**Surgical Ciliated Cyst of the Left Maxilla - A Case Report of Unusual Pathogenesis**

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**Abstract**

**Introduction:** Surgical ciliated cyst (SCC) is a type of true epithelial lined antral cysts, which mostly occurs in maxilla from the sinonasal mucosa and gland, comprising 20% of all oral cysts in Japan. It is also called as postoperative maxillary cyst or postoperative paranasal cyst. The etiology is believed to be post trauma or surgery in the region of maxillary antrum. **Patient concerns:** Patient’s concern is less evident in this cystic pathology, because this cyst even though an aggressive cyst, presents with fewer symptoms that can be uncomfortable to the patient or push them to seek medical attention. Often, this becomes an incidental finding during routine postoperative follow-up. **Rationale:** The rationale behind this report is to emphasize even a traumatic extraction of tooth can damage the floor of the sinus and lead to the cyst formation. **Report:** We report a case of SCC in a 76-year-old female of the left maxillary sinus possibly after a traumatic extraction of the tooth, for which enucleation of the cyst was done as a primary modality of treatment. **Diagnosis:** Diagnosis was made after a computed tomography (CT) scan and incisional biopsy. Diagnosis of such rare cases poses a great challenge due to its paucity in etiology, and it often requires a CT scan that becomes an essential gold standard investigation. **Outcomes:** Patient is currently under one year follow up and without any signs of recurrence. **Lessons:** It should be acknowledged that tampering trauma or with sinus mucosa can trigger the formation of a SCC of the maxilla, and it should always be considered as a possible sequel after any procedure of such potential in the maxilla.

**Keywords:** Delayed complication, extraction, maxilla, postoperative maxillary cyst, sinus surgery, swelling, trauma

**INTRODUCTION**

Postoperative maxillary cyst was originally described by Kubo in 1927, which is also known as surgical ciliated cyst (SCC), postoperative paranasal cyst, or respiratory implantation cyst. [1] Gregory and Shafer first reported these cystic lesions in English as “surgical ciliated cysts of the maxilla.” [2] By theory, entrapment of the sinus membrane during the closure of wounds after surgical procedures such as traumatic extraction [3] or trauma to face [4] or surgery in the maxillary sinus region, e.g., orthognathic surgery, [5] maxillary sinus floor augmentation, [6] and Caldwell-Luc radical antrostomy, [7] would result in an inflammatory process that would stimulate the epithelial proliferation and the expansion of the cyst, would be resulted by the osmotic pressure. [7]

In japanese literature, postoperative maxillary cyst has been reported as a delayed complication of Caldwell-Luc antrostomy for chronic sinusitis, as well as few reports as complication after events such as sinus lift procedures and traumatic extraction.

We report a unique case of SCC of the left maxilla in a 76-year-old female with a history of traumatic extraction. The uniqueness of this case is the development of maxillary SCC with a relatively short latency period and the fact that tampering or trauma of Schneiderian membrane of the sinus floor can give rise to a postoperative maxillary cyst.

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**Case Report**

A 76-year-old female patient presented to the Department of Oral and Maxillofacial Surgery with the chief complaint of swelling in the left palatal region for the past 2 months. The patient had a history of loss of the upper left second molar tooth three years ago when the patient herself plucked it out due to pain and presumably damaging the alveolar socket in the process. Further, the patient had a history of extraction of the first molar tooth 5 months back elsewhere, supposedly a traumatic extraction of extended duration, following which the patient has been informed of possible involvement of the sinus floor after the procedure by the previous dental practitioner. The patient’s past medical history revealed to be uncontrolled diabetic, hypertensive and under anticoagulants. There was no previous history of trauma, previous surgeries, or maxillary sinusitis.

On extraoral examination, mild swelling was evident in the left upper cheek region and the patient also had paresthesia on the left infraorbital region and left nose blockage for a duration of one month. On intraoral examination, palatal swelling of 2.5 cm × 3 cm was confined to premolar and molar region, not extending across the midline with obliteration of the left buccal vestibule and expansion of the alveolar segment over the molar region [Figure 1]. Skin overlying the swelling was of normal color, and the swelling was slightly tender on palpation but not pulsating. The premolar teeth were not tender to percussion. Periodontal pocketing was absent and surrounding gingiva was of normal color and appearance. All teeth were vital. No intraoral or extraoral sinus openings were identified.

