

Personalized subperiosteal implant-supported obturator for the rehabilitation of rhino-orbit-cerebral mucormycosis sequela: A case report

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Abstract. Severe atrophy of the maxilla occasionally renders it impossible to place standard endosseous implants to replace absent teeth. For such cases, personalized subperiosteal implants (PSI) are presented as a treatment alternative. Due to novel design and manufacturing technologies, PSIs are fitted closely to the bone structure of the patient, after defining the anchorage areas where the bone is of higher quality and allowing a passive dental prosthesis to be attached to restore function and aesthetics to the patient. The present case report documents a patient with severe bone defects as a sequela of rhino-orbit-cerebral mucormycosis. After a failed micro-vascular fibula flap reconstruction, the patient was treated with a removable implant-supported prosthesis attached to a PSI, which provided occlusion with the mandible of the patient and closed the oronasal-antral communication defect. At 18 months after treatment, the patient felt well, with no biological complications and the prosthesis was well adjusted and with good function. Consequently, we consider that in some cases such as this, a customized solution of this type can avoid complex reconstruction treatments.

Introduction

Rhinocerebral mucormycosis is an acute and rare disease caused by infection with the fungi from the *Mucoraceae* family, first described by Paltauf in 1885 (1). It typically affects patients with diabetic ketoacidosis or immunosuppression, where its clinical course is fulminant in the majority of cases (2). It most frequently begins in the nose and paranasal sinuses, with symptoms resembling those of acute rhinosinusitis that do not respond to treatment. It then rapidly progresses and can become fatal if not diagnosed and treated early (3). Extensive surgical debridement combined with systemic treatment with intravenous antifungals is key for controlling this disease (4). However, due to such an invasive surgical approach, survivors frequently experience significant sequelae in the maxillofacial region, with extensive involvement in the maxilla, nose and orbital regions (5), involving loss of vision, difficulty speaking and eating, hearing problems and aesthetic impairment. Such maxillectomy defects are typically treated with prosthetic obturation or autologous tissue reconstruction (6). Since each of these techniques has its advantages and disadvantages, the optimal approach remains subject to debate (7). Reconstruction has the advantage of closing the defect while avoiding the use of a removable prosthesis, but subjects the patient to surgery with a high morbidity rate, while an obturator avoids surgery but is uncomfortable and sometimes very difficult to adapt.

Nevertheless, over the past decade, advances in medical imaging (8) and computer-aided design/computer-aided manufacturing (CAD/CAM) technology (9) have made it possible to develop novel protocols for designing and manufacturing personalized implants that can aid in the reconstruction of these maxillary defects. These implants, known as personalized subperiosteal implants (PSIs), were first described in 1943 (10). Due to recent technological advances, numerous modifications, including reductions in size and thickness, and new connections, have improved the design and manufacturing processes of such PSIs (11,12), enabling such sequelae to be treated with notable results, with fewer exposures and

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Abbreviations: CAD/CAM, computer-aided design/computer-aided manufacturing; PSI, personalized subperiosteal implant; CT, computer tomography

Key words: mucormycosis, maxillectomy, prosthetic obturation, computer-aided design/computer-aided manufacturing, personalized implants

better prosthetic management (13). The present case documents the rehabilitation of a patient with rhino-orbit-cerebral mucormycosis sequela in the maxilla, using a removable implant-supported prosthesis attached to a PSI.

Case report

A 53-year-old male patient, a former smoker without any other relevant medical history, with a IIIB maxillary defect (14), right orbital exenteration and bilateral ethmoidectomy due to rhinocerebral mucormycosis, was referred to the Virgen Macarena University Hospital (Seville, Spain) in December 2021 from the University Hospital of Badajoz (Badajoz, Spain) after the failure of reconstruction using a microvascular fibula flap due to internal jugular vein thrombosis on postoperative day 3 (Fig. 1). Upon arrival at the Virgen Macarena University Hospital 1 year after the fibula flap failure in December 2021, the patient exhibited severe difficulties in oral intake and speech. In addition, the patient presented with significant aesthetic damage, with a sunken mid-facial third and microstomia due to the scar on the lip from previous surgeries (maxillectomy and deferred microvascular fibula flap reconstruction).

Following the unsuccessful reconstruction attempt, placing an obturator was considered for this patient. However, this option was dismissed due to the lack of support provided by the bone and soft-tissue defect. The significant collapse of the lip and nose posed challenges in manufacturing any type of prosthesis, compounded by the absence of support from intraoral tissues, leading to the rejection of this therapeutic option, even as a provisional solution. Soft-tissue reconstruction surgery using a microvascularized forearm flap to cover the defect was proposed, but having already undergone two surgical operations, for mucormycosis and the failed attempt at reconstruction, the patient did not want further reconstructive surgery, so expressed a preference for an alternative reconstructive approach rather than another microvascularized graft. Given the patient's rejection of any reconstructive therapeutic options and the impossibility of placing an obturator, PSI was then suggested as a support for the prosthetic obturator.

