The Journal of Laryngology & Otology

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Subperiosteal inferior maxillectomy in mucormycosis patients: case series

V Wadhwa, D Chaudhary, R Meher, P K Rathore, V Kumar, S Soni and N Kumar

Department of Otorhinolaryngology, Maulana Azad Medical College, New Delhi, India

Abstract

Objective. Mucormycosis is a rapidly progressive and fulminant fungal infection mainly affecting the nose and paranasal sinuses and often requiring aggressive surgical debridement, which commonly includes inferior maxillectomy. Conventional inferior maxillectomy involves removal of the bony hard palate and its mucoperiosteum. This can lead to formation of an oroantral fistula and thereby increase the morbidity in these patients leading to prolonged rehabilitation. Subperiosteal inferior maxillectomy involves sparing of the uninvolved mucoperiosteum of the hard palate. This flap is used for closure of the oroantral fistula, which preserves the functional capabilities of the patient, such as speech, mastication and deglutination.
Method. This case series describes the experience of using the technique of mucosa-preserving subperiosteal inferior maxillectomy in five patients with mucormycosis.
Results. With the technique used in this study, complete oronasal separation was achieved in all six patients. The overall surgery time was also decreased when compared with free tissue transfer. Patients also did not have to bear the weight of prosthesis.
Conclusion. Mucoperiosteal palatal flap-preserving subperiosteal inferior maxillectomy is an excellent approach for all patients with mucormycosis and healthy palatal mucosa.

Clinical Record

Dr D Chaudhary takes responsibility for the integrity of the content of the paper

Cite this article: Wadhwa V, Chaudhary D, Meher R, Rathore PK, Kumar V, Soni S, Kumar N. Subperiosteal inferior maxillectomy in mucormycosis patients: case series. *J Laryngol Otol* 2023;**137**:225–230. https://doi.org/10.1017/S0022215121002875

Accepted: 28 September 2021
First published online: 13 October 2021

Key words:
Imaging; Informed Consent; Swallowing

Author for correspondence:
Dr D Chaudhary, Department of Otorhinolaryngology, Maulana Azad Medical College, Bahadur Shah Zafar Marg, New Delhi 110002, India
E-mail: deepikachaudhary19@gmail.com

Introduction

Mucormycosis is a rare opportunistic fungal infection caused by fungi belonging to Mucorales order and Mucoraceae family. Various clinical presentations include rhino-orbito-cerebral, pulmonary and cutaneous forms, and less frequently, gastrointestinal, disseminated and miscellaneous forms.[1]

Immunocompromised patients are at the greatest risk for mucormycosis. Diabetes mellitus is the most common predisposing factor associated with mucormycosis.[2,3] A state of diabetic ketoacidosis further increases the risk of developing mucormycosis. Persistent hyperglycaemia impairs polymorphonuclear cell chemotaxis and intracellular phagocytosis ability. The acidic environment also leads to reduction in the binding of iron to transferrin, increasing free iron concentration which enhances fungal growth.[4] Other risk factors include patients with haematological malignancies, transplantation, neutropenia, corticosteroid therapy and use of deferoxamine.[2,5,6]

Factors critical for management of mucormycosis include: rapidity of diagnosis, reversal of the underlying predisposing factors, appropriate aggressive surgical debridement of infected tissue and appropriate antifungal therapy. Early diagnosis is important because small, focal lesions can often be surgically excised before they progress to involve critical structures or disseminate.[7]

Applied anatomy of palate and its blood supply

The palate divides the nasal cavity and oral cavity with the hard palate positioned anteriorly and soft palate posteriorly. The palatal mucosa is strongly adhered to the underlying periosteum, which is subsequently attached to the bone via fibrous tissue pegs known as Sharpey's fibres. The hard palate comprises the palatine process of the maxilla and the horizontal palatal lamina of the palatine bones. A longitudinal suture separates the maxilla in the midline; the palatal aponeurosis attaches to the posterior margin of the hard palate and is continuous with the tensor veli palatini laterally.[8]

