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# A successfully rehabilitated post-mucormycosis maxillary defect with guided quad-zygomatic implants: A case report

Akash Chakravarthy, Dinesh Sharma, Geyasri Vinnakota, Racheeta Ramanujam, Sulagna Chakrabarti, Deepika Reddy

## ABSTRACT

**Introduction:** Mucormycosis infection has received occasional attention because of the low number of cases in comparison with other frequent infections. With the emergence of the SARS-CoV-2 disease, the incidence of fungal infections like mucormycosis has increased. In mucormycosis, after surgical debridement, the defects that result are different from other maxillary defects. In terms of functional and aesthetic rehabilitation there are several treatment options for patients with maxillectomy defects. Amidst all, zygomatic implants are more favorable and viable solution for the above-mentioned defects. Since there is a scarcity in literature on the use of zygomatic implants in such cases, we presented a case report using the utmost advantage of these implants in rehabilitation of maxillary defects.

**Case Report:** In the present case report, a 38-year-old male patient presented with an absence of the maxillary alveolar bone and anterior nasal spine and minimal presence of hard palate. Such type of maxillary defect following mucormycosis was managed with quad zygomatic implants with computer-guided approach.

**Conclusion:** Thus, the author concluded through this case report that a severe maxillectomy defects can be managed well with the zygomatic implants. In the present article, the author preferred to perform under guide in order to avoid errors while placement of implants. Hence, proper diagnosis with definitive surgical approach can help in achieving precise treatment outcome and establishing patient's self-esteem.

**Keywords:** Maxillary rehabilitation, Maxillectomy defects, Mucormycosis, Post-mucormycosis maxillary defect, Quad zygoma, Zygomatic implant guided surgery

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## INTRODUCTION

Mucormycosis is a rapidly progressive, emerging fungal and life-threatening infection. In 1885, Paltauf was the first to report a case of mucormycosis and described it as mycosis mucorina [1]. The first effort to analyze all the available literature was made by Roden et al. in 2005 [2]. The incidence of mucormycosis has been increasing in recent decades, mainly due to the growth of the

number of severely immunocompromised patients [2, 3]. Mucormycosis, also known as black fungus, caused chaos in India during the calamitous COVID-19 epidemic's second wave (between April and June 2021) by a rapid and deadly surge with up to a 50% fatality rate. While the concrete reason for its sharp rise during the second wave is still unknown, it has been identified that diabetics and immunocompromised patients who had recovered from COVID-19 infection are more susceptible to mucormycosis [4]. Even patients who had a well-controlled diabetes with no underlying immunosuppressant risk factors are still at risk for mucormycosis infection [5].

Medical professionals were also facing challenges in the early diagnosis and treatment of this aggressive fungal infection during the COVID-19 pandemic. The degree of morbidity in patients surviving mucormycosis depends principally on how quickly the disease is diagnosed and treated which is challenging. Studies have shown that it increases survival [6], and it may also reduce the need for extent of surgical resection, disfigurement, and suffering [7].

Smith and Krichner criteria of 1950 are considered the gold standard for clinical diagnosis of mucormycosis [8]:

i. Black, necrotic appearance of turbinates that is easily misinterpreted as dried, crusted blood;
ii. Blood-tinged nasal discharge and facial pain on the affected side;
iii. Discolored and indurated periorbital or perinasal swelling;
iv. Complete ophthalmoplegia along with proptosis of the eyeball and ptosis of the eyelid; and
v. Multiple cranial nerve palsies unrelated to documented lesions.

