








Post-surgical ciliated cyst: a case report in two brazilians patients and literature review

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Aim: The aim of this study is to report two cases of post-surgical ciliated cysts (PSCC) in patients from Brazil with a history of trauma in the maxillary zygomatic suture region and traumatic tooth extraction. Additionally, we conducted a literature review of PSCC. **Methods:** We reported two clinical cases of edentulous patients diagnosed with PSCC, including information on patients medical and surgical history. Furthermore, we performed a literature review, identifying previous cases of PSCC and compiling relevant information. **Results:** In Case 1, the patient, a 64-year-old female, was edentulous and asymptomatic. The approach involved obtaining a panoramic radiograph, performing an incisional biopsy, followed by enucleation and curettage of the lesion. The patient is currently under follow-up, with no evidence of recurrence. In Case 2, the patient, a 61-year-old male, was also edentulous and had no painful symptoms. Imaging tests were ordered, and he subsequently underwent an excisional biopsy. Unfortunately, the follow-up was lost. According to the literature review, patient ages ranged from 17 to 76 years, with males comprising 52% and females 48% of the cases. Pain, swelling, and tenderness are common symptoms in the affected area. Le Fort I osteotomy was frequently associated with PSCC cases. Enucleation or curettage emerged as the predominant treatment approach for PSCC, demonstrating favorable outcomes with rare recurrences. **Conclusion:** Considering the diagnosis of PSCC is crucial in patients with a history of prior maxillary surgeries. Additionally, a comprehensive patient history is imperative for the assessment of maxillary cystic lesions.

Keywords: Cysts. Maxilla. Maxillary sinus. Postoperative complications.



Introduction

A post-surgical ciliated cyst (PSCC) is a benign uncommon lesion that develops after maxillary sinus surgery or trauma. It is also reported as a postoperative maxillary cyst, paranasal cyst, respiratory implantation cyst, surgical implantation cyst, and respiratory mucocoeles¹⁻³. This cyst was first described by Kubo in 1927 and recently was recognized as an odontogenic cyst⁴. Many cases of PSCCs have been reported in Asians, especially in the Japanese population, representing approximately 20% of all reported PSCCs⁵. This condition usually manifests in adults in the 4th and 5th decade without gender predilection⁶. Despite it being well-studied in Japanese literature, it is rare in the Western population⁷.

The most frequently affected oral site is the posterior maxilla. Commonly, the lesion appears as an expanding swelling in the maxillary sulcus with occasional pain and purulent discharge, if secondarily infected⁶. The histopathological presentation consists of a cyst covered by pseudostratified ciliated columnar epithelium or cuboidal epithelium with occasional goblet cells⁵. Areas lined by nonkeratinized stratified squamous epithelium can also be observed⁶. Radiographically, it is a well-demarcated unilocular or multilocular radiolucency with a sclerotic border⁵. The adjacent sinus wall could decrease in thickness and eventually puncture as the cyst enlarges³. The conventional treatment consists of enucleation or curettage of the cyst, with a good prognosis and rare recurrences^{3,6}.

This study aims to provide a comprehensive report of two cases of post-surgical ciliated cysts in patients from Brazil. Additionally, a literature review of post-surgical ciliated cysts is provided.

Materials and Methods

Case Report 1

A 61-year-old male patient presented to the Surgical Clinic of the Brazilian Dental Association in Governador Valadares for dental treatment. The reason for the visit was an asymptomatic increase of volume on the right side of the maxilla. The evolution time was unknown. A panoramic radiographic was performed, which showed a radiolucent image with cortical fracture on the right side of the maxilla. There was also fixation of the infraorbital rim due to trauma in the region of the maxillary zygomatic suture (Figure 1A). The patient mentioned that he had undergone maxillary surgery after a traumatic event several years earlier.

Incisional biopsy of the lesion was performed. Histopathologic analysis stained with hematoxylin and eosin (H&E) revealed a cyst lined by pseudostratified columnar ciliated epithelium accompanied by a fibrous cystic capsule (Figures 1B and C). The diagnosis was postoperative ciliated cyst. The patient underwent enucleation and curettage of the lesion by an oral and maxillofacial surgeon. The patient is currently being followed in the surgical service.

