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## A rare case of invasive zygomycosis in an immunocompetent trauma patient

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## ABSTRACT

Zygomycosis is an aggressive fungal infection occurred in immunocompromised patients commonly involving sinuses and lungs. zygomycosis has been reported in road traffic accidents and in farmland injuries. The mortality rate of zygomycosis is 40% to 80% due to angioinvasion. We report a case of immunocompetent young girl developing zygomycosis after road traffic accident in a urban area. The extensive infection patient survived after above knee amputation and antifungal therapy.

## 1. Introduction

Zygomycosis is an aggressive fungal infection found in farmland injuries and in immunocompromised patients[1,2]. Mortality rates for zygomycosis is about 40% to 80% of reported cases[1,2]. Risk factors include hematological malignancies, prolonged neutropenia, uncontrolled diabetes mellitus and systemic immunosuppressive therapy[1]. Early diagnosis, prompt antifungal therapy and radical surgical debridement, is needed to prevent mortality. Here, we report an immunocompetent 14-year-old girl who had road traffic accident and had a Gustilo Grade IIIA femoral fracture. She developed angioinvasive zygomycosis of right thigh. Patient survived after high above knee amputation. This case has been reported to have a high index of suspicion of fungal infection in compound fractures even without predisposing factors.

## 2. Case report

A 14-year-old girl sustained Grade IIIA fracture of right femur

after road traffic accident in urban area. She was referred to our hospital after wound debridement and external fixation. There were no associated injuries. On examination, the wound was about 10 × 15 cm over the lateral aspect of the thigh and 7 × 10 cm wound over anterior aspect (Figure 1). Repeat wound debridement was done and culture revealed pseudomonas infection for which patient was started with meropenem (Figure 2). Wound was healthy for 2 days after which patient developed progressive muscle necrosis (Figure 3). On repeat debridement, histopathology revealed extensive muscle necrosis with angioinvasive fungi consistent with zygomycosis (Figures 4, 5 and 6). Patient was started with amphotericin B injection. In spite of antifungal therapy, there was progressive muscle necrosis. On surgical exploration, there was extensive muscle necrosis involving hamstring and quadriceps muscle and there was no involvement of major vessels. However, high above knee guillotine amputation was done to save the life of the patient. Vacuum assisted closure dressing was applied to the stump following amputation. Wound size gradually reduced and secondary closure was done. Amphotericin B was continued for 3 weeks followed by oral posaconazole for 3 months duration. Wound healed completely after 3 months (Figures 7 and 8). Chest X-rays were taken at 6 months and 1 year interval normally. There was no recurrence of infection at 3 years follow up.

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**Figure 1.** Showing raw area of size of 10 cm × 15 cm and 7 cm × 10 cm over the anterior and lateral aspect of right thigh and knee exposing the deep fascia and quadriceps muscle.



**Figure 2.** Showing raw area of size of 20 cm × 18 cm over the lateral aspect of the thigh and 10 cm × 17 cm raw area of the anterior aspect of leg following wound debridement.



**Figure 3.** Showing progressive muscle necrosis after 3 days following wound debridement.



**Figure 4.** About 10% KOH of the tissue showing broad aseptate hyphae.



**Figure 5.** Lactophenol cotton blue mount of the culture (magnification 400×). Broad aseptate hyphae showed champagne glass appearance of the sporangial sac.



**Figure 6.** Culture grown on Sabouraud's agar showing a white cottony growth almost filling the tube.



**Figure 7.** Healed scar following right above knee guillotine amputation.



**Figure 8.** Six months post-op right above knee guillotine amputation.

### 3. Discussion

Necrotizing fasciitis following traumatic injuries is caused by bacterial infections. Fungi of the class Zygomycetes can cause necrotizing fasciitis in immunocompromised patients and trauma in farmland injuries. The decaying vegetable matter and moisture in farmland injuries predisposes severe traumatic injury to aggressive fungal infections. Angioinvasive fungal infections commonly affects rhinocerebral, orbital, pulmonary and gastrointestinal areas in immunocompromised patients[1,2]. Zygomycosis has been reported to occur after natural disaster like tsunami and earthquake victims[3]. This has been reported in polytrauma patients with zygomycosis Pulmonary zygomycosis is very rare in the absence of an underlying illness[4-6]. The cause of death in zygomycosis is due to pulmonary involvement by angioinvasion[7].

Our patient is a 14-year-old healthy girl sustained road traffic accident. The aggressive nature of these fungal infections is attributed to their angioinvasive property which causes local tissue necrosis and infarction[8-10]. She had no signs of pulmonary involvement in the chest X-ray and CT scan on admission. Diagnosis of zygomycosis is by tissue culture and microscopic examination. The 10% KOH of the sample showed broad aseptate hyphae (Figure 4). Culture grown on Sabouraud's agar showed a white cottony growth almost filling the tube (Figure 6). Lactophenol cotton blue mount of the culture (magnification 400×) indicated that broad aseptate hyphae showed champagne glass appearance of the sporangial sac (Figure 5)[11,12].

Zygomycosis presents clinically as superficial infection or deep infection. Superficial infection is rare. Deep infection manifests as gangrene. In our patients, there was a deep infection with large zones of blackened and necrotic foul smelling tissue when she presented to our hospital. Surgical debridement remains the mainstay of treatment. Repeated removal of necrotic tissue may be required to prevent dissemination. Current literature recommends radical debridement of the lesion till bleeding tissue is encountered and the margins are free of fungal growth. In our patients, even after radical debridement, the margins had positive growth after 2 days. This was due to local angioinvasion. The success of debridement depends upon the extent of angioinvasion. Although in our patient, we did not encounter major bleeding due to involvement of major vessels, there was a extensive muscle involvement and preventing thromboembolism on high above knee amputation was considered for this patient.

Amphotericin B and posaconazole are the most active agents against zygomycetes, though their activity varies among the different Mucorales families. In our patients, amphotericin B is administered for a period of 3 weeks. Although liposomal amphotericin B has fewer adverse effects, amphotericin B deoxycholate was administered because of less cost. Patients did not develop cardiac toxicity, however they developed twice hypokalemia and creatinine increased to 1.9 which returned to normal after stopping amphotericin B.

In all mentioned studies of either cutaneous or pulmonary zygomycosis, the best outcome was obtained when the diagnosis was made early, aggressive surgical debridement was used and high doses of amphotericin B were given.

Oral posaconazole therapy was started after 3 weeks of oral use of amphotericin B. There is no clear consensus on duration of posaconazole therapy. We gave posaconazole for a period of 3 months after discontinuing amphotericin B. We did not consider iron chelation therapy as patients had elevated creatinine levels

Hyperbaric oxygen therapy was not available to us and therefore could not be used. There was the infection of the wound with pseudomonas which settled with injection meropenam and vacuum suction dressing.

Angioinvasive fungal infection can rarely occur in immunocompetent patients without farmland injury. We believe that our patient can survive because of amputation combined with antifungal therapy.

### Conflict of interest statement

We declare that we have no conflict of interest.

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