The major blood supply of the hard palate comes from the greater palatine artery, which emerges from the greater palatine foramen located on the hard palate between the second and third maxillary molars. Further behind, there are the lesser palatine foramina where the branches of the lesser palatine artery emerge. These arteries supply the majority of the hard palate together with the soft palate. The branches of the greater palatine artery travel within the palatal bony groove divided into medial and lateral palatine grooves by the palatine spine. The medial palatine groove contains the greater palatine nerve, whereas the greater palatine artery lies in the lateral groove to supply the mucosa, periosteum and palatal gingiva before entering the incisive foramen to form an anastomosis with the nasopalatine artery. The nasopalatine artery enters the incisive canal to supply the anterior region of the hard palate.[9]

Table 1. Case series

Parameter	Case 1 (Figure 1)	Case 2 (Figure 2)	Case 3 (Figure 3)	Case 4 (Figure 4)	Case 5 (Figure 5)	Case 6 (Figure 6)
Gender	Male	Male	Female	Male	Male	Male
Co-morbidities	Type 2 diabetes mellitus + hypertension	Type 2 diabetes mellitus	Type 2 diabetes mellitus	Type 2 diabetes mellitus	Type 2 diabetes mellitus	Type 2 diabetes mellitus
History of Covid-19	Yes	Yes	Fever: yes. Not tested for Covid-19	Yes	Yes	Fever: yes. Not tested for Covid-19
Contrast-enhanced computed tomography of paranasal sinuses & orbit	Post-operative changes along with soft tissue content in the right maxillary sinus, soft tissue content in intraconal & extraconal compartment of right orbit & erosion of the hard palate	Mucosal thickening in left ethmoidal air cells, left maxillary sinus & left nasal cavity, & ill-defined heterogenous soft tissue in the left pterygopalatine fossa with erosion of medial wall of left maxillary sinus & left alveolar arch	Soft tissue content in the left maxillary sinus, soft tissue infiltration of left periantral fat planes & left pterygopalatine fossa, & multiple erosions involving the roof, the floor, anterior wall, medial wall & posterolateral wall of left maxillary sinus, superior alveolar arch & hard palate	Focal areas of bony erosions seen in anterior, medial & posterolateral walls of right maxillary sinus, right pterygoid process, hard palate & superior alveolar arch on right side	Soft tissue content in the left maxillary sinus & left pterygopalatine fossa along with focal erosions in all walls of left maxillary sinus & hard palate on the left side	Heterogeneously enhancing soft tissue infiltration in left maxillary sinus, pterygopalatine fossa & postantral region. Focal bony erosions seen in posterolateral wall of left maxillary sinus & left side of hard palate
Medical management	Injection of liposomal amphotericin B: 5 g	Injection of liposomal amphotericin B: 5 g	Injection of liposomal amphotericin B: 5 g	Injection of liposomal amphotericin B: 5 g	Injection of liposomal amphotericin B: 5 g	Injection of liposomal amphotericin B: 5 g
Surgical intervention	Left subperiosteal inferior maxillectomy with orbital exenteration	Left partial maxillectomy (subperiosteal)	Left subperiosteal inferior maxillectomy	1: Right functional endoscopic sinus surgery. 2. Right partial maxillectomy (subperiosteal)	1: Left partial maxillectomy via Weber Ferguson approach. 2: Left subperiosteal inferior maxillectomy	Left subperiosteal inferior maxillectomy
Post-operative speech rehabilitation	Yes	Yes	Yes	Yes	Yes	Yes
Post-operative swallowing difficulty	No	No	No	No	No	No

