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Invasive cutaneous mucormycosis in an extremely preterm infant

David Brooks[a,b,*], Shahab Abdessalam[a,b], H. Dele Davies[a,b], Aileen M. Aldrich[a,b], Jiri Bedrnicek[a,b], Nathan Gollehon[a,b]

[a] Children's Hospital and Medical Center, 8200 Dodge St, Omaha, NE, 68114, USA
[b] University of Nebraska Medical Center, South 42nd St & Emile St, Omaha, NE, 68198, USA

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ABSTRACT

An extremely preterm neonate developed a progressively worsening right axillary ulcerative lesion at the previous site of a temperature probe. Despite treatment with broad-spectrum antibiotic and antifungal therapy, the lesion worsened and required extensive surgical debridement. Histologic examination of the debrided tissue demonstrated the presence of invasive mucormycosis. With prompt surgical intervention and proper antifungal therapy, the patient survived. Here we present a unique case of invasive cutaneous mucormycosis in a preterm neonate, along with a review of the current literature on this rare infection.

1. Introduction

Mucormycosis is a broad term describing infections caused by fungi of the Zygomycota phylum [1]. While infections with these fungi are more common in adults and older children, it can affect neonates and is associated with a high mortality rate in this population [2]. Preterm neonates are at increased risk for invasive infection due to immaturity of both the innate and adaptive immune systems [2]. When infection does occur, it is typically treated with antimicrobial therapy with surgical intervention reserved for those refractory to medical management [3]. For control of soft tissue infections, the involvement of the surgeon ranges from aspiration of fluid collections to debridement of necrotic tissue.

2. Case presentation

A newborn female was born at 25 weeks' gestation via emergent cesarean delivery secondary to placental abruption with a birthweight of 806 g. The infant received antifungal prophylaxis with fluconazole from birth until her umbilical catheters were removed on day of life (DOL) 7. On DOL 3, bruising was observed on the right axilla where a temperature probe had previously been placed. On DOL 6, the lesion appeared increasingly discolored with new central necrosis. The infant was evaluated for sepsis and started empirically on vancomycin and tobramycin. A surface skin culture and punch biopsy of the lesion were obtained. The skin culture demonstrated *Enterococcus faecalis*, coagulase-negative *Staphylococcus* species, and Diphtheroids. A total of 7 days of vancomycin therapy was administered. Histologic examination of tissue from the punch biopsy revealed a presumptive diagnosis of Aspergillosis. Amphotericin B was initiated on DOL 7 and voriconazole was added on DOL 8. As the infection worsened, micafungin was added to the regimen on DOL 12 and amphotericin was discontinued on DOL 13.

The infection failed to show improvement, so on DOL 13 the patient was transferred to a surgical neonatal intensive care unit (NICU) for evaluation. Upon arrival, the infant was examined with extensive soft tissue loss noted over the lateral right chest, with exposed ribs and no discernible nipple-areolar complex (Figs. 1–3). Given these findings, the infant was immediately taken to the OR for extensive debridement of the infected skin, soft tissue, portions of the latissimus dorsi, pectoralis, and serratus anterior muscles. Portions of the right 6th and 7th ribs were removed as they were brittle and congruent with the surrounding infection (Fig. 4). The infant tolerated the procedure well and the chest wall remained open and covered with wet dressings postoperatively.

On DOL 15, pathology reported that the debrided tissue was, in fact, showing Zygomycete species, not Aspergillus as originally suspected (Fig. 5). Amphotericin B was restarted, voriconazole was discontinued, and micafungin was continued for its synergistic effect. The patient returned to the operating room on DOL 16 for repeat muscle biopsies, removal of an exposed portion of the 5th rib, and SurgiMend® mesh closure of the chest wall with interrupted 4-0 polydioxanone sutures (Figs. 6 and 7). Given the patient's small size and the potential for negative pressure on the lung, vacuum-assisted closure (VAC) was not used. The patient initially received wet-to-dry dressings with saline

* Corresponding author. University of Nebraska Medical Center, Department of Pediatrics UT 5146, South 42nd St & Emile St, Omaha, NE, 68198, USA.
E-mail addresses: david.brooks@unmc.edu (D. Brooks), sabdessalam@childrensomaha.org (S. Abdessalam), dele.davies@unmc.edu (H.D. Davies), aileen.aldrich@unmc.edu (A.M. Aldrich), jbedrnicek@childrensomaha.org (J. Bedrnicek), ngollehon@childrensomaha.org (N. Gollehon).

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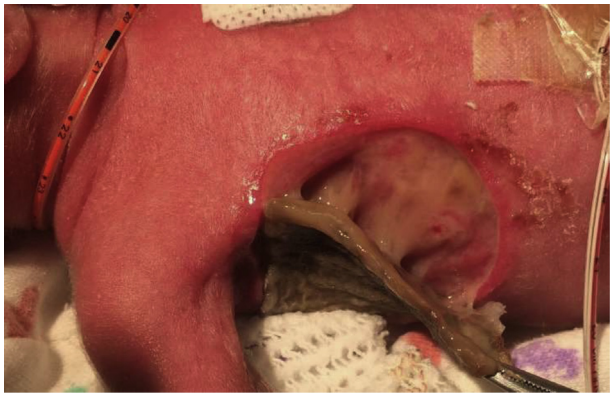

Fig. 1. Initial examination with elevation of dressing to demonstrate the purulence of the chest wall.

