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REVIEW ARTICLE

Surgical ciliated cyst after maxillary orthognathic surgery: a literature review and case report

Jose Golaszewski[1] · Ruben Muñoz[1] · Dionelys Barazarte[1] · Ligia Perez[2]

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Abstract

Purpose Surgical ciliated cyst of the maxilla is a rare complication following surgical procedures or trauma involving the maxillary sinus. The surgical ciliated cyst of the maxilla is a rare lesion and appears as a delayed complication after surgery in the maxillary sinus, midface osteotomies, traumatic tooth extraction and maxillary fractures.

Case report and literature review We report a case that occurred 5 years after a maxillary sinus surgery in a Caucasian Female. A well-defined unilocular radiolucency in the right anterior maxilla was noted in cone beam image associated to osteosynthesis material. The lesion was completely excised, and upon histological examination, findings were consistent with Ciliated cyst. The PubMed database was searched for PMC within the last 15 years.

Results Together with the current case, we found 18 reports including 21 patients describing PMC. It wasdiagnosed at a mean time of 22 years after causal surgery at a mean age of 47 years. The main radiologicalsign was a unilocular radiolucency.

Discussion This lesion may present histologically different epithelial linings, but respiratory epithelium was the most frequent. The most important clinical and pathological features of these conditions are also discussed.

Conclusion Although surgical ciliated cysts have only rarely been reported after orthognathic surgery, anincreased awareness of this possibility is necessary to avoid delays in diagnosis.

Keywords Ciliated cyst · Case report · Maxilla

Introduction

Surgical ciliated cyst was first described in 1927 in Japan by Kubo [1]. Very limited cases with this entity have been reported in English articles [2]. The lesion is more frequent in Japan [3]. Its prevalence in patients with 5 to 49 years of age who had a previous history of maxillary sinus surgery was estimated to be more than 20% in Japan [2].

Surgical ciliated cyst of the maxilla is a benign cyst of the maxillary sinus that formed after a period of time following surgery or trauma involving the Schneiderian membrane. It is also known as postoperative maxillary cyst, paranasal cyst, or respiratory implantation cyst [1, 3–5].

✉ Jose Golaszewski
jbgmaxilofacial@gmail.com

[1] Oral and Maxillofacial, Dr Angel Larralde Universitary Hospital, Carabobo University, Naguanagua, Venezuela

[2] Oral and Maxillofacial, Zulia University, Maracaibo, Venezuela

We now report the case of a patient with a very large surgical ciliated cyst that developed in the maxilla 5 years after a Le Fort I advancement osteotomy.

Case report

In 2011, a 22-year-old female patient underwent a Le Fort I maxillary impaction osteotomy to correct a dentofacial deformity**.** The patient was symptom free until May 2017, when she developed a painful swelling in the right maxillary area. The patient was medicated with antibiotics and analgesics for 7 days. An endodontic evaluation of teeth was made without positive symptoms. A volumetric computed tomography scan showed a unilocular cystic lesion without perforation of vestibular and lateral cortical bone in the pyriform area associated with one screw of rigid fixation (Fig. 1).

In June 2017, a biopsy of the cystic lesion was performed under local anesthesia through an incision in the labial sulcus. Intraoperatively, a submarginal flap was raised in order to avoid possibility of recession of the involved teeth; she underwent a

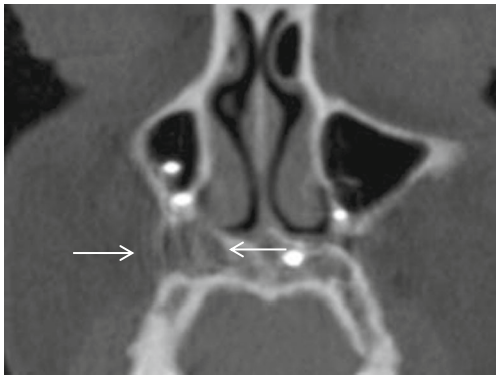

Fig. 1 Preoperative computed tomography (CT) study

surgical procedure to remove the osteotomy fixation plates from the anterior right side of the maxilla and the cyst was enucleated in total together with the sinus lining (Fig. 2).

The wound was then closed primarily with Vycril 3-0. Postoperatively, the patient was given diclofenac sodium 50 mg every 8 h and amoxicillin/clavulanic acid 625 mg every 12 h for 1 week. Healing was uneventful.

After surgery, the patient has been reviewed regularly. Healing has been satisfactory with no clinical complications.

Histopathological examination of surgical specimen showed a cystic cavity lined in some areas by ciliated pseudostratified columnar epithelium, while in others is simple cuboidal. A fibrous connective tissue wall is identified as well as a moderated inflammatory chronic infiltrated, which confirmed the diagnosis of a surgical ciliated cyst of the right maxillary sinus (Fig. 3).

Patient was reviewed on a regular basis. A cone beam computed tomography (CBCT) scan was taken at 6 months postoperative and compared with the preoperative CT scan. There was no abnormality detected both clinically and in the CT scan during the review visit.

