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Case Report

Idiopathic granulomatous mastitis: diagnostic dilemma

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ABSTRACT

We report a case of Idiopathic granulomatous mastitis in a 21 years old female who came to our hospital with complaints of painful lump in her left breast because the results of initial excision of fibroadenoma, when re-exploration was done to rule out any foreign body – excision biopsy was done. However, the final histopathological diagnosis was granulomatous mastitis with suture granuloma, no evidence of malignancy. A review of literature revealed that idiopathic granulomatous mastitis has a tendency to affect young women. Clinical diagnosis and imaging has often been difficult. Complete resection or corticosteroid therapy can be recommended as appropriate treatment. Since recurrence rate is as high as 50%, long term follow up is indicated in such patients.

Keywords: Idiopathic granulomatous mastitis, Corticosteroid therapy, Fibroadenoma

INTRODUCTION

Idiopathic granulomatous mastitis (IGM) is a rare uncommon chronic inflammatory disease of the breast with unclear, unknown etiology.[1] It was first described by Kessler and Wollock in 1972.[2] IGM most commonly occurs in the child-bearing age or with a history of oral contraceptive use.[2] It most commonly presents as a unilateral breast mass. Up to 25% of cases can involve both breasts.[1] IGM may be a self-limiting condition.[1] It may persist for a range of 2-24 months, but chronic conditions may last for several years.[3] Women often present with painful, unilateral, discrete breast masses that have a high recuurence rate.[4] IGM is also considered a variant of periductal mastitis by some experts.[3] The pathologic, clinical and imaging diagnoses of IGM remain difficult. Histologically, IGM demonstrates non caseating granulomas limited to the mammary lobules with or without associated micro abscesses resulting from a local immune process.[5] Possible etiologies include an autoimmune process, trauma, infection, oral contraceptive use, and prolactinemia.[6,7] IGM may also be confused with other conditions besides malignancy, conditions such as tuberculosis, sarcoidosis, erythema nodosum, and Wegener granulomatosis. Thus, histopathological examination is important to confirm evidence of IGM.[8] The ideal treatment of IGM also remains unclear. At present, there is still no universally accepted management strategy for IGM.[10-12]

Damage to the ductal epithelium produced by any of these etiological factors could permit luminal secretions to leak into the lobular connective tissue, thereby causing a localized immune reaction with lymphocyte and macrophage migration.[13]

Studies have demonstrated moderate success with varying options including observation, steroids, and immunosuppressants.[14] Often, surgical management is the last resort, although lesions may recur and result in poor aesthetic outcomes.[8]

CASE REPORT

A 21 year old female patient, unmarried, presented to our centre with complaints of lumps in bilateral breasts, gradually progressed in size, more so on left side over 5 to 6 months. She had no history of any evening rise of temperature, no loss of appetite or weight, no family history of breast carcinoma, no history of similar

complaints in past, no past surgeries pertaining to same. No menstrual cycle disturbances. No history of cyclical mastalgia. On physical examination there was palpable lump of 5×4 cms and 3×2 cms in left breast at upper outer quadrant (1 o'clock) and lower outer quadrant (4 o'clock) positions. In the right breast there was a palpable lump in the upper outer quadrant (12 o'clock) position. A sonomammogram was done for bilateral breasts and features suggested of bilateral multiple fibroadenomas with BIRADS II. Subsequently she underwent excision of fibroadenomas of the left breast. Both the fibroadenomas as mentioned earlier were excised and sent for histopathological examination.

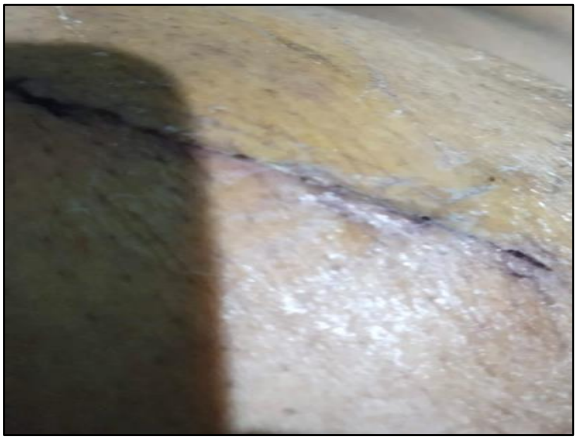

Figure 1: Infection and discharge from the sutured site.

