Painful Ulcerating Lesions on the Breast

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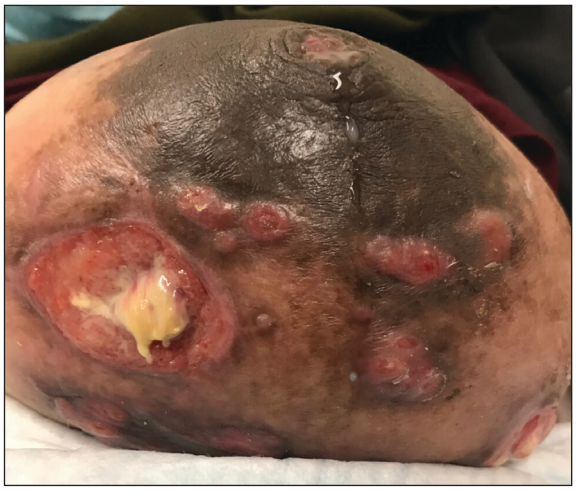

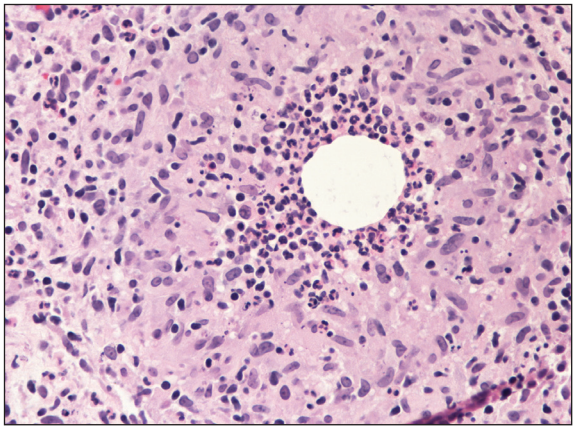

A 36-year-old puerperal woman presented with painful, unilateral, ulcerating breast lesions (top) of 3 months' duration that developed during pregnancy and drained pus with blood. No improvement was seen with antibiotics or incision and drainage. Biopsy of a lesion showed stellate granulomas with cystic spaces and suppurative lipogranulomas where central lipid vacuoles were rimmed by neutrophils and an outer cuff of epithelioid histiocytes (bottom). Acid-fast bacilli, Grocott-Gomori methenamine-silver, Gram, and Steiner staining did not reveal any microorganisms. Additionally, wound cultures were negative.

WHAT'S YOUR **DIAGNOSIS?**

a. cystic neutrophilic granulomatous mastitis
b. hidradenitis suppurativa
c. mycobacterial infection
d. pyoderma gangrenosum
e. sarcoidosis

PLEASE TURN TO PAGE 150 FOR THE DIAGNOSIS

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THE **DIAGNOSIS:** Cystic Neutrophilic Granulomatous Mastitis

The histopathologic findings in our patient were characteristic of cystic neutrophilic granulomatous mastitis (CNGM), a rare granulomatous mastitis associated with *Corynebacterium* and suppurative lipogranulomas. Although not seen in our patient, the lipid vacuoles may contain gram-positive bacilli.[1] The surrounding mixed inflammatory infiltrate contains Langerhans giant cells, lymphocytes, and neutrophils. Cystic neutrophilic granulomatous mastitis is seen in parous women of reproductive age. Physical examination demonstrates a palpable painful mass on the breast. Wound cultures frequently are negative, likely due to difficulty culturing *Corynebacterium* and prophylactic antibiotic treatment. Given the association with *Corynebacterium* species, early diagnosis of CNGM is essential in offering patients the most appropriate treatment. Prolonged antibiotic therapy specifically directed to corynebacteria is required, sometimes even beyond resolution of clinical symptoms. The diagnosis of CNGM often is missed or delayed due to its rarity and many potential mimickers. Clinically, CNGM may be virtually impossible to discern from invasive carcinoma.[1]

Our patient was treated with vancomycin and cefepime with incision and drainage as an inpatient. Upon discharge, she was started on prednisone 1 mg/kg daily tapered by 10 mg every 5 days over 1 month and doxycycline 100 mg twice daily. She was then transitioned to topical hydrocortisone and bacitracin; she reported decreased swelling and pain. No new lesions formed after the initiation of therapy; however, most lesions remained open. Cystic neutrophilic granulomatous mastitis remains a challenging entity to treat, with a variable response rate reported in the literature for antibiotics such as doxycycline and systemic and topical steroids as well as immunosuppressants including methotrexate.[2,3]

Cystic neutrophilic granulomatous mastitis can be distinguished from hidradenitis suppurativa clinically because ulcerating lesions can involve the superior portions of the breast in CNGM, whereas hidradenitis suppurativa typically is restricted to the lower intertriginous parts of the breast. Other mimics of CNGM can be distinguished with biopsy. Histology of pyoderma gangrenosum lacks prominent granuloma formation. Although sarcoidosis and mycobacterial infection show prominent granulomas, neither show the characteristic lipogranulomas seen in CNGM. Additionally, the granulomas of sarcoidosis are much larger and deeper than CNGM. Mycobacterial granulomas also typically reveal bacilli with acid-fast bacilli staining or via wound culture.

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