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UNUSUAL FORMS OF MASTITIS– A SERIES OF THREE CASES WITH REVIEW OF LITERATURE

Pathology

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ABSTRACT

Mastitis is a diverse and broad group of inflammatory condition of breast which is of varied etiology with overlapping clinicopathological features. Autoimmune mastitis is an unusual form of mastitis that is often misdiagnosed or underdiagnosed clinically. Among the various forms of autoimmune mastitis with overlapping histological features that require additional serological tests for confirmation, Diabetic mastopathy and IgG4-related mastitis exhibit characteristic microscopic features. In this article we report three unusual forms of Autoimmune mastitis- One case of Diabetic mastopathy and two cases of IgG4 related mastitis. One of the cases of IgG4 related mastitis is reported in a male patient which is of very rare occurrence. We present this case series to increase the awareness among the clinicians and pathologists about such rare and ever expanding types of Autoimmune mastitis that can be mistaken for malignancy thereby precluding appropriate treatment of the patient.

KEYWORDS

Diabetic mastopathy, IgG4 related mastitis

INTRODUCTION:

Inflammatory lesions of breast referred to as Mastitis are rare and are often seen among women of childbearing age [1] Mastitis can either present as an acute form or as a long standing recurrent chronic disease. The etiology and types of mastitis are of diverse nature posing significant diagnostic challenge to the clinicians and pathologists. Other than infectious causes, Autoimmune etiology is being implicated in many forms of chronic mastitis with varied and overlapping histological patterns and clinical presentations. [1] Unless the clinicians and pathologists are aware of this broad group, Autoimmune mastitis are often underdiagnosed or misdiagnosed thereby precluding proper curative treatment. In this article we report two unusual types of autoimmune mastitis in the form of a series of three case reports with review of literature.

CASE 1:

A 51-year-old female, a known diabetic on oral hypoglycemic drug with poor glycemic control presented with lump in the right breast for 4months duration, associated with few fever episodes and pain. On examination 2*2 cm tender lump that was not freely mobile was noted in the right breast and a clinical diagnosis of breast abscess was made. FNAC was inconclusive and USG revealed a suspicious lesion in right breast following which excision biopsy was performed. Microscopic examination showed breast parenchyma exhibiting areas of dense keloidal type collagenisation, periductal and perilobular lymphocytic infiltrate along with scattered epitheloid myofibroblasts (Fig 1-3). Histopathological features were consistent with Sclerosing Lymphocytic Lobulitis (Diabetic mastopathy).

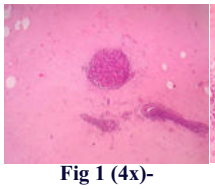

Fig 1 (4x)- Lymphocytic lobulitis with keloidal fibrosis

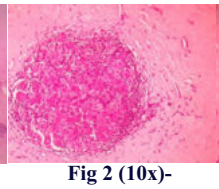

Fig 2 (10x)- Lymphocytic lobulitis

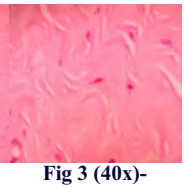

Fig 3 (40x)- Epitheloid myofibroblasts

CASE 2:

A 38-year-old female presented with lump in the right breast for 2 years duration associated with nipple discharge. On examination, 2x1 cm not freely mobile lump was observed in the right breast. There was nipple retraction without any active nipple discharge. USG revealed suspicious lesion and FNAC was inconclusive following which excision biopsy was done. Histopathological examination showed fibrofatty breast parenchyma with dense infiltrate of plasma cells and lymphocytes exhibiting concentric perivascular distribution obliterating the vascular lumen. There were extensive areas of storiform fibrosis distorting the lobular architecture (Fig 4-6). No dilated ductal structures made out. Microscopic features were consistent with Autoimmune mastitis related to IgG4 sclerosing disease. The patient could not afford further workup. Based on the histopathological report appropriate treatment was given and patient was relieved of her complaints.

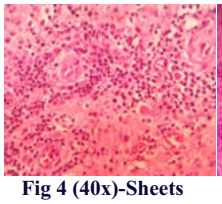

Fig 4 (40x)-Sheets of Plasma cells

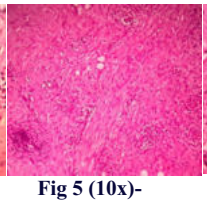

Fig 5 (10x)- Storiform fibrosis

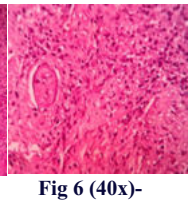

Fig 6 (40x)- Obliterative phlebitis

CASE 3:

A 43-year-old male patient presented with a painful lump in the left breast. On examination a tender lump measuring 1x1 cm was noted in the left breast. FNAC and USG were not done since it was suspected to be breast abscess clinically and the surgeon proceeded with excision biopsy. Histopathological examination of the biopsy sample showed fibrofatty tissue with dense lymphoplasmacytic infiltrate, storiform fibrosis and obliterative phlebitis (Fig 7-9). There was no neutrophilic infiltrate made out. Final diagnosis of Autoimmune mastitis related to IgG4 sclerosing disease was made. Subsequently the patient was treated with medications and was perfectly normal on follow up.