Covid-19 = coronavirus disease 2019

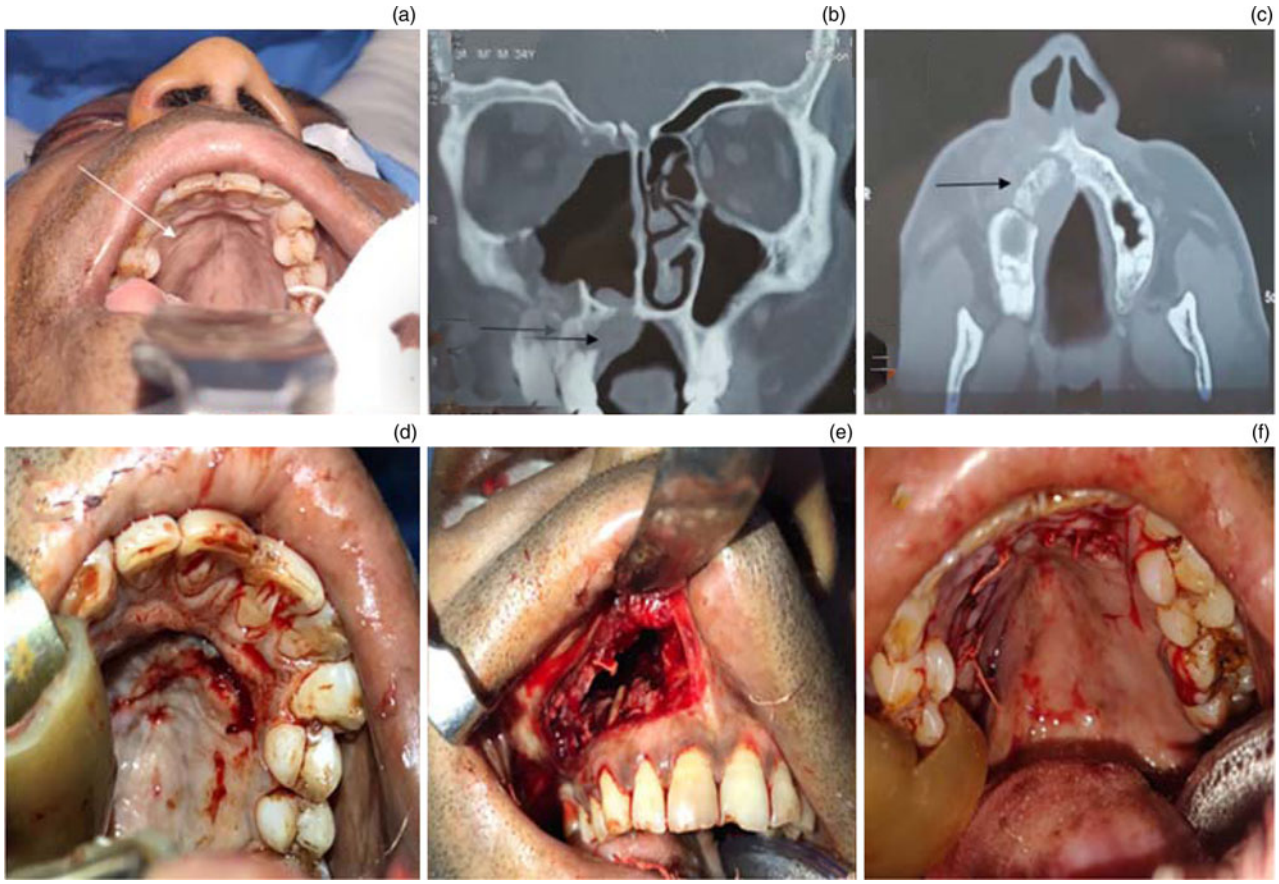

Fig. 1. Case 1. (a) Pre-operative image. White arrow indicating palatal bulge. (b) Coronal plane contrast-enhanced computed tomography image of the nose, paranasal sinuses and orbit. Black arrow indicates right hard palate erosion. (c) Axial plane contrast-enhanced computed tomography image of the nose, paranasal sinuses and orbit. Black arrow indicates right hard palate erosion. (d) Intra-operative image showing palatal incision. (e) Intra-operative image showing gingivolabial flap. (e) Intra-operative image after closure.