A progressive regeneration of the cavity was observed on cone beam computer images.

Review of the literature

A PubMed database literature search was performed to analyze the etiology, incidence, and latency (postoperative maxillary cysts (PMCs)). Search strategy contained the keywords "postoperative maxillary cyst", "surgical ciliated cyst", and "postoperative paranasal cyst". Results were limited to English, Portuguese, and German literature of the last 15 years. Out of the included articles, patients' age, gender, initial symptoms, location, etiology, histopathology, and length of follow-up examinations were extracted (Table 1) [6–22].

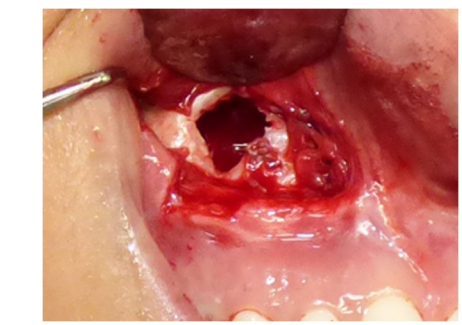

Fig. 2 Intraoperative photo showing the cyst in situ

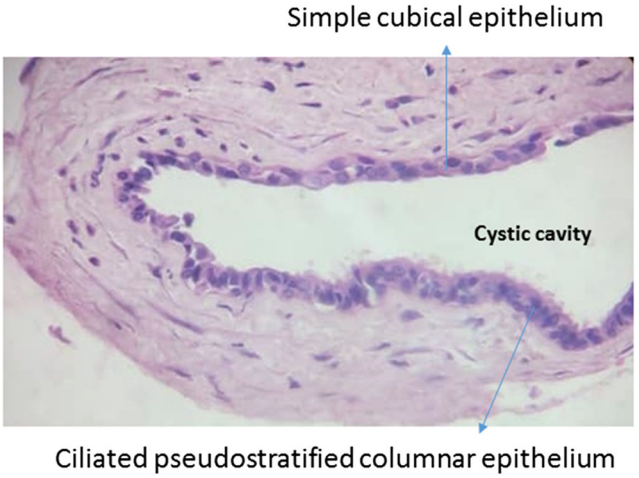

Fig. 3 Photomicrograph of the cyst showing pseudostratified ciliated columnar epithelium lining (original magnification × 20, H&E staining)

Results of the literature review

Ciliated cyst of the maxilla is a rare complication after maxillary surgery or maxillary sinus surgery. A total of 15 articles describing surgical ciliated cyst were found.

The male patients (10 patients) were affected slightly more often than females (9 patients).

The average time of latency for ciliated cyst of maxilla after first surgery was 15.83 years.

(min 2, max 55). Unilateral incidence was described more often (89%) than bilateral (11%). The etiology reported traumatic extraction (2 patient), Le Fort I osteotomy (4 patients), trauma (3 patients), and 9 patients under maxillary sinus surgery.

Discussion

The pathogenesis of the surgical ciliated or postoperative maxillary cyst is thought to be due to the entrapment of remnants of sinus mucosa in the wound after maxillary sinus surgery or early closure of the natural ostium before the sinus is completely filled with regenerating granulation tissue [6].

The maxillary sinus lining is invariably breached during orthognathic surgery, and it is unclear why there have not been more reported cases in the Western literature of surgical ciliated cysts after this type of elective surgery [7, 10, 12, 14, 19].

Table 1 Case reports published in literature in the last 15 years

Authors	Case	Sex	Location	Country	Surgery	Years after surgery
Rajkumar et al.,2003 [6]	1	F	Posterior maxilla	India	Traumatic extraction	5 years
Amin et al., 2003 [7]	1	M	Anterior maxilla	United Kindom	Le Fort I osteotomy	15 years
Bartnik and Bartnik-Krystalska, 2004 [8]	1	NI	Posterior maxilla	Poland	Maxillary sinus	26 years
Cano et al., 2009 [9]	1	F	Posterior maxilla	Spain	Maxillary sinus	3 years
Shakib et al., 2009 [10]	1	F	Anterior maxilla	United Kindom	Le Fort I osteotomy	7 years
Bulut et al., 2010 [11]	1	F	Posterior maxilla	Turkey	Maxillary sinus	8 years
Marano et al., 2012 [12]	1	F	Posterior maxilla	Brazil	Traumatic extraction	5 years
Leung et al., 2012 [13]	3	2 F, 1 M	Posterior maxilla	China	Maxillary sinus	2–26 years
Fernadez et al., 2013 [14]	1	M	Posterior maxilla	Brazil	Maxillary sinus	15 years
Jingang et al., 2014 [15]	1	M	Anterior maxilla	China	Facial trauma	10 years
Coviello et al., 2016 [16]	1	M	Anterior maxilla	Italy	Le Fort I osteotomy	12 years
Niederquell BM et al., 2016 [17]	1	F	Bilateral posterior maxilla	Germany	Maxillary sinus	55 years
Shima et al., 2017 [18]	1	M	Posterior maxilla	Iran	Maxillary sinus	30 years
Gonzalez et al., 2017 [19]	1	M	Bilateral posterior maxilla	USA	Trauma	22 years
Yamamoto et al., 2017 [20]	1	F	Posterior maxilla	Japan	Maxillary sinus	9 years
Lim et al., 2017 [21]	1	M	Posterior maxilla	Malaysia	Maxillary sinus	30 years
Gowthamnath, et al., 2018 [22]	1	M	Posterior maxilla	India	Trauma	2 years
Han, 2018 [23]	1	M	Posterior maxilla	Korea	Maxillary sinus	1 years
Current case report	1	F	Anterior maxilla	Venezuela	Le Fort I osteotomy	5 years