To reconstruct the maxillary defect, a PSI (Avinent Implant System S.L.U) with connections for a removable implant-supported prosthesis was proposed, which would provide occlusion with the mandible and close the oronasal-antral communication defect.

A virtual simulation of the obturator and maxillary prosthesis was first performed, using 3-matic Medical® 17.0 software (Materialise), based on the defect and the opposing dental arch (Fig. 2). Acquiring digital impressions with an intraoral scanner was not feasible due to the lack of intraoral references. Consequently, analog impressions were obtained instead using heavy silicone in two stages. Initially, impressions were made from the orbital defect towards the oral cavity, before impressions were then made from the oral cavity towards the orbital defect. Integrating the two impressions facilitated duplication of the defect, enabling the creation of a prototype with which to conduct the facial CT scan used for PSI planning [CT machine model: Revolution CT; supplier, GE Healthcare; Imaging parameters: Scan

mode, helical; collimation, 0.625 mm; slice thickness, 1 mm; reconstruction interval, 0.5 mm; tube voltage, 120 kVp; tube current, 100-200 mA (automatically adjusted by the system); field of view, 220 mm; reconstruction matrix, 512x512; reconstruction filter, bone plus (high-resolution bone kernel); reconstruction mode, multiplanar and 3D; rotation time, 0.4 sec; pitch, 0.8; contrast, no (contrast is generally not used for bone evaluations); patient position, supine, head first]. Radiopaque markers were placed on the resulting prosthesis prototype, along with locator-type connections on the orbital end to ensure proper prosthesis placement during the facial CT scan of the patient (Fig. 3). A cone beam CT scan of the prosthesis was also conducted (Kodak Carestream CS Imaging version 8.0.25; DICOM files voxel size 76x76x76 μ m). Images from both CT scans were segmented using Mimics 25.0 (Materialise) and merged using 3-matic Medical® 17.0 software (Materialise). In this manner, an implant model that conformed seamlessly to the unique contours of the remaining bone of the patient was crafted.

Using the latter software, a PSI made of a sintered Grade V titanium alloy (Ti6Al4V) with a thickness of 0.8 mm was designed together with Avinent Implant System S.L.U engineers and manufactured by Avinent Implant System S.L.U using an EOS M290 printer (EOS GmbH). The PSI was then meticulously tailored to accommodate both the defect and the adjacent anatomical structures where it is anchored. The implant included six universal external hex connections. The connection area was reinforced by increasing the implant thickness to 1.2 mm.

The present case employed a Weber-Ferguson approach (15) to access the malar area and the remaining right infraorbital rim. On the left side, an intraoral approach was used due to the lesser necessity for exposure of the left malar for the proper fixation of the PSI. The implant was fixed to both zygomatic bones and to the left maxillo-zygomatic buttress using self-drilling osteosynthesis screws with a thickness of 1.9 mm and a length of 6 mm (Fig. 4), leaving part of the PSI exposed in the oral cavity. No intraoperative complications or mucositis in the 2nd quadrant area were encountered. Only the immediate postoperative fabrication of a silicone protector for the area exposed to the PSI was needed to prevent mucosal contact with the upper lip. No provisional prosthesis was considered to avoid interference with soft tissue healing. At 2 weeks after placement, a removable implant-supported prosthesis was designed using six locators, completely obturating the defect and allowing the patient to speak and eat normally, whilst facilitating the hygiene of both the prosthesis and the oropharyngeal mucosa (Figs. 5 and 6). The present case designated 2 weeks as the time frame between PSI placement surgery and the prosthetic phase to avoid interference with tissue cicatrization. After checking for correct cicatrization, the prosthetic phase commenced.

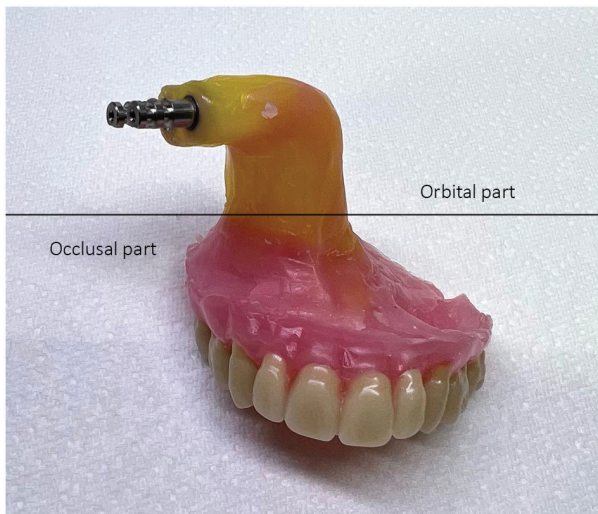
In January 2024, 18 months after treatment, the patient was doing well, the PSI remained exposed in the oral cavity without causing any problems and the prosthesis was functioning properly, with no mobility of the PSI and good adjustment of the prosthesis, ensuring that the defect was filled and the patient could therefore speak and eat normally.