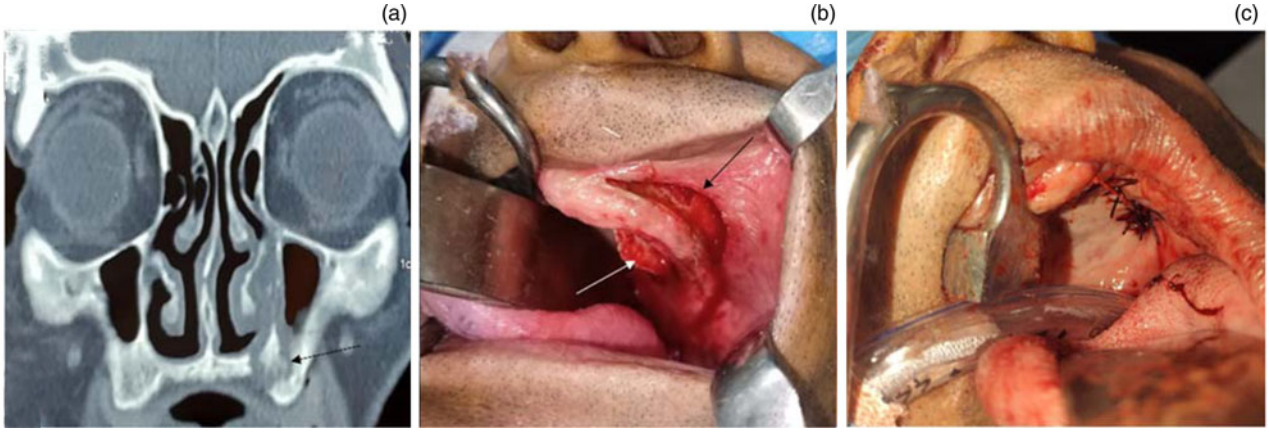

Fig. 2. Case 2. (a) Coronal plane contrast-enhanced computed tomography of the nose, paranasal sinuses and orbit. Black dotted arrow indicates left-sided hard palate erosion. (b) Intra-operative image (edentulous patient). White arrow indicating palatal flap. Black arrow indicating gingivolabial flap. (c) Intra-operative image after closure.

The main arterial supply of the soft palate is the ascending palatine artery, which most commonly arises from the facial artery but can occasionally arise from the external carotid, the ascending pharyngeal or the maxillary artery.[10] The ascending palatine artery runs inferomedially from the lateral pharyngeal space into the palate, dividing into the anterior and posterior branches.

Knowledge of the blood supply of the palate and the course of the palatal vasculature is important in oral surgical procedures to aid in flap design and may enable clinicians to avoid intra- and post-operative complications when planning oral surgery interventions by optimised incision and flap designs.

Case series

Sixteen patients with mucormycosis who were admitted to the ENT Department of Lok Nayak Hospital, Delhi, India, underwent inferior maxillectomy; of these patients, 6 underwent mucosa-preserving subperiosteal inferior maxillectomy after

https://doi.org/10.1017/S0022215121002875 Published online by Cambridge University Press

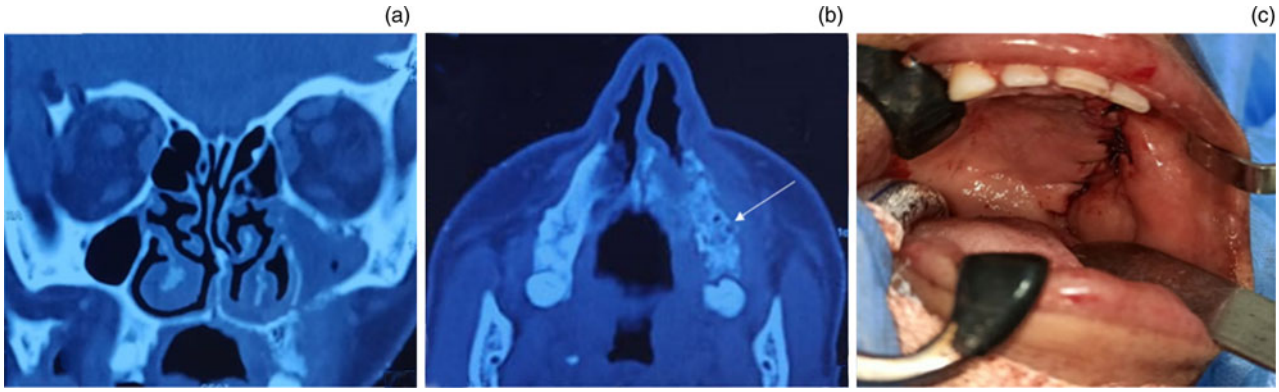

Fig. 3. Case 3. Contrast-enhanced computed tomography of the nose, paranasal sinuses and orbit in the (a) coronal and (b) axial view. White arrow indicates left-sided hard palate erosion. (c) Intra-operative image after closure.