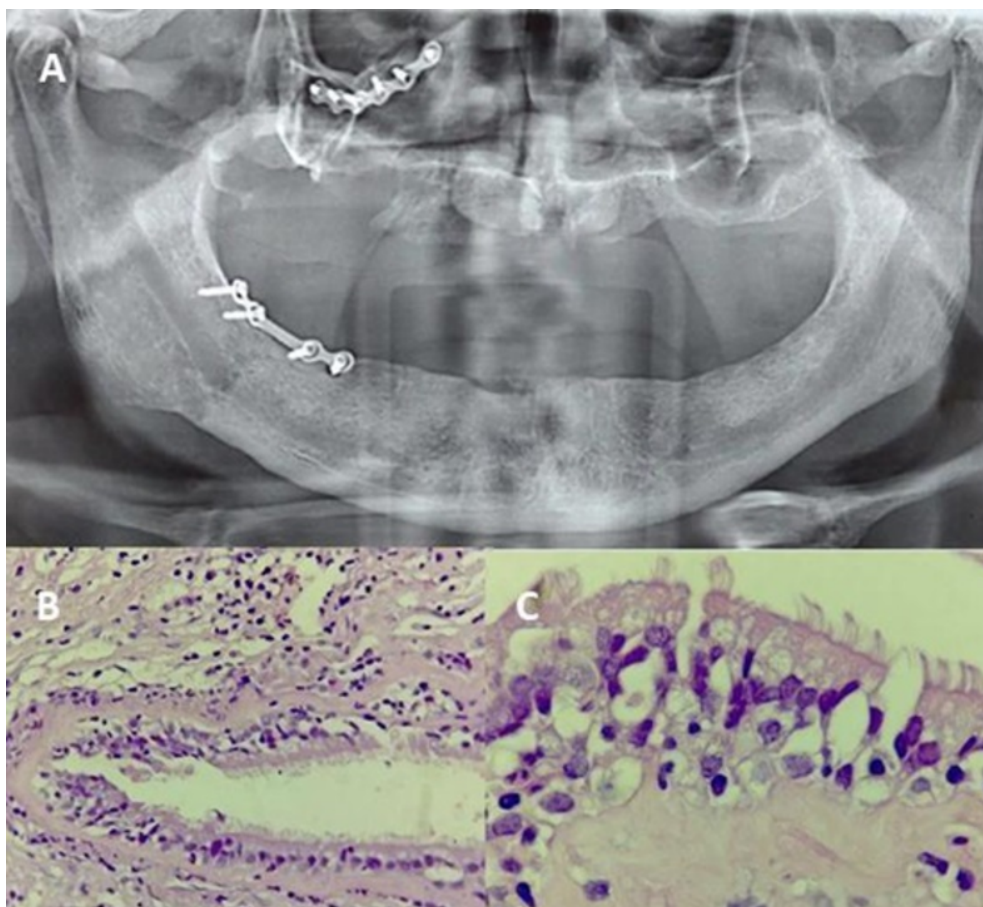


Figure 1. Case 1. 1A: Panoramic radiographic showing a radiolucent image with a ruptured cortical on the right side of the maxilla. Fixation of the infraorbital rim is also observed. 1B: Histopathological examination reveals a cyst lined by pseudostratified ciliated columnar epithelium and fibrotic capsule with minimal inflammation (10X, Hematoxylin, and Eosin). 1C: In detail, ciliated epithelium, and mucous cells (100X, Hematoxylin and Eosin)

Case Report 2

A 64-year-old female patient presented to the Brazilian Dental Association complaining of an asymptomatic increase in volume on the right side of the face, accompanied by intense suppuration. The lesion had been slowly growing over several years, with no specific timeframe provided by the patient. During the medical history, the patient mentioned a previous puncture performed by another healthcare professional. Then, a cone beam computed tomography was performed and revealed a hypodense lesion in the right maxilla (Figure 2A). An excisional biopsy of the lesion was subsequently performed.

