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Pathology

CYSTIC NEUTROPHILIC GRANULOMATOUS MASTITIS -A CASE REPORT

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ABSTRACT Cystic neutrophilic granulomatous mastitis (CNGM) is a rare subtype of granulomatous mastitis with a highly distinct histological pattern often associated with Corynebacterium species.(1) To date, in the literature, a total of 141 cases of CNGM presenting at a mean age of 35 years have been reported since 2002.(2) We report herewith a case of CNGM in a 26 year old female of Indian origin who presented to OPD with recurrent left breast abscess. USG guided biopsy was performed which revealed cystic neutrophilic granulomatous mastitis.

KEYWORDS : Cystic, Neutrophilic, Granulomatous, Mastitis, Breast.

INTRODUCTION

A 26 year old female of Indian origin presented to the OPD as a case of left recurrent breast abscess. She is G3P3. Last child birth was four years back and patient breastfed the baby for one year.

Physical examination revealed indurated mass measuring 4X4 cms with an obvious pustule in the upper outer quadrant of the left breast. There were two linear scars seen each 0.5X 0.5 cms of previous incision and drainage done 4 months ago.

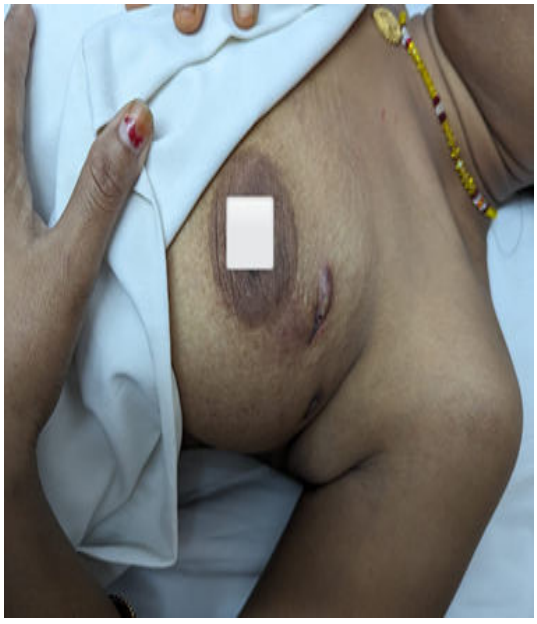

Fig : 1 Clinical Photograph Of The Lesion In The Left Breast Showing Swelling, Redness And Sinus Formation.

Initial left breast USG revealed evidence of two hypoechoic, irregular lesions , at 12 o clock position, measuring 11.5 X 10 mms , 5 cms from areolar region and at 1 o clock position, measuring 16 X 13 mms, close to areolar region. No calcific foci / cystic foci seen in both lesions. No axillary lymphadenopathy noted. Initial Impression on USG was Fibroadenomas of left breast.

Follow up USG a week later ,showed cystic liquefaction in previously noted lesions at 12 o clock and 1 o clock position. Two new cystic areas (12.7 X 7.2 mms and 13.1 X 5.1 mms) at 3 o clock position close to areolar margin were noted. No axillary lymphadenopathy noted. Imaging was suggestive of multiloculated breast abscess of left breast.

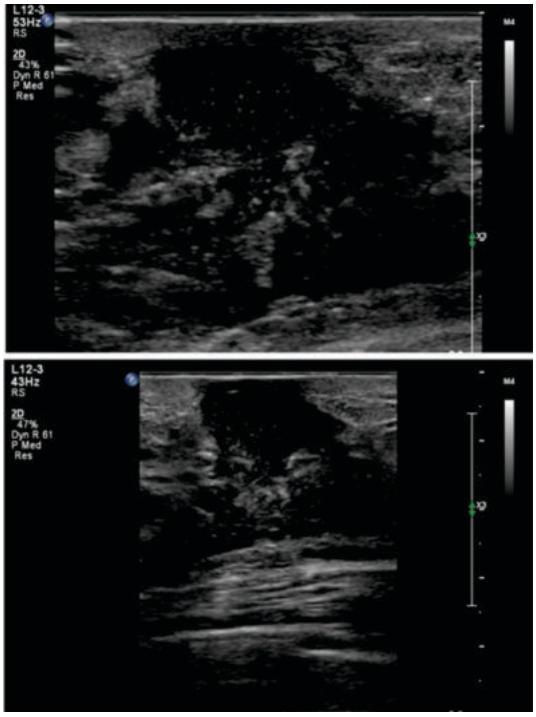

Fig : 2 Follow Up USG Showing Cystic Lesions With Internal Echoes And Posterior Acoustic Enhancement

Biopsy was performed in a private hospital and slides were sent for review which revealed suppurative lipogranulomas comprising of epithelioid cells, lymphocytes, multinucleated giant cells, in the centre of which was seen "empty" lipid vacuole rimmed by neutrophils.

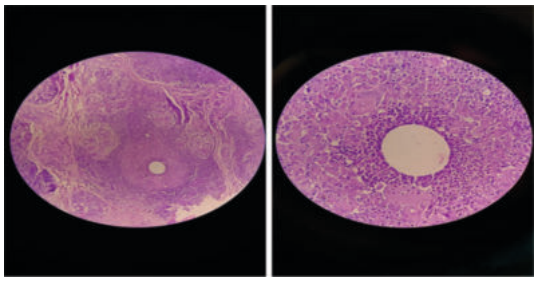

Fig : 3 Low Power View Of The H & E Slide Showing Necrotizing

Granulomas With Central Lipid Vacuole And High Power View Of The Same.