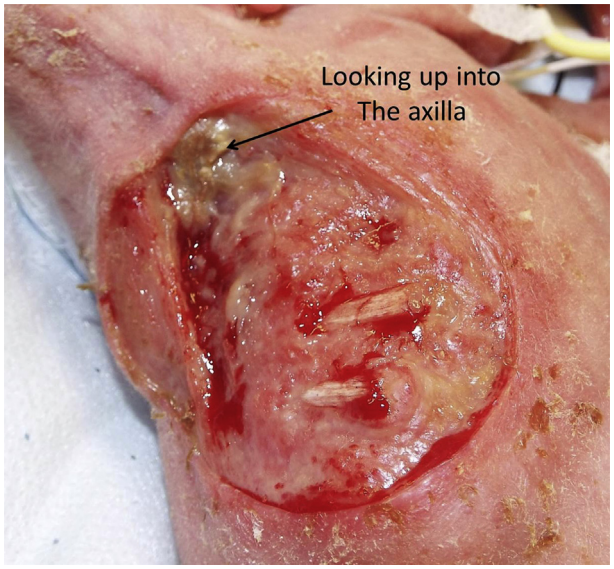

Fig. 2. Initial examination in the operating room. View is from the anteriolateral right chest.

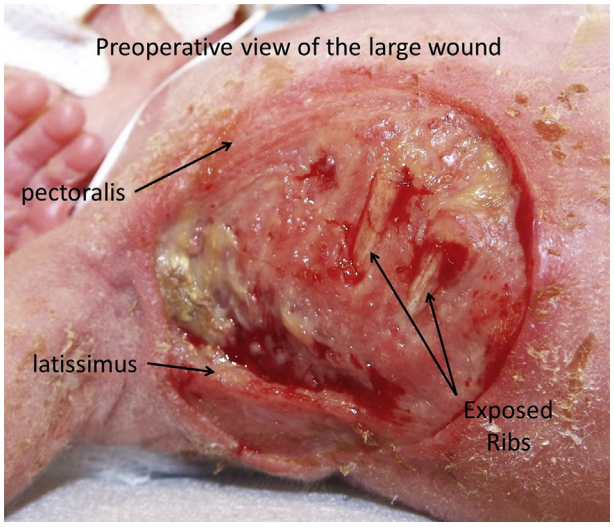

Fig. 3. Initial examination in the operating room. View is from the lateral right chest.

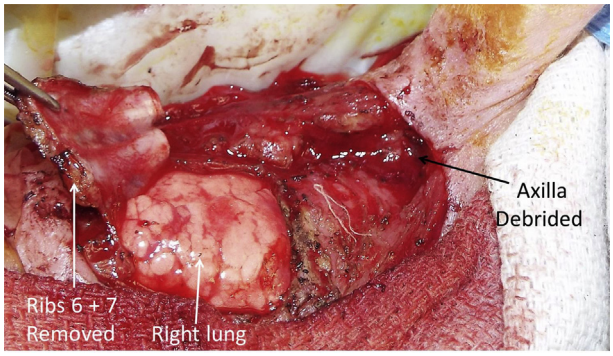

Fig. 4. Intraoperative debridement of muscle, axilla, and ribs of right chest.

over the mesh but developed a "slime" on the wound so was changed to betadine (wet-to-dry) for better destruction of fungal elements. After 5 days of betadine dressings, she was changed back to wet-to-damp dressing changes with saline to prevent desiccation of the SurgiMend® mesh. After 6 weeks, Dakins solution was added, due to concern for the presence of pseudomonas (green appearance around the wound), and was continued until complete epithelization of the wound at week 10.

Pathologic examination of all tissue biopsied at the second debridement showed no further Zygomycete, except for the 5th rib. The patient was treated with micafungin and amphotericin B therapy for 126 and 163 days respectively and experienced no major medication-related side effects. Complete epithelialization with residual cicatricial scarring of the wound occurred over the next 3 months (Fig. 8).

The patient required tracheostomy for long-term, home mechanical ventilation, and placement of a gastrostomy tube for nutrition and medication administration (Fig. 9). The patient was discharged on DOL 180 at a corrected gestational age 50 weeks and 6 days. A chest x-ray obtained 11 months postoperative shows good chest wall growth with some minor volume loss of the right chest (Fig. 10).

3. Discussion

Mucormycosis is a term that refers to infections caused by fungi of the Zygomycota phylum. The most common genera within this phylum that cause infection in humans are *Rhizopus, Mucor, Absidia,* and *Cunninghamella,* with *Rhizopus* being the genus of most clinical importance in humans [1]. These fungi are found worldwide and omnipresent in the environment. Despite its reputation for only affecting immunocompromised or severely ill patients, most documented infections caused by mucormycosis occur in otherwise healthy individuals [4]. The body's main defense against fungi are phagocytic cells which, in the above conditions, are often less functional than normal or decreased in number [2].

Mortality from mucormycosis infection depends on the level of host immunosuppression, time to diagnosis, and location of infection. Cutaneous involvement has the most favorable prognosis with a mortality rate of 25% (based on one case series), and gastrointestinal involvement has the least favorable prognosis with a mortality rate up to 85% [1]. Clinically, the most common locations of mucormycosis infection are: rhino-orbital-cerebral, gastrointestinal, cutaneous, pulmonary, and disseminated infection [1]. In neonates, the most common locations are gastrointestinal and cutaneous [5–7]. Cutaneous infection is often the result of direct inoculation of fungal spores into the skin and typically occurs in areas of trauma or burns in susceptible hosts. There are two clinical appearances of cutaneous mucormycosis: superficial and gangrenous. Superficial infection presents as asymptomatic vesicles or pustules that eventually ulcerate whereas gangrenous infection presents with painful papules that rapidly progress to necrotic plaques [8]. The hallmark finding of cutaneous infection, a black, necrotic eschar at the site of injury, may or may not be present in superficial