BIRADS) score of 3. An incision biopsy was performed with histopathology confirming the lesion as lymphocytic lobulitis with fibrocystic breast disease. The patient was symptomatically managed with analgesics and reassured. She was observed on regular follow up and is currently healthy. Lymphocytic lobulitis is a rare benign lesion which mimics carcinoma. Clinically it presents with ill defined single or multiple breast lumps in young or middle aged women with thickening and hardening of skin. Magnetic Resonance Imaging better differentiates this otherwise indolent lesion from malignancies. A histopathological examination is usually confirmatory and required to alleviate concerns of patients regarding presence of a malignancy which has a much fearsome physical and psychological implication.

Key Words : Lymphocytic Lobulitis , Diabetic Mastopathy, Sclerosing Lymphocytic Lobulitis

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1. INTRODUCTION

Lymphocytic Lobulitis also known as Sclerosing Lymphocytic Lobulitis is a relatively uncommon rare benign fibroinflammatory condition which was first described in 1984 by Soler and Khardori[1] . It usually occurs in young and middle aged women between 34-47 years[2]. It constitutes less than 1% of benign breast diseases. This condition mimics inflammatory carcinoma of breast clinically as well as radiologically. Patients usually present with a hard mass which is ill defined and a discrete lump may not be palpable.It may be associated with pain and may be bilateral. It is associated with type I (insulin dependent) diabetes or other autoimmune diseases like Hashimoto Thyroiditis, Graves disease, Sjogrens syndrome, rheumatoid arthritis, pernicious anaemia and SLE[3,4]. No particular HLA type has yet been found to have any association with the development of fibrous breast disease in insulin dependent diabetics[5].

2. CASE REPORT

A 61 year old female presented to the OPD with complaints of lump in the right breast for 3 months which was insidious in onset and gradually increased in size. It was associated with complaints for pain for the past 3 days. There was a history of whitish nipple discharge 3 months ago, with no discharge on presentation. There was a change in size of the lump on and off. No history of fever, loss of appetite, loss of weight. Patient was a known case of diabetes mellitus for 8 years and on medication but without good glycaemic control. There was a history of taking insulin injections for the same. On examination, the peau d'orange appearance of the left breast is seen, below the nipple areolar complex. Skin appeared thickened and edematous (Fig.1). On palpation, vague mass measuring 6x5 cm was felt in the lower inner quadrant of the right breast. Erythema , induration and tenderness was present over the lump with a peau d'orange appearance. It was non mobile and not fixed to the underlying structures. 2x1 cm right mobile , firm , axillary node was palpable in the anterior group with presence of multiple non specific apical nodes. White nipple discharge was noted. Examination of the right breast and axilla was normal. Her routine blood investigations did not show any abnormality. Mammogram showed diffusely increased breast thickening with microcalcifications suggestive of inflammatory carcinoma. USG breast showed diffusely thickened breast with irregular margins and right mammary duct ectasia BIRADS III with edematous changes. Patient was planned for incisional biopsy. On table, no specific breast mass was seen. Breast tissue was firm in consistency and biopsy was taken from the same for histopathological confirmation. Histopathology report showed breast parenchyma with lobules of ductal glands in tiny aggregates and groups with dilated glands surrounded by dense periductal, perivascular (Fig 2 & 3) and perilobular lymphocytic infiltration. Focal collection of foamy macrophages seen. Adjacent breast tissue shows dilated ducts lined by apocrine cells. Some duct shows epithelial hyperplasia and keloidal fibrosis. No evidence of atypia/malignancy. Impression was Lymphocytic lobulitis with fibrocystic disease.