Histopathologic examination revealed an irregular fragment measuring approximately 4.5 x 2.5 x 0.3 cm, with a brownish color and a firm, elastic consistency. Microscopic analysis revealed a cystic cavity lined by respiratory epithelium with ciliated areas and mucous cells, as well as areas of squamous epithelium and fibrous

capsule (Figure 2B and C). After histopathologic evaluation, the patient reported having undergone several traumatic tooth extractions in the region. Based on the histopathology report and its correlation with clinical and radiographic data, the diagnosis of postoperative ciliated cyst was made. Unfortunately, the patient was not followed up.

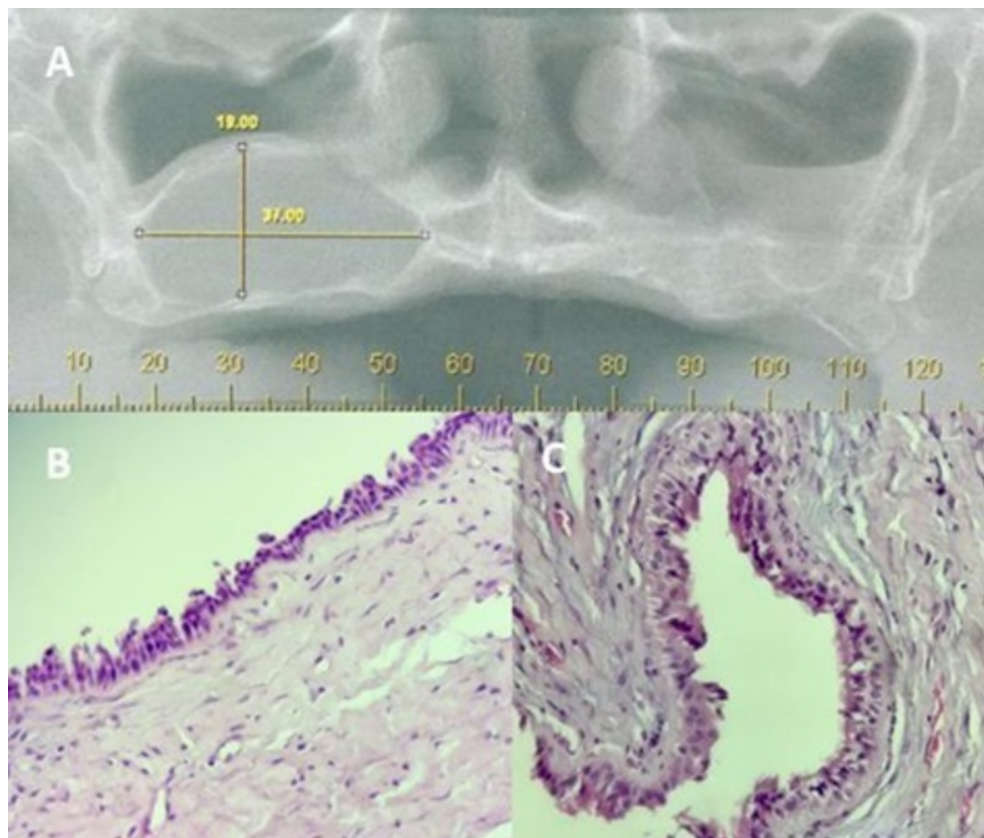


Figure 2. Case 2. 2A: Panoramic reconstruction showing a soft tissue density image on the right side of the maxilla. 2B and C: Histopathological examination reveals a cyst lined by respiratory epithelium and fibrotic capsule without inflammation (10X and 40x respectively, Hematoxylin and Eosin).

Literature Review

A literature search was performed on the electronic databases: Scopus, EMBASE, and Pubmed/MEDLINE. The search strategy contained the keywords 'surgical ciliated cyst jaw', 'surgical ciliated cyst maxilla', 'surgical ciliated cyst mandible', 'surgical ciliated cyst maxillary', and 'surgical ciliated cyst mandibular.'