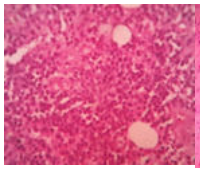

Fig 7 (40x)- Sheets of Plasma cells

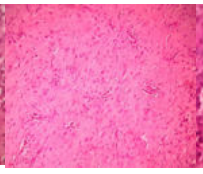

Fig 8 (10x)- Storiform fibrosis

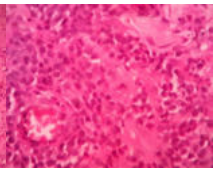

Fig 9 (40x)- Obliterative phlebitis

DISCUSSION:

Mammary gland can be affected by a wide range of autoimmune diseases. All types of systemic autoimmune diseases like diabetes, thyroiditis, SLE, vasculitides, IgG4 related disease etc can involve the breast while cases of breast specific autoimmune condition like Idiopathic granulomatous mastitis have been reported.[1]

Autoimmune mastitis is a rare and broad group of clinical conditions that can clinically be mistaken for other breast lesions like abscess or

malignancy due to its varied presentations. The clinical presentation can be in the form of recurrent mastalgia, painful breast lumps, nipple discharge or retraction.[1]

Autoimmune mastitis can exhibit overlapping histological patterns with the most common features being
i) Lymphocytic infiltrate
ii) Duct ectasia
iii) Granulomatous mastitis &
iv) Vasculitis.

Among the various forms of autoimmune mastitis with overlapping histological features that require additional serological tests for confirmation, Diabetic mastopathy and IgG4 related mastitis exhibit characteristic microscopic features thereby facilitating histopath ological diagnosis.

Diabetic mastopathy or **Sclerosing lymphocytic lobulitis** occurs in Type-1 or Type-2 diabetic patients requiring insulin treatment. Occasional cases also occur in those not receiving insulin therapy.[2] The autoimmune etiology of Diabetic mastopathy was first suggested by Soler and Khardori [3] based on their observation. Miura et al [4] demonstrated the presence of circulating anti-insulin antibodies cross reacting with breast duct epithelium. Tomaszewski et al [5] suggested that sustained hyperglycemia in diabetic patients leads to accumulation of Advanced Glycation End products (AGE) which are highly immunogenic, favouring cytokine production, B-cell proliferation and extracellular matrix remodelling due to increased collagen production all leading to breast inflammation.

Clinically Diabetic Mastopathy patients around the age of 40years present with a hard lump easily mistaken for malignancy. Histologically diabetic mastopathy shows specific and characteristic triad of features:

1) Tight circumscription of lymphocytic infiltrates around lobules (Lobulitis)
2) Keloidal fibrosis
3) Epitheloid myofibroblasts.

IgG4 related sclerosing disease is a recently described entity with varied clinical presentation and multi-organ involvement. It is a chronic inflammatory autoimmune condition and closely resembles a malignant condition clinically due to its tumefactive properties. The pathophysiology of IgG4 related diseases is related to complex immunological mechanism involving TH2 CD4 T cells, B-lymphocytes and plasmablasts resulting in excess IgG4 production.[6]

IgG4 related breast diseases affect mainly females with only few cases reported in males and clinically characterised by unilateral or bilateral hard painful breast lumps mimicking cancer. One of our cases was observed in a male patient which is of rare occurrence. Histopathology remains the gold standard for diagnosis[6] and is characterised by

1. Dense lymphoplasmacytic infiltration
2. Storiform fibrosis
3. Obliterative phlebitis.

An increased number of IgG4 positive plasma cells within the tissue is also part of classical histologic pattern but not essential for diagnosis.[1]

CONCLUSION:

Autoimmune mastitis related to diabetes and IgG4-related sclerosing disease should always be included in the differential diagnosis of breast lump. Awareness among the clinicians and pathologists about these rare and ever expanding broad group of condition will enable early diagnosis and institution of appropriate treatment thereby avoiding radical surgical procedures.

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