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Mirror-imaged correction of unilateral orbital deformation caused by a maxillary air cyst

Jan-Falco Wilbrand [a,*], Jens Peter Klussmann [b], Gabriele Krombach [c], Detlef Litzlbauer [c], Klaus-Heiko Wassill [d], Christine Langer [e], Ralf Wilbrandt [a], Hans-Peter Howaldt [a], Sebastian Boettger [a]

[a] University Hospital Giessen, Dept. for Cranio-Maxillofacial Surgery, Klinikstr. 33, 35385, Giessen, Germany
[b] University Hospital Cologne, Dept. for Otorhinolaryngology, Kerpenerstr. 62, 50931, Cologne, Germany
[c] University Hospital Giessen, Dept. for Radiology, Klinikstr. 33, 35385, Giessen, Germany
[d] University Hospital Giessen, Dept. for Ophthalmology, Friedrichstr.18, 35385, Giessen, Germany
[e] University Hospital Giessen, Dept. for Otorhinolaryngology, Klinikstr. 33, 35385, Giessen, Germany

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ABSTRACT

Purpose: To describe a case of hyperpneumatization of the maxillary sinus along with orbito ethmoidal Haller's cells leading to progressive upward dislocation and proptosis of the right globe.
Design: Interventional case study.
Methods: Using a web-based simulation of surgical resection, along with a mirror-imaged reconstruction of the right orbit by a custom-made titanium implant, transconjunctival surgical intervention was planned and accomplished under general anesthesia. Clinical evaluations regarding proptosis and upward dislocation of the right globe were collected. The total preoperative, simulative-, and postoperative orbital volumes were measured using Simpson's method.
Results: The orbital volume was significantly lower on the affected side than on the unaffected side. During simulation, approximations of the right and left orbital volumes were planned. Postoperatively, the right and left orbital volumes were effectively equal, but the simulative orbital volume was approximately 5 cm^3 higher than computed tomographic measurements of the orbital volume.
Conclusion: Hyperpneumatization of the maxillary sinus with orbital infliction is a rare event. Decompression along with a three-dimensional simulated alignment of the orbital volume to the healthy side leads to reproducible results and enhances intraoperative security.

1. Introduction

The pathological expansion of air-filled compartments of the skull is mostly located in the frontal and sphenoidal sinus. Pathological expansion of the maxillary sinus has seldom been described, with only 12 cases described with signs of orbital infliction [1]. Clinical terminology regarding this matter is inconsistent, and identification varies between hypersinus maxillaris, pneumosinus dilatans (PD), pneumocele [2], and air cysts, while the latter was introduced to cover all pathological expansions of the sinus [3]. The

* Corresponding author.
E-mail address: jan-falco.wilbrand@uniklinikum-giessen.de (R. Wilbrandt).

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formation of hyperpneumatization is not completely understood, but it can sometimes be connected with occult meningioma or fibro-osseous disease, or may arise from unidirectional valve effects [1].

Clinical findings caused by hyperpneumatization of the maxillary sinus may include facial pain or swelling and numbness in the region of the infraorbital nerve, but also ophthalmic issues such as proptosis, dislocation of the globe, and even loss of vision [1].

The therapy of choice when clinical impairment arises is surgical decompression of the sinus by creating a nasoantral window, involving plain resection of the protruding orbital wall, but also an antral wall turnover, greenstick fracture, or electrical burring [4,5]. The reconstruction of the resected areas is most important, e.g., the region of the orbital floor, to avoid prolonged functional ophthalmic and oculomotor problems [6].

The use of mirror-imaged custom-made titan implants in orbital reconstruction has increasingly played a role in facial surgery [7, 8]. Individually manufactured implants and osteosynthetic materials are used for reconstruction after tumor ablation [9] or trauma [10]. This relatively new technology facilitates a predictable and safe postoperative outcome, which is of utmost relevance in orbital procedures. However, it has not yet been described in the context of hyperpneumatization of the paranasal sinuses.

2. Patient and methods

A 20-year-old patient with slow progression of right side exorbitism and upward dislocation of the globe during the last two years presented to our University Center for Maxillofacial surgery (Fig. 1a and b). After ophthalmic, otorhinolaryngological, and radiological evaluations, a diagnosis of pneumosinus maxillaris dilatans along with Haller's cells in the orbitoethmoidal region was made (Fig. 2a and b).

