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Case Report

Cilliated Cyst of Maxilla: A Rare Case Report and Review of Literature

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ABSTRACT

A surgical ciliated cyst of the maxilla is a rare, benign lesion that arises in the maxillary sinus or surrounding areas, characterized by its lining of ciliated epithelial cells. These cysts are typically filled with mucous and are often asymptomatic but can cause problems due to their location, leading to symptoms like facial pain, sinus infections, or obstructed nasal airflow. Surgical removal is often necessary, especially if the cyst is large or symptomatic. We report a case of Surgical ciliated cyst of left maxillary sinus in 30 years-old male patient possibly after a surgical extraction of tooth, for which enucleation was done as a treatment modality resulting in good outcomes.

KEYWORDS

• Traumatic extraction • Maxillary sinus • Post surgical • Enucleation

INRODUCTION

A ciliated cyst also known as sinus mucocele/ postoperative maxillary cyst. It is an implantation type epithelial cyst that primarily develops in the maxilla.[1] The proposed pathogenesis is attributed to any traumatic event or surgical procedure involving the maxillary sinus causing the entrapment of sinus membrane or nasal mucosa into the bone.[2,3]

This pathology takes latency of 18 years on an average for development following

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any traumatic or surgical event.[4] Being asymptomatic in majority of the cases, typical age of diagnosis in 4th to 5th decade.[5] Various symptoms associated are pain, swelling and nasal stuffiness etc.[6]

Some cases have been reported in the mandible, caused by iatrogenic implantation of epithelium from maxillary sinus into mandible during simultaneous both Jaw surgery.[7] The diagnostic criteria based on the latest World Health Organization (WHO) Classification of Head and Neck Tumors (2022) are history of earlier surgery; radiographical evidence of a well-delineated radiolucency; and the histological presence of respiratory lining epithelium

Hereby, we present a iatrogenic case of ciliated cyst of left maxilla related to surgery, presenting with the chief complaint of frequent bleeding from nose.

CASE REPORT

A 30-years-old male patient reported to the Department of oral and maxillofacial Surgery with the chief complaint of frequent nasal bleed since 6 months. The patient gave history of increased frequency of bleeding with time and also noticed slight swelling on the left latral side of nose. All the possible causes for frequent nasal bleed were ruled out. On further exploration, he revealed history of surgical extraction of tooth from the same side of mouth 3 years back.

Figure 1: OPG showing missing left second premolar

Extraoral examination showed mild, soft, non tender, non pulsating uniformly diffused swelling over the mid face region on left side. Skin overlying the swelling was of normal colour. Intraorally there was obliterated vestibule w.r.t left maxillary canine, first premolar region. Second premolar was missing. These teeth were normal in appearance and non tender on percussion. Nointraoral signs of pus discharge or any other abnormality was detected. OPG depicts root resorption w.r.t left maxillary 1st premolar and 1st molar along with haziness in the left maxillary sinus (figure 1). Computed tomography on axial section shows unilocular expansile cystic lesion involving the left maxillary sinus with encroachment on the left lateral wall of nose, causing deviation towards right side (figure 2)

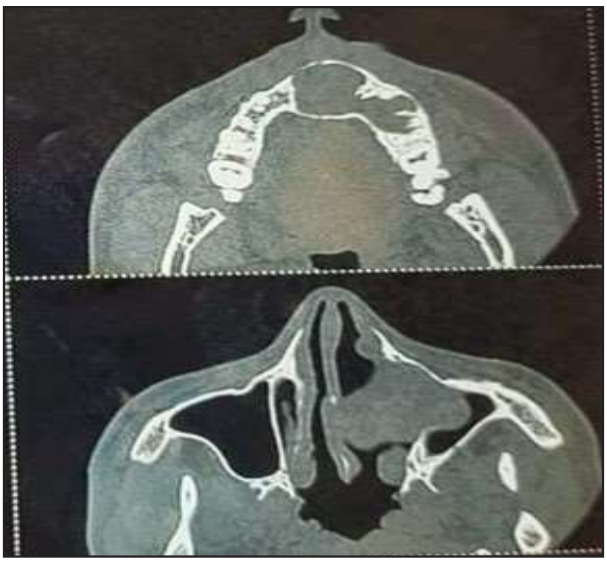

Figure 2: CT scan revealing large unilocular lesion involving left maxillary sinus

As patient gave the history of surgical extraction of the impacted maxillary second premolar, so there can be cystic lesion associated with impacted tooth probably the dentigerous cyst,which was not removed completely. An Incisional biopsy was performed to confirm the diagnosis. Reports suggesting surgical ciliated cyst of maxilla indicates the possibility of surgical trauma related to the removal of impacted maxillary second premolar.