Fig 1 - clinical presentation

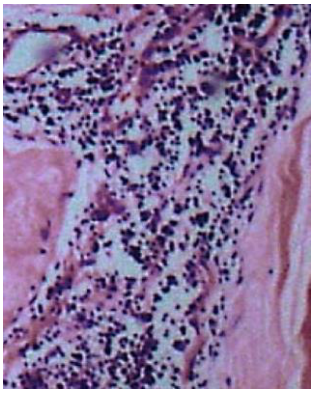

Fig 2 - periductal lymphocytic infiltrates

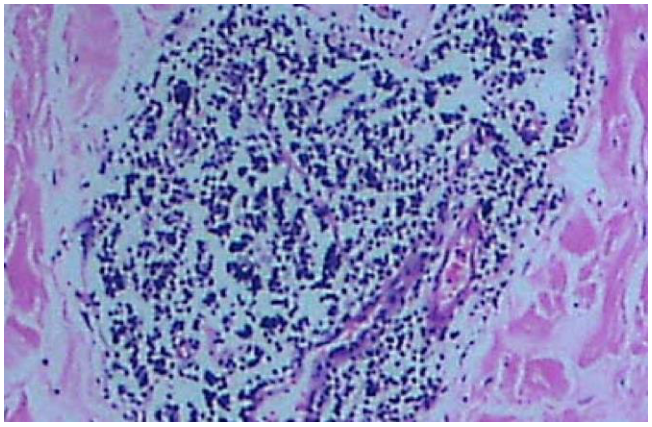

Fig 3 - Perivascular lymphocytic infiltrates

3. DISCUSSION

Fibrocystic disease represents clinical, mammographic and histologic findings. It is common during the fourth and fifth decades of life, usually lasting until menopause. An exaggerated response of breast stroma and epithelium to hormones and growth factors is characterized by breast pain, tenderness and nodularity. Clinical findings for fibrocystic changes in breast range from mild alterations in texture to dense, firm breast tissue with palpable masses and rarely with large palpable cysts. These changes are usually seen on mammography as diffuse or focal radiologically dense tissue. Histologically, in addition to macrocysts and microcysts, women with fibrocystic change have identified solid elements, including adenosis, sclerosis, apocrine metaplasia, stromal fibrosis and epithelial metaplasia and hyperplasia. Depending on the presence of epithelial hyperplasia, fibrocystic changes are classified as nonproliferative, proliferative without atypia, or proliferative with atypia. All three types of changes can occur alone or in combination and to a variable degree, and in the absence of epithelial atypia, these changes represent the histologic spectrum of normal breast tissue. However, epithelial atypia also known as atypical ductal hyperplasia [ADH] is a risk factor for the development of breast cancer. Atypical proliferations of ductal epithelial cells confer increased risk for breast cancer; however, fibrocystic change is not itself a risk factor for the development of breast malignancy. Lymphocytic mastopathy, also named fibrocystic mastopathy, diabetic mastopathy, or sclerosing lymphocytic lobulitis, is a benign clinicopathological entity which mimics and whose main differential diagnosis is breast carcinoma[6], affecting both young and middle-aged women[2]. The prevalence of DMP has been found to be less than 1% of benign breast diseases[7]and the differentials include invasive carcinoma, low grade fibromatosis-like metaplastic carcinoma, fibromatosis, Amyloidosis and MALT Lymphoma[7]. It is associated with the presence of Type 1 insulin dependent diabetes mellitus. Diabetic retinopathy and neuropathy are also associated with this disorder[8]. This condition is uncommon in patients with non insulin dependent diabetes[9], however prevalence is increasing. It may however be present in type 2 diabetics as well as the non diabetic population. Pre menopausal women with diabetic complications such as hand joint disease are also more likely to develop this condition[10] Clinically it may present as single or multiple, firm, palpable, nontender, ill defined mobile masses or diffuse nodularity unilaterally or bilaterally. In 1989, Logan et al[11] mentioned criteria for the radiographic diagnosis of diabetic mastopathy, i.e. a long-term history of IDDM, hard palpable masses, mammographic dense glandular tissue and strong ultrasound acoustic shadowing. Nevertheless, these radiologic findings can still be mistaken for malignancy. Mammography frequently negative, may show asymmetric density or dense breast tissue, without a discrete lump. Magnetic resonance imaging is superior to mammography and ultrasonography in the differentiation between lymphocytic mastopathy and malignant lesions, and may be utilized as an appropriate guidance for the management of the benign lesions. It has been hypothesized to occur due to hyperglycemia causing stromal matrix expansion and accumulation of glycosylated products[12] and a B cell inflammatory response and due to immunologic response to exogenous insulin . The exact pathogenesis is unknown. Ultrasound may show irregular shaped mass(es), indistinct margins,irregular, diffuse shadowing zones, sometimes hypoechoic attenuating nodules , parallel orientation to chest wall, absent vascularity and posterior shadowing[13]. Marked hypoechogenicity and posterior shadowing are probably because of the extensive fibrosis seen in DMP. Parallel orientation and absent vascularity of these lesions significantly differ from a malignant lesion which might be positive predictors of DMP from carcinomas. However, the other ultrasound features of DMP resemble those of invasive cancers, particularly low-grade or Luminal-A subtypes of invasive ductal carcinoma and invasive lobular carcinoma due to which these lesions are usually categorized as BIRADs 4 lesions[14]. Mammography usually describes such lesions as dense breast parenchyma, dense glandular tissue, asymmetric densities, and parenchymal deformity[15] with no spiculations or discrete lumps. CT is a better diagnostic tool for differentiating diabetic mastopathy from malignancy[16]. USG with colour doppler shows no signal as opposed to a malignancy that shows increased vascularity[17]. This corresponds to the avascular histology profile of diabetic mastopathy. Isomoto et al. suggest that MRI with diffusion-weighted imaging may be helpful in distinguishing diabetic mastopathy from malignancy[18]. Contrast MR shows a low, homogeneous, gradual and progressive uptake, without sudden washout[19]. It can be more accurately diagnosed following an ultrasound guided core biopsy or an incisional biopsy and should be correlated with presence of diabetes or other autoimmune conditions. Fine needle aspiration cytology is usually insufficient for diagnosis[20]. Histologic triad of keloidal type fibrosis, lymphocytic inflammation and epithelioid stromal myofibroblasts are seen. Periductal, perilobular and perivascular lymphocytic infiltrates and epithelioid stromal myofibroblasts are seen[21]. Epithelioid features of myofibroblasts may be subtle or absent. Stromal mitoses are usually absent. Tomaszewski et al. were the first to describe Epithelioid like fibroblasts (EFB). These are