The preoperative ophthalmic findings showed an 8 mm upward dislocation of the right globe with slight diplopia in abduction over 35°, with no decrease in vision. The patient did not suffer from any other symptoms like paraesthesia or pain in the facial region.

Preoperatively, an interdisciplinary web-based simulation session of the surgical correction was performed with the IPS Case designer® Software of KLS Martin Tuttlingen, Germany. In this context, the margins of bony resection to decompress the right orbit and a mirror-imaged custom-made titan implant corresponding to the contralateral orbital floor and medial wall were planned (Fig. 3a and b) and manufactured. Consent of the institutional ethic committee was obtained and surgery was performed under general anesthesia. The correction of the orbital boundaries via a transconjunctival approach was successful without any complications.

3. Results

The patient had mild swelling for a number of days, and a slight hypaesthesia in the infraorbital region after the surgery, which disappeared after 4 weeks. Exorbitism, upward dislocation of the globe, and diplopia were sufficiently corrected by this procedure (Fig. 4a and b). A postoperative ophthalmologic assessment obtained no signs of motility impairment, a remaining upward dislocation of the globe of 2mm with no exorbitism, normal visus and no more diplopia. The result was stable even after one year of follow-up. A

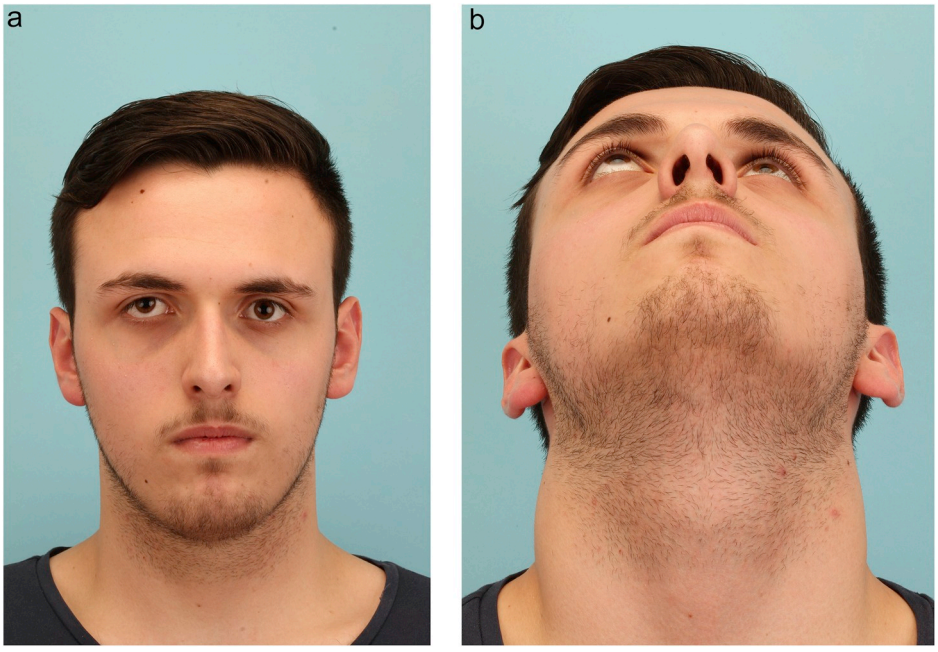

Fig. 1. Preoperative appearance in a patient with orbital infliction by hyperpneumatization of the right maxillary sinus. a: frontal view; b: basilar view.

Fig. 2. Preoperative CT-Scan of Patient with hyperpneumatization of the maxillary sinus along with a Haller's cell in the orbitoethmoidal region. a: coronal plane; b: sagittal plane.