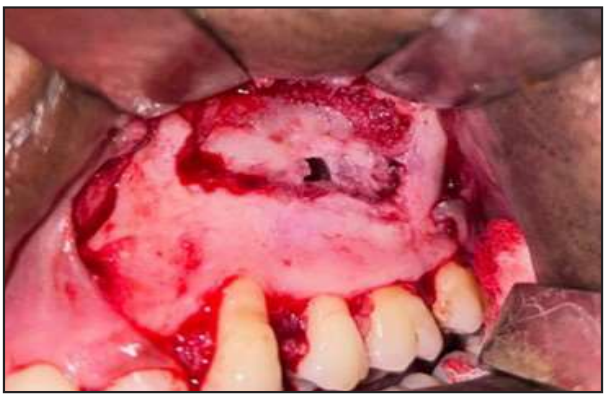

Figure 3: Exposure of cystic lining

After taking the informed consent, patient was planned for enucleation through intraoral vestibular approach using crevicular incision. A window was created in the anterior wall of maxilla (figure 3) and whole cystic lining was removed (figure 4) and cavity was irrigated and packed with iodoform gauze and primary closure done using 3-0 silk suture (Figure 5) and a small opening left for the removal of gauze after a week.

Figure 4: Cavity after removal of lesion

Figure 5: Primary closure with iodoform pack

The obtained specimen was sent for histopathological reporting. Microscopic examination showed a lining of pseudo stratified columnar ciliated epithelium and mild chronic inflammatory infiltrate and many seromucinous glands in the underlying stroma of the cyst wall, suggestive of ciliated cyst of maxilla (figure 6). The patient was followed up at regular intervals for a period of 3 months and healing was uneventful (figure 7). CT scan was repeated after 3 months which showed remarkable healing. (figure 8)

Figure 6: Postoperative Clinical picture

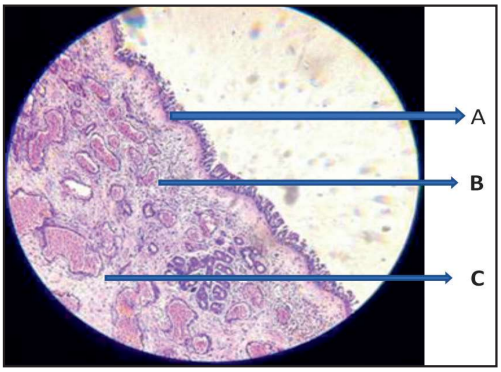

Figure 7: Histopathological Depiction
A. Pseudostratified ciliated Squamous epithelium· **B.** Inflammatory Infiltrate **C.** Blood vessels

