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Surgical ciliated cyst of the maxilla. Clinical case

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Abstract
Surgical ciliated cyst is uncommon in Western countries but frequently reported in Asian populations as a delayed complication of surgery, with inclusion in the bone of nasal or sinus mucosa. Isolated cases have also been reported in the mandible after orthognathic surgery. We report a case in the maxillary region three years after radical sinus surgery for chronic sinusitis. Intraoral examination revealed a small painless tumefaction with no color change in the surrounding tissues. Computed tomography demonstrated a well-defined intraosseous lesion lateral to the maxillary sinus. The lesion was completely excised, when histology demonstrated a pseudostratified ciliated epithelial lining. Differential diagnosis from other lesions such as an odontogenic keratocyst or inflammatory cyst were also made. This case report,uncommon in Western countries, of a surgical ciliated cyst illustrates the need for a meticulous surgical technique, proper management of complications, and routine, long-term follow-up of patients undergoing any type of sinus surgery.

Key words: *Ciliated cyst, maxilla.*

Introduction
Surgical ciliated cyst was first reported by Kubo in 1927 (1) as a posterior maxillary cyst after the surgical treatment of maxillary sinusitis. Very few reports on this entity have been published in the English language literature, but it is frequently described in the Japanese literature as a "postoperative maxillary cyst" or "paranasal cyst". It is one of the most common maxillary cysts in Japan, detected in up to 20% of patients after radical maxillary sinus surgery (2). It is a locally aggressive lesion that appears as a delayed complication of surgery in the maxillary sinus region, e.g. orthognathic surgery or Caldwell-Luc radical antrostomy (3). It may appear as a tumor, with expansion of vestibular or palatine bone cortical plates, pain in adjacent facial area and, in some cases, fistulization (4). There are also reports of mandibular localization in which the respiratory epithelium may have been included in the mandibular bone after bimaxillary orthognathic surgery (5). Three cases of mandibular cyst were also described after genioplasty with a basal osteocartilaginous graft (6). We report the case of a patient with surgical ciliated cyst in lateral maxillary region diagnosed after three years of radical antrostomy.

Case Report

A 56 year-old woman came to the Department of Buccofacial Medicine and Surgery (School of Dentistry-UCM-Madrid) reporting discomfort and inflammation in the right maxillary region at premolar level. The patient had no systemic disease of interest. Three years earlier, she had undergone surgery on the right maxillary sinus on the same side due to chronic sinusitis.

Intraoral examination revealed a small tumefaction, with no color change in surrounding tissue (Fig.1a). The patient reported some discomfort when the alveolar process in the lateral maxillary region was palpated. The upper right second premolar had been extracted due to caries. Teeth in the first quadrant were disease-free. A complementary CT scan showed an oval, unilocular, well-defined translucent area with radiodense border intraosseously localized and apical to the upper right first molar. The scan showed no perforation of medial cortex and no communication to maxillary sinus or nasal cavity was found (Fig.1b-d).

Surgery was performed under local anesthesia. After intracrevicular incision in molars and mesial discharge incision, a full-thickness mucoperiosteal flap was elevated, revealing a complete but thinned vestibular cortex (Fig.2a). The pathological tissue was completely excised by blunt dissection through the bone window. It was round (approximately 15 mm in diameter) with a yellowish mucoid content and thin wall, and it was readily detached from the bone (Fig.2b). The excised tissue was fixed in buffered formalin and sent for histological study (Fig.2c). No communication was observed with the sinus or nasal cavity and the bone cavity was not filled with material, closing it with interrupted simple suture.

The pathology report described a cystic formation internally lined with cylindrical ciliated epithelium, with presence of goblet cells and mucoid material and absence of atypia. The wall was composed by fibrous connective tissue with non-specific lymphoplasmocitary inflammatory component. It contained an amorphous material with foamy macrophages. The diagnosis was a non-odontogenic ciliated cyst (Fig.3a-b).

The patient was regularly followed-up after the surgery with no signs of clinical symptoms, and a progressive regeneration of the residual cavity was observed on CT images at one year (Fig.4).

Discussion

In Japan, the reported incidence of surgical ciliated cyst after radical maxillary sinus surgery ranges from 3% to 20%, and cases have reported at up to 49 years after the intervention. This appears to be due to a high prevalence of chronic sinusitis in that country and the use of surgical rather than antibiotic treatment before 1970 (2). The lesion is believed to be caused by sinus or nasal mucosa entrapment in the bone healing process after an osteotomy in the area. It has been recommended to suture any tearing of the sinus or nasal mucosa in orthognathic osteotomies before osteosynthesis and to clean the saw before performing new osteotomies after cutting sinus mucosa (6,7).

This type of cyst usually expands toward the anterolateral wall of the canine fossa, although it can also extend toward the nasal wall or sphenopalatine wall of the sinus. These cysts are occasionally more aggressive and occupy the orbit floor or ethmoidal air cells. Its structure can be unilocular or multilocular (8). Amin (3) re-