Inclusion criteria comprise cases and case series involving surgical ciliated cysts with sufficient clinical, radiological, and histological data to confirm. The literature search was not restricted to English-language literature. Meeting/conference abstracts and articles with no full text available were excluded.

The search was performed by two reviewers who selected relevant titles and abstracts based on inclusion criteria. Those that met the eligibility criteria were selected for full-text analysis. For each study included, the following data were extracted: authors' name and affiliation (country), year of publication, number of cases reported, age, sex, location, previous surgical history, the time between surgery and diagnosis, treatment, recurrence, and follow-up.

Discussion

In the literature, 62 patients were diagnosed with PSCC, with a slightly predominance of male (n=32). The age range of the patients was between 17 and 76 years old. In this study, one female and one male patient, both in their sixties, were reported. Regarding site, the maxilla was the most affected^{1-5,7-30} which is consistent with our reports, where both patients had lesions in the maxilla.

PSCC may present clinically as an expansion of the vestibular or palatal cortical bones and swelling in the adjacent facial region^{2,5,6,8-12,15,17,21,22,24-26,28-33}. Pain^{3,5-8,10,12,15,21,29-31} and purulent discharge³¹ are noted due to secondary infection as well as paresthesia⁶. Some patients were asymptomatic and discovered the lesion incidentally during radiographic examination^{5,15,34}. In Case 1, there is cortical expansion in the right maxillary region, which is associated with a history of trauma involving the maxillary zygomatic suture. In Case 2, the patient reported a history of traumatic tooth extraction and presented with facial swelling accompanied by intense suppuration.

Radiographically, PSCC usually presents as a well-demarcated unilocular radiolucency with sclerotic borders, as observed in case 1. Meantime, there are cases present with a multilocular radiolucency with an indistinct border^{11,17}. A Cone Beam Computed Tomography (CT) is a gold standard complimentary exam. In case 2, a Cone Beam CT showed a soft tissue density image on the right side of the maxilla.

Histologically, a respiratory-type pseudostratified ciliated columnar epithelium is observed, but transitions to simple, cuboid, or squamous columnar epithelium are also described^{11,14}. The pathogenesis of this cyst involves the entrapment of segments of the respiratory epithelium between the bony edges of the maxillary osteotomy, potentially leading to physical migration implantation of the sinus mucosa in an ectopic site^{1,8,14}. The enlargement of the cyst by the osmotic difference leads to the destruction of bone and adjacent structures¹¹.

PSCC typically develops nearly two decades after primary surgery, although there are reports of a relatively short latency period^{8,9}. This cyst is usually associated with a history of surgical intervention in the maxillary sinus, such as Caldwell-Luc^{11,16,20,23}, orthognathic surgery^{6,32}, traumatic tooth extraction^{2,12,29} and trauma^{13,23,27}. The most commonly reported prior surgical procedure was Le Fort I osteotomy^{1,3,5,8,10,14,24,29-31,33,34}. The time interval between any surgery and the diagnosis of PSCC ranged from 2 to 56 years. In the present report, both cases were associated with a history of surgical interventions in the maxillary region.

PSCC represents one of the most common maxillary cysts in Japan, exhibiting a prevalence close to 20% among adults in the 4th and 5th decade, with no gender predilection⁵. However, PSCC may be more common outside Japan than pre-

viously suggested, potentially attributable to underdiagnosis or misdiagnosis. A study conducted in England with 23 reports of PSCC showed that all patients reported a history of prior antral surgery, with the duration between this surgical intervention and the presentation of the cyst ranging from 7 to 39 years, with a mean interval of 20 years¹⁵. The prevalence of PSCC may improve due to the increasing number of cases of orthognathic surgery performed for aesthetic purposes³³. However, it is expected a reduction in the incidence of PSCCs secondary to maxillary sinus surgeries due to a recommended conservative management of sinusitis by endoscopic techniques^{1,3,16}.

