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Auricular Mucormycosis Following Burn Injury: A Case Report with Literature Review

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Case Report
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Abstract

Objective: Mucormycosis is a dreaded clinical entity caused by filamentous fungi of the order Mucorales. Traumatic mucormycosis mainly occurs in civilian trauma, burns and vehicle accidents. Herein, we present a rare case of burn-related mucormycosis in a 29-year-old healthy male patient.

Methods: The patient was referred to our hospital after being involved in a car accident with vehicle explosion and burn injury. 20 days after admission to our hospital, a new discharge from the left ear was noticed. Microbiological results revealed high suspicion of fungal infection and pathologic examination of necrotic cartilage revealed aseptate hyphae of mucormycosis

Results: A prompt parenteral liposomal Amphotericin B was initiated, local control of the disease by extensive surgical debridement was performed, including total excision of both auricles and cartilaginous parts of the external auditory canals. Excisions were done until a healthy, bleeding tissue was obtained. The wounds healed completely without any recurrence

Conclusions: Mucormycotic infections are uncommon in patients with burns; however, when they are present, they usually occur as a cutaneous burn wound infection and remain difficult to treat and is often lethal. This necessitates prompt and early diagnosis, control of risk factors, surgical debridement of necrosed tissue and antifungal drugs. Amphotericin B is the antifungal of choice for the treatment of mucormycosis. Since inoculation of soil-dwelling moulds into wounds can occur at the time of injury, the context in which burns occur needs to be considered to evaluate the risk of mucormycosis, especially in auricles which are likely at increased risk, probably because they protrude from the head and come more in contact with the ground.

Keywords: Fungal Infections, Auricular Trauma, Burn Injury, Mucormycosis, Head & Neck surgery

Abbreviations: CT: Computed Tomography; ICU: Intensive Care Unit; GMS: Gomori Methamine Silver; TBSA: Total Body Surface Area.

Introduction

Mucormycosis is a rare, highly aggressive, rapidly invasive fungal infection, with a high mortality rate. The pathogen is a filamentous fungus of the zygomycete class of the Mucorales order [1]. This life-threatening infection has a tendency for invading into vessels and lymphatics causing formation of Mucor thrombi leading to ischemia and infarction of the affected tissue [2]. Histologically, mucormycosis is characterized by fungi with thick-walled, non-septated hyphae, with branching at right angles.

Mucormycosis usually affects patients with alteration of their immunological system [3]. Recently fungi of the order Mucorales are increasingly recognized as important causes of necrotizing wound infections in the setting of military injuries, burns, natural disaster-related, and other civilian trauma [4,5].

The disease may be manifest in different anatomic locations, such as in paranasal sinuses, rhinoorbital and rhinoorbitocerebral regions, pulmonary system, gastrointestinal tract, cutaneous sites, and rarely as a disseminated disease [6]. Mucormycosis involving the ear is a rare occurrence.

Imaging techniques are not usually diagnostic, and cultures are not completely reliable. Definitive diagnosis is exclusively obtained by means of histological examination [3]. The ideal treatment includes the correction of underlying risk factors, antifungal treatment with amphotericin B, and aggressive surgery [2].

Even with optimal treatment, the disease has a high mortality rate. Better prognosis can be achieved with early diagnosis and multidisciplinary treatment. Here, we report a case of auricular and cutaneous mucormycosis following burn injury, that was treated successfully with combination of aggressive surgical excision and antifungal therapy.

Clinical Presentation

A 29-year-old male patient was admitted to a secondary referral hospital after being involved in a car accident with vehicle explosion. At the hospital there was a respiratory deterioration, for which he underwent intubation. Subsequently after stabilization, a computed tomography (CT) scan was performed which did not show any internal injuries. On the same day, he was transferred to our institution for management in the burn intensive care unit (ICU). On admission the patient was diagnosed with burns involving 55% of the body surface (mostly in the limbs). During the next few weeks, the patient underwent multiple skin grafts surgeries by the plastics department team.

