

Mucormycotic osteomyelitis of maxilla following maxillofacial trauma: The disease of the diseased

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ABSTRACT

Mucormycosis is a rare opportunistic fungal infection with acute, aggressive, and invasive nature, seen in immunocompromised/debilitated patients, especially with diabetes mellitus. The key to successful therapy is the early diagnosis of signs and symptoms of the disease, correction of the underlying medical disorder(s), and aggressive medical and surgical intervention. The case presented here is mucormycotic osteomyelitis of the nasomaxillary-zygomatic complex following trauma in a middle-aged man. Preoperative amphotericin B therapy along with surgical resection of necrotic tissue was done followed by prosthetic rehabilitation with an obturator. There was no sign of recurrence of the diseases for the follow-up period of 1 year.

Keywords: Amphotericin B, cavernous sinusthrombosis, hypoxia, mucormycosis, osteomyelitis

Introduction

Osteomyelitis of maxilla is an uncommon process due to its rich vascular supply, although it can occur due to bacterial, viral, and fungal infections, especially in immunocompromised patients. Mucormycosis is caused by saprophytic and aerobic fungi *Rhizopus*, *Rhizomucor*, and *Cunninghamella* genera of the family Mucoraceae which frequently colonize in the oral/nasal mucosa and paranasal sinuses. Mucormycosis was first reported as a human disease by Paultauf in 1885. Mucormycosis generally affects a wide age range with no definite sex or race predilection and having mortality rate of 50–85%.^[1,2] We present a case report of a middle-aged male who developed necrosis of maxilla due to infection after a traumatic injury that followed a chronic course and eventually led to fungal osteomyelitis of maxilla.

Case Report

A 42-year-old male patient reported to us with a chief complaint of pain, swelling, and pus discharge from left upper jaw. History

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revealed trauma due to blunt injury over the left cheek bone 3 weeks back. Medical history was nonsignificant. On clinical examination, there was a preseptal orbital cellulitis of lower eyelid [Figure 1]. Intraorally, there was a draining abscess in the left maxillary buccal vestibule in the canine–premolar region. Clinically, there was no displaced fracture of the maxilla or zygomatic bone. Noncontrast computerized tomographic scans showed an undisplaced fracture of the left orbital floor with intraorbital air foci, hyperdensity (HU 35–40) in the maxillary, ethmoid and sphenoid sinuses, and soft tissue swelling in the left infraorbital and maxillary region [Figure 2]. The patient was admitted and routine blood investigations and pus culture and sensitivity tests were ordered. The tests reported a random blood glucose level of 498 mg/dl, leucocytosis with neutrophilia, and increased ketone bodies in the urine. Pus culture showed a growth of *Klebsiella pneumoniae* stains. After 10 days, on intraoral examination, a necrotic and mobile bony segment of the left maxilla in the canine to molar segment was noted along with a palatal abscess in the first molar region and mobile teeth. Surgical debridement of the maxillary sinus region and lateral wall of the nose along with the extraction of involved teeth were done and multiple pieces

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of blackish escharotic bone were recovered [Figures 3 and 4]. Histopathology of the tissues using hematoxylin–eosin stain confirmed mucormycosis of the maxilla [Figure 5]. Intravenous injections of lyophilized amphotericin B (AmB) 50 mg were given for 6 weeks with monitoring of blood urea and creatinine levels as the drug is nephrotoxic. After 3 months of healing and complete epithelization, an interim obturator with a nasal and antral bulb was fabricated to prevent oronasal regurgitation.

The patient has been disease free for the follow-up period of 1 year [Figure 6].

Discussion

Mucormycosis is a rare opportunistic fungal infection caused by a saprophytic fungus (phycomycetes). The early clinical features include perinasal paresthesia, periorbital cellulitis, rhinorrhea, nasal crusting with stuffiness and epistaxis with or without complain of fever, arthralgia, and weight loss which is followed by eschar formation and necrosis of the naso-facial region. Strawberry gingivitis is one of the clinical features seen. Severe infection may cause life-threatening conditions like cavernous



Figure 1: Pre-operative view showing cellulitis of lower eyelid



Figure 2: Non-contrast computerized tomographic images showing an undisplaced fracture of the left orbital floor with intraorbital air foci

