Idiopathic granulomatous mastitis with erythema nodosum simulating breast abscess in pregnancy: A case report

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Abstract

Granulomatous mastitis is a rare benign inflammatory condition of the breast and is known to be associated with pregnancy. A 25-year-old Hispanic G2P1 at 17 weeks gestation presented to the emergency department with findings consistent of a breast abscess. The abscess failed to resolve with incision and drainage followed by multiple courses of antibiotic therapy. A biopsy was then obtained and yielded a diagnosis of granulomatous lobulitis. The patient was treated with steroids and her symptoms resolved. Granulomatous lobulitis may present with characteristics of various clinical entities including neoplasm or, as in this case, abscess. Clinicians should consider a diagnosis of granulomatous mastitis in cases of recalcitrant breast abscess.

Keywords

Complications, high-risk pregnancy, immunology, maternal–fetal medicine, neoplasm

Case report

A 25-year-old Hispanic G2P1 at 17 weeks gestation with an uncomplicated prenatal course, presented to the emergency department with fever, erythema nodosum, left breast erythema, tenderness, and swelling. Findings were consistent with a breast infection with a WBC of $X10^9/L$ and sedimentation rate of 89 mm/h. Urine and blood cultures were negative. She was treated with Nafcillin without improvement. A methicillin-resistant *Staphylococcus aureus* infection was suspected and antibiotics were changed to vancomycin and ceftazidime. Rheumatology consultation diagnosed the patient with erythema nodosum secondary to primary infection. There was no serologic evidence of lupus, vasculitis, or sarcoidosis. Breast ultrasound was consistent with an abscess. Surgery consulted and she underwent an incision and drainage of the left breast lesion. Postoperatively, the erythema nodosum and breast lesion resolved with prednisone 5 mg twice daily for one week followed by a two week taper.

Despite patient compliance with the medication regimen, her left breast abscess continued to drain. She returned at 20 and 27 weeks gestation and had a second and third incision and drainage. During both repeat occurrences, breast cultures remained negative. She received multiple course of antibiotics. Biopsy showed acute and chronic inflammation with necrosis consistent with abscess.

At 34 weeks, the patient presented with worsening breast symptoms. Her clinical course, labs, and pathology were reviewed (Figure 1). Upon review of the histologic specimens, a noncaseating granulomatous inflammation in the breast lobules composed of epithelioid cells and multinucleated giant cells was identified. The final diagnosis was determined to be granulomatous lobulitis.

Treatment with Prednisone was initiated with a dose of 40 mg daily with a taper to 10 mg daily. The patient had an immediate response, the lesions resolved, and no new areas appeared. Her symptoms flared when Prednisone was decreased to 5 mg daily and she remained on Prednisone 10 mg daily for the remainder of her prenatal course. She had an uncomplicated delivery.

Post-partum, she was stable on 5 mg of Prednisone daily. She was offered surgical resection and methotrexate therapy, both of which she declined. She takes Prednisone for short periods when she has an exacerbation. Five-year follow-up demonstrates no residual disease and well-healed breasts with scar tissue.

Discussion

Granulomatous mastitis is a rare condition. It typically presents in parous women as a hard, tender, extra-areolar breast lump occurring usually within two years post-partum. Nipple retraction, peau d'orange-like changes, and axillary adenopathy may be present.¹ Symptoms can appear as an infection and present with an associated periarthritis. Differential diagnosis includes infective mastitis, vasculitic lesions, plasma cell mastitis, duct ectasia post-partum, or reaction to foreign body (i.e. silicon leakage from breast implant).² Extra-mammary manifestations of granulomatous mastitis are very rare.³ The diagnosis often depends on exclusion of other granulomatous diseases including tuberculosis, sarcoidosis or a foreign body reaction¹ and should be differentiated from carcinoma.⁴ Biopsy is necessary to make a diagnosis.

The aetiology of granulomatous mastitis is currently unknown. Due to morphological similarities to granulomatous thyroiditis, allergic and immunologically mediated causes have been suggested.³ Concomitant erythema nodosum in several previously reported cases^{2,5,6} support altered immune status as a potential etiology. The immune reaction may be in response to extravasated milk secretion, breast trauma, or infection.^{7,8}

Histologically, features that have been described include necrosis, giant cells, epithelioid histiocytes, granulomas, and neutrophils present in the background.⁹ A discrete, characteristic noncaseating granulomatous lobulitis is common. The granulomata often coalesce and form microabscesses.² Despite these findings, definitive diagnostic cytological criteria for granulomatous mastitis remain poorly defined.

The clinical course of granulomatous mastitis may be self-limiting²; however, intervention is frequently required. After infective causes

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Figure 1. (a) Breast appearance at 34 weeks after the third incision and drainage procedure. (b) Breast appearance at 36 weeks after two weeks of steroid therapy. (c) Breast appearance at 38 weeks after four weeks of steroid therapy. (d) A low power view showing the granuloma next to a lobule. (e) A higher power showing the granuloma intimately associated with the lobule. (f) A high power view showing the classic granuloma composed of epithelioid cells, a Langhans type giant cell, and a scattering of acute and chronic inflammatory cells.

have been excluded, antibiotics and steroids are the mainstay of treatment. Steroids have a variable clinical response and recurrence may be problematic after steroid withdrawal.¹⁰ Treatment may require the addition of anti-inflammatory drugs or colchicine. The use of methotrexate has also been successful. In case of formation of abscesses, antibiotic therapy should be instituted before steroid therapy.⁷ Commonly, the clinical course has slow resolution and recurrence rates can be up to 50%. In resistant cases, surgical excision is indicated.¹¹

Competing interests

None declared.

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Ethical approval

Written consent for publication was given by the patient.

Contributions

RL, DG, and MIR researched literature and conceived the case report. TJM reviewed the slides and provided pathologic evaluation. RL and RLP wrote the first draft of the manuscript. All authors reviewed and edited the manuscript and approved the final version of the manuscript.

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