Idiopathic granulomatous mastitis: report of 3 cases and a review of the literature

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Abstract

Idiopathic granulomatous mastitis is a rare, inflammatory, and benign breast disease characterized by non-caseating granulomata and microabscesses limited to breast lobules. This condition presents as a firm breast mass that is clinically and radiologically indistinguishable from breast cancer, or as multiple or recurrent abscesses, or mastitis in a young non-lactating woman. Almost always, the diagnosis is made after surgical interventions to rule out other pathologies as differential diagnoses especially tuberculosis which is endemic in our country.

We are going to report 3 cases of granulomatous mastitis besides a brief review of the literature. We emphatically recommend that although this entity should be kept in mind as a rare differential diagnosis, it should not be considered as the first one.

Keywords: Chronic mastitis, Breast mass, Breast abscess, Granuloma, Idiopathic granulomatous mastitis

Introduction

Idiopathic granulomatous mastitis (IGM), first described by Kessler and Wolloch in 1972, is a rare benign inflammatory disease of the breast in young women. Clinically and histologically, it is a mimicker of acute infection or carcinoma of the breast.

Its etiology is unknown, and autoimmune pathogenesis has been advocated. Histologically, the disease is characterized by the occurrence of numerous granulomatous lesions with multinucleated cells of the Langhans type and focal central necrosis [1].

All specific infectious, granulomatous and neoplastic diseases must be excluded for a correct therapeutical approach [2]. Idiopathic granulomatous is mastitis (IGM) characterized by non-caseating granulomata and microabscesses limited to breast lobules. The disease is presented mostly by multiple and recurrent breast abscesses or a firm mass that is clinically and radiologically not distinguished from breast cancer. Frequently, patients are young and multiparous women, some, complain of mastalgia [1-4].

Almost always, the condition is diagnosed after surgical interventions (excisional or incisional biopsy). Two cases referred to the surgical ward and were candidates for mastectomy due to the high suspicion of breast cancer or inflammatory carcinoma. They were treated post-operatively as having tuberculosis (TB) and developed chronic sinus after antituberculous empiric chemotherapy. For this reason, this disease is explored and reviewed in the literature.

Although some organisms may be found in the lesions of granulomatous mastitis, they are...
not supposed to be the causative agents and as a
matter of fact, the disease is idiopathic [4]. This
condition must be differentiated from the other
granulomatous diseases such as tuberculosis.
Granulomatous mastitis has a great tendency to
recur but usually resolves spontaneously even
without treatment [2-4].

There is no consensus about the best treat-
ment for this disease but it is believed that prop-
er management and close follow-ups are the
preferred choices. Steroids have been used in
the treatment course and considered effective in
some recurrent or intractable ones but, drug
side effects or relapse may be inevitable. In a
study conducted in Australia, methotrexate was
trialed in refractory cases after surgery or a
course of steroid therapy and associated with
successful results [5]. It is obvious that surgical
intervention per se, can not prevent recurrence
of the disease and actually can lead to wound in-
fecction persistent wound discharge, suppurative
sinus formation or even wound nonhealing
[6,7]. On the other hand, it is necessary to have
histologic confirmation and also surgical sam-
pling is inevitable [8].

For abscesses, simple drainage accompanied
by incisional biopsy of the abscess wall is ade-
quate and constitutes the standard treatment
[8]; but for a breast mass that mimics cancer, it
is preferred to obtain a tissue sample by fine-
needle-aspiration (FNA) or core needle biopsy
[5,9]. Needle aspiration or a small incision to
drain abscesses is preferred if feasible.

Both the aforementioned procedures were
performed by the author and then reported on
this paper.

Case reports

Case 1:

A 41-year-old multiparous woman was re-
ferred to the surgical ward because of pain in
the right breast in addition to a palpable mass
with surrounding erythema. She also com-
plained of nipple purulent discharge. The symp-
toms were initiated one month prior to admi-
sion and she was febrile when referred. She had
a history of the same breast abscess drainage
last year, accompanying psychologic disorder
necessitating antipsychotics and biperiden. She
was not lactating. Ultrasonography revealed a
large retroareolar abscess measuring 8 centime-
ters in diameter. Abscess drainage with abscess
wall biopsy was performed under general anes-
thesia. It resembled malignant but on patholog-
ic examination, granulomatous perilobular
mastitis was reported. The patient suffered a de-
layed infection and recurrent contralateral
breast abscess formation which was treated by
recurrent aspirations. Abscess discharge culture
was negative for acid fast bacilli(ABF) and
polymerase chain reaction (PCR) confirmatory
test was also negative. Discharge culture how-
ever, revealed nonspecific bacterial growth.

