

Mucormycosis of the Palate and its Post-Surgical Management: A Case Report

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Abstract:

The aim of this article is to discuss a case report of palatal mucormycosis and its post-surgical management and to add another case in point to the existing literature. This is an interesting case report of a 47-year-old male who reported to our clinic with the complaint of ill-fitting denture. On edifying history, he was a known case of uncontrolled diabetes that was surgically treated by partial maxillectomy for palatal mucormycosis a year back. On examination, the patient presented with a large oro-nasal communication with an ill-fitting obturator. He was managed by replacement of the old obturator with a new denture bearing self-stabilizing obturator. To conclude it is ingenious for the dentist to consider mucormycosis in the differential diagnosis of palatal necrosis and perforation especially in elderly patients who are immunocompromised. The obturator serves the best treatment of choice in terms of immediate reconstruction, lack of complication such as osteoradionecrosis, patient acceptance, cheap and restoring the physiological functions.

Key Words: Diabetic ketoacidosis, mucormycosis, obturator

Introduction

Mucormycosis is an opportunistic frequently fulminating fungal infection that is caused by a saprobic organism of class zygomycetes. Spores liberated in air can be inhaled by the human host.^{1,2} Paltauf in 1885 first reported such a disease and coined the term "mycosis mucorina" which later changed as "mucormycosis."³

Mucormycosis of the hard palate is an unpropitious sign.⁴ Rich vascularity of the palate postpones its necrosis, but this fungus erodes the arteries leading to thrombosis and then to necrosis. Mucormycosis of the hard palate is generally seen as necrotic ulceration or sloughing of the palatal mucosa.⁵

Prior to HIV pandemic, diabetic acidosis was considered 50-70% as etiological factor. Mucormycosis is common in insulin dependent diabetes mellitus patients who are ketoacidotic. Ketoacidosis inhibits the binding of iron to transferrin allowing serum iron levels to rise.^{1,4} The growth of these organisms is enhanced by iron. 60% of the cases have oral, cranial, or facial involvement.¹

The palate which separates the oral cavity from the nasal cavity can be subjected to malignant and infectious lesions (fungal, viral, and bacterial) where surgical intervention like alveolectomy and palatotomy becomes inevitable. This can further worsen the situation by creating a communication between the oral and nasal cavity compromising many physiological activities such as chewing, swallowing, and phonation. Unintelligible speech, inability of the tongue to make contact with the solid surface, fluid leakage through the nose, acute and chronic episodes of sinusitis, collapse of middle third of the face, associated temporomandibular joint problems are all the post-surgical complications. It also has psychological implications on the patient.²

Maxillary bone maintains the esthetics of the nose, cheeks, and hemi-face.² Hence, restoring its function and aesthetics is the treatment goal. The scarred and tense post-surgical tissues exert strong dis-lodging forces.⁶ Palatal obturator is a simple and inexpensive post-surgical restoration especially in mucormycosis cases in terms of restoring the physiologic function and patient satisfaction.²

This article discusses a surgically treated case of mucormycosis who reported to our clinic with a palatal defect and was treated with a hollow-bulb denture bearing obturator.

Case Report

The 47-year-old male patient reported to our clinic with the complaints of difficulty in swallowing, nasal tone in speech, difficulty in breathing while lying down, severe snoring and food leakage through the nose while eating due to nasal regurgitation. On eliciting the past medical history the patient gave history of uncontrolled diabetes for which he

did not take regular medications and associated necrotic ulcerative lesion of the hard palate associated with stuffiness of the nose for which he was surgically treated. On checking his previous reports, it was clear that he was a treated case of mucormycosis of the palate. Partial palatotomy was done under general anesthesia a year back, and a temporary obturator was given. His previous diabetic reports revealed pp - 400 mg/dl. The histopathological report describes extensive necrosis with numerous non-septate hyphae at the periphery, and enormous neutrophilic infiltrate in the connective tissue (Figure 1). His computed tomography (CT) reports revealed complete opacification of the right maxillary sinus (Figure 2) and a lesion of soft tissue density in the nasal cavity, causing destruction of the nasal septum centrally, destruction of the medial walls of the antra bilaterally and perforation of the palate inferiorly (Figures 3a and b). Partial destruction of the floor of the orbit and ethmoidal sinus could be seen on the coronal CT images (Figure 2). CT scan of the brain was apparently normal. The patient has been treated with high doses of amphotericin