Early and broad surgical treatment is recommended for mucormycosis whenever possible, in addition to antifungal medications and correction of underlying predisposing factors [9]. Considering the cumulative number of cases and the principal involvement of the orofacial region during the current COVID-19 pandemic, we can expect to come across more patients with orofacial defects after surgery. Thus, there is an immediate need to provide maxillofacial prosthetic rehabilitation for patients with mucormycosis to improve their quality of life. Successful management of mucormycosis is based on a multidisciplinary approach, including early administration of active antifungal agents at the optimal dose, complete removal of all infected tissues and the use of various adjunctive therapies [10]. Rapid correction of metabolic abnormalities is mandatory in patients with uncontrolled diabetes and suspected of mucormycosis [11]. If required and possible, surgery must be aggressive. Not only necrotic tissues but also surrounding infected healthy-looking tissues should be removed, as the speed of the extension of the infection by the Mucorales hyphae is massive [11]. Wide en bloc resection often leads to communication between oroantral and oronasal compartments, which results in difficulty in mastication, speech, deglutition, articulation, and breathing. With such extensive surgeries, an implant-supported prosthesis is essential, but the most common problem is a lack of the significant bone necessary for anchorage. In such situations, distant bone structures like zygomatic bone or the palato-pterygoid fusion zone can be leveraged. On account of distance and postresection voids, the surface of such implants will not be fully embedded in hard tissue [12].

Zygoma implant reconstruction of extensive maxillary defects are a safe, predictable, and cost-effective treatment modality. Implant-supported prosthesis are necessary for loss of soft and hard tissue which often provides support to oral musculature and restore function [13]. For such a prosthesis, zygomatic and pterygoid implants for the rehabilitation of maxillary defects have been reported to be more successful than conventional implants [14, 15]. In this case report we aimed to exhibit how post-mucormycosis case with extensive bone resections could be managed with zygomatic implants.

## CASE REPORT

A 38-year-old male patient visited our clinic with a chief complaint of missing teeth, difficulty in speech, and mastication. On examination, intraorally an edentulous maxillary defect was observed. Past history revealed that the patient was tested COVID-19 positive about one year ago for which he was treated with medications by his family physician. Post-COVID he noticed pain, numbness, swelling for which appropriate treatment has done. He had received surgical bilateral subtotal maxillectomy along with wide local excision of area infected by mucormycosis, due to which extensive edentulous maxillary defect was detected (Figure 1).

The extraoral and intraoral examination revealed the absence of the maxillary alveolar bone and anterior nasal spine with the nasal floor, minimal presence of hard palate. His corners of mouth found drooped, and he had insufficient upper lip support (Figure 2). The lateral walls of the defect were lined with nonkeratinized mucosa along with evidence of scared tissues. Absence of clinical suppuration or swelling was noted. In the mandible, healthy teeth were present. His tongue function was normal, and pseudo-class III intermaxillary jaw relation was observed because of the maxillary defect and absence of the teeth. Radiographically, the pterygoid, zygomatic, and pyriform bones had been well-maintained bilaterally (Figure 3). The aim of the reconstruction was to restore functionally and aesthetically by re-establishing facial profile with implant supported prosthesis. The prosthesis would share the support and stabilization and while retention would be entirely dependent on the implants. But the reference for placing implants surgically is difficult due to the massive defects seen in mucormycosis cases, thus guide can lend a hand for error-free placement of implants.

Computed tomography (CT) scan was suggested with slice thickness less than 0.6 mm in all 3 axes. This improves the resolution of the image which helps in better diagnosis and planning. Implant planning was done in blue sky bio software in 3D segmentations (Figure 4). Arbitrary teeth were placed and implant positions were determined while planning. Guide for placing the zygomatic implants is fabricated by using direct metal laser sintering (DMLS) 3D printing. Virtual 3D stereolithographic models were then printed for mock surgery. The 3D metal guide is used to perform a mock surgery on the 3D printed model to check for any errors in planning. This metal guide simplifies zygomatic implant placement during surgery, minimizing angulation and position mistakes. The objectives of using the guide are: (a) Surgical accuracy, (b) Prosthetically driven implants, and (c) Complications free, i.e., to avoid implant failure.