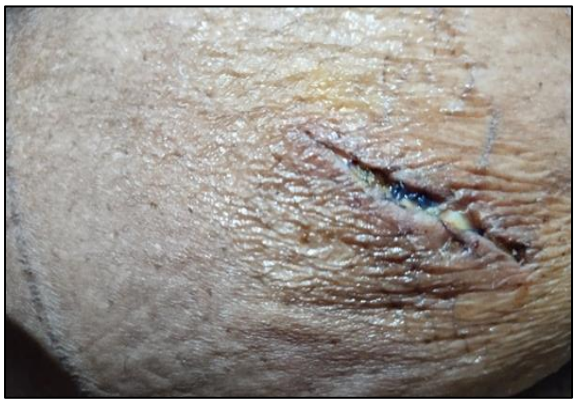

Figure 2: Pus discharge from the sutured wound with surrounding induration.

Subcuticular suture was placed. Postoperative period was uneventful, hence patient was discharged. The histopathological examination reported as fibroadenoma. After 3 weeks patient again came with painful lump in the left breast over the operated area (only at upper quadrant excised site) with pus discharge and surrounding erythematous skin changes, and nipple inversion.

The patient underwent multiple dressings and good antibiotic coverage Tab. Augmentin 625 mg BD was given for a week. After a week of antibiotic coverage the wound still did not show any improvement. A repeat sonomammogram was done. It showed features of post operative changes underlying incision site, with left axillary lymphadenopathy (1.4×0.4 cms). Patient was put on antibiotic coverage Tab. Taxim O 200 mg BD for another 10 days. Later after 7 days patient presented with painful lump at the same area. No fever history. Patient was planned for exploration of the operated wound site. On operating table, pale inflamed breast tissue with surrounding areas of induration was noted, the specimen showed small cavitatory areas with red inflamed tissue.

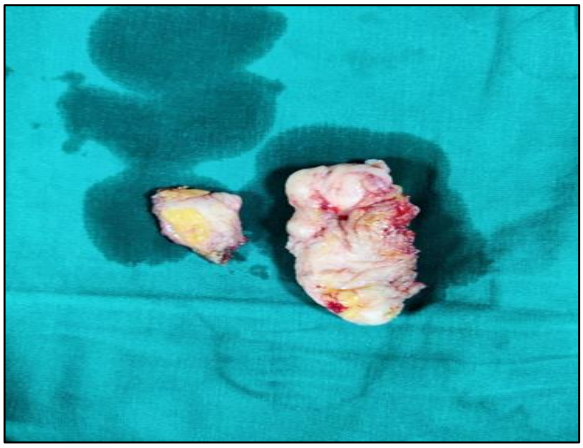

Figure 3: Operated specimen showing pale inflamed breast tissue with surrounding induration.

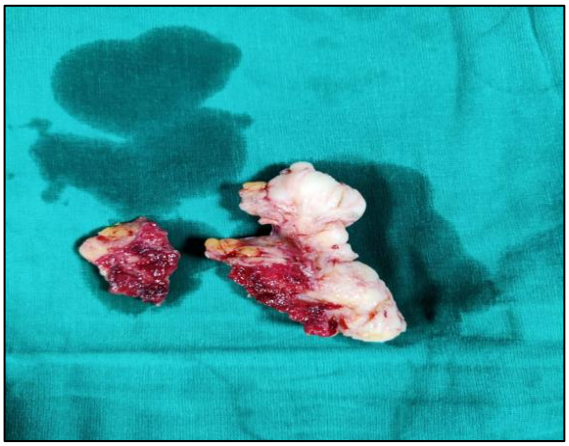

Figure 4: Cavitatory lesions with surrounding inflamed breast tissue suggestive of granulomatous mastitis.

Primary closure of the wound was done and a drain placed. Postoperative period was uneventful. Drain removed on day 2. Histopathological examination reported as chronic mastitis with inflammatory cell infiltrates. Thus, a diagnosis of idiopathic granulomatous mastitis was made after excluding all possible differentials. The patient was started on Tab. Wysolone 10 mg TID on post op day 3 and the and the dose tapered over a month. On postoperative day 8 the sutures were

removed and the breast examined, showed regression of lump and supple breast tissue. Patient responded very well symptomatically to oral corticosteroids therapy.