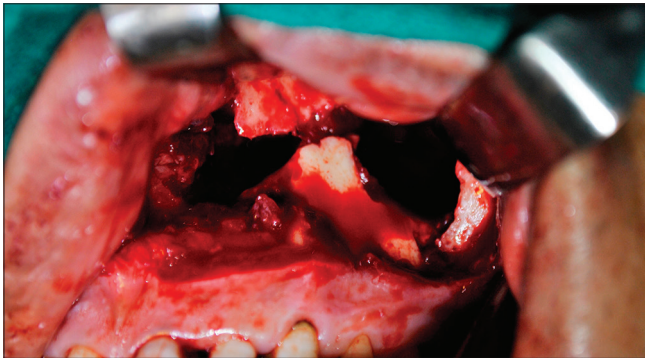


Figure 3: Surgical debridement of the maxillary sinus region and lateral wall of the nose

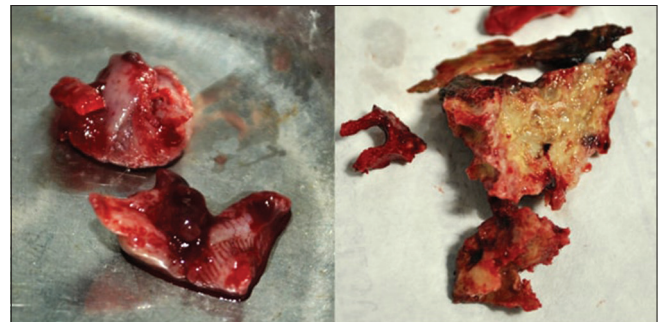


Figure 4: Excised soft and hard tissue

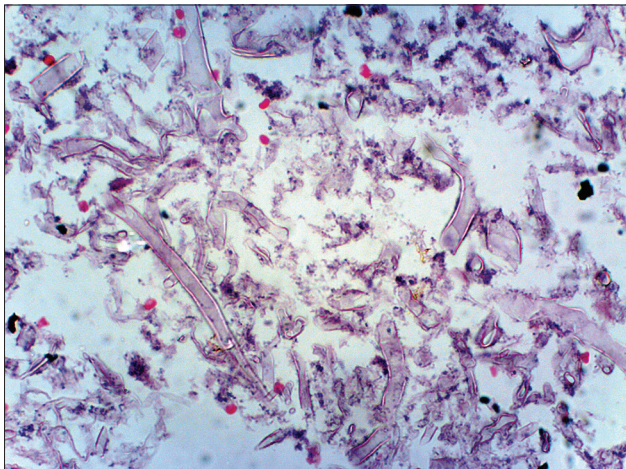


Figure 5: Hematoxylin–eosin staining showing non-septate hyphae



Figure 6: Post-operative view showing healed lesion

sinus thrombosis, thrombosis of carotid artery or internal jugular vein, and death.^[3]

Medical treatment is not alone effective because of poor drug concentration and availability to the infection site due to thrombosis of vascular system. Treatment consists of correction of underlying systemic abnormality, for example, control of diabetic state, stoppage or modification of immunosuppressant or corticosteroids are essential.^[4]

Treatment approach includes antifungal therapy combined with surgical intervention (sequestromy and debridement) and adjunctive therapy. Choice of antifungal drug is AmB therapy (1.0–1.5 mg/kg/day).^[3] Posaconazole 400 mg is to be taken twice daily.^[5]

Deferasirox therapy should be continued as 20 mg/kg for 2–4 weeks. Hyperbaric oxygen therapy should be given as 100% oxygen for 90 min–2 h at a pressure of 2.0–2.5 A.^[5]

Conclusion

Mucormycosis usually precipitated in immunocompromised/debilitated persons. Various health promotion programs need to be formulated regarding systemic disease like diabetes. The preventive measures, patient counseling, and education about complications like mucormycosis can be facilitated so that early diagnosis and prompt treatment of the lethal diseases can be done at various primary health-care settings. This case report will add a guide for primary caretaker in better management for patient with mucormycosis.

The main aim should be to recognize the clinical presentation of mucormycosis, early diagnosis, correction of the underlying systemic disorder, and surgical management along with administration of potent antifungal therapy and to look, with

auspicious sight, the exposed bone in immunocompromised patients.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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