

Post surgical immediate prosthetic reconstruction in patients with rhinocerebral mucormycosis: Case series

Kamya Mahajan, Monalisha Das, G. Ajay Kumar

Department of Prosthodontics and Crown and Bridge, Army College of Dental Sciences, Secunderabad, Telangana, India

ABSTRACT

Coronavirus disease has taken a toll on everyone's lives either physically or emotionally throughout the world. One of the most serious complications occurring currently in covid-positive and recovered patients is the deadly invasive mucormycosis. There are five clinical types of mucormycosis, out of which rhino-orbito-cerebral is the most common form (44%–49%). Management includes early diagnosis, control of the underlying diseases, and aggressive medical and surgical intervention. In patients undergoing maxillectomies, the use of immediate surgical obturators has become the gold standard. The rationale for its use is threefold—functional (matrix for the surgical dressing; allows the patient to speak and swallow), hygienic (separates the maxillary surgical site from contents of the oral cavity), and psychological (restores patient's self-image by reproducing lost oral structure). This article includes two case reports of patients diagnosed with rhinocerebral mucormycosis. The surgical debridement, medical management, and immediate reconstruction using palatal obturators have been explained.

Keywords: Coronavirus disease, mucormycosis, rhinocerebral mucormycosis, surgical obturator

Introduction

Mucormycosis is an infection caused by fungi *Rhizoglyphus oryzae* (70%) belonging to the order Mucorales.^[1] There are five clinical types of mucormycosis, out of which rhino-orbito-cerebral is the most common form (44%–49%).^[2] The infection begins in the nose and progresses to the paranasal sinuses, invading the CNS and orbit secondarily. The management includes early diagnosis, control of the underlying diseases, and aggressive medical and surgical intervention. This article emphasizes the immediate prosthetic reconstruction in patients operated for rhinocerebral mucormycosis along with their medical management, which must be known to all general primary care providers and physicians to avoid any delay in treatment.

Address for correspondence: Dr. Kamya Mahajan,
Department of Prosthodontics, Army College of Dental Sciences,
Secunderabad - 500 087, Telangana, India.
E-mail: mails4kamya04@gmail.com

Received: 17-06-2021

Revised: 04-10-2021

Accepted: 08-10-2021

Published: 31-01-2022

Access this article online

Quick Response Code:



Website:
www.jfmpc.com

DOI:
10.4103/jfmpc.jfmpc_1192_21

Case History

Case report number 1

A 57-year-old male patient reported to the Department of Prosthodontics with the chief complaint of difficulty in swallowing and speech since a month.

The medical history of the patient revealed type II diabetes mellitus since 17 years and was on oral hypoglycemics. The patient had tested positive for COVID-19 4 months back and was diagnosed with rhino-orbito-cerebral mucormycosis of the right side extending to the right temporal lobe, right nasal cavity up to the hard palate. Three months back, he underwent neurosurgery of the temporal region and was monitored closely for any post-surgical complications. Two months post surgery, the mucormycosis invaded the right lateral nasal cavity involving the middle, superior, and inferior turbinates and the maxilla up to the palate.

On extraoral examination, the right half of the face was swollen extending from the supraorbital region to the corner

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Mahajan K, Das M, Kumar GA. Post surgical immediate prosthetic reconstruction in patients with rhinocerebral mucormycosis: Case series. J Family Med Prim Care 2022;11:379-85.

of the lip [Figure 1]. There was inability to open the right eye [Figures 1 and 2].

On intraoral examination, the patient presented with a white crusty lesion on the right side of the palate extending till the midline medially and the first molar laterally [Figure 3]. Grade II mobility was present in relation to the molar on the affected side. The mouth opening of the patient was reduced to 2 fingers, that is, 25 mm [Figures 4 and 5].

Endoscopic debridement of the fungal debris and palatal reconstruction with a surgical obturator was decided as the treatment of choice. A multidisciplinary approach involving an anesthetist, an otorhinolaryngologist, and a maxillofacial prosthodontist was planned. After the treatment plan was formulated, it was explained to the patient and his son. They were educated about its shortcomings such as difficulty in speech and swallowing initially before the treatment was initiated.

Fabrication and insertion of the surgical obturator

Before the surgery, maxillary and mandibular arch impressions were made with irreversible hydrocolloid [Figure 6]. The impressions

were poured after blocking the teeth that would undergo extraction and clasps were fabricated. Complete wax-up was done for the upper arch and flaked [Figure 7]. Clear acrylic PMMA was used for the fabrication for ease of visibility on insertion [Figure 8].