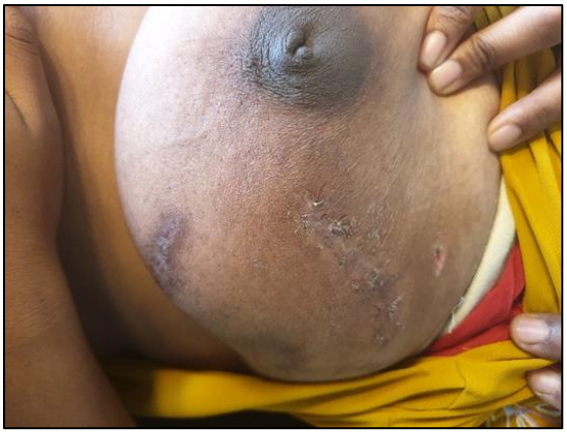

Figure 5: Post-operative healed wound with corticosteroid therapy.

DISCUSSION

Kessler and Wolloch first described IGM in 1972 as a rare benign chronic inflammatory disease of the breast, characterized by presence of non caseating granulomas confined to the breast lobules.[15-18]

The exact pathogenesis of IGM is unknown, although it is thought to be immunologically mediated.[15-19] IGM affects young healthy parous women and is often associated with pregnancy and lactation.[15-18] In our case patient was unmarried young adult female. IGM is diagnosed after exclusion of infection, malignancy, foreign body reaction and other autoimmune disorders.[15-18]

The absence of caseating necrosis and a predominantly neutrophilic background with multiple cavities on histopathology were important clues favoring a diagnosis of IGM.[19] Association between fibroadenoma and IGM is not known and needs further evaluation.

The principle treatment options include antibiotics, surgery, corticosteriod agents, or a combination of them.[16,13,20] Some investigators suggest that systemic corticosteroid therapy may be the initial treatment of choice in IGM, whereas surgical resection should be reserved for recurrent lesions.[20,21]

Immuno- suppressive therapy should be continued until a complete remission is reached, as recurrence rates as high as 50% have been reported.[13] A recent prospective cohort study of 49 women with IGM found that the treatment period required to achieve a full remission varied from 3 to 18 months.[20] Alternatively, some investigators propose that IGM should be treated with wide local excision at the onset of disease, citing a lesser chance of recurrence with surgical therapy.[20] However, review of the literature finds that relapses can still occur despite surgical resection, and it may require repeated, potentially disfiguring surgeries to cure the condition.[18,20] Prompt diagnosis and medical treatment of this rare condition is important, as it may prevent patients from undergoing potentially disfiguring surgery.

Hani et al pointed out that the largest reported series of IGM came from developing countries.[17] Because of this, they suggested that IGM might be the reflection of underdiagnosis of tuberculosis mastitis. In these countries, tuberculosis is endemic and Bacille Calmette-Guerin (BCG) vaccination is done routinely in our patient, there is no history suggestive of or clinically symptomatic for tuberculosis.

Corticosteroids are one the primary options for treating this disease, but the results of previous studies concerning their efficacy have been controversial.

Combination approach may result in a lower rate of recurrence and side effects in IGM patients rather than corticosteroid therapy alone.

A high index of clinical suspicion and multidisciplinary approach is required. Granulomatous mastitis is considered to be idiopathic and the pathogenesis is poorly understood. many patients require careful judgment to ensure optimal type and sequencing of treatments. It can mimic carcinoma of breast. Establishing a diagnosis can be challenging and requires a high index of suspicion with exclusion of infective and autoimmune breast diseases.[9]

Steroid therapy was effective in the treatment of IGM by reducing the lesion size and extent. With regard to the current treatment options available for IGM, surgical excision with combined steroid therapy.[22] Seems the better treatment option compared to steroid therapy without surgical excision. This treatment sequence reduces the rate of recurrence.

CONCLUSION

After treating the patient with anti-biotics and surgical excision, the author administered corticosteroid 10 mg TID for 10 days with a tapering dose of 10 mg BD for 10 days and 10 mg OD for 10 days. On assessment there was disappearance of lump and surrounding inflammation clinically. Hence the author recommends administration of corticosteroid to achieve symptomatic relief.