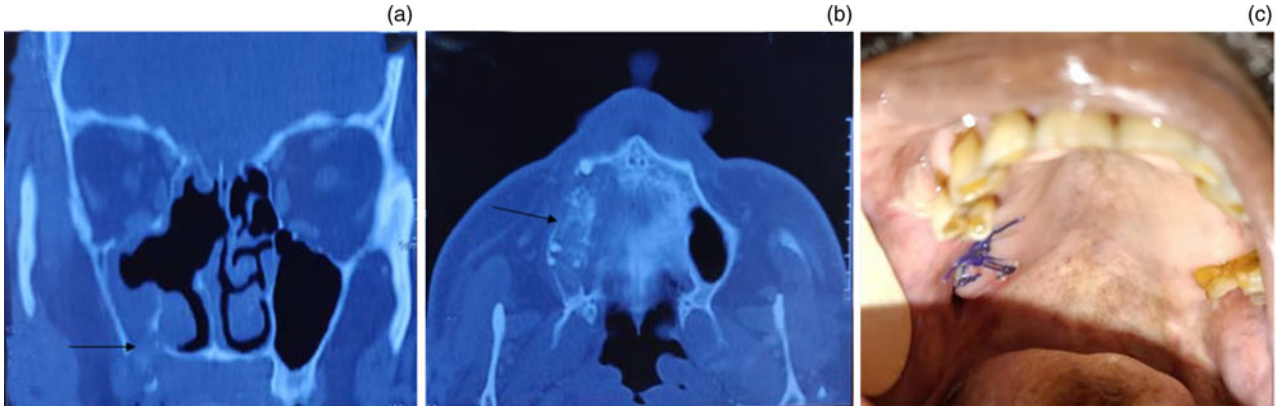

Fig. 4. Case 4. Contrast-enhanced computed tomography of the nose, paranasal sinuses and orbit in the (a) coronal and (b) axial view. Black arrow indicates right-sided hard palate erosion. (c) Post-operative image.

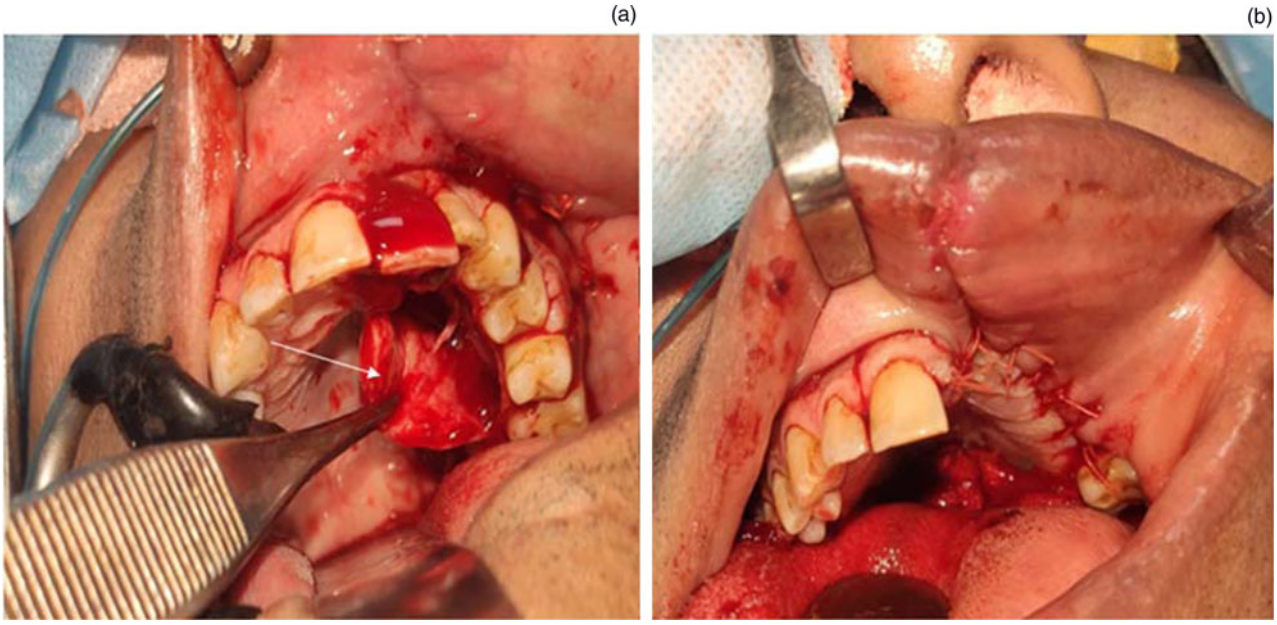

Fig. 5. Case 5. (a) Intra-operative image. White arrow indicates palatal flap. (b) Intra-operative image after closure.

informed consent. The palatal mucosa was clinically normal in all patients, who all had a positive prick test (i.e. healthy blood supply to the palate). After thorough history and clinical examination, all patients were assessed by radiological imaging using contrast-enhanced computed tomography (CT) of the nose, paranasal sinuses and orbit (Table 1).