Patient came for further management to the Surgery OPD at our institution. Subsequently FNAC was performed which showed suppurative granulomatous inflammation.

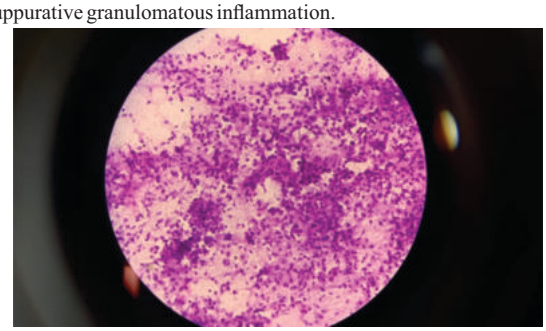

Fig: 4 FNAC Smear Showing Suppurative Granulomatous Inflammation.

The pus was sent for bacterial culture and sensitivity, which came sterile, CBNAAT for Mycobacterium tuberculosis which was negative and Fungal culture which was also sterile.

DISCUSSION

CNGM is a rare subtype of mastitis with a distinct histological pattern that is associated with the Corynebacterium species like Corynebacterium kroppenstedtii. (2).However the organism is fastidious and difficult to culture.(3).The first well-documented compilation of disease associated with this species was published in 1997 by Funke et al.(4).The association of Corynebacterium species with mastitis was first postulated in a review of mastitis cases by Taylor et al in 2003(5).In 2011, Renshaw et al coined the term cystic neutrophilic granulomatous mastitis to draw attention to the distinct pattern of Corynebacterial infection in the breast that includes enlarged vacuoles within neutrophilic inflammation and the presence of Gram positive bacilli within the cystic spaces.(6).

CNGM is usually unilateral, although 8.5 % of patients have presented with bilateral disease. Breast mass, nipple inversion and sinus formation are the most common manifestations. Of the 122 patients reported by Jessie M WU et al , at least 64 (52.5 %) presented with a breast mass. Other symptoms included pain (at least 11.5 %),nipple discharge (10.7 %), erythema (10.7 %) and abscess (12.3 %) (1).Our patient had multiloculated recurrent left breast abscess.

Ultrasound is the preferred imaging modality in which the most common presentation is a mass (72.2%), followed by dilated ducts (11.1%), abscesses (5.6%), edema (5.6%) and fluid collection (5.6%) (1).

Definitive diagnosis is made on histopathological examination. The differential diagnosis for CNGM includes infectious and non infectious diseases.

The Infectious causes include tuberculosis and fungal infection. The granulomas of primary tuberculosis of the breast are well formed, necrotizing and lack the neutrophils and central lipid vacuole that characterize CNGM .Tuberculous granulomas of the breast also involve both ducts and lobules, while CNGM is confined to the lobule. Ziehl- Neelsen stain, PCR and culture would support the diagnosis of tuberculosis and other mycobacterial infections of the breast. Fungal infection will not show central lipid vacuole and fungal culture will be positive.(2).

Non infectious causes of lipogranulomatous inflammation, including fat necrosis and silicone implants can be distinguished from CNGM by the absence of abundant neutrophils and the presence of polarizable material in giant cells. Autoimmune causes of necrotizing granulomatous inflammation, including graulomatosis with polyangiitis and rheumatoid arthritis have been reported in the breast. Granulomatosis with polyangiitis can be distinguished microscopically from CNGM by the presence of necrotizing vasculitis. Rheumatoid nodules presenting as granulomatous mastitis have a central area of fibrinoid necrosis palisaded by histiocytes and plasma cells. Abundant neutrophils have been described in rheumatoid nodules, but lipogranulomas are not present. Serology for ANCA antibodies and rheumatoid factor supports the diagnosis of these specific autoimmune causes of granulomatous mastitis. Sarcoidosis is an idiopathic cause of granulomatous inflammation that is usually multisystemic and involves the breast in <1% of cases. Sarcoid granulomas are well formed, typically non necrotizing and are composed mainly of epithelioid histiocytes and Langhans giant cells. (2). Idiopathic granulomatous mastitis is characterized by the presence of non necrotizing granulomas confined to breast lobules, in which no microorganisms are identified. The disease is immunologically mediated. It is a diagnosis of exclusion.(7)

The first line of treatment for CNGM is antimicrobial medications comprising of doxycycline and sulfamethoxazole. Some women are treated with immunosuppressants (eg. Prednisone) alongside an antibiotic. In some cases, sustained use of these medications can result in disease remission. However , cases that are refractory to medical treatment may have to proceed to surgical resection of breast tissue. One promising immunosuppressive agent , in particular, is Adalimumab, a tumor necrosis factor-alpha inhibitor. Additionally, dapsone may provide benefits by its antimicrobial and anti inflammatory properties. (8).

Our patient is currently on doxycycline 100 mg tds and is responding well.

CONCLUSION

As CNGM is a relatively new entity with few cases reported in literature, this diagnosis is often made after excluding other possible causes and with compatible findings on biopsy. Optimal management of CNGM is unknown, and clinicians often have to draw on the previous experiences with related conditions such as idiopathic granulomatous mastitis or granulomatous lobar mastitis. These may include expectant management, antibiotics, corticosteroids, immunosuppressive medications and surgery. (9).

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