B and surgical debridement. On clinical examination, the patient had a huge oro-antral communication of about 5 cm x 6 cm in diameter on the right side, extending to the left side crossing the midline. Right maxillary alveolus was missing and teeth 18-22 were missing (Figure 4). There was dental caries 35 and 46 which was treated by root canal treatment before fabrication of obturator. We advised for a panoramic radiograph (Figure 5) which revealed missing maxillary teeth on the right side with a radiolucency extending into the maxillary sinus. There was a mild pathological extrusion of mandibular teeth due to missing counterparts, and it was orthodontically corrected before obturator fabrication. A thorough clinical examination was made to rule out any new lesion, and his post prandial glucose level was checked which was 128 mg/dl. The patient was under insulin therapy. The patient was managed by fabricating an obturator. First, the primary alginate impression (Figure 6a) was taken by closing the nostrils of the patient with cotton. The impression was poured using Type III gypsum (Figure 6b) product. In the cast a custom tray was fabricated using auto-polymerising resin material, and border moulding was done with heavy body addition silicone impression material and a secondary impression (Figure 6c) was taken with light body impression material as they have dimensional stability. Teeth setting was done, and a hollow bulb denture bearing obturator was fabricated (Figure 7a-c). On delivery of the obturator occlusal adjustments were made, and any interferences were noted and corrected.

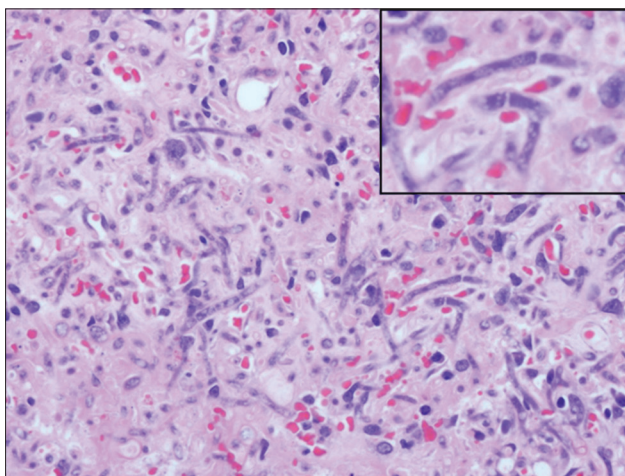


Figure 1: Neutrophilic infiltrate in connective tissue with non-septate hyphae (H and E, x10).

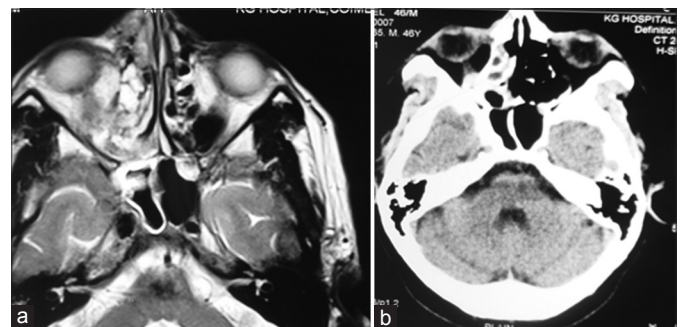


Figure 3: (a and b) Axial computed tomography images showing right antral opacification.

