

Non-Surgical Ciliated Cyst of the Maxilla – An Unconventional Variant

Abstract

The term surgical ciliated cyst of the maxilla is a designation for cysts of the maxillary sinus conventionally associated with surgery and trauma. Surgical ciliated cysts with a noncontributory history of surgery or trauma can pose a diagnostic challenge. We report an interesting case of ciliated cyst of the maxilla in a 54-year-old male patient. The present case provides a plausible explanation for the occurrence of ciliated cyst of the maxilla lacking history of surgery or trauma.

Keywords: *Cyst, dental caries, endodontic inflammation, epithelium, maxillary sinus, oroantral fistula*

Introduction

Surgical ciliated cysts of the maxilla have been conventionally reported to be associated with a history of surgery or trauma to the maxilla. However, cases resembling the histopathological diagnosis of surgical ciliated cyst of the maxilla without a prior surgery or trauma need to be critically discussed. To the best of our knowledge, an extensive literature search revealed that only two similar cases have been reported.^[1,2] Here, we present an interesting case of (non)-surgical ciliated cyst of the maxilla in a 54-year-old male and a novel plausible explanation for the occurrence of the same. We intend to report this case as it is an unusual presentation of common pathology which has an important clinical implication, along with a view of suggesting a novel pathogenesis for its occurrence.

Case Report

A 54-year-old healthy male presented with a chief complaint of pain and swelling in the right maxillary posterior region for 3 months. The patient denied any history of sinusitis, surgery, or trauma involving the right maxillary sinus [Table 1]. Extraorally, no facial deformity, draining sinus, or cervical lymphadenopathy was noted; additionally, the overlying skin appeared normal. Intraorally, a solitary, well-defined soft cystic swelling measuring

about 2 cm × 3 cm, extending from the permanent maxillary right first premolar to the permanent maxillary right first molar obliterating the buccal vestibule, was observed. No right maxillary sinus involvement was evident. The overlying mucosa was inflamed, nonulcerated without any intraoral sinus drainage. The permanent maxillary right second premolar presented as a nonvital, grossly carious root piece with pain on percussion. The permanent maxillary right first premolar and permanent maxillary right first molar were vital on electric pulp testing. Based on the clinical findings, differential diagnosis of radicular cyst, odontogenic keratocyst (OKC), ameloblastoma, and a benign fibro-osseous lesion (BFOL) were considered. The orthopantomograph showed a well-defined unilocular radiolucency associated with the root apices of the permanent maxillary right first premolar, second premolar, and first molar. The floor of the maxillary sinus was raised [Figure 1]. Cone-beam computed tomography showed corticated radiolucency, bicortical expansion, thinning, and inward displacement of the right lateral nasal wall. A radiographic differential diagnosis of radicular cyst, OKC, and BFOL was contemplated [Table 2]. Based on the clinico-radiologic features, a provisional diagnosis of infected dental cyst was rendered.

The lesion was surgically enucleated under general anesthesia and no direct communication, or sinus perforation was observed during surgery. Gross examination

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Table 1: Timeline of history, diagnostic assessment, intervention, and follow-up

Clinical feature	Timeline of occurrence
Painful swelling in the right maxillary posterior region	1 month (September 2020)
Gradual increase in size	Next two 2 months (October–November 2020)
Clinical visit, detailed history taken, and provisional diagnosis of infected dental cyst based on clinical diagnosis	November 3, 2020
Panoramic radiographic findings	November 6, 2020
Consent followed by enucleation and specimen sent for histopathological diagnosis	December 1, 2020
Histopathological diagnosis	5 days after excisional biopsy (December 5, 2020)
Patient kept under follow-up	2 weeks postsurgery showed uneventful healing (December 18, 2020)
	6 months postsurgery – no recurrence (June 5, 2021)

Table 2: Clinical and radiographical differential diagnosis

Clinical diagnosis	Radiographic diagnosis
Radicular cyst	Radicular cyst
OKC	OKC
Ameloblastoma	BFOL
BFOL	

OKC: Odontogenic keratocyst; BFOL: Benign fibro-osseous lesion

revealed a single, soft, brownish cystic specimen measuring about 3 cm × 2 cm in size. No mural growth was noticed within the lumen. The histopathological examination revealed a cystic lumen lined by the maxillary sinus epithelium comprising pseudostratified columnar ciliated type with few mucous cells and infected dental cyst lining in a minor area. The cyst wall contained intense chronic inflammatory cell infiltrate and cholesterol clefts. The final diagnosis of nonsurgical ciliated cyst of the maxilla was rendered [Figure 2]. The etiology thus proved to be an endodontic infection. The patient was kept under follow-up for 2 weeks. The postoperative healing was uneventful.

Patient perspective

A misdiagnosis could have led to persistent maxillary sinusitis and subsequent extensive involvement of the sinus, hence accurate diagnosis through histopathology is warranted.

Review of literature

A PubMed database literature search was performed to analyze the etiology and latency of the postoperative maxillary cyst. Search strategy contained the keywords: “postoperative maxillary cyst,” “surgical ciliated cyst,” and “postoperative paranasal cyst.” A total of 424 articles were found, out of which only 18 were of the maxilla and 2 cases were of the mandible. Results were limited to English, and only cases without a history of trauma or surgery were included.

