Necrotizing cutaneous fungal infection of the breast in a patient with breast implants

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Bacterial infections of the breast are common. However, in nonlactating women, breast mycoses are rare. Herein, we describe a rare case of cutaneous fungal infection in a nonlactating woman.

CASE PRESENTATION

A 50-year-old woman with a medical history of hypothyroidism and right breast cancer presented with left breast skin changes. Her breast cancer had been treated with right mastectomy and transverse rectus abdominal myocutaneous flap reconstruction 11 years before presentation; one year before presentation she underwent bilateral breast augmentation and mastopexy. She initially presented to her primary care physician (PCP) with an edematous, erythematous skin lesion of the left breast that was warmer than surrounding skin and a low-grade fever. Her PCP prescribed cephalexin for presumed mastitis. Her symptoms improved; however, two months later, the erythema returned.

She then developed two white areas on the breast skin that reportedly grew in size and eventually overlapped in area (Figure 1). She presented to another hospital with a 2 cm white eschar and surrounding cellulitis. According to the report, there were no signs of fluctuance or abscess formation. Ultrasound revealed minimal fluid beneath the lesion (Figure 2).

She was managed at that facility with excision and biopsy of the involved skin. Pathology demonstrated no malignancy. There was evidence of subepidermal bullous formation with foci of reepithelialization. Periodic Acid-Schiff (PAS) stain was negative for fungal organisms.

Approximately 8 h after surgery, she developed another necrotic lesion; her surgeon immediately opened the lateral aspect of the wound for concern of purulence; however, there was none. The wound was left open without packing. She was prescribed vancomycin while in hospital, and she completed an oral course of levofloxacin as an outpatient. While on antibiotics, the infection progressed and she developed a new similar skin lesion. She was then referred to the authors' tertiary care facility for further work-up.

Key Words: Bipolaris; Breast; Cutaneous; Disseminated; Immunocompetent; Necrotizing

Bipolaris species are dematiaceous fungi; cutaneous or disseminated infections with Bipolaris species have rarely been reported in immunocompetent patients. Unresectable or recurrent lesions should be treated with antifungals. Cutaneous necrotizing breast fungal infections should be considered in patients who do not respond to antibacterial therapy.

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Examination of the breast showed a healed scar at the excision site with a 4 cm area of skin breakdown and underlying granulation tissue extending laterally from the scar and involving the areola. Slightly inferior to this was a 4 cm full-thickness eschar. Lateral to the eschar was an area of white ischemic skin consistent with nonviable skin. There were no other areas of evident ischemia. Initial management consisted of seven days of observation, after which it became evident that the ischemic area was a full-thickness eschar, and all nonviable areas were debrided (Figure 3). Postoperatively, the wound was managed with wet-to-dry dressings.

On pathological review of the debrided material, Gomori methenamine silver stain and PAS stain confirmed the presence of branching septate fungal hyphae invading the epidermis, dermis and subcutaneous adipose tissue, associated with extensive tissue necrosis (Figure 4). A Gram stain revealed clumps of Gram-positive cocci, present only on the surface of the ulcer. The final diagnosis was invasive filamentous fungal infection with cutaneous necrosis and ulceration. Fungal and bacterial cultures grew 2+ Bipolaris species and 4+ Staphylococcus epidermidis, respectively.

The patient was treated with voriconazole 200 mg twice per day for one month for the fungal infection, and there was no intervention for the Staphylococcus because it was judged to be a colonizing bacterium only.

At one week of follow-up, she had excellent granulation tissue at the wound base with a small amount of fibrinous exudate. She later reported resolution of the lesion and healing of her wound.

At five months following completion of antifungal therapy, the patient reported a new lesion on her back similar to those she had on her breast. Her PCP again referred her to the authors’ centre. The lesion was a 2.5 cm full-thickness eschar with appearance similar to her breast lesions (Figure 5A). It was presumed she had disseminated fungal infection from her breast. Her back was further debrided and samples were sent for culture and pathology. Pathology demonstrated focal sparse filamentous structures suspicious but not definitive for fungi similar to those previously found in her breast on Gomori methenamine silver stain. The structures were also focally highlighted by the PAS stain. Because she had presumed disseminated disease, she was further evaluated with chest x-ray to assess for pulmonary manifestations and echocardiogram to rule out endocarditis, both of which were negative. A new lesion developed directly adjacent to the debrided portion on her back two days later. She was started on voriconazole empirically before results of pathology and microbiology returning. Final fungal cultures yielded no growth. She was seen one week after initiation of voriconazole, and progression of the process at the new site had halted, showing only partial thickness involvement, not requiring further debridement (Figure 5B).