rounded epithelioid cells with abundant cytoplasm and oval vesicular nuclei, which are individually isolated by collagen and distributed similarly to spindle fibroblasts. It was stated that they appear uniquely in diabetic mastopathy and are diagnostic when found, although they are not present in all cases [22]. Mature lymphocytic infiltrates are non clonal, predominantly B lymphocytes and lack germinal centers. Recurrence rates may vary; 10 - 30% and it may be prone to single or multiple recurrence in the same breast or the contralateral breast[23]. Surgical excision may lead to recurrence therefore conservative management may be of more benefit in such patients. Unilateral lesions may be managed with excisional biopsy. A 2017 literature review of surgical management of DMP found that, of 178 cases of DMP, 160 patients (89.9%) underwent lumpectomy, four patients (2.2%) had mastectomies and two patients underwent excision as part of a reduction mammoplasty[24]. Allué et al reported performing a nipple sparing mastectomy for a patient with diabetic mastopathy[25]. There have been no reports of associated increased risk of breast carcinoma[26] or lymphoma in these patients. There has been a single report of regression of the lesion. There is no standard management protocol of these lesions. If DMP is histopathologically confirmed, annual physical, mammographic, and ultrasonographic examinations should be done. Some patients present with secondary lesions usually within 5 years of first diagnosis[27]. Literature reports about 42–80% relapse within 5 years after the excisional biopsy. These lesions are usually expanding and may be bilateral[28].If the patient presents with expanding or newer lesions, serial fine needle aspiration cytologies or core biopsies are suggested to rule out malignancy. Excision biopsy is not recommended if clinically and radiologically features are suggestive of DMP[29] [13]. Patients can be reassured and followed up. Surgical excision may be considered if symptomatic, especially if multiple and bilateral masses present.

4. CONCLUSION

This case report highlights a rare benign condition which mimics a varied group of differential diagnoses including invasive carcinoma, low grade fibromatosis-like metaplastic carcinoma, fibromatosis, Amyloidosis and MALT Lymphoma. We reiterate that triple assessment is often not enough to diagnose this condition and a biopsy confirmation is always warranted. As per our report, the patient was counseled and on follow up did not require any invasive procedures. The knowledge of diabetic mastopathy is important to rule out breast carcinoma and thereby not over treat the patient in an otherwise benign condition.

AUTHORS' CONTRIBUTION: Dr. Tannistha Chakraborty conceptualized and collected data and prepared the original and final draft of the manuscript. Dr. Shreya Shetty and Dr. Tannistha Chakraborty were involved in the examination, procedures, management and follow up of the patient. Dr. K.S Ravishankar provided valuable inputs regarding management of the patient, editing and designing of the manuscript.

5. CONFLICT OF INTEREST

Conflict of interest declared none.

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