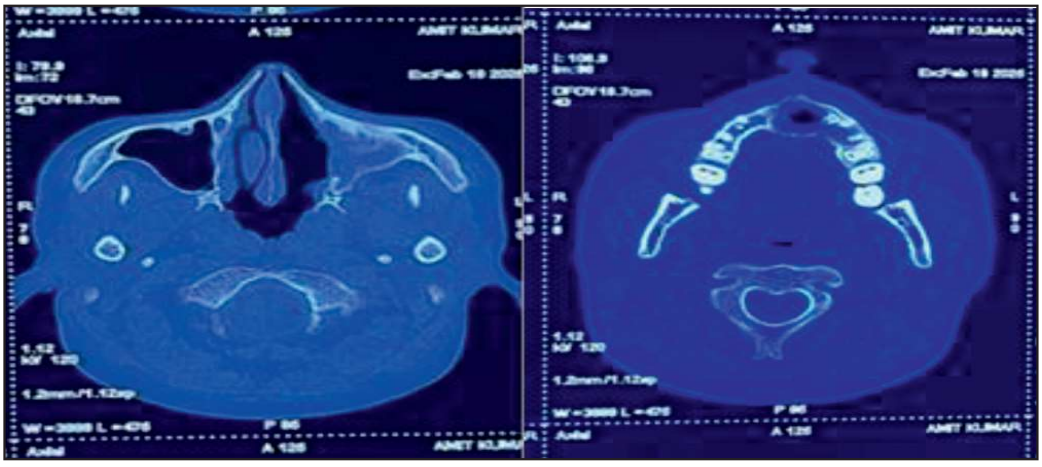

Figure 8: Post-operative CT Scan

DISCUSSION

Postoperative ciliated cyst of maxillary sinus was first described by Gregory and Shafer in 1958.[7] It was more common in Japan for many years after World War II. The surgical ciliated cyst of maxilla is currently added into the 5th edition WHO classification of head and neck tumours (2022). It forms due to trapped maxillary sinus or nasal mucosal epithelium in the jaw bones potentially due to trauma or surgical procedures like sinus augmentation, extraction, endodontic surgery, orthognathic maxillary surgery, Caldwell luc antrostomy.

Considering the possible causes, surgery of maxillary sinus or any trauma (According to Neville *et al*. trauma due to difficult extraction can cause its development), the probable cause of cyst development in our case seems to be related with surgical extraction of impacted maxillary second premolar, which was otherwise absent both clinically and radiologically.

It is difficult to differentiate the lesion clinically from other cysts or tumors of maxillofacial region. The differential diagnosis of surgical ciliated cyst are Dentigerous cyst. Antral mucocele, odontogenic keratocyst, unicystic ameloblastoma, primordial cyst and malignant tumor of maxillary sinus. Although the incisional biopsy report suggests the presence of surgical ciliated cyst of maxilla.

Histopathological examination of surgical ciliated cyst does not shows seromucinous glands, which are seen in the mucosal cyst of antrum[8,9], but this case's histopathology shows abundance of seromucinous glands.

Maruyama *et al* studied 360 epithelial lining of postoperative maxillary cyst and found that 66 % its length contains pseudo stratified ciliated epithelium, 28% transition epithelium, 6% squamous epithelium.[10] This case's microscopic examination corresponds to the majority feature found in the above study showing pseudo stratified ciliated columnar epithelium in addition to many seromucinous glands found in the underlying stroma.

The diagnosis of this case was challenging due to variation in the clinical, radiological as well as histopathological findings.

To our knowledge, only 5 cases of Surgical ciliated cyst after traumatic extraction have been reported in literature[11-15] till date.

The treatment of surgical ciliated cyst includes Caldwell-Luc operation, marsupialization, and endoscopic surgery[16] depending upon the site and size. In the present case, we opted for Caldwell luc procedure with excellent postoperative results.

REVIEW OF LITERATURE OF SURGICAL CYSTIS DEVELOPMENT FOLLOWING EXTRACTION

Author	Site	Age and Sex	Years after Extraction
Rajkumar *et al*., 2003	Posterior maxilla	27/F	5
Fernandes *et al*., 2013	Posterior maxilla	63/M	15
Ramakrishnan *et al*.	Posterior maxilla	76/F	5 months
Robert S. Redman *et al*., 2020	Anterior maxilla	53/M	2 weeks
Dey S *et al*., 2024	Posterior maxilla	30/M	2 months

CONCLUSION

In conclusion, a surgical ciliated cyst of the maxilla is a rare, benign cystic lesion that typically arises in the maxillary sinus and is characterized by the presence of ciliated epithelium. It may present with symptoms such as facial pain, swelling, or sinus congestion, nasal bleed though it can also be asymptomatic and discovered incidentally during imaging. The primary treatment for this condition is surgical excision, which is necessary to remove the pathology and prevent recurrence or its complications. While these cysts are not common, early detection and appropriate management are crucial for a favorable outcome. Regular follow-up after surgery is important to monitor for any signs of recurrence

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