On the day of surgery, surgical debridement was done and a palatal perforation was visible [Figures 9 and 10]. It presented as a classic class II defect according to Aramany's classification.^[3] The surgical obturator was inserted immediately while the patient was unconscious [Figure 11]. Amphotericin B was administered at 5 mg/kg for 14 days, which was later replaced by oral posaconazole at 300 mg every 12 hours on the first day followed by 300 mg once daily for 2–3 weeks.

He was recalled 4–5 days post surgery for a review. Complete obturation was checked with closed mouth water rinsing test. It was observed that when the patient held water in his mouth without the obturator, it would regurgitate through the nose; however, after placing the obturator, it would not, ensuring a proper seal. Regular telephonic conversations were made with



Figure 1: Profile view



Figure 2: Right lateral view- temporal sutures suggestive of previous surgery



Figure 3: Yellow-white crust seen on right side of palate extending till the first molar



Figure 4: Mouth opening measured by measuring the interincisal distance



Figure 5: Reduced mouth opening (25 mm)



Figure 6: Impressions made with hydrocolloid impression material



Figure 7: Wax-up



Figure 8: Surgical obturator

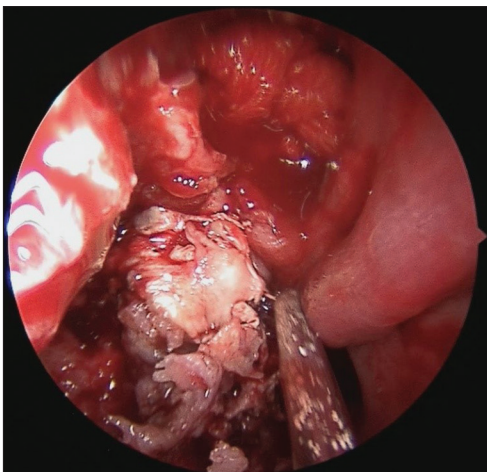


Figure 9: Fungi seen during endoscopic debridement



Figure 10: Maxillary defect

the patient's guardian to enquire about his health. After 3 weeks, the patient was recalled for a review [Figure 12]. The general health of the patient had improved considerably. As the surgical obturator was fitting well, a soft liner was placed in the region of the defect and altered as the interim obturator.

Case report number 2

A 60-year-old male reported with the chief complaint of tenderness over the region of the maxillary sinus and the orbit since 3 days. He also complained of loss of vision in the left eye since 2 days.

The medical history of the patient revealed uncontrolled diabetes mellitus (type II) since 10 years. No history of fever, breathlessness, or any other comorbidity was noticed. The viral markers were negative.

On examination, swelling of the left side of the face involving the orbital cavity and the left temporal side was noticed [Figure 13]. There was no movement of the left eye, no pupillary reflex, and the bulbar conjunctiva was congested. Intraoral examination revealed a white crusty lesion involving the left side of the palate [Figure 14]. The mouth opening of the patient was 26 mm [Figures 15 and 16]. Nasal endoscopic examination revealed black-yellow crusts involving the left nasal cavity and palate.

The MRI reports revealed soft tissue enhancement with edema suggestive of inflammation within the sinuses. Focal thickening and enhancement of the meninges were noted at the base of the temporal lobe. Part of the internal carotid artery showed minimal narrowing.

The patient was diagnosed with rhinocerebral mucormycosis (left side) extending to the cavernous sinus, middle cranial fossa, orbit, and hard palate. The treatment plan included endoscopic

debridement of fungal debris, followed by palatal reconstruction with an immediate surgical obturator.

Fabrication and insertion of the surgical obturator

The steps of fabrication of the obturator were similar to the first case [Figures 17–19]. During the surgery, it was observed that the mucor had invaded the left side of the palate completely. Partial maxillectomy was done creating a class I defect according to Aramany's classification of maxillectomy [Figures 20 and 21]. The obturator was dipped into 2% glutaraldehyde and first secured by clasps on the unaffected side. Four interrupted sutures were placed to secure the obturator to the affected side [Figure 22]. A Ryle's tube was inserted to aid in feeding.

