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Recurrence of an odontogenic keratocyst in the zygomatic bone with orbital floor involvement: a case report

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Case Report

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Abstract

Background

An odontogenic keratocyst (OKC) is a benign but locally aggressive intraosseous lesion arising from remnants of the dental lamina or the odontogenic epithelium. It has a marked tendency to recur, and patients with early-stage disease are often asymptomatic; swelling is the most common presenting sign. Involvement of the zygomatic bone and orbital floor is exceptionally rare, since OKC typically arises in the mandibular ramus and molar region.

Case presentation

32-year-old woman presented with pain, swelling, and pus discharge below the right eye, with mild orbital dystopia and a scar below the lateral canthus of the right eye. She had a history of multiple surgical procedures for a right maxillary cyst dating back to adolescence. Intraoral examination revealed missing upper right second and third molars and mobility of the premolars. Cone-beam computed tomography (CBCT) demonstrated a well-defined radiolucency involving the infraorbital rim, zygoma, and floor and lateral wall of the right orbit, extending from the maxillary sinus. She underwent cyst enucleation, peripheral ostectomy, and chemical cauterization with modified Carnoy's solution, together with orbital floor reconstruction using titanium mesh. Histopathology confirmed odontogenic keratocyst. At 7 months' follow-up there was no clinical or radiological evidence of recurrence, and vision and ocular motility in the operated eye remained unchanged.

Conclusions

Recurrence of OKC in the zygoma and orbital floor is rare and illustrates how residual satellite cysts left after incomplete removal can reach anatomically inaccessible sites over time. Accurate clinical and radiological diagnosis, combined with enucleation plus an adjunctive measure such as peripheral ostectomy and chemical cauterization, is important to reduce further recurrence, and long-term follow-up remains essential.

Background

Odontogenic keratocyst (OKC) is a benign yet aggressive and locally recurrent lesion of the maxillofacial region. It is a histopathologically and behaviorally distinct entity, showing characteristics of both a cyst and a benign tumor [1]. OKCs arise from remnants of the dental lamina or the basal cells of the overlying oral epithelium [2]. The lesion is most common in the second and third decades of life, with a maxilla-to-mandible ratio of approximately 1:2. OKCs occur most frequently in the mandible, accounting for 65–83% of cases, while fewer than 1% arise in the maxillary sinus [5]. The maxillary sinus lies in close proximity to the developing tooth germs and root apices of the premolars and molars and is therefore

vulnerable to odontogenic infection, cysts, and tumors. Diagnostic imaging in the maxilla is challenging because of the overlap of anatomical structures on radiographs [6]. Reported recurrence rates for OKC range from 20% to 60% [4].

The optimal treatment of odontogenic cysts remains a matter of debate, and several surgical approaches have been described, including marsupialization, enucleation (with curettage or peripheral ostectomy), enucleation with chemical cauterization, and enucleation with cryotherapy using liquid nitrogen [3].

Because of its high recurrence rate, patients with OKC frequently undergo multiple procedures at the same site or in its vicinity, and recurrent lesions can occasionally extend beyond the primary site to involve unusual, distant locations.

In this report, we describe a case of OKC of the maxilla that recurred in the body of the zygoma, eroding the orbital floor.

Case presentation

32-year-old woman presented with pain, swelling, and pus discharge below the right eye. Her medical history was otherwise unremarkable, but she reported multiple previous surgical procedures for treatment of a jaw cyst. She recalled first noticing an upwardly displaced right eye and persistent nasal congestion in 2003, at 15 years of age. She was evaluated at a local hospital and diagnosed with a dentigerous cyst associated with an impacted tooth in the right maxillary sinus; the cyst and tooth were surgically removed under local anesthesia later that year. Symptoms recurred in October 2004 and were managed with intranasal inferior meatal antrostomy. Twelve years later, in November 2016, her original complaints recurred. Computed tomography (CT) at that time showed a well-defined cystic lesion (2 x 2 cm) in the right maxilla protruding into the inferolateral aspect of the right orbit. She underwent cyst enucleation with extraction of tooth 17, and histopathology confirmed an odontogenic keratocyst of the right maxillary sinus. Similar episodes of pain and swelling recurred over subsequent years and were treated as osteomyelitis of the right maxillary sinus, with intraoral curettage and extraoral incision and drainage below the right eye. The patient did not report any family history of a similar condition, nor did they have a medical history related to the disease.