Intraoral defect

Left maxillo-malar tuberosity and buttress remain

Figure 1. Initial view. The intraoral defect visible in the image communicates with the orbital cavity due to the absence of the right maxilla and orbital contents. Only the left maxillo-malar tuberosity and buttress remain.



Occlusal part

Orbital part

Figure 2. Prosthesis prototype for planning. A prototype was created, consisting of an occlusal part that properly positioned the teeth in relation to the mandibular arch and upper lip and an orbital part that supported and immobilized the prototype during the CT scan study.

Discussion

Mucormycosis is a fungal infection that is contracted through the inhalation of fungus spores. It typically affects immunocompromised patients, leading to severe conditions that can be life-threatening (3,16,17), but it is rare for healthy individuals to be affected (16), as in the present case study. In recent years, an increase in cases has been described, particularly associated with the use of corticosteroids during the coronavirus disease-19 pandemic (18-20). Treatment with antifungals and early surgical intervention are crucial for the survival of the patient (17).



Figure 3. Prosthesis placement during the facial CT scan of the patient using locator-type connections on the orbital end. Locators were added to the orbital extension of the prototype to maintain its correct position, whilst the patient underwent the CT scan.



Figure 4. Preoperative planning of the personalized subperiosteal implant on the bone of the patient. This planning was conducted with a segmentation software (Mimics 25.0; Materialise), using a 3D model obtained from the patient's CT scan.



Figure 5. Implant exposed in the oral cavity at 18 months after treatment.

Defects associated with the surgical treatment of rhinocerebral mucormycosis present a therapeutic challenge for maxillofacial surgeons. Difficult-to-reconstruct sequelae are often encountered in the maxilla, nose and orbit (14).



Figure 6. Prosthesis in occlusion.

A combination of surgical and prosthetic rehabilitation is preferred in cases of large midfacial defects to adequately restore the patient's functional and aesthetic needs (21). However, there remains to be a lack of valid recommendations regarding the optimal procedure, especially in terms of the quality of life (22,23).

When the situation of the patient does not allow for reconstruction, the least invasive solution is to treat these sequelae using an obturator (22), which provides notable results for patients who are not good candidates for major reconstruction surgery or who reject this type of surgery. Specifically, it entails closing the communication between the oral cavity and the sinuses and nose, allowing the patient to eat and speak whilst using the prosthesis. In such cases, this can provide a temporary solution until the defect has undergone reconstruction or a definitive solution in patients where communication cannot be closed off (24,25). The prosthesis is typically retained by metal hooks anchored to the remaining teeth, if there are any (26). In terms of the multiple classifications of obturator use in existence, depending on the type of maxillary defect, there is a consensus that the main challenge for patients is the stability and retention of such devices (27). The use of CAD/CAM in the design and manufacture of obturators has improved their adaptation to the intraoral defect (28).

When the option of a removable obturator is not feasible, such as in patients with larger and more complex defects where prosthesis adaptation and fit are not straightforward, it is necessary to find anchor points that can allow for proper closure and stable fixation. PSIs can provide such anchor points by attaching them to areas adjacent to the defect, such as the nasal and zygomatic buttresses, which are typically preserved in these types of sequelae (10). PSIs are customized to the bone anatomy of the patient and include the prosthetic connections in the implant design itself (11,12,29-32). The use of PSIs in oral cavity defect reconstruction is well established, which is frequently combined with microvascularized flaps (33) to reconstruct soft tissues and close the oromaxillary communication. In these cases, the PSI replaces the placement of endosseous implants, as it provides the connections necessary for optimal prosthetic rehabilitation. However, its use as

an alternative to reconstruction (13), as in the case described in the present report, is less developed. Although advances in locoregional and microvascular reconstruction have led to successful surgical outcomes, not all patients are candidates for surgical reconstruction (34). The patient in the present case was referred from another center after a failed microvascular fibula graft and declined a second reconstructive surgery operation.

The option of placing an obturator was considered but dismissed, even as a provisional solution, due to the lack of support provided by the bone and soft-tissue defect. Given the patient's refusal of any reconstructive therapeutic options and the impossibility of placing an obturator, PSI was then suggested as a support for the prosthetic obturator.

The advantages of using a PSI, compared with reconstructing the defect, are that it avoids a second bone and soft-tissue reconstruction, which would entail greater morbidity and a new donor area, simplifies the surgery and avoids a third surgical operation to place conventional implants (33). PSI surgery provides a solution that then only awaits the prosthetic rehabilitation. However, with this option the patient must then always use the prosthesis as an obturator to cover and close off the defect. In addition, one zone of the PSI will remain exposed to the oral cavity where it is unknown how it will develop in the long term.

