

Mammary subcutaneous basidiobolomycosis in a male

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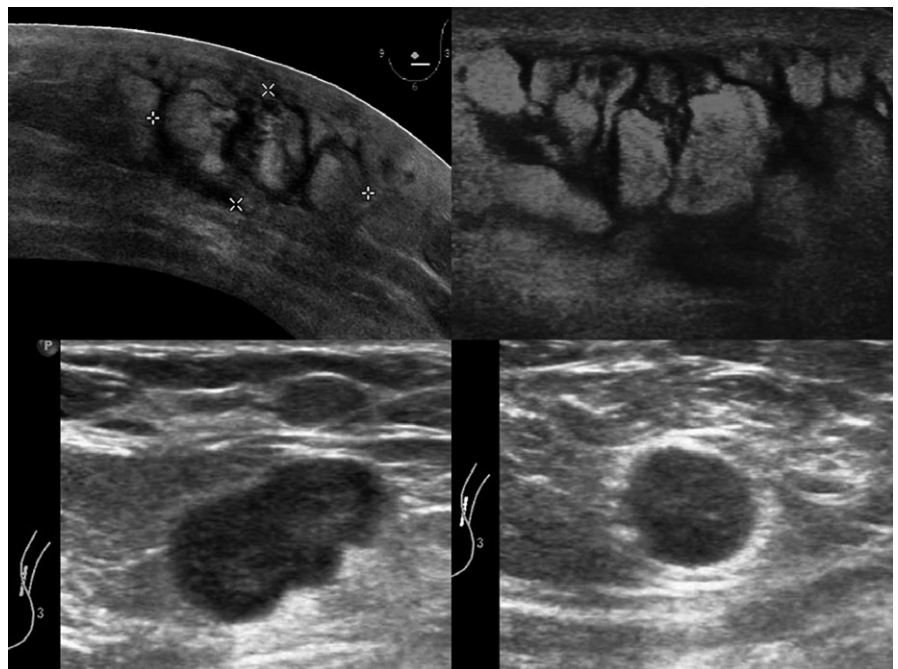
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A 27-year-old Thai farmer presented with a huge painful lump in his left breast for 4 months. Three days prior to admission, he underwent incisional drainage. He was given the diagnosis of breast abscess and treated with oral amoxicillin and clavulanic acid for 10 days. Physical examinations showed an ill-defined rubbery firm lump in his left breast. Ultrasound revealed a subcutaneous lesion with hyperechoic material interspersed with multiple slit-like fluid collections. The lesion size was 5.9×2.7×5.5 cm. Marked surrounding soft tissue swelling and overlying skin thickening were noted (Figure 1, upper images). There were multiple reactive left axillary

lymph nodes with cortical bulging and replaced fatty hilae. They ranged from 0.5 to 1.8 cm (Figure 1, lower images). Core needle biopsy under ultrasound guidance was performed. The histopathology of the left mammary tissue revealed chronic granulomatous inflammation and tissue eosinophilia with broad mycelial filaments disclosed pauci-septation and surrounded by Splendore-Hoeppli reaction (Figure 2). Gomori methenamine silver (GMS) stain showed short, broad fragmented hyphal elements. Tissue polymerase chain reaction (PCR) for 18S ribosomal ribonucleic acid (rRNA) revealed *Basidiobolus meristosporus*. The final pathologic diagnosis was mammary

FIGURE 1 At the most inflamed area at the lower part of left breast, ultrasound reveals a subcutaneous lesion with hyperechoic material interspersed with multiple slit-like fluid collections. The lesion size is 5.9×2.7×5.5 cm. Marked surrounding soft tissue swelling and overlying skin thickening are noted. Two dominant left axillary lymph nodes, level I, show bulging cortex and replaced fatty hilae. A panoramic and targeted ultrasound is shown in the upper images, an axillary view is shown in the lower images