Discussion

The surgical ciliated cyst first described by Kubo in 1927, usually originates after a surgical procedure involving the



Figure 1: Panoramic radiograph showing a well-defined unilocular radiolucency associated with the root apices of the permanent maxillary right first premolar, second premolar, and first molar raising the floor of the right maxillary sinus

maxillary sinus, like the Caldwell-Luc surgery or dental extraction associated trauma.^[3] Its attribution to a surgical history, was due to a high incidence of this cyst among Japanese subjects who underwent sinusotomy during World War II when antibiotics were unavailable.^[4]

The most accepted theory is the postsurgery iatrogenically implanted respiratory epithelium that contaminates, proliferates, and develops into a cyst.^[3,5,6] Yamamoto *et al.* suggested that this cyst could have developed by the entrapment of the Schneiderian membrane without its visible perforation or tearing post sinus augmentation.^[5] Similar mechanism can be proposed during the self-healing of oroantral communication. Only one case of a surgical ciliated cyst of maxilla being developed without a history of trauma or surgery, has been reported in the previous literature. Herein, the author had suggested three mechanisms like, the migration of cells from the maxillary sinus or nasal cavity, metaplasia of stratified squamous epithelium and differentiation from totipotent cells for its pathogenesis.^[1]

The review of literature of surgical ciliated cyst of the maxilla showed that there were two cases without a history of trauma, and were associated with nonvital teeth [Table 3].^[1,2]

In the present case, there was no history of surgery, the tooth associated with the lesion was carious and nonvital

Table 3: Previous literature review of surgical ciliated cysts without previous history of trauma or surgery involving the maxillary sinus

Author	Year	Age (years)	Gender	History
Miller <i>et al.</i>	1988	21	Male	Endodontic treatment and apicectomy
Martinelli-Kl�ay C P <i>et al.</i>	2017	35	Male	Endodontic periodontal lesion; tooth negative for vitality
Present case	2021	54	Male	Nonvital tooth with apical periodontitis

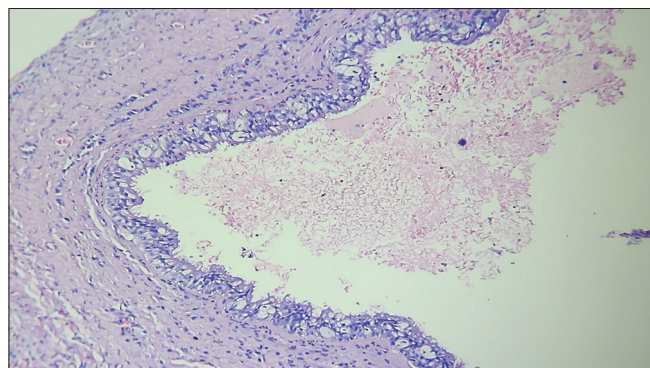


Figure 2: Histologic examination showing cystic lumen lined by pseudostratified ciliated columnar epithelium covering chronically inflamed connective tissue capsule (H and E, x10)

on electric pulp testing. The histological examination revealed features of infected dental cyst in a confined area and the rest of the lining composed of the maxillary sinus epithelium. Our case questions the development of surgical ciliated cyst of the maxillary sinus associated with the history of surgery or trauma involving the maxillary sinus. Hence, the word “surgical” in the term of surgical ciliated cyst needs to be critically discussed. We hypothesize that chronic periapical infection could have resulted in a microscopic oroantral communication leading to a prolapse and merging of the maxillary sinus epithelium with the infected dental cyst lining as observed in our case [Figure 3]. This being only the third case to be documented in the literature of a surgical ciliated cyst of the maxilla developing without previous trauma or surgery of the maxillary sinus, distinguishes it as a rare phenomenon which needs to be considered.

A clinical differential diagnosis that was considered was radicular cyst, OKC, ameloblastoma, and BFOL. A radiographic differential diagnosis contemplated for the periapical radiolucency, with well-defined border, was radicular cyst, OKC, and BFOL [Table 2]. The presence of a decayed and nonvital tooth along with a well-defined periapical radiolucency favored the consideration of radicular cyst as the provisional diagnosis.

Conclusion

We would like to propose the term “(Non)-surgical ciliated cyst of the maxilla” to designate those variants of surgical ciliated cysts of the maxilla associated with infectious and nonsurgical etiology.

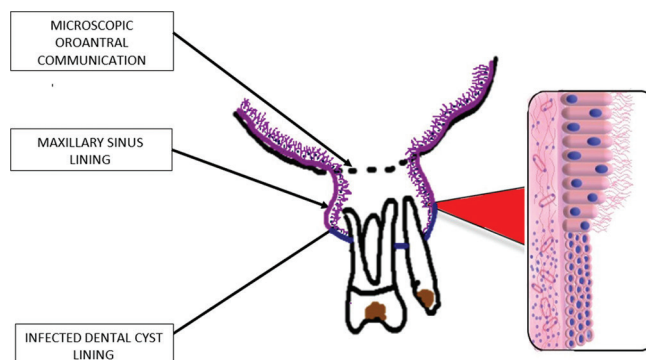


Figure 3: Schematic diagram depicting merging of pseudostratified ciliated columnar epithelium of the maxillary sinus with infected dental cyst lining

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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