Case 2:

A 34-year-old woman with a history of nor-
mal vaginal delivery at the age of 29, was re-
ferred because of a tender mass accompanying
a sense of heaviness in her right breast. She was
not lactating and there was no history of oral
contraceptive (OCP) consumption. She had a
history of taking levothyroxin for 15 years be-
fore admission because of a benign goiter be-
sides taking antipsychotic agents. On ultra-
sonography, the mass was detected as solid and
heterogenous in echo pattern and on mammog-
raphy, a hyperdense mass with irregular board-
ers suspected of infiltrative malignancy. On
pathologic tissue evaluation, chronic mastitis
with granulomatous changes was reported. Af-


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therapy, breast masses with sonographic features suspected of malignancy were detected but pathologic evaluation resulted in the same diagnosis. Afterwards, the patient was followed-up, for her condition.

Case 3:
A 24-year-old girl was referred with left periareolar breast abscess. On examination, the whole breast was indurated and inflamed. Sonographically, the abscess was confirmed. Hence, surgical drainage was done and a biopsy was taken from the inflamed abscess wall but revealed granulomatous mastitis. Tissue smear and culture for AFB was negative. There was a history of convulsive disorder and antiepileptic agent (eg. phenytoin & valproate sodium) consumption since childhood. Following abscess drainage, a suppurative sinus complicated the surgical site and it took a long time to be healed. In less than 6 months, the breast abscess recurred and was managed by needle aspiration.

Discussion
Granulomatous mastitis was first described by Kessler & Wolloch in 1972. They reported 5 cases with the initial diagnosis of malignant breast mass who developed recurrent abscess formation containing granuloma subsequently. They emphasized that this rare disorder had a tendency to recur chronically [5].

Other systemic granulomatous disorders like Wegener’s granulomatosis or sarcoidosis must be ruled out to establish the final diagnosis of granulomatous mastitis[3,4]. In primary granulomatous mastitis, the inflammatory process is confined to breast lobules and microabscesses may occupy the whole lobule. Areas of necrosis may also develop. Giant cells and non-caseous epithelioid cells constitute the nonspecific granulation tissue in granulomatous mastitis and those composed of foreign body cells and the Langhans’ cells. In the center of the granuloma, polymorphonuclear cells (PMNs) are more frequent than other cells and duct ectasia may be absent [1,3].

The most important granulomatous disorder which must be differentiated is tuberculosis (that is especially endemic in our country). In all 3 of the aforementioned cases, tissue acid fast staining and the PCR test were negative for tuberculosis. It is noteworthy that we did not find any caseating necrosis (the key point to de-

Fig. 1. Photomicrograph shows completely affected breast lobules by chronic inflammation and granulomatous formation. (Hematoxylin and eosin; magnification × 100.)
termine tuberculosis) in our tissue specimens. In all suspected cases, inflammatory carcinoma must be ruled out; especially because it affects nonlactating women. Hence, pathologic evaluation of the lesion is strongly suggested.

The Granulomatous mastitis etiology is unknown, and more common in young women. No correlation between breast feeding or OCP consumption and the mentioned disorder has been found so far. Kessler & Wolloch have supported the autoimmune etiology mostly because microscopic features are similar to other granulomatous disorders (like granulomatous thyroiditis or orchitis).

Treatment of the granulomatous mastitis is controversial [12]. Surgery is not abruptly beneficial and in most of cases, simple drainage and incisional biopsy are adequate [8]. De-Hertogh and colleagues showed that short courses of high dose prednisolone was efficient although the follow-up period was shorter to precisely evaluate recurrence rate [12].

In several studies, the diagnostic accuracy of FNA has been reported as 87-90%.

The aforementioned cases of granulomatous
Mastitis who all presented with distinct clinical features of the disease, are characteristic examples of clinical diagnostic pitfalls and the necessity of histologic diagnosis to rule out other benign and malignant pathologies of the breast.

Granulomatous mastitis predominantly occurs in nonlactating women [3,4], so, it is more probable to assume the clinical feature as a malignant process [1,2,3], rather than an inflammatory disorder like mastitis or breast abscess. It must be considered as a rare differential diagnosis (but not the first one) in breast specialists approach.

Conservative and symptomatic treatment should follow the surgical one at least to definitively diagnose the disorder and rule out the differential ones. Medical treatments such as steroids or methotrexate, are under survey [1,8,12]. The probability of drug resistance, recurrence of the disease after drug discontinuation and the risk of drug side effects should be considered when medical therapy is selected [8,11].

By reviewing the literature, granulomatous mastitis is more common in Eastern countries like China and Turkey where it can be epidemiologically significant. Performing similar studies in Iran can also be helpful.

In addition, in all 3 cases of this paper, there was a history of psychoactive drug consumption that can be noteworthy but further studies with a larger sample-size are required to determine the probable correlation. As a whole, more clear evidence is needed to reveal its pathogenesis.

**Conclusion**

It is necessary to consider granulomatous mastitis as a rare differential diagnosis from other pathologies and not the first one. More common benign and especially malignant disorders of the breast should be ruled out prior to making a definite diagnosis.

**References**