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REFERENCES

1. Erozgen F, Ersoy YE, Akaydin M, Memmi N, Celik AS, Celebi F, et al. Corticosteroid treatment and timing of surgery in idiopathic granulomatous

mastitis confusing with breast carcinoma. Breast Cancer Res Treat. 2010;123:447-52.
2. Kessler E, Wolloch Y. Granulomatous mastitis: A lesion clinically simulating carcinoma. Am J Clin Pathol. 1972;58:642-6.
3. Al-Khaffaf B, Knox F, Bundred NJ. Idiopathic granulomatous mastitis: A 25-year experience. J Am Coll Surg. 2008;206:269-73.
4. Diesing D, Axt-Fliedner R, Hornung D, Weiss JM, Diedrich K, Friedrich M. Granulomatous mastitis. ArchGynecol Obstet. 2004;269(4):233-6.
5. Tse GM, Poon CS, Law BK, Pang LM, Chu WC, MaTK. Fine needle aspiration cytology of granulomatousmastitis. J Clin Pathol. 2003;56(7):519-21.
6. Erhan Y, Veral A, Kara E, Ozdemir N, Kapkac M,Ozdedeli E, et al. A clinicopthologic study of a rareclinical entity mimicking breast carcinoma: idiopathicgranulomatous mastitis. Breast. 2000;9(1):52-6.
7. Imoto S, Kitaya T, Kodama T, Hasebe T, Mukai K. Idiopathic granulomatous mastitis: case report andreview of the literature. Jpn J Clin Oncol. 1997;27(4):274-7.
8. Asoglu O, Ozmen V, Karanlik H, Tunaci M, Cabioglu N, Igci A, et al. Feasibility of surgical management inpatients with granulomatous mastitis. Breast J. 2005;11(2):108-14.
9. Godazandeh G, Shojaee L, Alizadeh-Navaei R, Hessami A. Corticosteroids in idiopathic granulomatous mastitis: a systematic review and meta analysis. Surg Today. 2021;51(12):1897-905.
10. Wilson JP, Massol N, Marshall J, Foss RM, Copeland EM, Grobmyer SR. Idiopathic granulomatous mastitis: in search of a therapeutic paradigm. Am Surg. 2007;73:798-802.
11. Going JJ, Anderson TJ, Wilkinson S, Chetty U. Granulomatous lobular mastitis. J Clin Pathol. 1987;40:535-40.
12. Heer R, Shrimankar J, Griffith CDM. Granulomatous mastitis can mimic breast cancer on clinical, radiological and cytological examination; a cautionary tale. Breast. 2003;12:283-6.
13. Lai EC, Chan WC, Ma TK, Tang AP, Poon CS, Leong HT. The role of conservative tretament in idiopathic granulomatous mastitis. Breast J. 2005;11:454-6.
14. Azlina AF, Ariza Z, Arni T, Hisham AN. Chronicgranulomatous mastitis: diagnostic and therapeuticconsiderations. World J Surg. 2003;27(5):515-8.
15. Adams DH, Hubscher SG, Scott DG. Granulomatous mastitisearare cause of erythema nodosum. Postgrad Med J. 1987;63(741):581-2.
16. Bes C, Soy M, Vardi S, Sengul N, Yilmaz F. Erythema nodosumassociated with granulomatous mastitis: report of two cases. Rheumatol Int. 2010;30(11):1523-5.
17. Bani-Hani KE, Yaghan RJ, Matalka Ismail I, Shatnawi NJ. Idiopathic granulomatous mastitis: time to avoid unnecessary mastectomies. Breast J. 2004;10(4):318-22.
18. Goldberg J, Baute L, Storey L, Park P. Granulomatousmastitis in pregnancy. Obstet Gynecol. 2000;96(5 Pt 2):813-5.
19. Akcan A, Akyildiz H, Deneme MA, Akgun H, Aritas Y. Granulomatous lobular mastitis: a complex diagnos-tic and therapeutic problem. World J Surg. 2006;30(8):1403-9.
20. Pandey TS, Mackinnon JC, Bressler L, Millar A, Marcus EE, Ganschow PS. Idiopathic granulomatous mastitisea prospective study of 49 women and treatment outcomes with steroidtherapy. Breast J. 2014;20(3):258-66.
21. Sheybani F, Sarvghad M, Naderi HR, Gharib M. Treatment forand clinical characteristics of granulomatous mastitis. Obstet Gynecol. 2015;125(4):801-7.
22. Karanlik H, Ozgur I, Simsek S, Fathalizadeh A, Tukenmez M, Sahin D et al. Can steroids plus surgery become a first linetreatment of idiopathic granulomatous mastitis. Breast Care (Basel). 2014;9(5):338-42.

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