The patient was in the ICU for the next 2 days and was closely monitored. Medical management included amphotericin B (liposomal) at 5 mg/kg body weight along with posaconazole 5 ml BD for 2 weeks and serum creatinine maintained below 2.5 mg/ml. The Ryle's tube was removed on the third day and the patient was kept on a semiliquid diet. No nasal regurgitation was reported. Postoperative instructions for maintenance were



Figure 11: Insertion of Surgical obturator immediately after surgery



Figure 12: Three weeks post-op



Figure 13: Profile view



Figure 14: Yellow-white crust seen on the left side of the palate



Figure 15: Mouth opening measured by measuring the interincisal distance

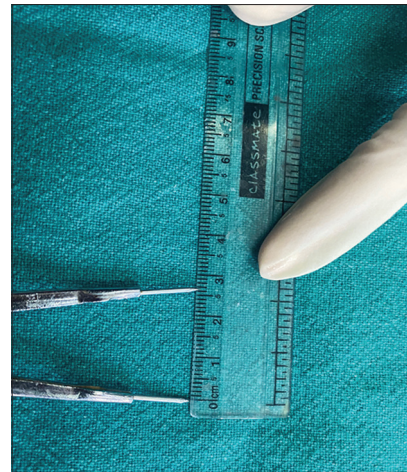


Figure 16: Mouth opening (26 mm)

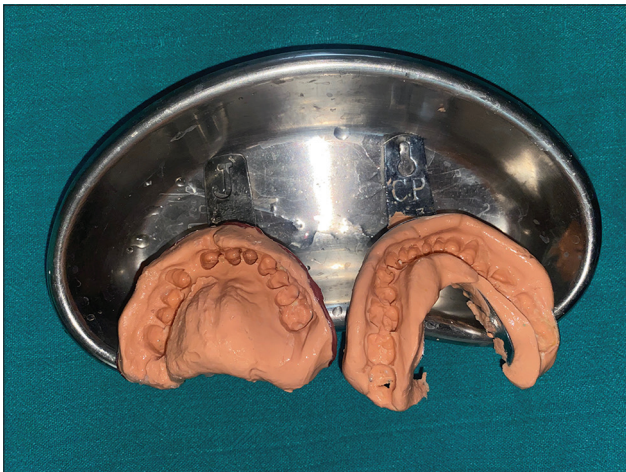


Figure 17: Impressions made with hydrocolloid impression material



Figure 18: Wax-up



Figure 19: Surgical obturator

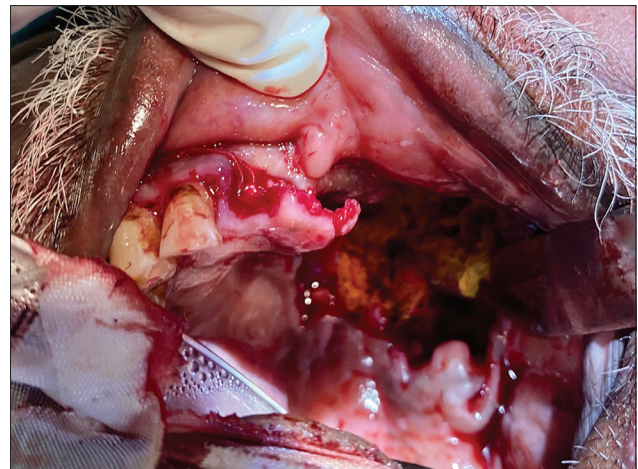


Figure 20: Partial maxillectomy of the left side

given to his guardian on the day of surgery. They were asked to clean the suture site repeatedly to avoid any infection. Regular telephonic conversations are still being made to the patient's guardian to enquire about his health.

Discussion

Rhinocerebral mucormycosis being a fulminant disease with a formidable mortality rate needs to be managed aggressively. Despite aggressive management in the form



Figure 21: Resected tissue

of surgical debridement and prompt antifungal therapy, the mortality of mucormycosis is >50% and reaches 100% in immunocompromised and patients with neutropenia.^[4] Major risk factor includes injudicious use of corticosteroids, organ or bone marrow transplantation, diabetic ketoacidosis, humidifier bottle contamination, increased serum iron levels, neutropenia, and malignant hematologic disorders.^[4,5] In diabetics, phagocytosis and polymorphonuclear neutrophil (PMN) chemotaxis is impaired, and macrophages are unable to phagocytose the fungal spores, thereby resulting in free spores in the tissues. The main portal of entry is the inhalational route. The fungi grow, resulting in vascular invasion and hematogenous dissemination.^[6] Antifungal medication such as amphotericin B is the drug of choice. Effective blood glucose and renal function monitoring due to nephrotoxicity of amphotericin B are essential. Additionally, hyperbaric oxygen along with the liposomal form of amphotericin B are used.^[7-9] New drug, isavuconazole, covers some of the gaps in the management of patients with renal and hepatic compromise.^[10]