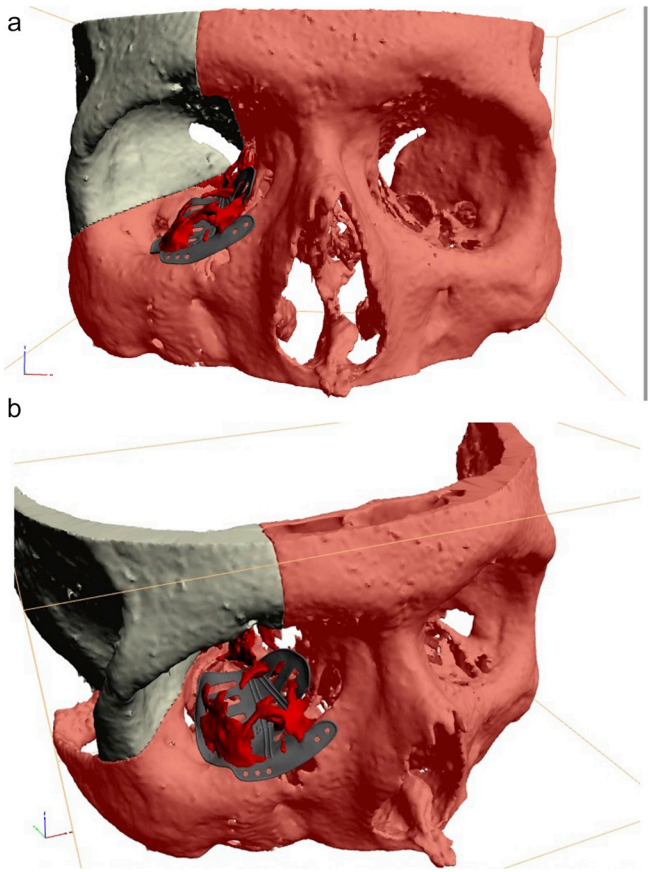

Fig. 3. Virtual planning of the orbital bony margins to be resected and mirror-imaged reconstruction of the orbital floor and the medial orbital wall. a: frontal view; b: oblique view.

postoperative CT-Scan showed the proper positioning of the patient-specific implant (Fig. 5a–d).

The respective volumes of the right and left orbits were measured using a preoperative computed tomography (CT) scan, during the preoperative web-based simulating session and in the postoperative CT scan using Simpson's rule. The CT-scans were measured by a radiologist. The simulative orbital volume was measured by an IT-specialist with the Software IPS Case designer® by KLS Martin. The orbital volume in the preoperative CT-scan was 27.8 cm^3 on the affected right side, and 30.4 cm^3 on the left side. During the simulating web-session, the "pre-correction" orbital volume on the affected right side was 33.1 cm^3, while on the contralateral (unoperated) side the total volume was 35.3 cm^3. The simulated "post-correction" orbital volume on the mirrored right side was 35.5 cm^3. A post-operative CT scan showed an effective postoperative orbital volume of 30.2 cm^3 on the unaffected side and 29.1 cm^3 on the affected side. Alignment of orbital volume has thus been achieved with high accuracy. From our point of view, the volumetric differences between simulation and reality can best be explained by the fact that the simulative measurements were carried out by a different

Fig. 4. Postoperative appearance in a patient with orbital infliction by hyperpneumatization of the right maxillary sinus. a: frontal view; b: basilar view.

