

Orbital floor reconstruction with patient-specific implant – sequelae to the excision of traumatic ciliary cyst of the maxilla

Parva A Patel, Kannan Balaraman, Devdutt Thomas, Vimalambiga Ramani, Keerthana Bhat, Ravindra Bharathi, S Raja Sabapathy

Orbital floor defects are challenging to repair, but with the advent of virtual surgical planning and the ability to print patient-specific implants (PSI), the reconstruction becomes predictable. Ciliated cysts of the maxilla often go undiagnosed, and sometimes, due to late presentation, they involve the orbital walls. Removal of the pathology and reconstruction of such defects with the help of Virtual Surgical Planning (VSP) and PSI have been discussed in this paper.

Key words: Orbital floor reconstruction, Patient specific implant, Traumatic ciliated cyst of maxilla, virtual surgical planning

Extensive defects of the orbital wall and floor are difficult to reconstruct due to the complexity of the anatomical region involved and the limited intraoperative view. Successful repair of the orbital skeleton restores function and cosmesis by normalizing globe position and allowing full motility of the extraocular muscles. Posttraumatic defects can be successfully repaired with standard implants. Excision of pathologies of the orbital floor can result in defects that are extensive, irregular, with volume deficiency, and hence challenging to repair. The development of patient-specific implants (PSI) offers an additional tool in such cases.^[1] Surgical ciliated cysts of the maxilla have been conventionally reported to be associated with a history of surgery or trauma to the maxilla. The pathogenesis of the surgical ciliated cyst is generally thought to be due to the entrapment of remnants of sinus mucosa in the wound after maxillary sinus surgery or the early closure of the natural ostium before the sinus is filled with regenerating

granulation tissue.^[2] Herein, we report our experience using PSI for orbital floor reconstruction of a defect secondary to excision of a ciliated cyst of the maxilla.

Case Report

A 48-year-old healthy male presented with complaints of discomfort and pain in the left infraorbital region for 1 month and persistent swelling in the same region for the past 6 years [Fig. 1].

He gave a history of facial trauma before 20 years ago, for which no treatment was done. Swelling was seen in the left infraorbital region; it was approximately 3 × 3 cm in size and firm. The skin over the swelling was normal. Intraorally, the swelling was bulging into the left maxillary buccal vestibule, which was firm, and the mucosa was normal. Vision was normal, and