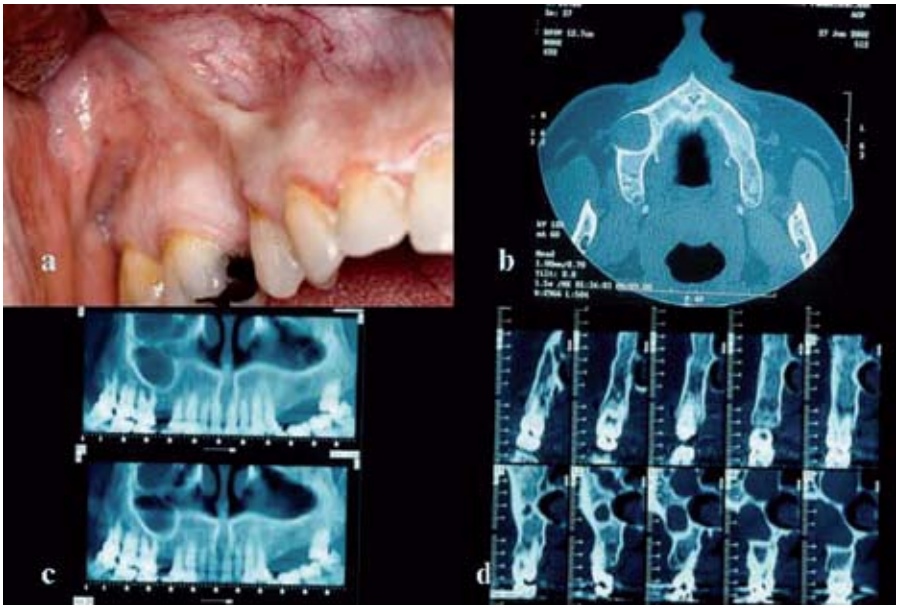

Fig. 1. (a) Intraoperative clinical view; (b,c,d) Preoperative computed tomography (CT) study.

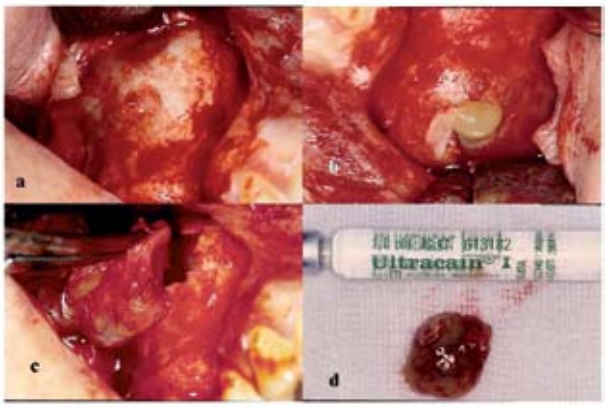

Fig. 2. (a) Mucoperiosteal flap; (b,c) Lesion excision (d) Tissue obtained for histological study.

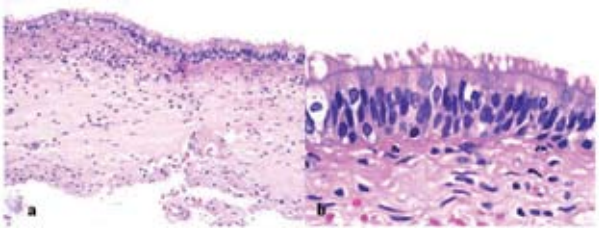

Fig. 3. (a) Histology at lower magnification; photomicrograph of lesion (HE, x200) showing capsule of connective tissue with thin epithelial lining; (b) Histology at higher magnification; photomicrograph of lesion (HE, x400), showing connective tissue lined with pseudostratified ciliated columnar epithelium.

Fig. 4. CT study at one year after surgery.

ported a lesion that appeared at 5 years after maxillary advancement surgery, with evident upper displacement of the sinus floor, communication with nasal fossa, and perforation of palatine bone. Because of the large size of the cyst, the cavity was filled with gauze and iodine-formalin paste for 48 hrs before primary closure.

The most common localization in the maxilla is the lateral wall, although they have also been reported at the infraorbital ridge and medial canthal region (4). One case of post-traumatic ciliated cyst was also reported at 6 months after a sinus floor elevation with particulated bone harvested from the skull. A 1-cm diameter lesion was observed in the center of the grafted area, which was treated with total enucleation, filling the cavity with iliac crest particulate graft. The presence of a sinus mucocele before the first graft procedure appears to have facilitated mucosal entrapment (9).

Maruyama et al. (10) conducted a histological and immunohistochemical study of 360 ciliated cysts: 66% of their length was pseudostratified ciliated epithelium, 28% transition epithelium, and 6% squamous epithelium. Goblet cells were abundantly present in all cysts except in areas with squamous epithelium. The number of goblet cells was correlated with the presence of inflammatory cells. It was also observed that sialylated glycoconjugates derived from lecithins in goblet cells were correlated with cyst wall inflammation and cyst growth.

Surgical ciliated cyst should be differentiated from mucous retention cyst (pseudocyst or mucocele) of the maxillary sinus, which appears as an accumulation of inflammatory exudates, presents a lining of pseudostratified ciliated columnar epithelium, and is sometimes correlated with a history of trauma (11). However, it is not found in an intraosseous location, shows a more benign behavior, and is usually symptom-free (3). Respiratory epithelium can also be observed in odontogenic cysts, attributed to the pluripotentiality of odontogenic epithelium. In this case the presence of this ciliated columnar of epithelium, not a stratified squamous epithelium, and other clinical aspects, specially the absence of inflammatory signs were required for the differentiation with an inflammatory cyst.

Odontogenic keratocysts can also have foci of ciliated epithelium but are distinguishable from ciliated cysts because the latter do not usually have keratinizing capacity or keratinized epithelial lining in any area (12), and the histopathological features of the odontogenic keratocyst are clearly distinguished from other types of odontogenic and non odontogenic cysts.

This uncommon case report, in Western countries, of a surgical ciliated cyst illustrates the need for a meticulous surgical technique, proper management of complications, and routine, long-term follow-up of patients under any type of sinus surgery.

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