The development of PSCC is rare in the mandible^{6,31-35} and can be attributed to the entrapment of the Schneiderian membrane by surgical instruments³⁴. A reported case of PSCC in the mandible was described with a history of simultaneous maxillary and mandibular orthognathic procedures. They theorize that the maxillary tissue, which includes respiratory epithelium, was implanted in the mandible. The authors show that the possibility that the cystic lesion was of odontogenic origin was ruled out and conclude that in the absence of information on the location of the cyst, a pathologist could easily misinterpret the findings³².

The diagnosis is not possible with only clinical or radiological features, as these may resemble other cystic lesions, especially odontogenic cysts¹⁵. It is challenging due to the scarcity of cases described in the literature and due to the similarity with other cystic lesions. Odontogenic keratocyst, incisive canal cyst, median palatine cyst, and antral mucocele are considered differential diagnoses of PSCCs². Mucous retention cysts (pseudocysts) should be considered in the differential diagnosis, as they are also lined by a pseudostratified ciliated columnar epithelium. Cystic expansion that affects adjacent teeth makes radiological characteristics like odontogenic inflammatory cysts¹¹. The respiratory epithelium can also be observed in radicular cysts, due to the pluripotential of the odontogenic epithelium¹⁸. Vitality tests are necessary to clarify the origin of the cyst in cases of doubt between traumatic or endodontic origin. Thus, the association of clinical, radiological, and histopathological data is required for diagnosis.

In most cases, surgical treatment is chosen. This typically involves curettage^{6,33}, enucleation^{1-3,5-7,9,11-14,16,18,20-30,32,34,35}, or a combination of both techniques^{8,10,31}. The prognosis is favorable, with recurrence considered rare and was described only in 3 cases^{6,10,30}. Typically the recurrence results from incomplete excision leading to residual cyst lining^{5,19}. In the present cases, enucleation was performed via an intra-oral approach and in case 1 recurrence was not observed.

Clinicians must always be concerned about radiolucent lesions involving the jaws. Regardless of the origin and cause, an active investigation is necessary aiming at an adequate treatment⁷. In the two reported cases, the patients are edentulous, and both have a previous history of local surgery. A comprehensive and detailed patient history through anamnesis is paramount to guiding the final diagnosis¹⁶.

In conclusion, the reported cases represent two cases of PSCC, a female and a male patient, in the 6th decade of life. In both cases, the patients were edentulous, and the site of involvement was the maxilla. History of traumatic tooth extraction and

post-trauma surgical procedures were reported by patients. According to the literature review, the patients' ages range from 17 to 76 years. Considering gender, 48% are female and 52% are male. Le Fort I osteotomy is the most cited in previous surgical history and clinically and some patients are asymptomatic or discovered the lesion accidentally (radiography). Swelling, pain and tenderness are the symptoms most frequently reported by patients.

According to evolution, it is not possible to define a pattern of age and time of development of the PSCC since there is variation in these data in the literature. There are numerous radiographic and histopathological variations related, which may mimic other cystic lesions. Thus, it is necessary previous knowledge to include PSCC as a diagnostic hypothesis, especially in cases with a history of previous surgery. Thus, it further reinforces the importance and necessity of carrying out a detailed and complete anamnesis.

Conflict of Interest

None.

Data Availability

Datasets related to this article will be available upon request from the corresponding author.

Author Contribution

Iara Vieira Ferreira: Conceptualization, data curation, formal analysis, methodology, writing – original draft. **Laís Heringer Mendes Coelho:** Conceptualization, data curation, formal analysis, methodology, writing – review & editing. **Francielle Silvestre Verner:** Formal analysis, methodology, writing – review & editing. **Rose Mara Ortega:** Formal analysis, methodology, writing – review & editing. **Larissa Stefhanne Damasceno de Amorim Póvoa:** Formal analysis, methodology, writing – review & editing. **Celso Henrique Najjar Rios:** Formal analysis, methodology, writing – review & editing. **Sibele Nascimento de Aquino:** Conceptualization, data curation, formal analysis, methodology, writing – review & editing.

All authors actively participated in the manuscript's findings and have revised and approved the final version of the manuscript.

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