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Short Report

Granulomatous Mastitis

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Abstract – *The aim of the study was to describe the clinical, radiological and pathological features of granulomatous mastitis, highlight the problem of diagnosing such a rare disease, examine the outcome and review treatment options. A retrospective analysis was made of five patients who had granulomatous mastitis diagnosed at the King Khalid National Guard Hospital from 1993 to 1999. In this group the mean age at presentation was found to be 42- years and the mean size of the mass was 5.3 cm. The initial clinical and radiological impression was that of malignancy. The diagnosis was confirmed in one patient using fine needle aspiration and in four by excisional biopsy. Although granulomatous mastitis is a rare disease, physicians should be made aware of its benign nature, and its histological and cytological features in order to make the correct diagnosis.*

Key Words: Breast, Granulomatous, Mastitis.

Granulomatous mastitis, a rare, chronic necrotising granulomatous lobulitis of uncertain aetiology, clinically mimics carcinoma of the breast and has been reported to be persistent or recurrent in over half of the cases in several series.[1] It affects the breasts of relatively young patients and occurs in a lobular distribution.[2]

The first definitive report of granulomatous mastitis was made by Kessler and Wolloch in 1972.[1] Since then numerous cases have been reported. Not long ago the majority of surgeons who dealt with breast disease perceived their role as little more than to differentiate benign conditions from malignancy and to treat the latter. Patients now demand specific management of benign disorders, which were previously ignored.

The main aim of this paper is to create an awareness of the inflammatory process, which may clinically mimic carcinoma, as well as to study the natural history of the disease and describe different management options.

Material and Method

A total of five patients with granulomatous mastitis were seen in King Khalid National Guard Hospital from 1993-1999. Patient notes were obtained and the clinical data noted. Slides of all biopsy specimens were obtained, and where necessary, the blocks were recut and restained. Special stains (Gram, Ziehl-Neelsen, and Grocotts methenamine silver method) were used to identify organisms. Microbiological tests were done on specimens submitted from each patient. Mammography was performed for all cases.

Table 1. Clinical data in patients with granulomatous mastitis

No.	Age	Parity	Years Since last delivery	Size of lesion (cm)	Side	Clinical diagnosis	Pain	Bacteriology	Mammography	Lymphadenopathy	Method of diagnosis	Other data including drug use
1	44	G9P9+0	3y	3	Rt	Malignant	Yes	Negative	Highly suspicious of malignancy	Negative	F.N.A	Mantoux test Negative
2	38	G3P3+0	2y	5	Rt	Malignant	Yes	Negative	Highly suspicious of malignancy	Negative	biopsy	Bromocriptin Discharging sinus
3	30	G1P1O+0	Pregnant	5	Lt	Malignant	Yes	Negative	Highly suspicious of malignancy	Positive	biopsy	–
4	56	G3P3+0	14y	10	Lt	Chronic breast abscess	No	Negative	Abscess	Negative	biopsy	–
5	41	G4P4+0	3Y	3.5	Lt	Chronic breast abscess	Yes	Negative	Abscess	Negative	biopsy	Discharging sinus

Figure 1 : Spiculated, 2.5 cm, uncalcified mass, superimposed over the glandular tissue in the superio-lateral aspect of the left breast. The radiological diagnosis is highly suspicious of malignancy.

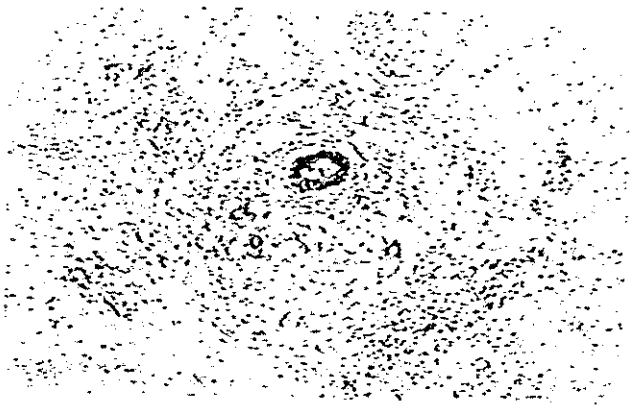

Figure 2 : Low power view of the typical histological features of granulomatous mastitis. An ovoid granuloma with multinucleated giant cells. The surrounding tissue shows a sprinkle of mixed inflammatory cells. The granuloma is situated around a cross section of a terminal ductule. H &E Stain

Follow-up information for periods of 3 months to 6 years was available for all patients. None of these cases had recurrences or developed other granulomatous disease during the follow-up period.

Results

As shown in Table 1, the age of presentation ranged from 30 to 56 years with a mean of 42 years. All patients except one were in the childbearing age. All patients had a history of multiple pregnancies, use of contraceptive pills and breast feeding. Physical examination was unremarkable except for lymphadenopathy in one patient. The size of the masses ranged from 3 to 10 cm with a mean of 5.3 cm. Four out of five patients complained of pain at the site of the lump.