On examination, there was mild diffuse swelling in the right infraorbital region, soft in consistency and mildly tender. Orbital dystopia and a small scar below the lateral canthus of the right eye were noted. Intraoral examination revealed missing teeth 17 and 18 and a surgical scar.

Cone-beam computed tomography (CBCT) showed a hypodense lesion involving the right maxillary sinus, zygoma, and the floor and lateral wall of the right orbit. Based on the clinical and radiological findings, a provisional diagnosis of recurrent odontogenic keratocyst of the zygomatic bone involving the orbital floor was made (Fig. 1).

Under general anesthesia, a transconjunctival incision with lateral canthotomy was performed. Malleable and orbital floor retractors were used to protect the globe. A small (5 mm) lateral canthotomy incision was extended to the crow's-feet area to provide adequate exposure, and the incision was carried through the palpebral conjunctiva. Following dissection, the tissue plane was developed superficial to the orbital septum and deep to the orbicularis oculi. Through a subperiosteal dissection, the cystic lesion was visualized and enucleated in toto, and the specimen was sent for histopathological examination (Fig. 2).

Peripheral ostectomy was performed, followed by chemical cauterization with freshly prepared modified Carnoy's solution (75% dehydrated ethanol and 25% glacial acetic acid, without chloroform), applied with care to provide adequate margin control while preserving cosmetic outcome. The lateral wall and floor of the orbit were reconstructed with titanium mesh. After achieving adequate hemostasis, the surgical site was closed with 5 – 0 Vicryl and 5 – 0 Prolene sutures (Fig. 3).

Postoperative healing was uneventful. Mild orbital dystopia persisted, with no change in vision or eye movements. Histopathology revealed a cystic lesion with a lumen, epithelial lining, and connective tissue capsule; the epithelial–connective tissue junction was flat, with basal cells showing a typical tombstone appearance. A few satellite cysts were noted within the connective tissue stroma, consistent with odontogenic keratocyst.

Postoperative CBCT confirmed the titanium mesh implant in place. At 7 months' follow-up there was no clinical or radiological evidence of recurrent disease; vision in the operated eye was unchanged and ocular motility was grossly full (Fig. 4). Surveillance for recurrence is ongoing.

At 5-year follow-up, CBCT continued to show the orbital mesh in place with good bone formation in the infraorbital region and no evidence of recurrence; vision in the operated eye remained unchanged and ocular motility was grossly full (Fig. 5).

Discussion and Conclusions

Reported recurrence rates of OKC are variable and depend mainly on the surgical approach used. As expected, segmental resection carries the lowest recurrence rate because it eliminates adjacent daughter cysts and involved mucosa [7]. However, resection of the zygomaticomaxillary complex in a young patient, typically through a Weber–Ferguson approach, would be debilitating, affecting globe position, ocular movement, and facial aesthetics. In a systematic review and meta-analysis, Al-Moraissi et al. (2017) reported recurrence rates for various surgical approaches: resection (8.4%), decompression followed by cystectomy (14.6%), enucleation with liquid nitrogen (14.5%), enucleation with Carnoy's solution (11.5%), enucleation with curettage (17.4%), marsupialization alone (32.3%), and enucleation alone (23.1%) [8]. A combined transnasal endoscopic approach with oroantral communication repair has also been described as a safe and effective procedure for closure of large oroantral fistulas and eradication of secondary sinus disease; for large cysts, transnasal modified medial maxillectomy is the primary approach, with a transoral approach added when tooth extraction or oroantral communication repair is required [9]. Follow-up in that series was performed by transnasal endoscopic inspection of the

maxillary sinus and intraoral examination of the alveolar ridge, with imaging reserved for suspected recurrence [9].

In our case, the orbital septum was preserved, and there were no clinical disturbances in eye movement or vision. Because a defect remained in the orbital floor, reconstruction was planned as part of the same procedure. Stoelinga et al. (1971) theorized that these cysts arise from proliferation of basal cells of the oral mucosa, or from hamartomas. With each successive curettage in our patient, residual satellite cysts appear to have persisted in and around the walls of the maxilla, zygoma, and infraorbital region. The choice of approach was itself a challenge: a Caldwell–Luc or transnasal approach alone would likely have left cystic remnants within the zygoma. Combining enucleation with an adjunct such as Carnoy's solution, together with orbital reconstruction using titanium mesh, represented a different approach for this recurrent, extragnathic presentation.