On day 20 after admission to our hospital, the treating physicians noticed a new discharge from the left ear. Ear swabs cultures were done, and several days later the microbiological results revealed high suspicion of fungal infection by mucormycosis. The otolaryngology head and neck team were consulted. Personal past medical history revealed no background medical conditions and no known immunosuppressive state.

On clinical examination, the patient was vitally stable and afebrile. A wet gangrenous black-to-brownish discoloration of the both auricles was noted (Figure 1). In addition, otoscopic examination showed an edematous external auditory canal with narrowing of the meatus. In addition, two necrotic lesions in the forehead, about 2 cm in diameter each, were noted. Examination of the nasal and oral mucosa was normal. The patient underwent brain CT scan which revealed opacity in left middle ear cavity, mastoid and canal, without any bone destruction (Figure 2).

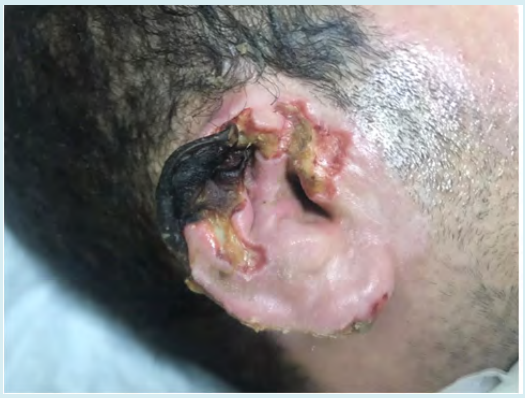

Figure 1: Patient's right ear upon presentation.

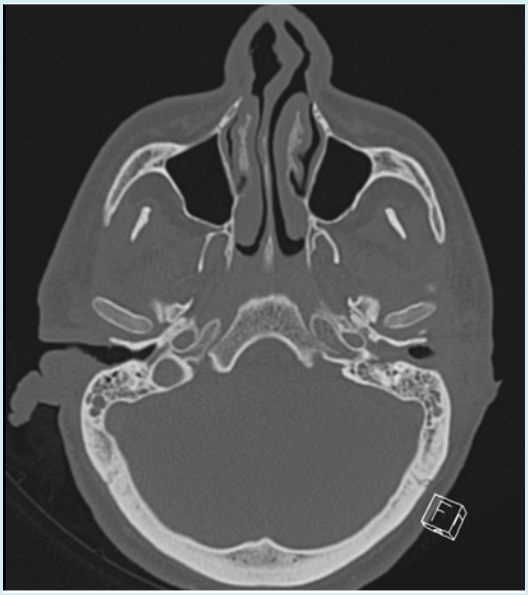

Figure 2: Preoperative CT scan with contrast showing opacity in left canal, middle ear cavity and mastoid cavity without any bone destruction.

Excisional biopsy specimens from the superior-posterior part of left auricular cartilage were sent for pathologic examination which showed clear evidence of skin and cartilage with massive necrosis and infiltration by non-septated fungal hyphae (Figure 3).

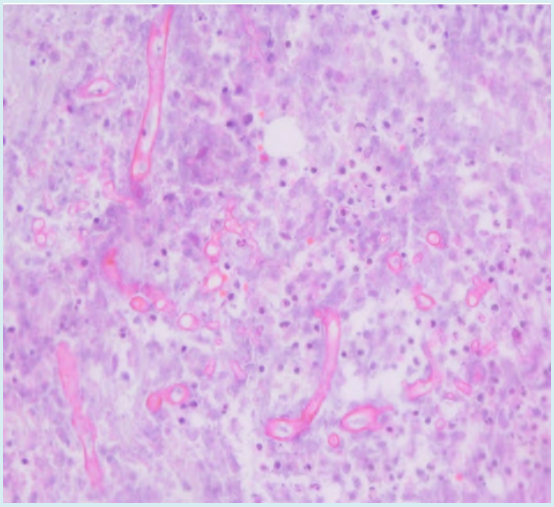

Figure 3: Broad aseptate hyphae, typical for those species belonging to the Mucorales, Hematoxylin and Eosin (H&E) Stain, original magnification x 40.

