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Rehabilitation of the Surgical Defect Secondary to Mucormycosis: A Case Report

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Abstract---Aim: To report a case of Mucormycosis of right maxilla due to uncontrolled Diabetes Mellitus along with its surgical management and immediate Prosthetic rehabilitation. Background: Phycomycetes is the causative agent leading to the deadly fungal infection called Mucormycosis. Predisposing factors involve patients with uncontrolled diabetes mellitus, immunosuppressive therapy, organ transplant. The survival rate of the patients depends upon how quickly the disease is diagnosed and treated. Case Description: This article presents a case report of a 54-year-old female patient reported with the complaint of pain on the right side of the face along with headache and difficulty in breathing. Patient was diabetic and not under medication. After routine investigations, patient was diagnosed for Mucormycosis. The treatment involved excision of the necrotic tissues present in the right maxilla. Patient was referred to Department of Prosthodontics before surgery. Following hemi-maxillectomy, closure of the defect was done to prevent regurgitation due to oro-antral communication using surgical obturator and inserted immediately following surgery. To reduce the weight, a hollow bulb surgical obturator was the prosthetic choice in this case report. Conclusion: Surgical obturator can be a treatment of choice for immediate closure of defect secondary to Mucormycosis.

Keywords---case report, diabetes mellitus, hollow bulb obturator, mucormycosis, surgical obturator.

Introduction

Neglect starts out as an infection and then becomes a disease. Mucormycosis or zygomycosis, refers to an opportunistic, deep mycotic infection which has life-threatening potential if not diagnosed and treated in the early stage. Paltauf first noticed this infection in 1885 and termed it as “mycosis mucorina” which is currently called as “Mucormycosis.”¹ Patients suffering from uncontrolled diabetes and immunocompromised individuals are more prone to such infection leading to necrosis of the hard and soft tissue. Based on the type of tissue involved it can be differentiated into fasciitis, necrotizing cellulitis and myonecrosis.² In most of the cases, human infection occurs due to Mucorales, especially *Rhizopus*, *Mucor*, *Absidia*, *Cunninghamella*, *Cokeromyces*, and *Syncephalastrum*.³

There are various types of Mucormycosis, in which Rhinocerebral type is most prevalent. It begins soon after the fungal spores are inhaled and may involve nasal mucosa, turbinate bones, paranasal sinuses, orbit. It can also spread into intracranial tissues. It causes thrombosis and necrosis of hard and soft tissue by penetrating blood vessels and arteries.⁴ Due to lack of available treatment, it had very poor prognosis. However, with the rapid advancement in medical science, survival rates have increased significantly up to 80%.⁵

When maxillary bone is involved, surgical removal of necrosed tissues is recommended which may lead to oro-antral communication or defects in hard and soft palate or alveolar ridges.⁶ This case report includes prosthetic rehabilitation of the surgical defect secondary to Mucormycosis with hollow bulb surgical obturator in an uncontrolled diabetic patient, immediately following the removal of infected and necrosed tissues.

Case description

A 54-year-old female patient was reported to the Department of Otolaryngology with the chief complaint of pain and swelling on right side of the cheek and eye along with headache and difficulty in breathing. The patient's medical history revealed that she was suffering from Diabetes Mellitus and was not under medication for last 3 years. On extra-oral examination the right mid face was tender on palpation. On intraoral examination, Patient was completely dentulous with respect to maxilla, with grade I mobility noted in relation to all maxillary teeth on the right side. A small depression with blackish discoloration was noted in the right side of the hard palate (Fig 1). Patient was sent for routine investigations. On diagnostic Nasal Endoscopy, necrotic tissues were seen on the medial wall of maxillary sinus.



Figure 1. A small depression with discoloration seen on the right side of the hard palate

Plain and contrast MRI (magnetic resonance imaging) of head was performed using T1W, T2W, FLAIR, DWI, SWI sequences in various planes. Findings were as follows:

- Right orbital cellulitis with preseptal inflammation.
- Thickening of the intra-orbital segment of the right optic nerve along with perineural sheath enhancement showing diffusion restriction suggestive of right optic neuritis.
- Chronic right maxillary, ethmoid and frontal sinusitis.