Zhou et al. reported an OKC affecting the right maxilla and zygoma without extensive intraosseous loculation; the cyst and associated third molar were removed via a Caldwell–Luc approach, with peripheral ostectomy and cryotherapy performed without reconstruction, and no recurrence was detected at 8 months [10]. Jackson et al. described a case of multiple recurrent mandibular OKC that ultimately invaded the zygoma, maxillary sinus, orbital floor, lateral wall and roof, bony external auditory canal, and skull base [11].

Zygomatic involvement by OKC is rare. Accurate clinical and radiographic assessment combined with histopathological examination is essential for the preoperative diagnosis of this challenging presentation in an unusual site. Given the difficulties encountered in reaching a working diagnosis in such cases, surgical management should be planned with the lesion's recurrence potential in mind. Although OKC is not a malignancy, several of its behavioral characteristics warrant a correspondingly cautious approach. Long-term clinical and radiographic follow-up is mandatory, since recurrence may occur even years after treatment.

We have described an aggressive OKC that recurred in a rare extragnathic location approximately 15 years after initial treatment, requiring exposure of the inferolateral zygoma in proximity to the orbital floor. Orbital floor reconstruction was performed with titanium mesh, and chemical cauterization with modified Carnoy's solution was used with careful technique as an adjunct to peripheral ostectomy to reduce the risk of further recurrence. We acknowledge the relatively short initial follow-up interval reported here and continue to monitor the patient for recurrent disease; follow-up to 5 years has shown no evidence of recurrence.

Abbreviations

OKC
Odontogenic keratocyst
CBCT
Cone-beam computed tomography

CT
Computed tomography.

Declarations

Ethics approval and consent to participate

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests

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Authors' contributions

S. Vishwak contributed to data collection, literature review, preparation of clinical records, manuscript drafting, and revision. **Ravi Veeraraghavan** conceived and supervised the study, contributed to patient management, critically reviewed the manuscript, and approved the final version. **Jaeson Mohanan Painatt** participated in patient management, interpretation of clinical findings, and manuscript revision. **Giri Sankar** contributed to surgical management, manuscript editing, and critical review. All authors read and approved the final manuscript.