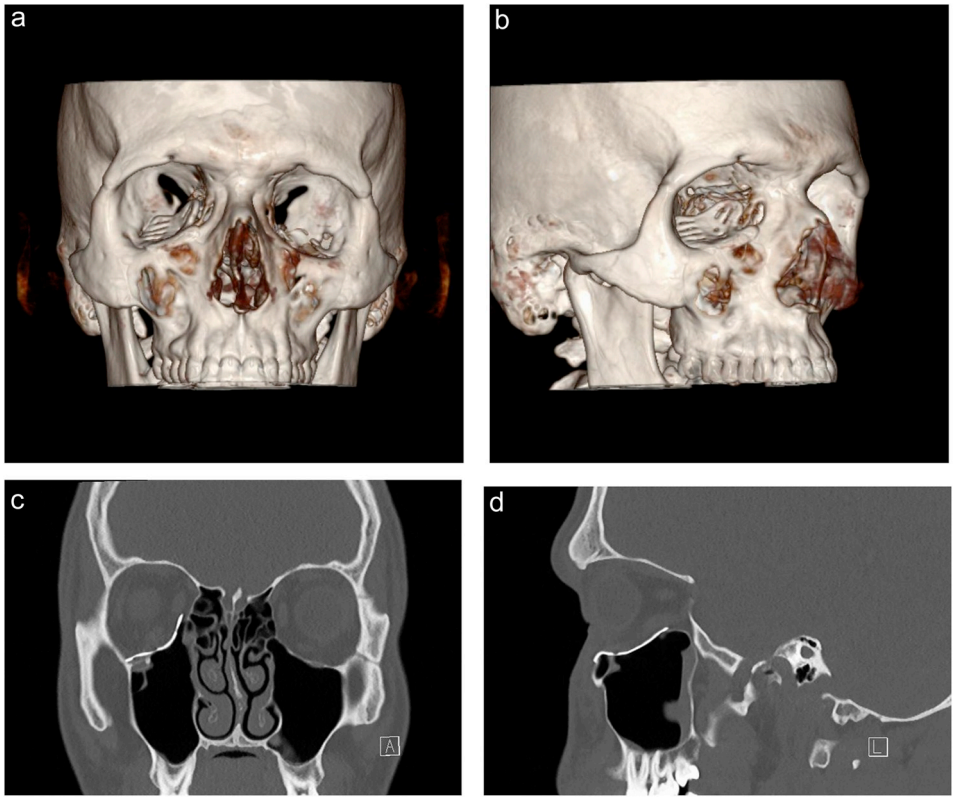

Fig. 5. Postoperative 3D-CT-Scan of Patient with hyperpneumatization of the maxillary sinus along with a Haller's cell in the orbitoethmoidal region. a: frontal view; b: oblique view; c: exemplary coronal plane; d: exemplary sagittal plane.

examiner (radiologist vs. IT specialist).

3. Discussion

The clinical picture of a hyperpneumatized paranasal sinus can remain clinically unapparent (hypersinus), leading to symptomatic expansion of the sinus boundaries with a regular (PD) or irregular configuration and thinning of the bone (pneumocele) [1]. To cover all of these possibilities and to minimize ambiguity in terminology, Tovi et al. proposed the term “air cyst” [3] in 1991.

The majority of clinicians believe in a unidirectional valving effect at the natural ostium of the maxillary sinus and consecutive elevation of internal pressure to be the main pathomechanism for hyperpneumatization [11,12] This effect was assumed to be operative in the patient we treated, because we found excessive labyrinthine convolution of the paranasal sinus, the orbito ethmoidal region, and the natural antrum on the right maxillary sinus. Some authors have suggested that a connection may exist between hyperpneumatization in the orbito ethmoidal region and occult meningiomas or fibro-osseus disease [13]. Other clinicians have hypothesized connection with hormone dysregulation [14], infection with gas-forming organisms, or mucocele discharge [15]. However, the exact etiology of PD is not yet fully understood, and definitive determination for the pathomechanism could not be made from the patient in this report. No signs of Meningeoma, fibro-osseus disease or hormone dysregulation were found in the presented case. One possible factor leading to hyperpneumatization of the maxillary sinus could be the presence of Haller’s cells in the region of the natural ostium and the ethmoid infundibulum. Haller’s cells are frequently found in a healthy sinus, and usually do not lead to any clinical symptoms. In some cases, onset of sinusoidal discomfort of headaches has been described [16]; these cells may block drainage routes [17], which can lead to a unidirectional valving effect and amplification of hyperpneumatization.

Five different treatment options for PD have been proposed [4]. While simple decompression of the sinus by a Caldwell-Luc approach and creation of a nasoantral window [1] have been described by some investigators, other investigators have advocated endoscopic resection of protruded bony areas towards the orbit [18]. Antral wall turnover, greenstick fracture, or electrical burring to correct accompanying deformities of the maxilla, the cheek, and the face have also been reported [4]. The plain resection of portions of orbital wall bones without any reconstruction, however, presents a risk for oculomotoric disturbances or enophthalmos [6]. This might be particularly applicable when more extensive resections are required, as in the present case. Transconjunctival open decompression of the constricted orbit, resection of the protruding margins of the paranasal sinus, along with immediate custom-made mirror-imaged orbital reconstruction implants to prevent postoperative enophthalmos or oculomotoric impairment have never been specified in past reports. Definite management, including preoperative simulation and perioperative measurement of the orbital volumes along with mirror-imaging of the contralateral orbit, is thought to lead to a predictable outcome, enhance intraoperative security, and define postoperative alignment of the orbital volume.

The production of mirror-imaged implants manufactured with the help of a preoperative thin-layer CT scan with slices <1 mm allows accurate determination of the new orbital margins, and already is the state of the art treatment for complex periorbital tumor surgery [9] or traumatology [10]. Therefore, the use of preoperative planning in decompressive non-oncologic periorbital resection, and for the treatment of orbital deformity, should be pursued.

Conflicts of interest

There are no financial or other conflicts of interests to declare, there was no funding and no grants.

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