Such personalized solutions render it possible to connect the area of residual bone, where the implant will be fixed, to the area where connections are needed for the prosthesis that the patient will wear. Reconstructing the soft tissues without needing bone reconstruction for endosseous implant placement, as previously proposed by Korn *et al* (35), would be a valid option for closing the oronasal-antral communication caused by bone loss. However, the present case shows that the PSI option is equally valid when the patient either cannot or will not undergo reconstruction. In the present case, the difficulties lay in the lack of bone support in the maxilla-orbit and the failure of previous attempts at soft-tissue reconstruction.

The PSI needs to be meticulously tailored to accommodate both the defect and the adjacent anatomical structures where it is anchored (35). In instances where bone quality is compromised, the objective in the present case was to devise the implant in a manner conducive to anchoring it in regions distal to the defect, leveraging the presence of cortical and trabecular bone architecture to ensure the robust stability of the PSI. Through strategic design and surgical approaches, a PSI capable of adapting to a spectrum of defects, even those as severe as those exhibited by the present patient, was successfully manufactured.

To the best of our knowledge, there is no previous reference in the literature regarding patients with low bone quality in whom a PSI has been placed to support an obturator with part of the PSI framework exposed to the oral cavity without a soft-tissue covering. In any event, since the majority of the PSI is fixed to the nasal and malar buttresses, where the bone is highly cortical, observations from the present case resulted in a hypothesis that the fixation can remain stable even in patients with poor maxillary bone quality.

We consider that the PSI provides sufficient stability to support a removable implant-supported prosthesis that can occlude the defect without the need for soft-tissue

reconstruction. The stable fixation minimizes obturator mobility and fit issues, in addition to being removable to allow the area to be cleaned. Planning and fabricating the implant from a prosthetic perspective allows the incorporation of 3D connections that are ideal for achieving the most precise functional and aesthetic fit for the prosthesis, even in extensive and complex defects such as those faced in the present case.

Unlike a subperiosteal implant used for bone atrophy, the present implant was thicker in the area that is exposed to the oral cavity, reaching critical thicknesses of 1.5 mm in areas that were considered important for force distribution, instead of the usual 0.8-mm thickness of these PSIs. To the best of our knowledge, minimum thickness data for this type of implant were not found in the literature. Regarding the fixation of the implant with osteosynthesis screws, a similar diameter to that described by Korn *et al* (35) was used, specifically between 1.9 and 2.2 mm. Furthermore, in the present case, it was hypothesized that more screws needed to be placed than in conventional PSI cases and a more distant anchorage was required, utilizing the nasal and zygomatic buttresses and extending to the zygomatic arch if necessary, as indicated by Korn *et al* (35) for a higher Brown's class 1 case.

Although an intraoral surgical approach is adequate for the rehabilitation of severe maxillary atrophies in cases of PSI to ensure optimal fixation (31), in other situations, such as the present case, it is necessary to combine the intraoral approach with an extraoral counterpart to gain access to stable bony fixation areas for PSI, such as the malar, zygomatic arch and orbital rim.

In cases such as that of the present patient, the traditional alternative would have been a microvascularized flap to provide external bone and soft tissues, followed by rehabilitation with osseointegrated implants. The cost of the PSI does exceed that of serial osteosynthesis plates for securing the flap bone, including plates combined with osseointegrated implants. However, since the costs associated with the surgical procedure and hospital stay were substantially lower with the PSI option, the expense was significantly lower overall compared with the traditional alternative. A cost-effectiveness analysis will be needed to confirm this hypothesis in the future.

In conclusion, taking into account the limitations of the present case, it may be concluded that PSIs are a valid option for prosthetic rehabilitation in patients with extensive defects of the maxilla. In some cases, they may even remove the need for reconstruction. However, further studies will be necessary to evaluate the medium and long-term performance of the area of the PSI that is exposed to the oral cavity.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

JPH and JHL performed the surgery and were responsible for study design. ATP was responsible for PSI design. EMC performed the surgery. DMG was responsible for the manufacturing and design of the prototype and the prosthesis, while GCC helped with the prosthesis design. ARM helped to design the study. JHL and JPH confirm the authenticity of all the raw data. All authors participated in the clinical case and contributed to writing the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The present clinical case is part of the 'CLIN25_Impact on quality of life and observational clinical follow-up of Avinent Subperiosteal Personalized Implants' study, approved by the Virgen Macarena and Virgen del Rocio University Hospital Ethics Committee of Seville (approval no. 01082023).

Patient consent for publication

The patient gave written informed consent for the publication of any associated data and accompanying images.

Competing interests

The authors declare that they have no competing interests.

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