Immediately after receiving the histopathological results, parenteral liposomal amphotericin B was initiated, and the patient was transferred to the operating room. Extensive surgical debridement was performed, including total excision of both auricles and cartilaginous parts of the external auditory canals. The forehead lesions were also excised. Excisions were done until a healthy, bleeding tissue was obtained. The wounds were left open, and surgery sites were dressed with sterile gauze. No middle ear surgery was performed as there were no clinical signs of involvement during the operation. Tissue taken for histopathological examination where Gomori Methamine Silver (GMS) staining revealed invasive fungal hyphae consistent with Mucormycosis in both auricles and the forehead lesions. The surgical wound underwent daily washing with Hydrogen Peroxide and supplementation of local amphotericin was used while doing daily dressing. Suspicious lesions were debrided and sent for pathological examination, with no evidence of recurrent infection.

At 60 days after the operation the patient was clinically stable with no evidence of invasive fungal infection. The resected areas were healing by secondary intention (Figure 4) with narrowing of the external auditory meatuses. Stenting of the external canal with tampons sponge (Ivalon®, Medsorb Dominican) was performed.s

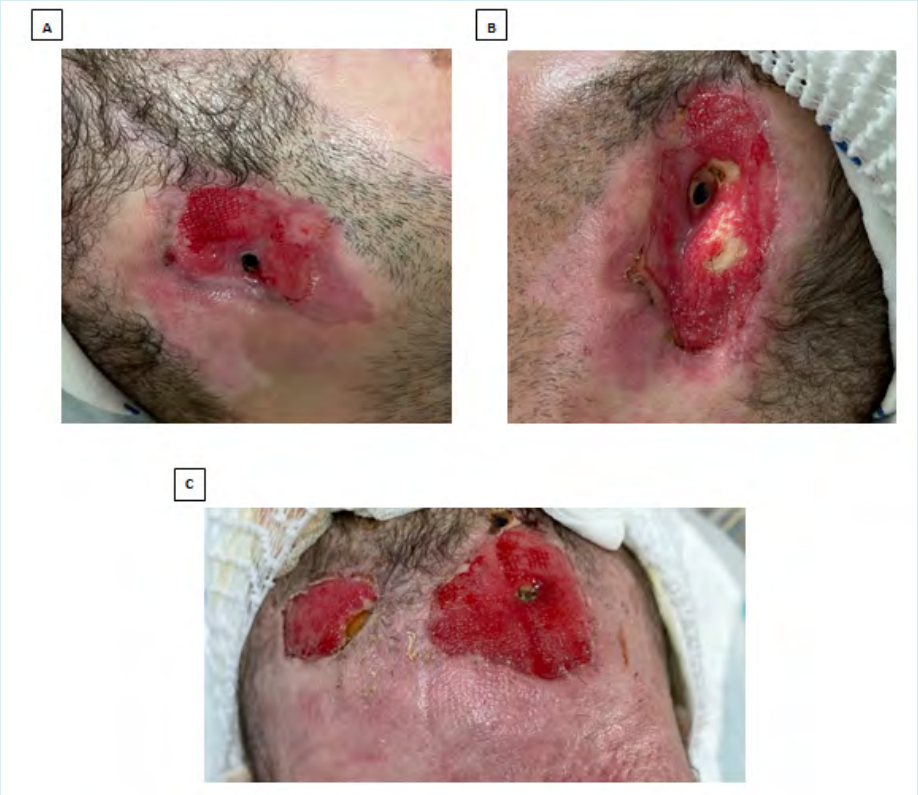

Figure 4: Day 60 postoperatively, showing complete healing of the wounds with no discharge or inflammatory signs. A, right ear. B, left ear. C, forehead.

Discussion

Mucormycosis is a devastating invasive fungal infection with estimated mortality rate ranging from 23%-100% [7]. It is the third most common invasive fungal infection, following aspergillosis and candidiasis [6,8,9]. This fungus exist widely in nature and their spores may be found in soil, air, spoilt food, and other decaying organic material. Healthy people are rarely affected, while immuncompromised patients are at an elevated risk for this infection.