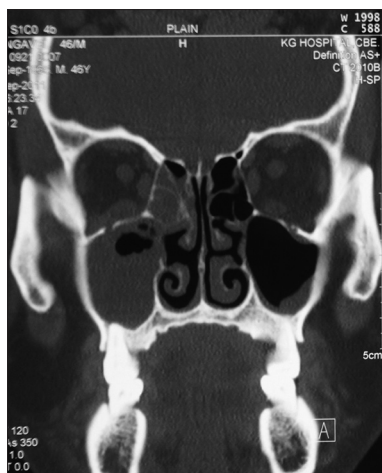


Figure 2: Coronal computed tomography showing complete opacification of the maxillary sinus, partial destruction of floor of the orbit, and walls of the ethmoidal sinus.

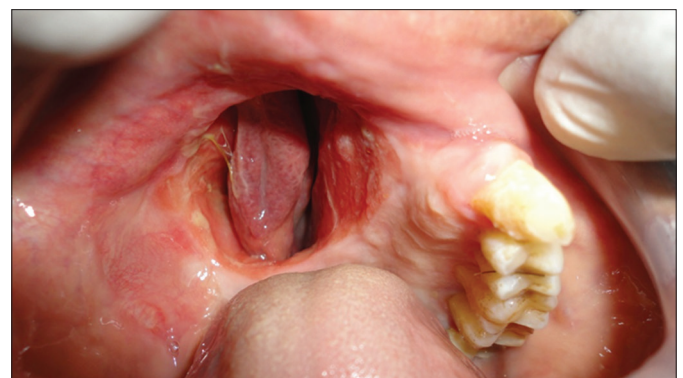


Figure 4: Intraoral photograph showing large oro-antral communication.

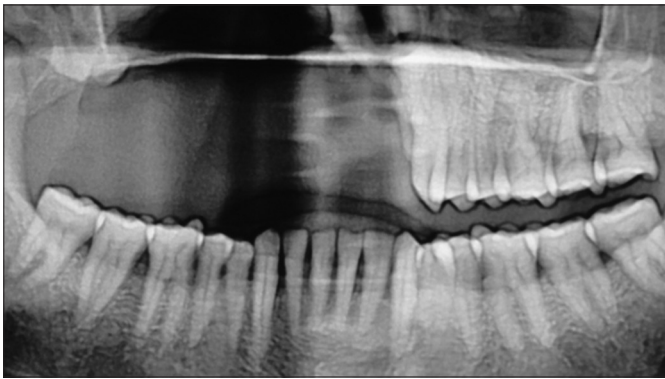


Figure 5: Panoramic radiograph.

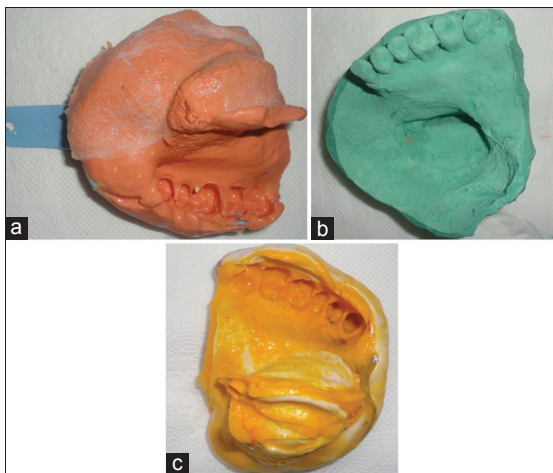


Figure 6: (a) Primary impression, (b) maxillary cast, (c) secondary impression.

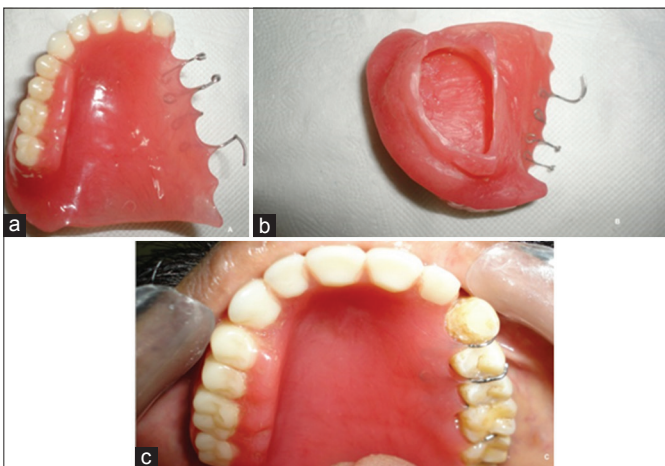


Figure 7: (a) Denture bearing obturator with clasps, (b) hollow bulb ventral surface that fits the maxillary defect, (c) patient wearing the obturator.

