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Idiopathic granulomatous mastitis masquerading as a postbiopsy abscess

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Idiopathic granulomatous mastitis (IGM) is a rare condition of unknown etiology with nonspecific findings. We present an unusual case of IGM manifesting after breast biopsy in a 42-year-old Turkish woman. IGM should be considered in the differential diagnosis when mastitis, carcinoma, and systemic diseases have been excluded and especially in the setting of a postbiopsy infection that is not responsive to antibiotic therapy.

Introduction

Idiopathic granulomatous mastitis (IGM) is a rare condition of unknown etiology. It mainly affects young women of childbearing age but has been reported in men and elderly women as well. What makes IGM particularly important is its propensity to resemble breast cancer (1).

Although IGM is poorly understood, with a variable duration, it does appear to have some predilections. IGM typically occurs in young parous women within a few years of childbirth (2). A significant proportion of these women have a history of breastfeeding for more than one year. Some studies have described a predilection for Hispanic and Asian women in IGM (3), while others have refuted any ethnic predispositions (4). Women often present with painful, unilateral, discrete breast masses that have a tendency to recur (5). In addition, many women present with inflammation and skin ulceration, including fistula formation and axillary lymphadenopathy, resembling inflammatory breast cancer. IGM is also considered a variant of periductal mastitis by some experts (2).

The pathologic and imaging diagnoses of IGM remain difficult. Histologically, IGM demonstrates noncaseating granulomas limited to the mammary lobules with or without associated microabscesses resulting from a local immune process (6). On ultrasound, IGM presents as hypoechoic lobulated or irregular masses. Mammography may be normal or may demonstrate ill-defined masses, or areas of focal asymmetry with or without associated architectural distortion; enlarged axillary lymph nodes may or may not be visualized (7-9). On MRI, IGM may mimic inflammatory breast cancer. It is often difficult to distinguish IGM from breast cancer based on imaging findings (7-8).

The cause of IGM remains to be elucidated. Possible etiologies include an autoimmune process, trauma, infection, oral contraceptive use, and prolactinemia (10-11). IGM may also be confused with other conditions besides malignancy, conditions such as tuberculosis, sarcoidosis, erythema nodosum, and Wegener granulomatosis. Thus, it is important to confirm evidence of IGM on histopathology (12).

The ideal treatment of IGM also remains unclear. Studies have demonstrated moderate success with varying options including observation, steroids, and immunosuppressants (13). Often, surgical management is the last resort, although lesions may recur and result in poor aesthetic outcomes (12).

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Case report

A 42-year-old asymptomatic Turkish woman, para 2, presented with areas of focal fibroglandular asymmetry in the right breast on screening mammography. There was no personal or family history of breast carcinoma. She delivered her last child 4 years before presentation and had breastfed continuously for 2 years. Sonographic evaluation revealed oval, well-defined, benign-appearing isoechoic-hypoechoic masses suggestive of fibroadenomas, at 6, 9, and 10 o'clock (Fig. 1). The lesion in the 6 o'clock subareolar location was palpable to the breast surgeon.

pathologic findings were thought to represent a chronic process. Clinical followup was the agreed-upon course of management.

Five weeks later, the patient returned with erythema, swelling, and drainage of purulent fluid through several skin ulcers and fistulas in her right breast. These findings began a few days following the core biopsies (Fig. 2). Antibiotic treatment for postbiopsy mastitis was initiated. Ultrasound revealed a 5cm complex fluid collection with sinus tracts centered between the 6 o'clock and the 9 o'clock positions (Fig. 3). Fluid aspirated from the collection was

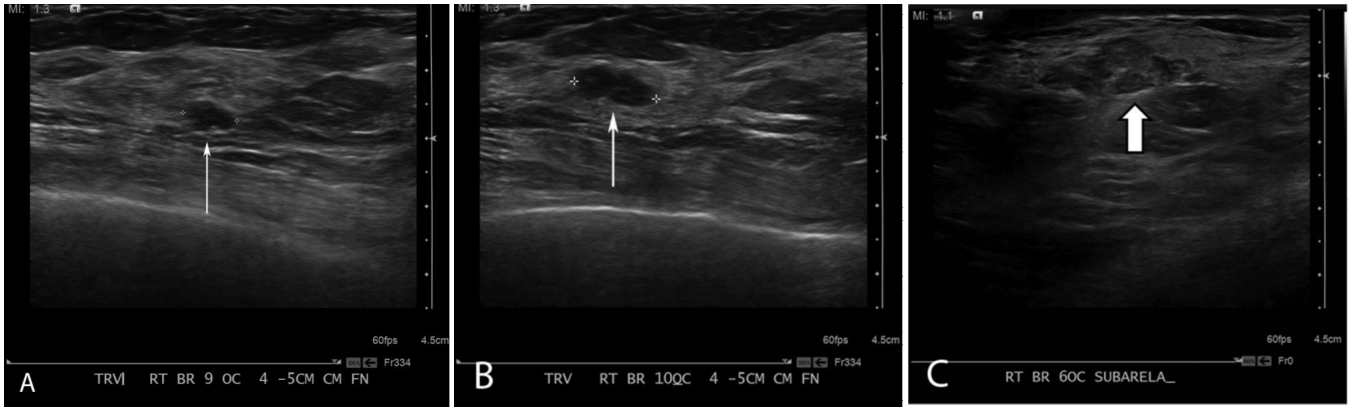

Figure 1. 42-year-old woman with idiopathic granulomatous mastitis. (A) A 0.8cm solid oval mass (arrow) in the 9 o'clock location. (B) A 1.2cm solid oval mass (arrow) in the 10 o'clock location. Both of these lesions were proven by biopsy to be benign breast tissue with sclerosing adenosis and stromal fibrosis. (C) A 1.5cm lobulated oval solid mass in the 6-to-7 o'clock subareolar region (arrow), the area of the palpable abnormality. Biopsy showed benign breast tissue with marked inflammation, multinucleated giant cell reaction, and abscess formation.