Surgical technique

Patients underwent debridement using subperiosteal mucosa-preserving inferior maxillectomy under general anaesthesia. Palatal incision was performed 5–6 mm away from the alveolar margin, and the mucoperiosteal flap was elevated. In cases of

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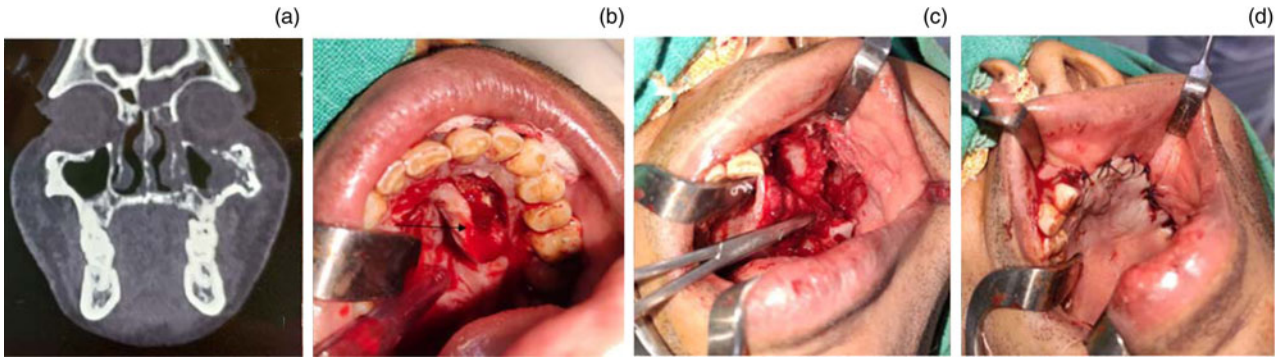

Fig. 6. Case 6. (a) Coronal plane contrast-enhanced computed tomography of the nose, paranasal sinuses and orbit. (b) Intra-operative image. Black arrow indicates raised palatal flap. (c) Intra-operative image. (d) Intra-operative image after closure.

larger defects, the palatal flap was raised until the junction of the hard and soft palate. The ipsilateral greater palatine artery was preserved. A separate sublabial incision was given and a gingivolabial flap was raised, which was subsequently made continuous with cheek flap. The diseased tissue of the underlying hard palate was removed. Removal of hard palate bone provided access to the maxillary antrum, pterygopalatine fossa and infratemporal fossa, which was cleared of the disease if found. After adequate disease clearance and haemostasis, the palatal mucoperiosteal flap was reposited and sutured with upper gingiva-labial flap using 3-0 vicryl sutures.

Discussion

Mucormycosis often involves the hard palate, requiring debridement in the form of inferior maxillectomy. Loss of hard palate in inferior maxillectomy leads to significant morbidity to the patient in terms of speech and deglutition problems. Subperiosteal inferior maxillectomy is a modification of conventional inferior maxillectomy in which the uninvolved mucoperiosteum of the hard palate is preserved with the ipsilateral greater palatine artery. This flap is used for reconstruction of the oroantral or oronasal defect. This technique can be adopted in patients wherein the palatal mucosa is normal and not ulcerated with a good blood supply, which can be confirmed by bleeding margins of the mucosal incision and a prick test (looking for brisk bleeding on pricking).

The preservation of the palatal mucoperiosteal flap is to be considered in all mucormycosis patients, in contrast to patients with malignancy of the maxilla, where adequate oncological margins are important. In all, 16 patients with mucormycosis who had associated palatal involvement were admitted to the ENT Department of Lok Nayak Hospital and subsequently underwent inferior maxillectomy. Among them, 6 patients were taken up for mucosa-preserving subperiosteal inferior maxillectomy and were included in our case series. Other patients had extensive palatal bony and mucosal involvement; therefore, palatal flaps were not available for reconstruction of oroantral defects.