Diagnostic Computed tomography of the patient showed widening of right maxillary sinus ostium along with thickening of mucosa in the right maxillary sinus. Erosion and irregular thinning were noted in the anterior, medial, lateral walls of the maxillary sinus which is suggestive of chronic right maxillary sinusitis. Frontal, ethmoid and sphenoid sinusitis was also noted (Figure 2).



Figure 2. Computed tomography showing opacification of the sinus along with the erosion and thinning of walls of the sinus

Diagnostic KOH MOUNT test was then performed which showed fungal colonies with aseptate hyphae suggestive of mucormycotic infection. Following investigations patient was diagnosed for Mucormycosis involving the right maxilla, inferior and middle turbinate. Fungal sinusitis involving right maxillary sinus. The treatment of choice was total right maxillectomy under general anesthesia. The excision was planned up to palatal midline medially and with the posterior extension up to 3rd molar region. Prior to the surgery patient was referred to Department of Prosthodontics, for the fabrication of a prosthesis to close the defect immediately after surgery. Hence it was decided to fabricate a Hollow bulb surgical obturator.

Diagnostic impressions were made with hydrocolloid impression material. The diagnostic maxillary cast obtained was duplicated, and a working cast was made in Plaster of Paris or Type II gypsum product. Based on the occlusal bite registration record, the casts were mounted on a semi adjustable articulator. According to the planned surgical procedure a defect was created on the working cast by arbitrarily scrapping the cast (Fig 3). The modelling wax was adapted to the defect area and maxillary rim was fabricated on the right side. Teeth arrangement was done, Circumferential Clasps were fabricated and adapted on to the left side premolars and molars using 21-gauge orthodontic wire for added retention.



Figure 3. Scrapping of working cast was done according to the size of the defect.

The surgical obturator was fabricated in conventional manner, using clear heat cure acrylic resin. To make it hollow, putty material was used. After complete acrylization, prosthesis was finished and polished (Fig 4 a, b). The prosthesis was then sterilized.

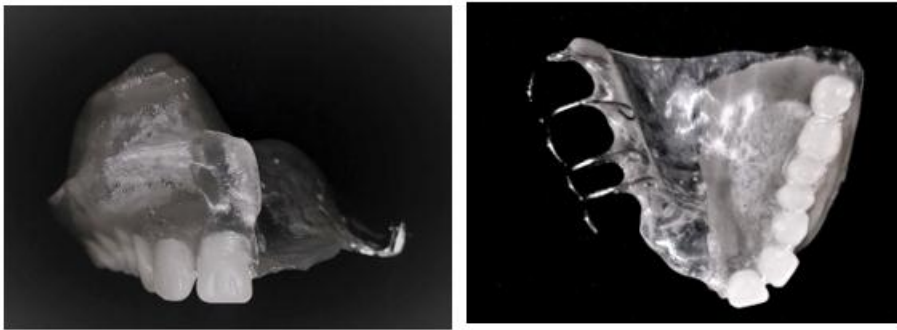


Figure 4. Hollow bulb surgical obturator
a. Front view depicting the hollow bulb; b. Occlusal view

The surgery was performed under general anaesthesia. Necrotic tissues were resected (Fig 5) and the surgical defect was closed by a hollow bulb obturator which was inserted immediately after surgery (Figure 6,7). It also provided a nasomaxillary seal to prevent regurgitation. The prosthesis was relined 4th week following the surgery with auto-polymerizing acrylic resin. (Figure 8)

The patient was advised to wait for three months as the rate of recurrence is high (reference) followed by which a definitive prosthesis would be delivered.