Figure 3) Preoperative cutaneous eschar

Figure 4) A Branching septate fungal hyphae invading the epidermis and dermis. B Branching septate fungal hyphae with vesicular swellings in the subcutaneous adipose tissue. (Gomori methenamine silver stain; original magnification ×600)

Figure 5) A Preoperative back eschar. B. Partial thickness involvement of eschar postoperatively
Necrotizing cutaneous breast fungal infection

DISCUSSION
This patient had an unusual presentation of fungal infection. There have been few reports of fungal infections in nonlactating women in the breast literature. While there have been previous reports of necrotic breast masses, to our knowledge, the present report is the first to describe a necrotizing cutaneous breast fungal infection with Bipolaris species.

Bipolaris species are dematiaceous fungi that produce phaeohyphomycosis. Dematiaceous fungi have dark-coloured walls secondary to melanin (2,3). More than 100 fungal species cause phaeohyphomycosis. The two most common species are Bipolaris spicifera and Exophiala jeaneselmei. Phaeohyphomycoses can present as superficial cutaneous or subcutaneous infections, as paranasal sinustis or as systemic infection. Typically, cutaneous infections begin as a small papule caused by unrecognized trauma and evolve into a suppurative subcutaneous cyst or nodule (2). The exact incubation time is unknown (3). It is most commonly observed on the extremities (2,3). Biopsy typically shows suppurative subcutaneous inflammatory infiltrate and possibly a granulomatous reaction. The organisms are found within the cyst (2).

Bipolaris spicifera has previously been reported as the cause of cutaneous infection of the ankle in an immunocompromised patient following a minor trauma to the region (4). Straka et al (4) also describe two other reports of cutaneous infections with B spicifera in immunocompromised patients from the 1970s. Interestingly, our patient was immunocompetent; her cancer had occurred >10 years previously, and she had no signs or symptoms of recurrence or of other immunocompromised states. Infections with Bipolaris species in immunocompetent patients have rarely been reported (5,6).

Often surgical excision is sufficient for treatment, and antifungals are not necessary. If the tissue is not entirely removed, infection can recur. Unresectable lesions can be treated with intravenous amphotericin and oral ketoconazole. Brandt and Warnock (3) also report itraconazole and voriconazole to be effective against many dematiaceous molds in vitro.

Truppman et al (7) reported a case series of fungi-associated breast infections after 700 augmentation mammoplasties over a nine-year period. The majority of these were subpectoral saline implants that contained fungal growth within the implant capsule and the prosthesis itself without cutaneous involvement at time of exchange. Only two of the patients’ cultures showed fungal growth. All four patients in this series had unilateral infection, and the authors could not determine a source of the infections. They speculated it may have been commensal rather than pathogenic, because all the implants were exchanged for mechanical reasons and the fungal-containing fluid was incidental.

Our patient did not have any known trauma to the affected region of the breast. This does not exclude the possibility of a minor trauma to the integrity of the epidermis allowing for entry of the fungal organisms. Fortunately, her infection did not compromise the integrity of her implant. The development months later of a new lesion caused us to presume she developed disseminated infection, making our case unique. We postulate that the fungus was travelling through her local breast lymphatics and disseminated hematogenously to a distant location, in the present case, her back. Disseminated cutaneous sporotrichosis, caused by the fungus Sporothrix schenckii, has shown similar behaviour in an immunocompetent individual (8).

CONCLUSION
Necrotizing cutaneous fungal infection of the breast is rare. It should be in the differential diagnosis of necrotizing infections that do not respond to antibacterial therapy. Debridement with fungal cultures followed by treatment with appropriate antifungal therapy was effective for this patient and is the recommended treatment option for such patients.

ACKNOWLEDGEMENTS: The authors acknowledge Cory Poetzl for her assistance with photography and photograph formatting.

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