Preoperative computed tomographic (CT) scan showed an extensive radiolucent unilocular cystic lesion within the left maxillary sinus measuring 35 × 31 × 34 mm, which obliterated the left osteomeatal complex superiorly till the left infraorbital foramen. Resorption of both anterior and posterior wall of the maxillary sinus was evident [Figure 2]. Expansion of palatal and buccal cortex is noted. Straw-colored fluid was aspirated from the lesion and the size of the lesion was reduced. An incisional biopsy was performed through the buccal vestibule, and the result suggested the presence of unspecified odontogenic cyst. Provisional diagnosis was made as an odontogenic keratocyst, and the decision was made based on the above findings to perform enucleation of the cyst under general anesthesia. On preoperative investigations, electrocardiogram reports of the patient showed abnormalities, and the patient was declared unfit for surgery under general anesthesia. Over the next 1½ months, her blood glucose and blood pressure were brought under control and the cardiologist opinion and fitness were obtained to perform surgery under general anesthesia. The size of the cyst remained the same.

As planned previously, enucleation of the cyst in toto with the sinus lining was done under general anesthesia. The fluid from the cyst was dark, dirty colored, and the specimen was sent for histopathological examination [Figures 3 and 4]. The bony cavity was packed with Bactigras to prevent hematoma formation or secondary infection. The wound was then closed primarily with a small opening left for removal of gauze after a week. Healing was uneventful.

Excisional biopsy reported a cyst lined by thin epithelium composed mainly of pseudostratified ciliated columnar type, underlying dense connective tissue showed areas of hyalinization and entrapped squamous cells and few areas of chronic inflammatory cells which confirmed the diagnosis of surgical ciliated cyst of the left maxillary sinus [Figures 5 and 6]. The patient was reviewed on a regular basis. There was no abnormality detected both clinically and radiographically during the review visit after 6 months.

**Discussion**

In the early 1970s in Japan, the reported incidence of SCC after radical maxillary sinus surgery ranges from 3% to 20%, and cases have been reported from six months and up to 49 years after the intervention. The SCC is a locally aggressive lesion, usually developing as a delayed complication many years after radical surgery in the maxillary sinus region or mandibular region. It is theorized that with bi-jaw surgeries, usage of same instruments for the maxilla as well mandible may lead to entrapment of maxillary sinus epithelium in mandible resulting in post-operative cyst formation in mandible.

Various other causes are maxillary osteotomies (5.5%), odontologic events (9%), fracture, angiofibromas, sinus lift procedures, and trauma. The lesion is believed to be caused by sinus or nasal mucosa entrapment in the bone healing process after an osteotomy in the area. Occurrence of postoperative cyst of the maxillary sinus varies from 0.1% to 19.5% among the jaw cysts. Clinical characteristics of the postoperative maxillary cysts are swelling or pain in the buccal region and discomfort of the maxilla or maxillary teeth. This cyst may remain asymptomatic for many years.

It was difficult to arrive at the pathogenesis of development of the cyst in this case due to variations in the incisional and excisional biopsy histopathological report. Development of SCC usually occurs as a delayed complication postsurgery. The time period between the patient’s history of extraction and development of this cyst was short. The duration between the presumed etiology which is traumatic extraction and the development of the cyst makes this case a peculiar one in comparison to other case reports. Hence, this particular case stands out for its unusual duration of pathogenesis and its cause, as stated by few theories: traumatic theory states that traumatic injury to the sinus lining initiates the cyst formation and inflammatory theory states that the continuous inflammatory process inside the congested nonaerated sinus after surgery can lead to cyst formation. The pathogenesis of our case could be after the traumatic extraction of second molar tooth, leading to entrapment of sinus lining and its proliferation. Cyst formation would have been due to difference in osmotic pressure in a relatively short latency period. As such, it is questionable if patients are susceptible to such phenomenon.
Sufficient evidence in existing literature is lacking regarding this notion. To our knowledge, only two cases of SCC after traumatic extraction have been reported in literature.\cite{3,16}
Table 1: Review of literature of surgical cysts due to traumatic extraction

| Author                | Number of cases | Site               | Age and sex | Years after extraction |
|-----------------------|-----------------|--------------------|-------------|------------------------|
| Rajkumar *et al.*, 2003 | 1               | Posterior maxilla  | 27/female   | 5                      |
| Fernandes *et al.*, 2013 | 1               | Posterior maxilla  | 63/male     | 15                     |
| Present case          | 1               | Posterior maxilla  | 76/female   | 5 months               |

