Rehabilitation of Acquired Maxillary and Mandibular Defects Secondary to Mucormycosis - A Case Series

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Abstract

Rationale: To suggest a directing algorithm for rehabilitative management of complex soft and hard tissue defects due to mucormycosis. **Patient Concerns:** An utmost need for surgical, reconstructive and rehabilitative options; functional, aesthetic, acceptable to the patient and enduring rehabilitation. **Diagnosis:** Different cases of rhinomaxillary mucormycosis pertaining to the facial skeleton, diagnosed with the help of radiographic as well as fungal cultures. **Treatment:** Thorough medical assessment, and antifungal prescription; followed by surgical resection of the affected areas, rigorous follow-up scheme and later rehabilitation with the help of implants and immediate loading. **Outcomes:** Regular follow-ups for at least one year and successful rehabilitation providing acceptable functional outcomes. **Take-away Lessons:** Unconventional pathologies like rhinomaxillary mucormycosis infecting the jaws require novel, unprecedented and elaborate procedures both surgical and reconstructive based on sound scientific principles. There must be a vision for the rehabilitation of such cases right from the commencement of the surgical treatment.

Keywords: Rhinocerebral mucormycosis, zygomatic implant, pterygoid implant, patient specific implant, obturator

Introduction

The most common type of invasive fungal infection is rhino-orbital-cerebral mucormycosis (ROCM) with morbidity reaching 85% even with quickly implemented treatment. Mucormycosis of the mandible is very rare occurrence with only 23 cases reported worldwide in the last 50 years.[1-3] In patients with ROCM, the poor penetration of antifungal agents into the infected area has been reported; thus, sinus surgery is essential, but its extent varies from limited to radical resection. Lack of support, retention and stability are common prosthodontic treatment problems for patients who have had a maxillectomy.[4,5] Surgical placement of implants is also challenging because of the lack of available bone. Therefore, implant placement into buttress sites such as zygoma has been advocated. Very limited literature has discussed the use of pterygoid/pterygomaxillary implants in patients undergoing maxillectomy.

Case Series

Case 1

A 26-year-old male patient had mucormycosis involving maxilla; surgical treatment for the same involved removal of left lateral nasal wall and parts of anterior maxilla, bone below the left zygomatic buttress extending from left lateral nasal wall up to the left-sided pterygoid plates laterally [Figure 1]. He was given posaconazole for one month after amphotericin B therapy, in consultation with an infectious diseases' specialist. Intravenous amphotericin B (lipid formulation) is the drug of choice for initial therapy. After soft tissue healing, an interim removable prosthesis was fabricated.

Contrast-enhanced computed tomography scans and virtual surgical planning were done after six months. The volume of bone and presence of anterior teeth

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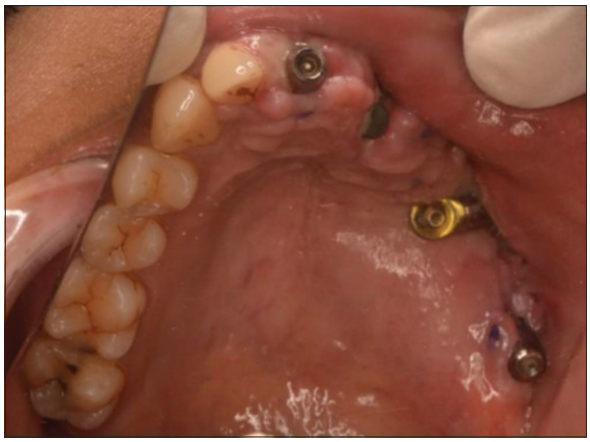

Figure 1: Post-operative intraoral image

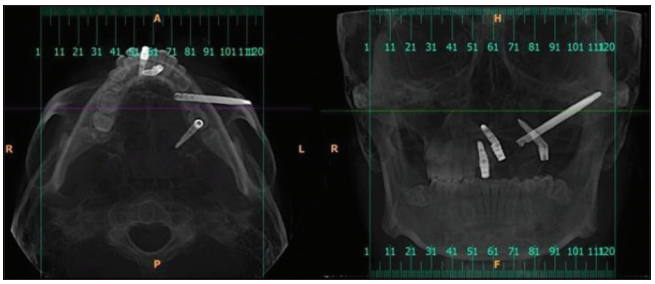

Figure 2: Post-operative computed tomography scans showing a conventional endosseous implant, a tilted implant, a zygomatic implant and a pterygoid implant are placed

