Mucormycosis of maxillary sinus invading maxilla: A case report
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Abstract
Mucormycosis is a fungal infection caused by saprophytic fungus that can be found in soil, fruits, and decaying vegetables. Mucormycosis is usually noted in immunocompromised individuals, patients on systemic steroid therapy, in patients having malignant hematologic conditions, poorly controlled diabetic patients, and organ transplant patients. In this report, a case of 49-year-old male with well-controlled diabetes who was diagnosed with mucormycosis of the left maxillary sinus invading maxilla is discussed. Systemic antifungal therapy along with debridement followed by the extraction of teeth and removal of necrotic bone was carried out. Complete debridement was achieved with well-healed maxillary sinus and alveolus with no signs of recurrence of the disease.

Keywords: Diabetic, fungi, immunocompromised, mucormycosis, maxillary sinus, oral mucosa

Introduction
Mucormycosis is an angioinvasive opportunistic fungal infection caused by saprophytic, terrestrial, rapidly growing fungi Mucorales. In 1885, Paltauf mentioned the first case of mucormycosis in humans. In 1942, Gregory et al. reported three cases of mucormycosis of the central nervous system. From then on many cases have been reported in literature. We present a case of maxillary mucormycosis in a 49-year-old patient with well-controlled diabetes.

Case Report And Results
A 49-year-old male patient presented to the outpatient department of Oral and Maxillofacial Surgery, Kashibai Navale Medical College and Hospital, Nalhe, Pune, with a chief complaint of mobility of the upper anterior teeth and occasional pus discharge from upper left posterior tooth. The patient gave a history of pain and pus discharge with 26, 4 months back for which he had undergone root canal treatment in private clinic, but the pus discharge did not subside. There was no pain or tenderness present. There was approximately 1 cm × 1 cm swelling present on the palate when the patient reported to us.

Computed tomography (CT) nose and paranasal sinuses coronal and axial views showed hypodense soft tissue density lesion completely occupying the left maxillary sinus. The left osteomeatal unit is widened and blocked by the lesion. The lesion is seen involving the left anterior ethmoidal air cells. The left frontal sinus draining pathway is blocked by the thickening, which has caused erosion and destruction of hard palate, perpendicular plate of ethmoid, inferior and anterior walls of the left maxillary sinus, and alveolar arches of upper incisors, canine, and premolars. Floor of the orbit is normal.

Incisional biopsy was taken in private clinic. Hematoxylin and eosin section showed broad aseptate fungal hyphae with the right angle branching morphologically consistent with mucormycosis. Granulomas composed of epithelioid cells, giant cells, and histiocytes were also seen. Periodic acid–Schiff stain showed similar fungal profiles. Methenamine silver stain highlighted similar black-colored argyrophilic fungal profiles.

Immediately after the diagnosis was established patient was started on Amphotericin B (2 mg/kg.d). 7 days later, the dose of liposomal amphotericin B was raised up to 4 mg/kg/day. During the entire course of the treatment, the patient was closely monitored for signs and symptoms of drug toxicity. After which patient was undertaken for sinus debridement and extraction of teeth 11 to 27 was done and necrotic bone was removed. Obturator was placed after 4 days postoperatively. Injection liposomal amphotericin B 4 mg/kg/day is given over 7 consecutive days postoperatively as well. Plain CT scan axial and coronal was done at the 12th post-operative week which showed well-healed cavity with no residual disease. The patient was given dental prosthesis after 2 months.
Discussion

In 1885, Paltauf mentioned the first case of mucormycosis in humans. Mucormycosis can be manifested in various forms such as rhinocerebral, pulmonary, abdominal, cutaneous, and systemic. A recent meta-analysis conducted by Roden et al., analyzed 929 cases since 1885, and reported that sinus infection was (39%), pulmonary infection (24%), and cutaneous infection (19%). A factor predisposing to mucormycosis is diabetes complicated by ketone acidosis which accounts for >50% of all infected cases. Management of mucormycosis should be untimely in diabetic patients, surgical debridement along with effective antifungal therapy should be started immediately. In our case, the patient had diabetes (well controlled) with no other comorbidities. There was no palatal perforation. Early diagnosis was established by CT scan and biopsy. The patient was immediately started on injection amphotericin B and the surgical debridement was done with preservation of maxilla and orbital floor which was followed by post-operative antifungal therapy of amphotericin B.

In an analysis of literature review by Mignogna et al., revealed that India was affected the most with 94 (44.3%) patients. The spores of this fungus enter the system due to inhalation from the atmosphere giving rise to pulmonary and rhinocerebral infections. In patients who are immunocompromised, the nose and/or maxillary sinuses are the predominant source of infection.

In case of our patient, the fungi may have come in contact with the damaged epithelium and may have invaded the sinus causing erosion of the bony walls of the sinus. Failure to establish diagnosis would have increased the chances of its spread to orbital region.

Furthermore, since the availability of amphotericin B, the mortality due to mucormycosis has decreased. It acts as the first-line therapy in the treatment of mucormycosis. Three lipid formulations of amphotericin B are clinically available: Liposomal amphotericin B, amphotericin B lipid complex, and amphotericin B colloidal dispersion. As it has less nephrotoxicity as compared to amphotericin B deoxycholate, higher doses can be administered with successful outcomes.
Conclusions

The present case report focuses on early diagnosis and surgical management of a case of mucormycosis with sufficient clinical outcome. Although this case can be treated by superficial hemimaxillectomy, early diagnosis and prompt systemic treatment along with debridement resulted in successful outcome of this conservative treatment.

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The authors report no conflicts of interest related to this study.

References


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