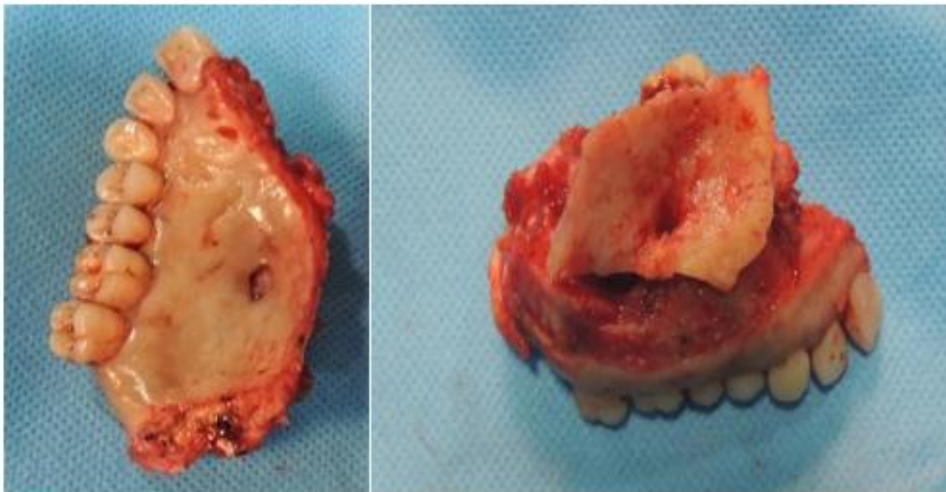


Figure 5. Surgically excised necrotic tissues involving right maxilla

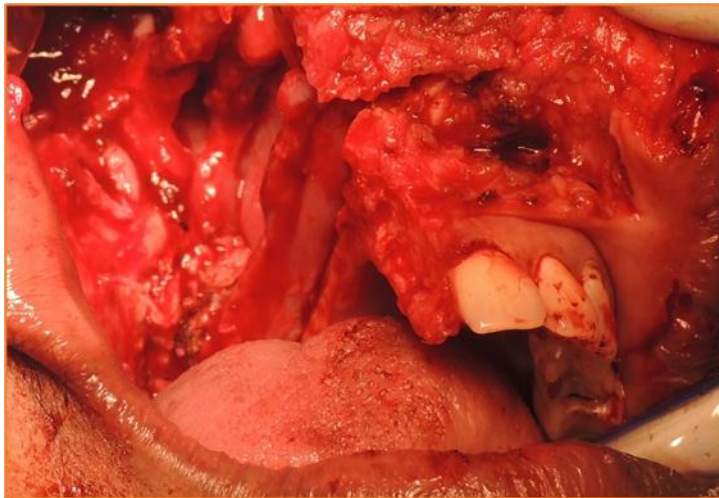


Figure 6. Surgical defect after excision

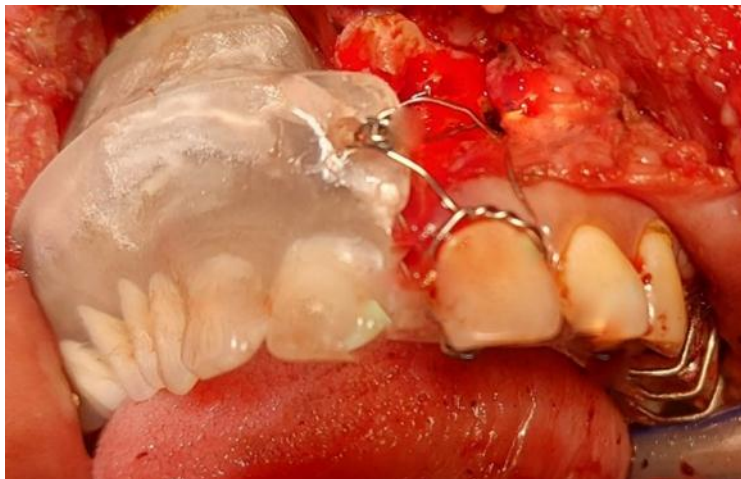


Figure 7. Insertion of hollow bulb surgical obturator. Prosthesis was ligated to the adjacent maxilla