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Figures

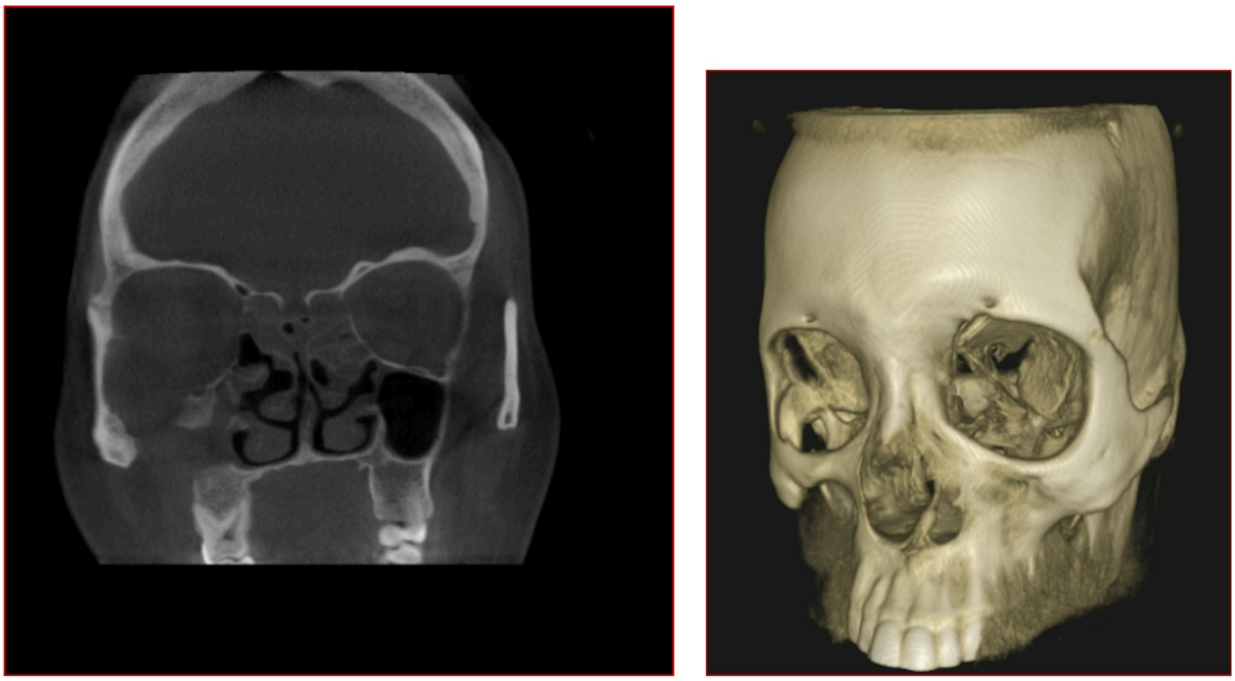


Figure 1

Preoperative CBCT, coronal and three-dimensional volume-rendered views, showing a well-defined radiolucent lesion involving the right zygoma and floor of the orbit

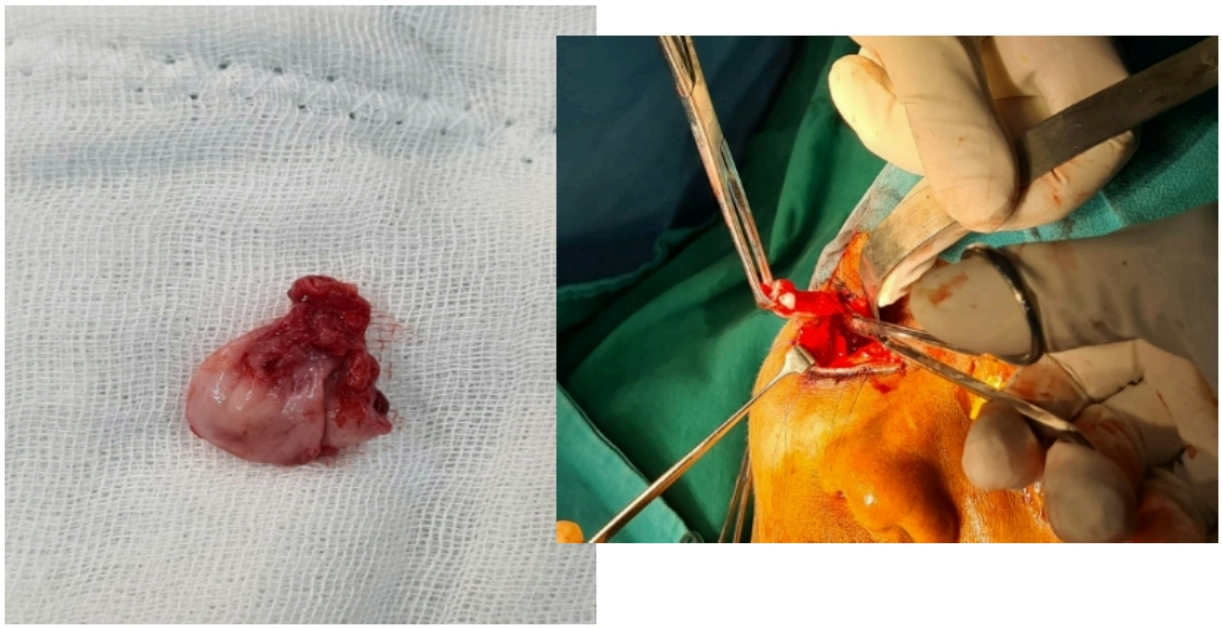


Figure 2

Intraoperative view showing the transconjunctival, lateral canthotomy approach and subperiosteal exposure of the orbital floor defect