Patients with extensive burns are an important group at risk for cutaneous mucormycosis. The first two cases of mucormycosis in burn patients were reported in 1961 by Rabin, et al. [10] Several reviews on cutaneous mucormycosis have been published, showing mostly localized infection or accompanied with deep extension, but few have shown dissemination [4]. Clinical presentation is characterized by rapid progression to necrosis and a better prognosis than other clinical forms of mucormycosis (31% mortality vs. 50-80%) [11]. Incidence of auricular mucormycosis is very rare. This is the first case reported in English literature on invasive mucormycosis involving auricular cartilage following burn injury, to the best of our knowledge.

Why and how this fungus has entered the auricular cartilage is debatable. Our assumption as we analyzed the circumstances of this burn injury is that the patient rolled with soil to extinguish the flames, and was consistent with an inoculation with the fungi at the time of the burn injury, as was described in previous reports of mucormycosis following burn injury [12]

The diagnosis is based on the combination of clinical examinations and mycological, histopathologic and radiological investigations [10]. Definitive diagnosis generally requires the demonstration of the organism in infected tissue or biopsy specimen [9]. Treatment is 3-fold: Aggressive surgical debridement to reduce fungal load and facilitate drug access, prescription of antifungals, and correction of other risk factors in immunodeficient patients [13]. Amphotericin B, preferably in liposomal form (AmBisome®, Gilead Science), is the preferred antifungal in mucormycosis. The liposomal form shows less nephrotoxicity, allowing higher doses, for a longer period of time.

Chamilos, et al. [14] quantified the benefit of early initiation of Amphotericin B antifungal therapy. They reported that if treatment was initiated within 5 days of diagnosis of mucormycosis, survival was markedly improved compared to initiation of polyene therapy at ≥6 days after diagnosis (83% vs. 49% survival). Therefore, urgent fungal and surgical therapies are the main methods for increasing survival.

In a recent review study by Devauchelle, et al. [5] the epidemiology of mucormycosis in burn patients was investigated. The median percentage of the total body surface area (TBSA) affected by a burn was 42.5% to 65%. Mortality in this review ranged from 29%-100%. Kyriopoulos and colleagues reported 6 cases of trauma-associated mucormycosis with review of literature. TBSA of burns ranged from 45-71%. Their treatment protocol for suspected mold infections of burn wounds stipulates rapid diagnosis and extensive surgical debridement accompanied by amphotericin B in treatment of cases of mucormycosis [12].

In our case, intravenous liposomal amphotericin B was started just after appearance of suspicious clinical presentation and cultural findings supporting mucormycosis. Then, the patient underwent surgical debridement on the second day of antifungal treatment. On reviewing the literature, the use of hydrogen peroxide in open wounds with mucor was documented [15]. Topical application of amphotericin along with hydrogen peroxide was performed. The dressings were changed twice every day till healthy granulation tissue was seen all around.

While rhinocerebral mucormycosis in the immunocompromised patient is a well described entity in the otolaryngology world, invasive fungal infection in a burn victim in not as frequently seen. The aim of this study was to describe this entity because survival is directly correlated with rapid diagnosis, which in turn depends on a high clinical suspicion.

Conclusion

In conclusion, mucormycotic infections are uncommon in patients with burns; however, when they are present, they usually occur as a cutaneous burn wound infection and remain difficult to treat and is often lethal. This necessitates prompt and early diagnosis, control of risk factors, surgical debridement of necrosed tissue and antifungal drugs. Amphotericin B is the antifungal of choice for the treatment of mucormycosis. Since inoculation of soil-dwelling moulds into wounds can occur at the time of injury, the context in which burns occur needs to be considered to evaluate the risk of mucormycosis, especially in auricles which are likely at increased risk, probably because they protrude from the head and come more in contact with the ground.

Conflicts of Interests

Authors declared no potential conflicts of interests with respect to the research, authorship, and/or publication of this article.

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