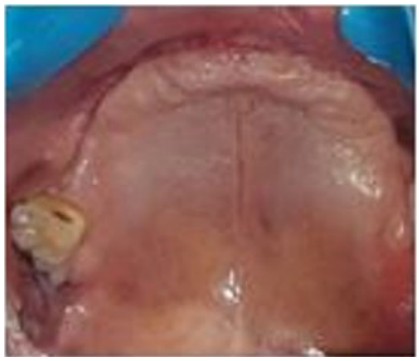

Figure 1: Intra oral pre-operative picture.

Figure 2: Pre-operative smile.

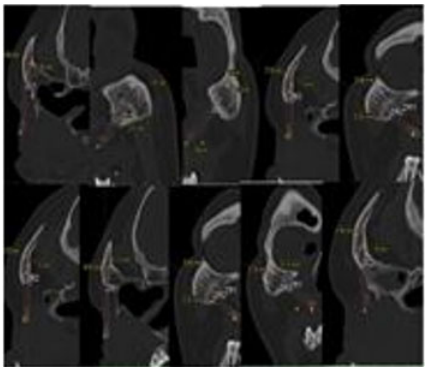

Figure 3: Pre-operative CT scan.

The surgical procedure was conducted under general anesthesia, incision given, full thickness flap reflected till the origin of the zygomatic arch, where the masseter muscle tendon is inserted (Figures 5 and 6). After the mucoperiosteal flap was reflected, guide was secured at the place with small titanium screws (Figure 7). The implant site preparation was performed with zygomatic implant kit drills mounted on a contra-angled handpiece (Figure 8). Healthy bone was seen on the drills after osteotomy site preparation (Figure 9). After osteotomy sites were prepared (Figures 10 and 11), zygomatic implants of size 4.2×57.5 mm in relation to zygoma right 1(ZR1), 4.2×45 mm in relation to ZR2, 4.2×55 mm in relation to ZL1, 4.2×40 mm in relation to ZL2 were placed (Figure 12). Multi-unit abutments were placed aligning with the teeth present in the mandible to achieve a supporting polygon and to avoid any offset loading as a result of a cantilever (Figure 13). Healing caps were placed (Figure 14). Flaps were approximated with 3-0 vicryl sutures placed. After patient recovered from general anesthesia, post-operative orthopantomogram (OPG) has taken followed by digital impressions which were recorded with ICAM scanner. Milled Titanium Bar supported prosthesis with PMMA supra-structure was delivered to provide rigid splint to the implants immediately (Figures 15–18). Post-operative instructions and medications prescribed and also instructed patient how to clean the prosthesis. The patient is evaluated at the follow-up visits (1 week, 4 weeks, 3 months, 6 months, and 12 months) for the periodic maintenance. One year follow-up examination reveals implants were stable and prostheses functioned as envisioned (Figure 19).

Figure 4: Pre-operative planning.

Figure 5: Flap elevated first quadrant.

Figure 6: Flap elevated second quadrant.

Figure 7: Guide placed.

Figure 8: Preparing osteotomy site.

Figure 9: Healthy bone seen.

Figure 10: Osteotomy sites prepared (first quadrant).

Figure 11: Osteotomy sites prepared (second quadrant).

Figure 12: Zygomatic implants placed.

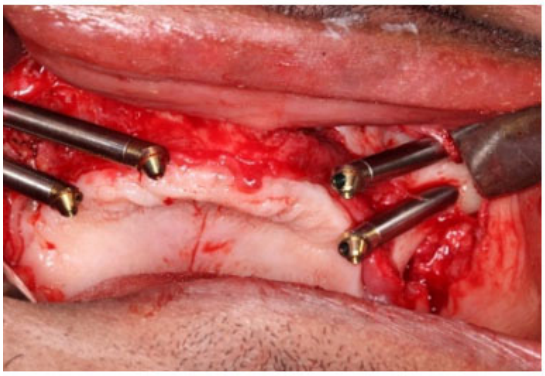

Figure 13: Multi-unit abutments placed.

Figure 14: Healing caps placed.