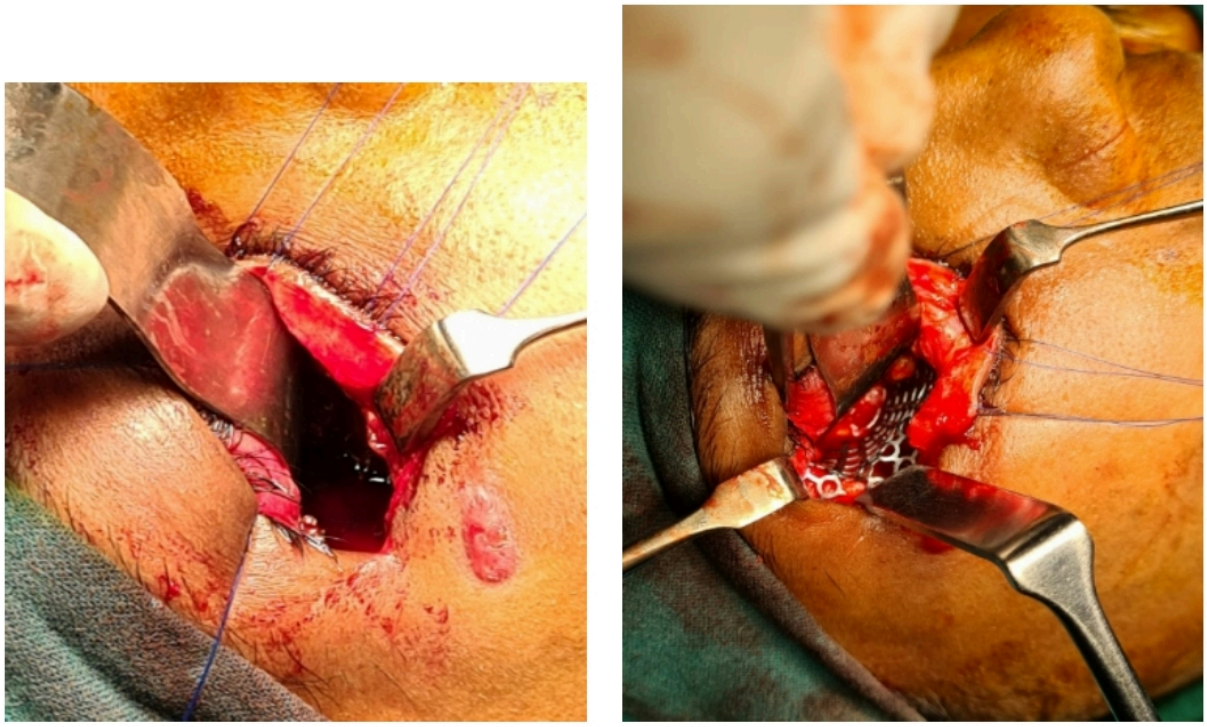


Figure 3

Intraoperative view after peripheral ostectomy, chemical cauterization, titanium mesh reconstruction of the orbital floor, and wound closure