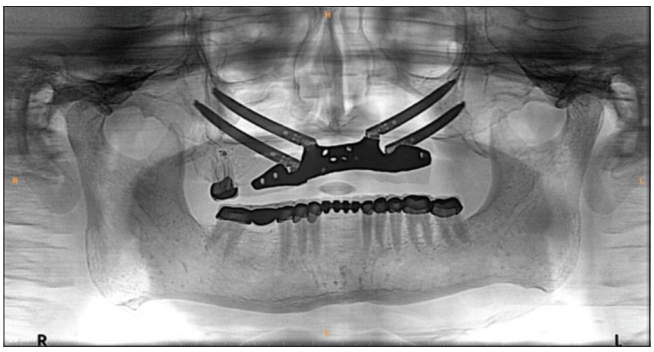

Figure 3: The post-operative orthopantomogram showing the zygomatic implants

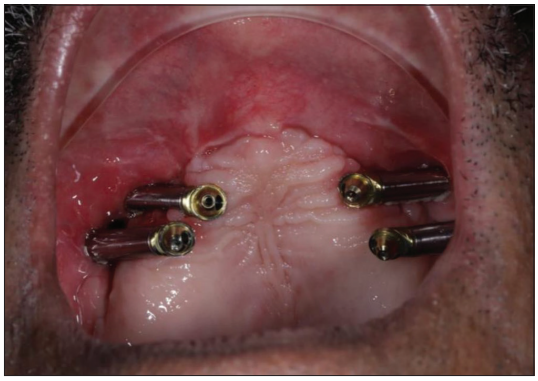

Figure 4: The clinical intraoral image of the four zygomatic implants with healthy mucosa

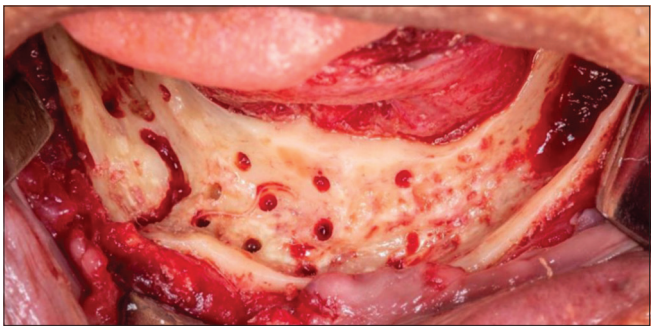

Figure 5: Intraoral clinical image of the residual cavity and healthy bone and the holes drilled into it to promote angiogenesis

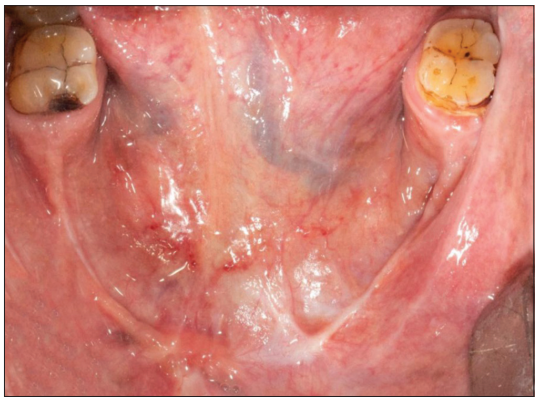

Figure 6: Intraoral clinical image of the healed mandibular arch and ready for rehabilitation

on the contralateral side prevented the placement of a patient-specific implant (PSI) without removal of the sound teeth. Osstem endosseous conventional implants, Norris's pterygoid (4.2 mm × 20 mm) and zygoma implants (4.2 mm × 37.5 mm) were placed [Figure 2]. Immediate prosthetic rehabilitation with a hybrid acrylic metal prosthesis was developed as soon as the soft tissue healed. The non-functional prosthesis was kept for another five months, after which a final fully functional computer-aided design/computer-aided manufactured (CAD-CAM) prosthesis was delivered. The patient has been followed up for one year since without any complaints.

Case 2

Another case was of a 62-year-old male patient with bilateral maxillary defect post-mucormycosis was rehabilitated with four zygomatic implants, two on each side, i.e. quad zygoma as there was no native maxillary bone available for conventional implants and prosthesis [Figure 3 and 4].

Case 3

This case was a unique one of a 52-year-old male patient of mucormycosis of the mandible. Initially, it was felt that the defect would be small, but there was a lot of affected bone, which was porous, leading to a huge central defect

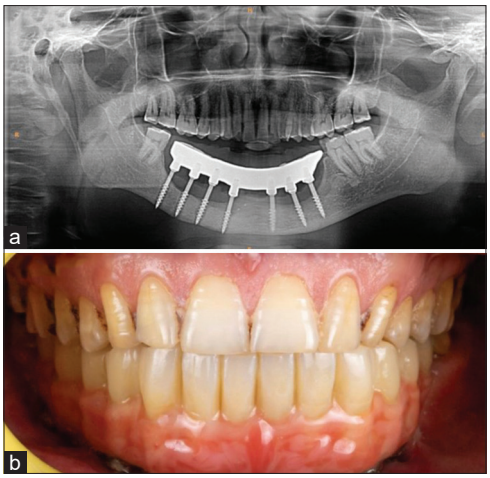

Figure 7: (a) Orthopantomogram showing the rehabilitated mandibular arch using seven basal implants and successfully mounted prosthesis. (b) Intraoral image showing the occlusion of the final prosthesis

of the mandible. The lower border was well preserved, and platelet-rich fibrin was placed *in situ* to accelerate the healing [Figures 5-7].