Figure 8. 4 weeks following insertion of the prosthesis, relining was performed

Discussion

In the current pandemic situations, we get to hear a lot about Mucormycosis. It has emerged as a secondary infection to COVID-19, especially during the second wave. The death rate has also been increased due to the infection as there were no correct treatment options available. According to the literature, it is an opportunistic fungal infection. Various predisposing conditions include poorly controlled diabetes mellitus, case of organ transplant, malignancy, long term use of steroids and other immunosuppressive drugs, prophylactic voriconazole therapy for transplant patients, prolonged ICU stay and blood dyscrasias.^{7,8} Patients with Uncontrolled diabetes mellitus produce the enzyme ketoreductase, which utilizes the ketone bodies and will alter the immunologic response due to reduction in phagocytic activity of polymorphonuclear leukocytes.⁵ Due to peripheral vascular disease in such patients, local tissue ischemia, thrombosis of internal maxillary artery or descending palatine artery and further infections are most common observational findings.⁹

After careful evaluation, suspected cases of mucormycosis should be subjected to suitable imaging which is strongly advised to document extent of the disease and then followed by surgical intervention. In present case, patient was subjected to MRI and CT scan. The infected tissue should be completely removed followed by the administration of active antifungal agents at optimal doses and use of various adjunctive therapies.

In the present case the patient had a history of uncontrolled diabetes mellitus, and the right side of the maxilla was completely necrotized. Successful management of mucormycosis is based on a multimodal approach, including reversal or discontinuation of underlying predisposing factors. Hence, immediate correction of metabolic disorders is a must in patients with uncontrolled diabetes and who are suspected of mucormycosis. Liposomal amphotericin B, enriched with two newer azoles (posaconazole and isavuconazole), is the drug of choice for the initial antifungal treatment, according to guidelines by ECIL-6 and

ECMM/ESCMID.¹⁰ Hence both surgical and anti-fungal therapy had been advised along with various adjunctive therapies. Present case followed the same protocol.

An obturator was advised post-operatively to prevent oronasal regurgitation and immediate closure of the surgical defect. A hollow bulb surgical obturator was fabricated for immediate post-surgical insertion. The obturator usually lacks bone support; therefore, retention is affected adversely.¹¹ The size of the defect determines the prognosis and if the defect is large, it adds more bulk to the prosthesis. We chose to fabricate a hollow bulb design of the surgical obturator. Hollow design reduces the weight of the prosthesis.¹² There are various methods described in the literature for fabrication of hollow bulb obturator.¹³⁻¹⁵ In this case, elastomeric impression material (condensation silicone putty) was incorporated into the mould during processing, to make it hollow which was removed after acrylization of the prosthesis. Hollow bulb also enhances resonance in speech.¹⁶

In the present case report, along with the surgical debridement systemic antifungal therapy, teeth were also incorporated in the hollow bulb surgical obturator prosthesis to improve the chewing efficiency of the patient. Routine follow-up was done to check for function and patient satisfaction, and it was found satisfactory.

Conclusion

A hollow bulb surgical obturator can be considered as a treatment of choice for immediate closure of the surgical defect, especially when dealing with Mucormycosis cases. It aids various oral functions and Prosthodontists play a very important in evaluation of such cases and for fabrication of the obturator prosthesis.

Clinical significance: Mucormycosis was very common complication seen during second wave of COVID-19 and uncontrolled Diabetes Mellitus was a major predisposing factor in these patients. Every case is unique and challenging for surgeon as well as for Prosthodontist. But with the advancement in Medical and Dental field, treatment and immediate post-surgical rehabilitation is possible in order to maintain patient's oral function.

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References

1. Garg R, Gupta VV, Ashok L. Rhinomaxillary mucormycosis: A palatal ulcer. *Contemp Clin Dent* 2011;2(2):119-23. <https://doi.org/10.4103/0976-237X.83080>.
2. Leitner C, Hoffmann J, Zerfowski M, Reinert S. Mucormycosis: necrotizing soft tissue lesion of the face. *J Oral Maxillofac Surg*. 2003;61(11):1354-1358. [https://doi.org/10.1016/s0278-2391\(03\)00740-7](https://doi.org/10.1016/s0278-2391(03)00740-7).