Ultrasound-guided core biopsies of all three lesions were performed. The pathology results were benign, with the pathology from the palpable lesion at 6 o'clock remarkable for findings of benign breast tissue with marked inflammation, multinucleated giant-cell reaction, and abscess formation. As the patient was asymptomatic, and the imaging appearance was not concordant with an acute abscess, the

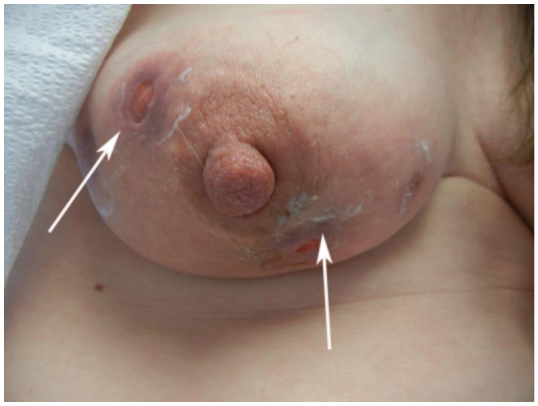

Figure 2. 42-year-old woman with idiopathic granulomatous mastitis. Dime-sized areas of induration and erythema at the sites of prior biopsies (arrows). Pus drainage was reported.

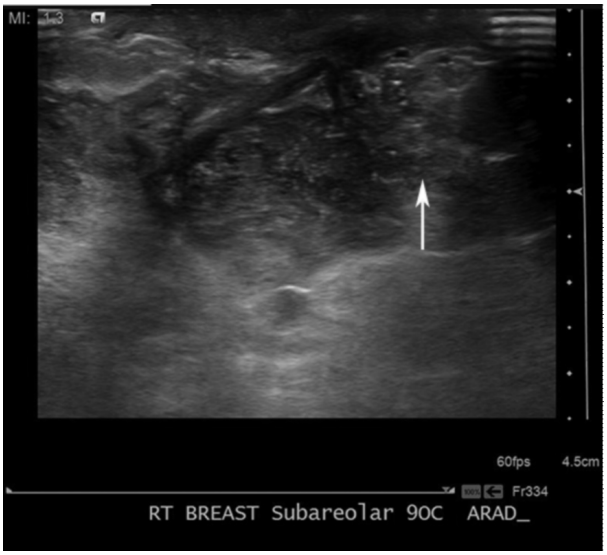

Figure 3. 42-year-old woman with idiopathic granulomatous mastitis. Antiradial right breast subareolar ultrasound demonstrates a 5cm complex fluid collection (arrow) at the site of the previous biopsy. Initially this was thought to be a postbiopsy abscess.

negative for malignant cells or bacterial growth. As the mastitis worsened, methicillin-resistant Staphylococcus aureus (MRSA) was considered. The patient was switched to the appropriate broad-spectrum antibiotics. Repeat aspiration did not reveal any organisms.

Evaluations for tuberculosis and sarcoidosis were negative. As the swelling, erythema, and fistula formation persisted, the decision was made to undergo surgical debridement. Surgical pathology revealed marked non-necrotizing granulomatous inflammation, predominantly periductal in location, with an exuberant acute and chronic organizing inflammatory response. The patient's clinical condition improved markedly following the debridement.

Discussion

This is the first reported case of IGM occurring either secondary to or associated with a percutaneous biopsy.

IGM is an exceedingly rare disease with nonspecific clinical findings. It is a diagnosis of exclusion, made after malignancy and other known granulomatous diseases such as mycobacterial infections and sarcoidosis have been ruled out (2, 6, 10). Studies have shown that IGM is associated with a history of childbirth and prolonged breastfeeding within the previous 5 years (14-15), consistent with our patient's history. It has been hypothesized that prolonged breastfeeding results in long-term distention of the acini and the ducts, facilitating rupture of these structures and inducing a granulomatous response (16). Similarly, we suspect that our patient may have had a previous subclinical granuloma associated with a chronically clogged duct, and that this was inadvertently opened during the biopsy procedure, with subsequent inflammation and the development of full-blown granulomatous mastitis. The absence of caseating necrosis and a predominantly neutrophilic background on histopathology were important clues favoring a diagnosis of IGM (17).

As the fluid drained from the abscess site was sterile, we also considered whether this could be a reaction to the titanium marking clips typically placed following biopsy procedures. Problems with such clips are rare but may occur. To the best of our knowledge, there has been only one reported case of an allergic reaction to titanium clips placed during breast surgery (18).

Infection following percutaneous biopsy, as seen in this case report, is an infrequent but known complication. IGM is a rare and diagnostically challenging condition. It should be considered in the differential diagnosis when mastitis, carcinoma, and systemic disease have been excluded, and should also be considered in the setting of a postbiopsy infection, unresponsive to antibiotic therapy.

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