The rationale for the use of an immediate surgical obturator is threefold, namely functional (matrix for the surgical dressing placed in the maxillary defect and allows the patient to speak and swallow), hygienic (it separates the maxillary surgical site from the contents of the oral cavity), and psychological (restores patient's self-image by reproducing the contours of the lost oral structure).^[11]

In the cases discussed above, the patients presented with typical symptoms of tenderness over the sinuses and orbital region with loss of vision. As the disease is fast spreading, emergency debridement was done. The first case was a post-covid case wherein the spread of the disease in the oral cavity was limited to involve the center of the palate. After 3 weeks, an interim obturator was fabricated for the patient and a reduction in the size of the defect was seen. Regular follow-ups showed considerable improvement in the general health of the patient.

On the other hand, the second case was not covid-positive but presented with uncontrolled diabetes, which lead to mucormycosis.

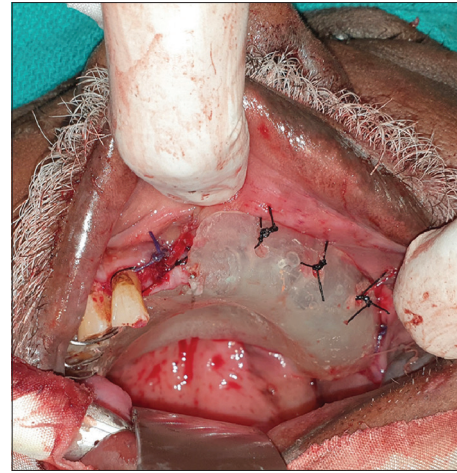


Figure 22: Surgical obturator secured with four interrupted sutures immediately post surgery

He had already lost the vision in his left eye, and the spread in the oral cavity extended to involve partial maxilla of the left side. An aggressive surgery including partial maxillectomy was performed.

Therefore, the presence of coronavirus disease does not ascertain the development of mucormycosis. Early diagnosis, control of the underlying diseases, and aggressive medical and surgical intervention remain the gold standard for managing such cases. Moreover, repeated clinical and radiographic assessments are essential to prevent the recurrence of the infection. Thus, we as doctors must diagnose all these factors and treat the patients to the best of our ability to save them from this devastating disease.

Key Messages

Immediate surgical obturators restore function, hygiene, and psychological state of a patient undergoing surgical resection due to rhinocerebral mucormycosis.

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.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Hibbett DS, Binder M, Bischoff JF, Blackwell M, Cannon PF, Eriksson OE, *et al.* A higher-level phylogenetic classification

- of the Fungi. *Mycol Res* 2007;111:509-47.
2. Swain SK, Sahu MC, Baisakh MR. Mucormycosis of the head and neck. *Apollo Med* 2018;15:6.
 3. Aramany MA. Basic principles of obturator design for partially edentulous patients. Part I: classification. *J Prosthet Dent* 1978;40:554-7.
 4. Spellberg B, Edwards J Jr, Ibrahim A. Novel perspectives on mucormycosis: Pathophysiology, presentation, and management. *Clin Microbiol Rev* 2005;18:556-69.
 5. Ibrahim AS, Spellberg B, Walsh TJ, Kontoyiannis DP. Pathogenesis of mucormycosis. *Clin Infect Dis* 2012;54(Suppl_1):S16-22.
 6. Rammaert B, Lanternier F, Poirée S, Kania R, Lortholary O. Diabetes and mucormycosis: A complex interplay. *Diabetes Metab* 2012;38:193-204.
 7. Price LJ, Stevens LD. Hyperbaric oxygen in the treatment of rhinocerebral mucormycosis. *Laryngoscope* 1980;90:737-47.
 8. Couch L, Theilen F, Mader JT. Rhinocerebral mucormycosis with cerebral extension successfully treated with adjunctive hyperbaric oxygen therapy. *Arch Otolaryngol Head Neck Surg* 1988;114:791-4.
 9. Fisher EW, Toma A, Fisher PH, Cheesman AD. Rhinocerebral mucormycosis: Use of liposomal amphotericin B. *J Laryngol Otol* 1991;105:575-7.
 10. Chakrabarti A, Singh S. Management of mucormycosis. *Curr Fungal Infect Rep* 2020;29:1-3.
 11. Huryn JM, Piro JD. The maxillary immediate surgical obturator prosthesis. *J Prosthet Dent* 1989;61:343-7.