One possible reason is the delayed clinical presentation of this lesion, with most reported cases having presented 10 to 55 years after the initial surgery [7, 8, 13–19, 21]. In this case, the patient developed symptoms approximately 5 years after a Le Fort I osteotomy.

Radiographically, it is a well-defined unilocular radiolucency closely associated with then maxillary sinus [7]. The cyst may be surrounded by a zone of sclerosis. As the cyst enlarges, adjacent sinus wall may become thin and eventually perforated [8]. Although it is usually unilocular, multilocular variants have also been reported [9]. In our report, the wall were not perforated.

In order to determine the extent of the lesion, a maxillofacial CT [22] is important to outline the characteristics of a POMC, which will become very important when planning surgery [2, 5]. In our report, the lesions was evaluated with volumetric CT scan, the orthopantomography does not show any image of the lesion.

The histology of a POMC could be potentially confusing because of the extensive fibrous repair of a previous trauma and could render the diagnosis difficult since the presence of respiratory epithelium is a mandatory finding in the maxillary sinus [12, 13]. Other lesions of the maxillary sinus that must be considered in the diagnosis of a POMC are mucous retention cysts and maxillary pseudocysts [7, 10, 11].

The pathogenesis of the surgical ciliated cyst is thought to be caused by sinus or nasal mucosa entrapment in the bone healing process after maxillary sinus surgery or by the early closure of the natural ostium before the sinus is completely filled with regenerating granulation tissue [7, 11]. In the case reported by Hayhurst et al. [23], it was supposed that during the healing process, small segments of nasal mucosa were entrapped between the bony edges of the maxillary osteotomy. Many years later, this complication resulted in the cystic degeneration and enlargement of the entrapped nasal mucosa. For this reason, many authors [7, 10, 16] and we suggest to suture any tearing of the nasal mucosa in orthognathic osteotomies before osteosynthesis begins and take a preventive measure for the occurrence of this type of cyst may be a more careful attention during surgery to avoid entrapment of epithelium at the surgical site.

The literature describe many cases associated with sinus surgery or midface osteotomies, traumatic tooth extraction, maxillary fracture, or a complication of Caldwell-Luc procedures [6–23]. In our study, we present a case of one patient with postoperative of maxillary orthognathic surgery.

The clinical presentation ranges from asymptomatic to an indolent enlargement causing an esthetic problem or acute swelling and pain, due to secondary infection [25].

In view of its benign nature, surgical ciliated cyst is adequately treated with enucleation of the cyst. This has remained the treatment of choice whenever clinically possible [7, 22]. In cases of large unilocular cyst with extensive bony perforation, marsupialization may be performed.

Recurrence has not been widely described by literature [22]. The incidence of surgical ciliated cyst of the maxilla

secondary to maxillary sinus surgeries should be declining as a result of conservative management of sinusitis and the advent of endoscopic techniques to treat antral disease [2324]. On the contrary, the incidence due to orthognathic surgery may increase as a result of increasing demand for the jaw corrective surgeries.

Conclusions

Although surgical ciliated cysts have only rarely been reported after orthognathic surgery, an increased awareness of this possibility is necessary to avoid delays in diagnosis and this lesion should always be included in the differential diagnosis of symptomatic patients who have previously performed antral or maxillary orthognathic surgery.

Precise studies are necessary to determine the incidence of surgical ciliated cyst in Latin America and Venezuela. Since surgical ciliated cyst of the maxilla is believed to result from entrapment of the epithelium of sinus in the maxilla during surgical procedure, a preventive measure such as careful suturing and patient follow-up is highly recommended.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical disclosure protection of people and animals The authors declare that procedures conformed to the ethical standards of the responsible committee on human experimentation and in accordance with the World Medical Association Declaration of Helsinki.

Confidentiality of data The authors declare that they have followed the protocols of their workplace on the publication of data from patients and that all patients included in the study have received sufficient information and have given their written informed consent to participate in the study.

Right to privacy and informed consent The authors have obtained the informed consent of patients and/or subjects referred to in the article. This document is in the possession of the author of correspondence.

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