Gram stains, Z.N. stains and cultures were performed in all samples, with negative results. Mammography was

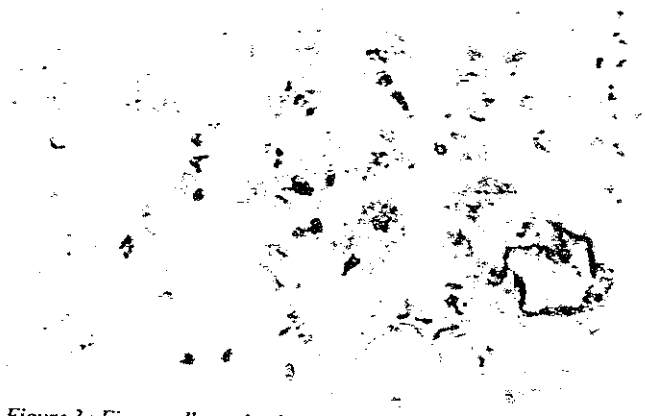

Figure 3 : Fine needle aspiration, cytomorphology of granulomatous mastitis showing a tissue fragment with foot-shaped, epitheloid histocytes. Diff Quik.

highly suspicious of malignancy in three patients and suggestive of abscess in another two. *Figure 1* shows a mammography of one of the patients, which was reported as highly suspicious of malignancy.

Microscopic Findings

In cases 2,3,4 and 5 the diagnosis was made by histological examination of the excised tissue. These showed the classical features of Granulomatous Lobular Mastitis.[2,3] The findings *Figure 2* were generally similar. The predominant lesion was necrotising granulomatous reaction with no caseation. The mixture of inflammatory cells comprised mainly epitheloid histiocytes, lymphocytes and plasma cells as well as a few multinucleated giant cells. These were centred in breast lobules. In case 2 the same process was also periductal. Focal suppuration was also present in all the aforementioned cases. Special stains for *Mycobacteria* (Zeihl-Neelsen and Gram stain) and for fungi (PAS and Grocot's methenamine silver) were made and were all negative.

In case 1 the diagnosis was made on Fine Needle Aspiration material *Figure 3.* The specimen showed a moderately cellular lesion consisting of multiple aggregates of typical foot shaped epitheloid histiocytes with multinucleated giant cells. These were admixed with mature small lymphocytes. No caseation was identified.

Discussion

Until recently benign disorders of the breast were regarded as relatively unimportant, far more attention being given to breast cancer. This resulted in many patients with benign breast diseases receiving very little attention from clinicians. There has been relatively little academic investigation into these disorders. Benign breast disease has also suffered from the major disadvantage of a

hopelessly confusing terminology, inadequate classification, and poor correlation between clinical, radiological, and pathological features.

Granulomatous mastitis, a rare, chronic necrotising granulomatous lobulitis of uncertain aetiology, clinically mimics carcinoma of the breast and has been reported to be persistent or recurrent in over half of the cases in several series.[1] It affects the breasts of relatively young patients in a lobular distribution.[2]

Histologically, the hallmark[2,3] is a necrotising but non-caseating granulomatous reaction. Granulomas are often accompanied by suppuration especially when they coalesce. The process is confined to the perilobular areas unlike that seen in other granulomatous disorders. A variable number of multuinucleated giant cells, lymphocytes, neutrophils and plasma cells may be seen. The histological differential diagnosis[2,3] should include tuberculosis and other infections such as fungal and protozoal diseases. Sarcoidosis should also be considered and can be excluded or confirmed by a constellation of typical clinical findings and exclusion of known causes of granulomas. Poorly formed histiocytic aggregates seen in fat necrosis should not be confused with this condition.

Fine needle aspiration cytology[4-6] findings usually reflect the above histological features and are distinctive in certain cases (as in case 4). However, were the mixed inflammatory component of plasma cells, lymphocytes and neutrophils preferentially sampled, the picture would be non-specific. The differential diagnosis would be conventional breast abscess and fat necrosis.

Granulomatous breast disease (GBD) may present with a clinical picture similar to malignancy,[1-12] as seen in three of our patients but can also present as breast abscess,[9] as in two of our cases, or as mammary fistula.[9,10] Mammography has no specific features; there may be thickening and calcification sometimes mimicking breast carcinoma.[9]

There are conflicting reports on the association of GBD with recent pregnancy,[4,7] breast feeding[1,9] and oral contraceptives.[1,9] A case of granulomatous mastitis in association with hyperprolactinaemia has also been reported.[11] All the patients in this study breast-fed their infants and had contraceptive pills, and all but one did not have a history of recent pregnancy.

The aetiology of this disorder is unknown but there are many theories, as to possible causes including an autoimmune process[13] and an infective agent.[8]

In the past there was a tendency to excise all lumps, and an excessive amount of unnecessary surgery was performed for benign diseases. The main problem from the woman's point of view is fear that such a lump may be cancerous. The clinician must therefore provide a high degree of diagnostic accuracy, while at the same time ensuring that an excessive biopsy rate is prevented. It is now easier to exclude cancer with the development of diagnostic aids such as mammography, ultrasonography, and aspiration cytology. With the increasing use of fine needle aspiration in the diagnosis of breast lesions, cytologists should become familiar with the existence of granulomatous mastitis.[14]

Because of its rarity, no preferred form of therapy has been described. Surgical resection[9-14] of the affected tissue has often been the preferred method of treatment, but many patients have experienced recurrences. DeHertogh and co-workers[10] were the first to advocate the use of corticosteroids for the treatment of granulomatous mastitis. Four of our patients had surgical resection with no recurrence during a follow-up period of 2 to 4 years. One patient was treated conservatively and was discovered to be pregnant just before steroids could be started but fortunately she did well and the mass regressed in size. Provided a definitive diagnosis is made on biopsy, conservative management is the treatment of choice, as it is associated with less episodes of recurrence and wound discharge compared with those treated by more extensive surgery.[15,16]

We concluded that although granulomatous mastitis is a rare disease, physicians should be aware of its existence, its benign nature, and its typical histological and cytological features to make the correct diagnosis.

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