[Table 1]. However, in the second case,[16] the patient had both a history of extraction (no clear history of traumatic force) and maxillary sinus surgery 15 years back.

Surgical ciliated cyst of maxilla (SCCM) has been detected anywhere from 6 months to 49 years following initial surgery.[17] The time and type of surgical procedure influences the age at which the SCC becomes apparent. The age ranges from 21 to 72 years.[18] In our present case, the duration of the cyst formation would be roughly 5 months from extraction. Considering the size and duration of the cyst, it is quite aggressive in nature, with perforation of palatal bone and extending superiorly to the infraorbital foramen. Usually, in radiographs, it is seen as a well-defined radiolucent lesion separated from the sinus lining.

Ciliated columnar epithelium is found in 42% of cyst walls, and is the basic form of epithelium found in SCCM. Other findings include metaplasia of the laminated squamous epithelium (27.8%) and degeneration or disappearance of epithelial cells (30.2%). Manyama *et al.* studied 360 epithelial lining of postoperative maxillary cysts and found that 66% of their length was pseudostratified ciliated epithelium, 28% transition epithelium, and 6% squamous epithelium.[19] A finding of ciliated columnar epithelium is not a definitive finding to prove a lesion as a postoperative maxillary cyst. In the present case, the cyst is lined by pseudostratified ciliated columnar epithelium.

Diagnosis of such rare cases poses a great challenge due to its similarity to other cysts or tumors of the maxillary sinus. The differential diagnoses of SCC which have similar clinical features are odontogenic keratocyst, incisive canal cyst, median palatal cyst, and antral mucocele. The mucosal cyst of the antrum, which is commonly caused by trauma usually consists of a lining of seromucinous glands. Seromucinous glands are not observed in SCC of the maxilla.[20,21] Odontogenic keratocysts can also have foci of ciliated epithelium, but they can be differentiated as SCC does not have keratinizing tendency.[22] For a diagnosis of postoperative maxillary cyst, it is important to consider the history and clinical findings of the patient. Thus, our case stands an outlier in age, duration, and etiology of the SCC in the maxilla.

Treatment of choice is usually enucleation according to site and size.[23] With consideration to its benign nature, the most accepted treatment is enucleation of the cyst, and in cases of extensive cases, marsupialization is advocated. Recently, endoscopic intranasal surgery has gained popularity in the treatment of postoperative maxillary cyst. However, some authors have reservations about the endoscopic treatment of Post-operative maxillary cyst (POMC), especially when the lesion extends outside the sinus because drainage can fail, possibly leading to recurrence.[24,25] Recurrence of these cysts is fairly uncommon unless it is infected.

SCCs have rarely been reported; an increased awareness of the above-discussed possibility of unusual cause and pathogenesis is necessary to avoid delay in diagnosis and should always be considered for the differential diagnosis of cystic lesions of the jaw. Hence, from our report, it is evident that even traumatic extraction of teeth can have rare and debilitating complications like SCCs.

**Limitations**

Even though this an unique presentation of SCC in regard to pathogenesis and duration, the evidence of this particular phenomenon should be validated with other such instances, thus limiting the evidence of this report.

**Conclusion**

SCC is one of the rare sinus pathologies that have got multiple etiological factors. Iatrogenic trauma to the sinus lining is one of the reasons for its occurrence, which happened in our case. Surgical ciliated cyst usually develops from a few months to twenty years after the primary maxillary surgery. This case demonstrates maxillary SCC development with a relatively short latency period. Substantial variations are found about the etiology, pathophysiology, and duration of development for SCCM, and this case emphasizes how less we are yet to comprehend about the cyst.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her 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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