Discussion

Mucormycosis of the palate is a rare opportunistic fungal infection. Jain *et al.*⁷ suggested both surgical and anti-fungal combination. Amphotericin B is given. It has minimal side effects. It is given in dextrose 5% in water intravenous (IV) at a dose of 1-1.5 mg/kg daily for 3 months. Mucorales show no pathogenesis to normal individuals when compared

to immunocompromised. There are six main types of mucormycosis, rhinocerebral, rhinomaxillary, pulmonary, cutaneous, gastrointestinal, and central nervous system. In this rhinocerebral and pulmonary forms are common in diabetes patients. Rhinocerebral form occurs by inhalation of air borne spores.⁸

Immediately, after inhalation or entering subcutaneous, tissue there is the action of mononuclear and polymorphonuclear phagocytes. As in diabetes ketoacidosis patients, rhizopus produces ketoreductase an enzyme which makes use of the patients ketone bodies leading to reduced phagocytic activity.⁹ Early diagnosis and early management prevent complications. Mucormycosis can be fatal. Recently, IV liposomal amphotericin, IV lipid complex, hyperbaric oxygen therapy are used.⁷

Obturator are classified as solid, open hollow, and closed hollow as to the nature of their extension into the defect site. Both open and closed hollow obturators allow for the fabrication of a lightweight prosthesis that can be tolerated by the patient.¹⁰

Barrak¹¹ treated four cases of mucormycosis with palatal perforation and reported two deaths. A full course of treatment and frequent debridement and care did not favor healing in these cases.

Rolston and Kontoyiannis¹² reported 15 cases of mucormycosis in patients with hematological malignancy and stated, oral soreness reported in patients with hematologic malignancy or organ transplantation is an alarming sign for immediate oral examination and screening for mucosal alterations related to fungal infections especially mucormycosis. Similar association between acute myeloid leukemia and mucormycosis was reported by Bany-Yaseen *et al.*¹³ Manjunatha *et al.* suggested that even simple procedures like dental extractions could initiate a fulminating disease like mucormycosis.^{14,15}

Iron is an essential element for cell growth and development, contributing to many vital processes of the cell. Therefore, successful pathogens use multiple processes for obtaining iron from the host. Recent data demonstrates that the level of available unbound iron in serum plays a critical factor in uniquely predisposing patients with diabetic ketoacidosis to mucormycosis.¹⁶

Artis *et al.*¹⁷ proved that acidosis occurring during a diabetic ketoacidosis crisis disrupts the fungal growth inhibitory function of serum, which can lead to a major defect in host defense and mucormycosis if not rapidly corrected.

The obturator is a common and wiser choice in cases of post-surgical palatal reconstruction. Obturator lacks bony base and posterior palatal seal which compromises retention.¹⁸ In a

dentulous patient retention is easier to achieve when compared to the edentulous patients as support can be gained from the remaining teeth. The success of an obturator depends on the volume of the defect. The size of the obturator should be as light as possible. A hollow bulb design was chosen to reduce the bulk of the obturator. It also adds resonance in speech which improves quality. In this frequent follow-ups of the patient was made to assure the stability of the obturator and ensure patients satisfaction in bringing back the physiologic functions.

Conclusion

Dentist should be alert and suspicious in cases of palatal perforation especially in elderly patients who are immunocompromised which will be of great benefit to the patient in terms of preventing wide spread surgical resection, post-surgical complications and loosing physiologic functions.^{19,20} The obturator serves the best treatment of choice in terms of immediate reconstruction, lack of complication like osteoradionecrosis, patient acceptance, cheap and restoring the physiological functions

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