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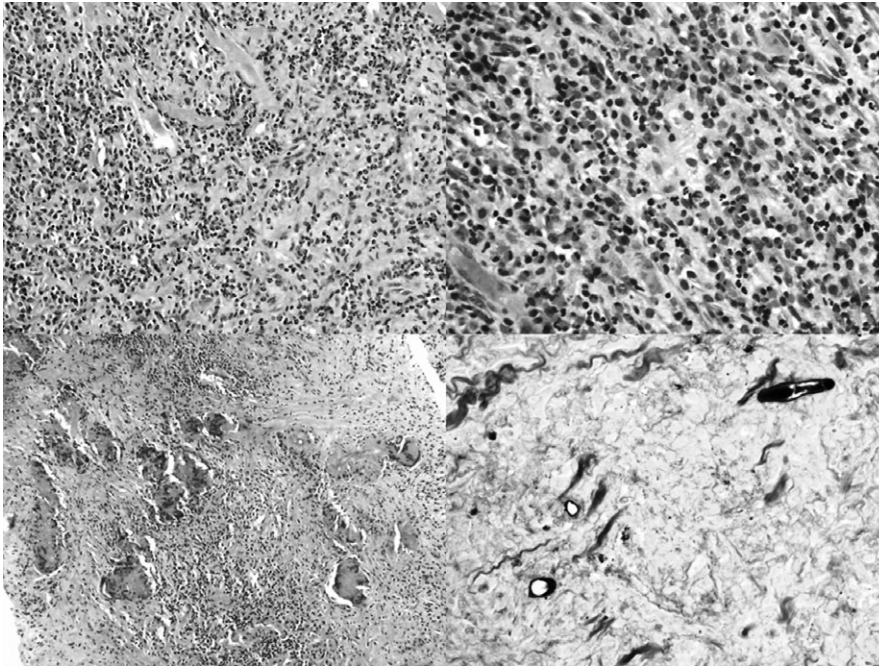


FIGURE 2 Section of the mammary tissue shows granulation tissue with tissue eosinophilia (H&E, $\times 100$, $\times 200$). The granulomatous inflammation with eosinophilic Splendore-Hoeppli materials are noted (H&E, $\times 100$). The GMS staining section shows short, broad fragmented hyphal elements ($\times 400$)

subcutaneous basidiobolomycosis. The medical therapy included itraconazole solution (200 mg twice a day) for 4 weeks. The mass rapidly decreased in size 2 weeks after the treatment.

Subcutaneous basidiobolomycosis is a chronic mycotic infectious disease predominantly reported in the tropical and subtropical regions of the world. The first case report of the human infection caused by *Basidiobolus ranurum* was reported in 1956 by Joe et al. Subcutaneous basidiobolomycosis shows a male predilection of approximately 3:1. The ages of patients range from 6 months to 79 years, predominantly in young adults (80% below the age of 20 years). The occupational history is usually an agricultural lifestyle. The disease is usually seen in immunocompetent individuals. However, there were reports of basidiobolomycosis in human immunodeficiency virus-infected patient and renal transplant patient.

Basidiobolomycosis usually presents with slowly progressive diffuse thickening of subcutaneous tissue and prominent distortion due to formation of chronic granulomatous inflammation involving the subcutaneous tissue. The lesion is occasionally mistaken clinically for malignancy. Eosinophilic granuloma, angiolymphoid hyperplasia with

eosinophilia, and Kimura disease are three major histopathologic differential diagnoses. The characteristic histopathology reveals fungal elements showing short, broad fragments and pauci-septation with right angle irregular nondichotomous branching nonparallel broad ribbon hyphae. The fungal hyphae of *Basidiobolus* spp. are diameter vary from 2 to 6 μm and are ensheathed by granuloma or amorphous intense eosinophilic Splendore-Hoeppli material, which is believed to be an antigen-antibody-mediated immune response. The adjacent stromal connective tissue consists of granulation tissue that is rich in eosinophils. Recent advances in technology including molecular identification by 18S rRNA tissue PCR sequencing might help to identify the fungal species.

The treatment of choice is oral antifungal therapy. The medications for entomophthoromycosis include potassium iodide, trimethoprim/sulfamethoxazole, amphotericin B, terbinafine, ketoconazole, itraconazole, fluconazole, miconazole, voriconazole, and hyperbaric oxygen. Itraconazole solution with a dosage of 200 mg twice a day is recommended for 4 weeks. This is an efficient and easily administered therapeutic regimen. In extreme cases, drug therapy has been combined with surgical intervention.