Discussion and Literature Review

Posaconazole or isavuconazole is used as a step-down therapy for patients who have responded to amphotericin B. They can also be used as salvage therapy for patients who are non-responders or cannot tolerate amphotericin B, for salvage therapy. When switching to oral posaconazole, we favour the use of delayed-release tablets (300 mg 12 hourly on the first day, then 300 mg once daily) taken with food if possible. A serum trough concentration of posaconazole should be checked after one week of therapy; we suggest a goal trough concentration >1 mcg/mL, but higher levels are preferred for the treatment of this serious infection.[5]

When providing bicortical implants, a few rules need to be respected. As these implants have smooth surfaces and rely on osseo-fixation, the main goal is to properly anchor them in cortical bone.[6,7] The implant placement used the available distant bone, but the definitive abutments should be carefully placed to achieve the so-called supporting polygon, which may be executed by bending the implant head. Such polygon is created in the maxilla by abutment of the implant in the canine region and the most distant implant usually in the tuberosity region.

Obturator was perhaps the only available resource for rehabilitating these defects previously. The merits of obturators are easily made, light weight and no surgery required with demerits being the need of bony engagement or undercuts.

The advent of zygomatic implants brought about a new era in rehabilitation of cases with complex defects. Merits: time tested, durable, excellent solution to provide teeth with aesthetics and function. Demerits: in unilateral defects, placement might be

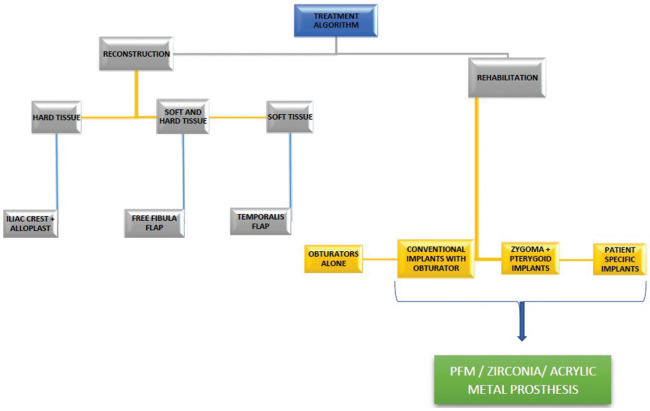

Figure 8: The treatment algorithm suggested by the authors

an issue owing to the presence of teeth on the opposite side due to which the angle of placement would be compromised. Furthermore, the healthy anterior teeth might need to be sacrificed to give cross arch anchorage while placing zygoma implants in unilateral defects. In such cases of unilateral defects, PSI can be preferred over the zygomas. In a review of English-language scientific journals,[8] 32 studies presenting clinical outcomes with a zygomatic implant were found. The publications included 1031 patients and 2131 zygomatic implants with a follow-up period of six months to 12 years. In total, 42 implants were reported as failures, giving an overall survival rate of 98.1%. The zygomatic implant technique is highly predictable and results in good clinical outcomes.

The pterygoid implant technique is associated with less overall morbidity, lower treatment costs and shorter healing times. From a prosthetic point of view, dental rehabilitation with pterygoid implants has the advantage of eliminating long distal cantilevers, due to the emergence of pterygoid implants in the second molar region. Although cleaning of the pterygoid implants prostheses that emerge in the posterior region of the maxilla may be a concern for both patients and professionals, this factor was not reported in any of the included studies. Curi and Penarrocha also reported a high degree of satisfaction of patients related to the final prosthesis rehabilitation.[9] The treated cases had intact pterygoids and with proper virtual surgical planning, engaging the same lead to a better stability for the prosthesis along with zygoma implants (Case 1). Cases having mutilated pterygoids necessitated the use of quad zygomas (Case 2).

PSIs were used for the first time in the 1940s.[5] They are readily printable and have the advantage of easy sterilization, antimicrobial properties, and regenerative capability with modifications provide for very exciting possibilities.[10] The drawbacks if any were the high cost to fabricate a polished implant and associated prosthesis. The lack of evidence with regard to long-term implant survival is another drawback. There are also reported incidences of implant exposure/ infection where the implants had to be removed; even 4-6 months after complete healing post-surgery. Lack of bony support and soft tissue paucity make them vulnerable to exposure and consequent infection.

Conclusion

Countering such a deadly fungal infection, of the rhino-orbito-maxillary and mandibular region which is highly important requires thorough knowledge of the pathogenesis, comorbid conditions, pharmacology of necessary drugs, surgical and endoscopic principles to effectively limit the spread and conserve as much tissue tissue as possible; followed by strictly regular follow-ups and then rehabilitating with functional, aesthetic and acceptable option in the best form possible. Henceforth, comprehensive planning and execution in accordance with sound scientific principles can lead to more predictable, durable, functional and aesthetically successful solutions. The authors have suggested an algorithm for such cases for ease in selection of treatment [Figure 8].

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.

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Conflicts of interest

There are no conflicts of interest.

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