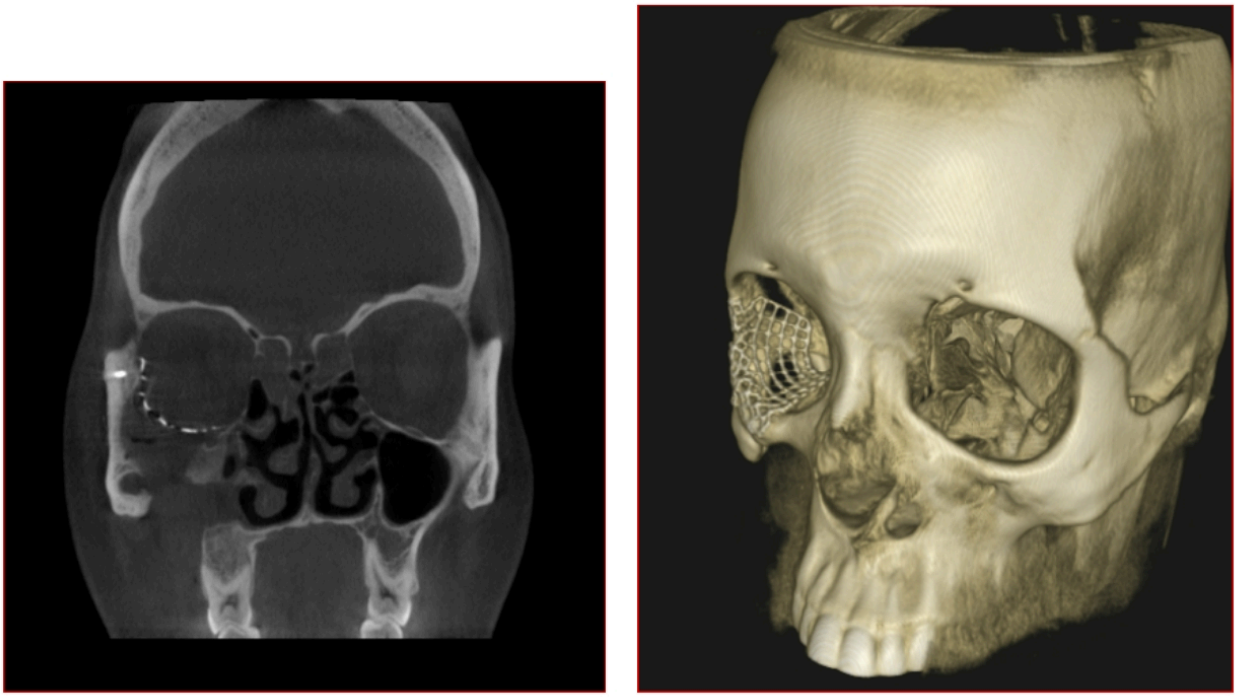


Figure 4

Postoperative CBCT at 7 months showing the titanium mesh in place with no evidence of recurrence

At 5-year follow-up, CBCT continued to show the orbital mesh in place with good bone formation in the infraorbital region and no evidence of recurrence; vision in the operated eye remained unchanged and ocular motility was grossly full (Fig. 5).

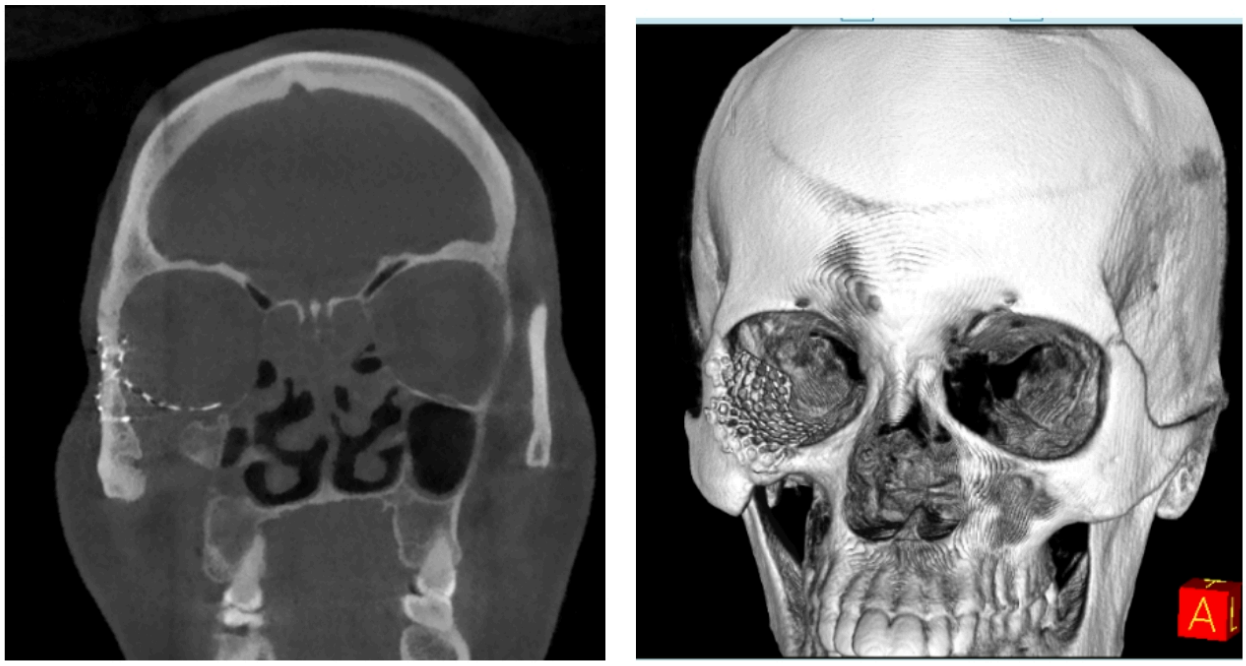


Figure 5

Postoperative CBCT at 5-year follow-up showing the orbital mesh in place, good bone formation in the infraorbital region, and no recurrence

Supplementary Files

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