3. Ribes JA, Vanover CL, Baker DJ. Zygomycetes in human disease, *Clinical Microbiological Reviews*, 2000;13: 236-301. <https://doi.org/10.1128/CMR.13.2.236>.
4. Nilesh, K., & Vande, A. V. (2018). Mucormycosis of maxilla following tooth extraction in immunocompetent patients: Reports and review. *Journal of clinical and experimental dentistry*, 10(3), e300–e305. <https://doi.org/10.4317/jced.53655>.
5. Manjunatha, B. S., Das, N., Sutariya, R. V., & Ahmed, T. (2012). Mucormycosis of the hard palate masquerading as carcinoma. *Clinics and practice*, 2(1), e28. <https://doi.org/10.4081/cp.2012.e28>
6. Shah RJ, Katyayan MK, Katyayan PA, Chauhan V. Prosthetic rehabilitation of acquired maxillary defects secondary to mucormycosis: clinical cases. *J Contemp Dent Pract*. 2014;15(2):242-9. <https://doi.org/10.5005/jp-journals-10024-1522>.
7. Jones AC, Bentsen TY, Fredman PD. Mucormycosis of the oral cavity. *Oral Surg Oral Med Oral Pathol* 1993;75(4):455-460. [https://doi.org/10.1016/0030-4220\(93\)90170-9](https://doi.org/10.1016/0030-4220(93)90170-9).
8. Kofteridis DP, Karabekios S, Panagiotides JG, Bizakis J, Kyrmizakis D, Saridaki Z, Gikas A. Successful treatment of rhinocerebral mucormycosis with liposomal amphotericin B and surgery in two diabetic patients with renal dysfunction. *J Chemother* 2003;15(3):282-286. PMID: 14579660.
9. Kumar JA, Babu P, Prabu K, Kumar P. Mucormycosis in maxilla: Rehabilitation of facial defects using interim removable prostheses: A clinical case report. *J Pharm Bioall Sci* 2013;5:S163-5. <https://doi.org/10.4103/0975-7406.114322>.
10. Skiada A, Lass-Floerl C, Klimko N, Ibrahim A, Roilides E, Petrikos G. Challenges in the diagnosis and treatment of mucormycosis. *Med Mycol*. 2018;56(suppl_1):93-101. <https://doi.org/10.1093/mmy/myx101>.
11. Bagis B, Aydogan E, Hasanreisoglu U. Rehabilitation of a congenital palatal defect with a modified technique: A case report. *Cases J* 2008;1(1):39. <https://doi.org/10.1186/1757-1626-1-39>.
12. Rani S, Gupta S, Verma M. Hollow bulb one piece maxillary definitive obturator - A simplified approach. *Contemp Clin Dent* 2017;8:167-70. https://doi.org/10.4103/ccd.ccd_887_16.
13. Didier M, Laccoureye O, Brasnu D, et al. New surgical obturator prosthesis for hemimaxillectomy patients. *J Prosthet Dent* 1993;69:520–3. [https://doi.org/10.1016/0022-3913\(93\)90163-i](https://doi.org/10.1016/0022-3913(93)90163-i).
14. McAndrew KS, Rothenberger S, Minsley GE. 1997 Judson C. Hickey Scientific Writing Awards. An innovative investment method for the fabrication of a closed hollow obturator prosthesis. *J Prosthet Dent* 1998;80:129–32. [https://doi.org/10.1016/s0022-3913\(98\)70098-8](https://doi.org/10.1016/s0022-3913(98)70098-8).
15. Habib BH, Driscoll CF. Fabrication of a closed hollow obturator. *J Prosthet Dent* 2004;91:383–5. <https://doi.org/10.1016/j.prosdent.2004.01.020>.
16. Blair FM, Hunter NR. The hollow box maxillary obturator. *Br Dent J*. 1998;184(10):484–7. <https://doi.org/10.1038/sj.bdj.4809669>.