Figure 1: Clinical picture

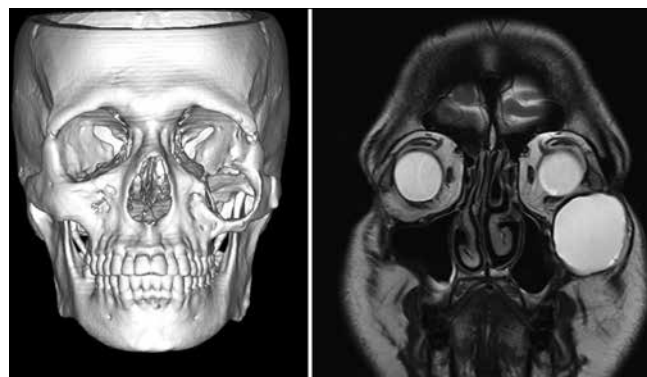


Figure 2: Pre operative CT scan and MRI

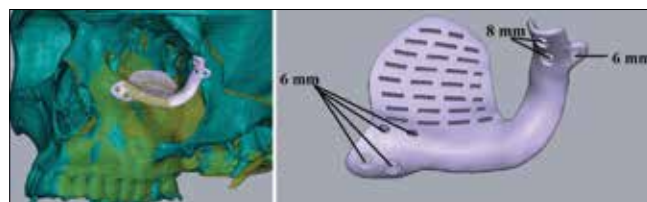


Figure 3: Virtual surgical planning

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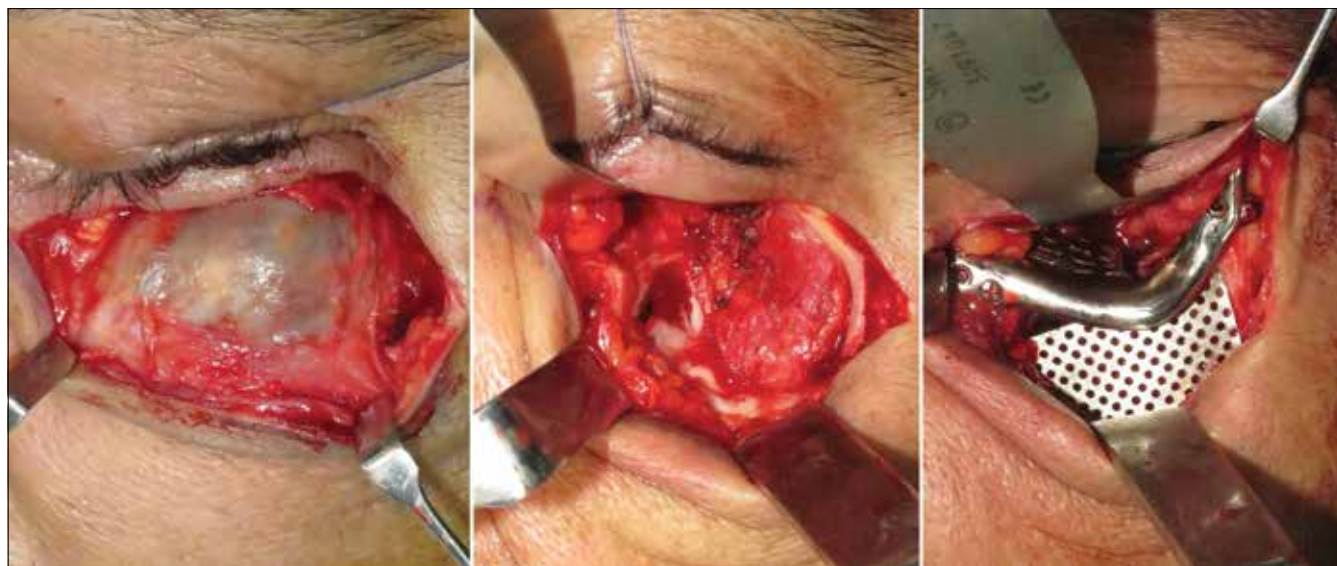


Figure 4: Intra operative pictures

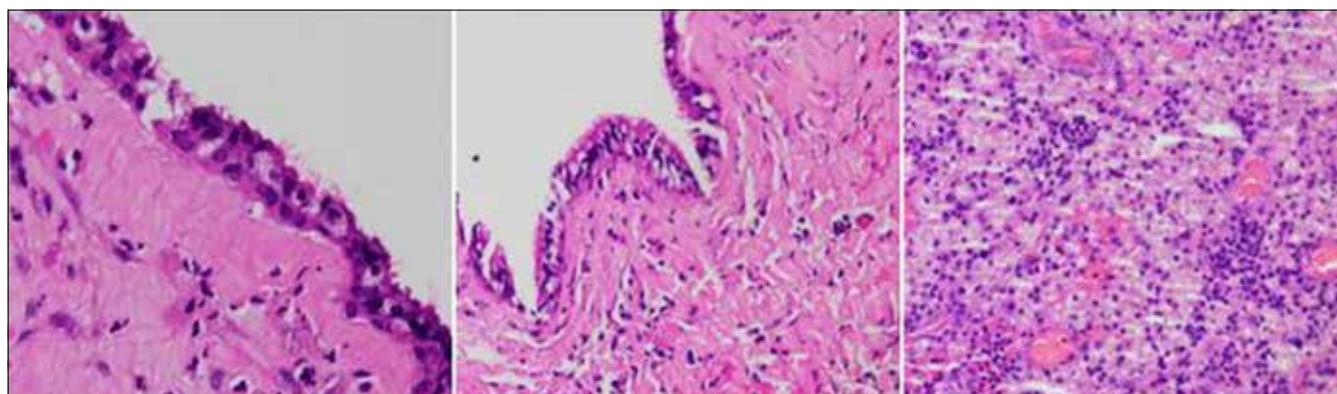


Figure 5: Histopathological images



Figure 6: 2.5 Year Follow up

there was no paraesthesia or discharge. Differential diagnosis could be mucocele, schwannoma, odontogenic cyst, or tumour. CT showed a well-defined hypodense cystic lesion in the left infraorbital region measuring $4 \times 3.9 \times 3.5$ cm, with resorption of the orbital floor, thinning of the left maxillary sinus anterior and lateral walls, and the lesion was bulging into the orbit.