Figure 15: Intra-oral image of prosthesis occlusal view.

Figure 16: Post-operative image.

Figure 17: Post-operative smile.

Figure 18: Post-operative OPG.

Figure 19: 12-months follow-up OPG.

## DISCUSSION

Maxillary defects with oronasal and oroantral communications can therefore cause masticatory problems, hypernasal speech, fluid leakage, and cosmetic concerns that can impair the normal form and function of stomatognathic system, thus affecting the patients' quality of life [16]. The adaptive capabilities of the patients are highly compromised, thus, prosthetic rehabilitation also plays a significant challenge in such cases. The purpose of the present case report was to present the possibilities of implant treatment in a case of severe hard and soft tissue deficiencies, for the long-term successful prosthetic rehabilitation and survival.

Multiple reconstructive options are available for reconstruction of maxillofacial defects. Various factors defining the selection of reconstruction of defect are dimensions of the defect, presence of infra-orbital rim, extent of the palatal involvement, underlying skin, and aesthetic considerations of patients [17]. In such cases, reconstruction of palatal and orbital floor can be done following overdentures or fixed framework with attachment for removable component (obturator/palatal plate) can be placed. In the present case patient is having intact palatal and orbital floor with no oroantral communications, so, further surgical approaches are not required. When considering the different methods for maxillary reconstruction, bone grafting methods, obturators with or without dental implants, zygomatic implants can be a solution to the lack of maxillary

bony support. With the introduction of the zygoma implant, prosthesis retention has significantly enhanced, translating into better stability overall [18, 19]. In the present case, maxillary defect was reconstructed using zygomatic implants which are rehabilitated with titanium bar-supported prosthesis. A study by Schmidt et al. concluded that combination of zygomatic and standard endosseous implants can be used to reliably reconstruct patients after extensive resection of the maxilla which is in accordance with our case report in which zygomatic implants alone are used to reconstruct the maxillary defect [20].

The best prosthetic option to replace composite defects in which teeth, gingiva, alveolar bone, and maxillary bone are missing is by using a fixed-hybrid or a bar-retained prosthesis [21]. Zygomatic implants are suitable for offset loading but not against axial loading; therefore, when applied, they need to be splinted with the anterior or posterior rigid implants [22]. A study conducted by Ujigawa et al. [23] compared functional stress distribution of zygoma implants with and without connected dental implants supporting a superstructure in a severely atrophic maxilla. They observed that the stress was mostly concentrated in the zygoma bone and the middle of the implant in the connected implant model. Whereas, the stress in the single implant model concentrated in zygoma bone, middle of the implant, maxillary alveolar bone, and implant-abutment joint. These findings presented that a better distribution of forces occurs when all the implants are splinted with the prosthetic reconstruction. In the present case report, zygomatic implants were splinted with titanium milled bar supported prosthesis which is also in accordance with our case.

We have manifested that zygomatic implants offer a reliable method to retain, support and provide the patient with highly favorable speech and aesthetics particularly in cases like mucormycosis which is a rapidly progressive and life-threatening infection.

## CONCLUSION

As mucormycosis is an aggressive opportunistic fungal infection which constraints early diagnosis and treatment intervention in which multidisciplinary approach is recommended to improve patients' quality of life. The maxillary defects that results are different and difficult to manage by conventional methods; therefore, it demands a thorough knowledge to attain function and aesthetics. Thus, we have exhibited a case with post-mucormycosis maxillary defect that was managed with zygomatic implants and a titanium bar supported prosthesis. As well, computer-guided approach contemplates to offer more predetermined final prosthetic outcome with the probable, cautious, and faster implant placement. Therefore, compared to freehand approach, digitally planned implants offer higher success level. Properly executed treatment not only rehabilitates the defect but also rejuvenates self-confidence and improves overall quality of life.

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## Conflict of Interest
Authors declare no conflict of interest.

## Data Availability
All relevant data are within the paper and its Supporting Information files.

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