In 2000, Brown *et al.* classified the defect following maxillectomy into vertical and horizontal components. The vertical defect, which involves resection in the vertical plane from dentition to the skull base, is classified into class 1–4. A letter (a, b or c) is added depending on how much of the upper alveolus has been removed, which is considered to be the horizontal component of the defect (Tables 2 and 3).[11]

The maxilla bears the maxillary teeth, transmits masticatory forces and provides a partition between the oral and nasal cavities and maxillary sinuses. It therefore assists in critical functions of mastication, speech and deglutition.[12] Loss of the maxilla is associated with significant functional morbidity. Palatal defects in isolation lead to formation of oroantral fistula, which results in difficulty in swallowing, speech and also creates cosmetic impairment and psychological consequences for patients.

- Mucormycosis is a fulminant disease of the nose and paranasal sinuses requiring aggressive surgical debridement
- Patients with sinonasal mucormycosis with palate involvement often require inferior maxillectomy
- Subperiosteal inferior maxillectomy is a modification of the conventional inferior maxillectomy wherein the healthy palate mucosa is preserved
- Oronasal separation is achieved, and speech and swallowing functions are preserved using this technique
- Mucoperiosteal palatal flap-preserving subperiosteal inferior maxillectomy has better functional and aesthetic outcomes than conventional inferior maxillectomy

Reconstruction of the maxillary is a significant challenge because the three-dimensional midface architecture has both functional and aesthetic functions. It requires the presence of a healed wound, separation of oral and nasal cavities, restoration of maxillary buttresses, restoration of functional dentition, mastication and deglutition, maintenance of a patent nasal airway, support and suspension of adynamic facial soft tissue and the restoration of an adequate and symmetrical facial form on the contralateral side, and psychological well-being.[13] Various techniques have traditionally been used for

Table 2. Classification of vertical defect

Class	Description
1	Maxillectomy without an oroantral fistula
2	Low maxillectomy (not including orbital floor or contents)
3	High maxillectomy (involving orbital contents)
4	Radical maxillectomy (including orbital exenteration)

Table 3. Classification of horizontal defect

Horizontal defect	Description
A	Unilateral alveolar maxillectomy
B	Bilateral alveolar maxillectomy
C	Total alveolar maxillary resection

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the reconstruction of inferior maxillectomy defects, including palatal prosthesis, local flaps and microvascular tissue transfers. The choice of a specific surgical procedure mainly depends upon the site and size of the defect present and the amount and location of palatal tissues available for repair.

Various studies depicting the role of obturators and local and free flaps for maxillary reconstruction following inferior maxillectomy are described in the literature, some of which are discussed briefly here. Choung *et al.*[14] described the use of ipsilateral or bilateral temporalis muscle with or without a segment of attached calvarial bone. This flap could be passed into the oral cavity by performing anterior and posterior osteotomy procedures on the zygomatic arch and mobilising it to its attachment on the coronoid process. Alternatively, Hatoko *et al.*[15] reported success in reconstruction of inferior maxillectomy procedures with a fasciocutaneous radial forearm free tissue transfer in three patients who had not tolerated obturators well. Futran *et al.*[16] reported the use of the fibula free flap in 27 patients with defects, including defects of the palate that were not amenable to the use of a conventional prosthesis. All these above studies looked at malignancy of maxilla where oncological margins are to be carefully considered. On the other hand, surgical debridement for mucormycosis is aggressive only for involved areas.

In our technique of subperiosteal inferior maxillectomy, we raised a subperiosteal palatal flap along with a superior gingivolabial flap. The diseased tissue of the antrum and the palatal bone was removed, and the flaps were sutured. In cases of larger defects, the subperiosteal flaps were raised up to the hard and soft palate junction, without damaging the greater palatine artery. The complete oronasal separation was achieved in all six patients, and this resulted in better aesthetic and functional outcomes. The overall surgery time was also decreased when compared with free tissue transfer. Also, the patient does not have to bear the weight of prosthesis. The technique allowed excellent palatal function and prompt rehabilitation allowing better patient outcomes without compromising disease removal.

Conclusion

Mucoperiosteal palatal flap-preserving subperiosteal inferior maxillectomy is a desirable approach for all patients with mucormycosis and healthy palatal mucosa, leading to better functional and aesthetic outcomes for the patient.

Acknowledgements. The authors would like to thank all the faculty and staff of the Department of Otorhinolaryngology at Maulana Azad Medical College and Associated Lok Nayak Hospital for their generous help and support.

Competing interests. None declared

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