Magnetic Resonance Imaging (MRI) showed a cystic lesion with peripheral enhancement and no solid component within the lesion. Due to the cystic nature of the lesion, the choice of

surgery was enucleation of the lesion with reconstruction of the defect [Fig. 2]. Excision alone without reconstruction can result in gross enophthalmos and hence, simultaneous reconstruction was also planned. Computed Tomography- Digital Imaging and Communications in Medicine (CT-DICOM) data was sent to the planning company (CTARS), and the defect was delineated on the virtual surgical platform (Materialise). The areas that needed reconstruction included the infraorbital rim, part of the lateral inferior rim, orbital floor, and anterior wall of the maxilla. We decided to use a patient-specific implant for orbital floor reconstruction, and titanium mesh was used to reconstruct the anterior maxillary wall [Fig. 3]. Through a subciliary incision, the lesion was exposed and enucleated. The ensuing defect was greater than 50% of the infraorbital rim, and the lateral orbital rim was found to be resorbed, the anterior maxillary sinus wall resorbed, and the lateral part of the orbital floor was found to be resorbed [Fig. 4]. The specimen was sent for histopathological examination. The implant was fixed on the rim and floor defect with the help of screws, and a titanium mesh was fixed for the anterior maxillary wall defect. The Histopathological Examination (HPE) report reveals an inflamed ciliated (respiratory epithelial) cyst with squamous metaplasia, negative for malignancy [Fig. 5]. The patient has been regularly followed up, and CT scans were taken, which show no recurrence and maintained facial symmetry in the recent 2.5 years follow-up [Fig. 6].

Discussion

There are multiple reconstruction options such as autologous grafts, allogenic, and alloplastic materials for orbital floor reconstruction. Of late, titanium mesh implants are gaining popularity because of their thin profile, pliability, and biocompatible nature. The ability to contour the material to achieve optimal reconstruction is dependent on the skill of the surgeon.^[3] Tumour or cyst excision results in irregularly shaped orbital defects that off-the-shelf implants do not address without significant modification. A PSI can be fashioned to perfectly fit this shape to restore missing orbital volume and allow for normalization of the orbital floor.^[4] VSP can also help design the location of fixation holes for the implant. The use of PSI for the reconstruction of the defect resulted in functional as well as aesthetic improvement, with no enophthalmos or diplopia in this patient. The diagnosis of surgical ciliated cyst is extensively used for a cystic lesion resulting from surgical invasion to the maxilla, such as radical maxillary sinus surgery, orthognathic surgery, implant-related surgery, and fracture surgery. However, in our case, as there was no history of surgery, we suggest a hypothesis wherein, subsequent to the previous trauma, the sinus mucosa could have been entrapped between the fractured ends, forming the cyst.^[5]

Conclusion

Benign lesions of the orbit which result in bony destruction will need reconstruction after the excision of the lesion. Recent advances like VSP and PSI provide a fantastic option for good outcomes especially regarding form and function. The ability to design the implant and determine the fixation points helps in optimal reconstruction of extensive defects of the floor. Though expensive as of now, regular usage will bring down the costs in the future. We would also like to propose the term “traumatic ciliated cyst of the maxilla” for those variants of ciliated cysts of the maxilla associated with untreated trauma.

Authors' contributions

Parva A Patel- Registrar. Kannan Balaraman- Chief Operating Surgeon. Devdutt Thomas- Ophthalmologist. Vimalambiga Ramani- Part of the team. Keerthana Bhat- Part of the team. Ravindra Bharathi- Part of the team. S Raja Sabapathy- Director of the Institute.

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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An upper eyelid discharging fistula in allergic fungal rhinosinusitis with frontal sinus osteomyelitis: A case report and review of the literature

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Allergic fungal rhinosinusitis (AFRS) is a noninvasive form of fungal sinus disease, often associated with nasal polyposis and olfactory dysfunction. Frontal sinus osteomyelitis is a rare but serious complication, involving infection of the frontal bone. Although upper eyelid fistulas can occur in severe sinus infections, they remain an uncommon manifestation in AFRS and frontal sinus osteomyelitis. We report the case of a 52-year-old female presenting with purulent discharge from the lateral upper left eyelid, just below the supraorbital margin, due to a fistula associated with

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