

AN EXTRA-ANTRAL PRESENTATION OF SURGICAL CILIATED CYST: A RARE CASE REPORT

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ABSTRACT

The surgical ciliated cyst, also known as the "postoperative maxillary cyst" or "implantation cyst," occurs as a result of iatrogenic implantation of respiratory epithelium into a noncontiguous surgical site after sinus surgery. It is a locally aggressive lesion that appears as a delayed complication of surgery in the maxillary sinus region. 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. Surgical ciliated cysts are rarely reported in the non-Japanese population. It typically presents as a well-defined radiolucency in the maxilla in young adults. Histopathologically, the cyst is lined by ciliated columnar, cuboidal, or pseudostratified squamous epithelium with mucous cells. We report a unique case report, in Asian subcontinent, of a surgical ciliated cyst of antrum on left side which was extraantrally presented after twenty years of surgery.

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INTRODUCTION

Surgical ciliated cyst was first reported by Kubo in 1927 as a posterior maxillary cyst after the surgical treatment of maxillary sinusitis.[1] 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 anrostomy.[2] Very few reports on this entity have been published in the English language literature, but it is frequently described in the Japanese population 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. [3]

We report a unique case of a patient with surgical ciliated cyst on left side presenting extra-antrally diagnosed after twenty years of surgery.

Case report

A 22-year-old female patient reported to the department with a chief complaint of swelling over left infraorbital region with existing scar seen just beneath the swelling. She had undergone a surgery at the age of 2 years in the same region but exact history of type and reason of surgery was not known to the parents.

The medical history of surgical procedure as told by the patient was non-contributory for the examination. On extraoral examination, face was asymmetric with diffuse swelling

approximately 3× 2 cm in size and extending from left infraorbital region to nasolabial fold with depression seen just below the swelling. A scar was present in the same region of approx. 3×2cm. There was an extraoral sinus scar just below to zygomatic buttress region but no history of draining pus or serous fluid was noted.(Fig 1) On palpation, intraorally no swelling was seen or palpated. There was no evidence of lymphadenopathy or neurological signs.



Fig 1 showing extraantral presentation of surgical ciliated cyst

A complementary CT scan showed an oval, unilocular, well-defined translucent area with radiodense border intraosseously localized in left maxillary antrum region. The scan showed no perforation of medial cortex and no communication to maxillary sinus or nasal cavity was found.

Surgery under general anesthesia was planned for the excision of lesion. Surgical access was achieved through existing scar. (Fig 2) The pathological tissue was identified and completely excised by blunt dissection which was round in shape and of 3×2cm in size with a yellowish mucoid content and thin wall,

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and it was readily detached from the bone. (Fig 3) During surgery, there was no communication seen between the lesion and sinus or nasal cavity. The excised tissue was sent for histopathological examination. Histopathological 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 histopathological diagnosis was non-odontogenic ciliated cyst. The patient was regularly followed-up after the surgery for 1 year duration.



Fig 2 showing surgical access through existing scar



Fig 3 showing excised cyst of approx. 3x2cm

DISCUSSION

A surgical ciliated cyst usually occurs in the maxilla and is a complication associated with sinus surgery.[3,4] Several theories of the etiopathogenesis exist. The widely accepted theory is that iatrogenically implanted respiratory epithelium from the sinus surgery (eg, the Caldwell-Luc procedure or Le Fort I osteotomy) contaminates the maxillary wound, proliferates, and undergoes cystic changes and enlargement.[4,5] In our case the history of previous surgery at the childhood of patient which was unclear might be the reason for the presentation of extra-antral surgical ciliary cyst of maxillary antrum.

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.[3] Surgical ciliated cysts are rarely reported in the non-Japanese population.[5] The most common site in the maxilla is the lateral wall, although they have also been reported at the infraorbital ridge and medial canthal region.[6] Mandibular

ciliated cyst are also rarely found.[5] This unique case report that surgical ciliated cyst presenting extra-antrally in maxilla.

Radiographs show a well-demarcated, unilocular radiolucency with a sclerotic border. Histopathologically, the cyst is lined by ciliated columnar or cuboidal epithelium, with occasional goblet cells; areas lined by nonkeratinized stratified squamous epithelium may be present.[7] The underlying connective tissue is sometimes hyalinized.[2] Maruyama et al 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. It was also observed that sialylated glycoconjugates derived from lecithins in goblet cells were correlated with cyst wall inflammation and cyst growth.[8]

Treatment is curettage or enucleation. Marsupialization is recommended if the lesion is extensive and involves the cortical bone. The prognosis is good, although residual disease can progress if not completely removed.[5]

CONCLUSION

We report a unique case report, in Asian subcontinent, of a surgical ciliated cyst of antrum which was extraantrally presented illustrates the need for a meticulous surgical technique, proper management of complications, and routine, long-term follow-up of patients under any type of surgery.

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References

1. Kubo I. A buccal cyst occurred after a radical operation of the maxillary sinus. *Z F Otol Tokyo*. 1927;3:896-7.
2. Amin M, Witherow H, Lee R, Blenkinsopp P. Surgical ciliated cyst after maxillary orthognathic surgery: report of a case. *J Oral Maxillofac Surg*. 2003; 61:138-41.
3. Kaneshiro S, Nakajima T, Yoshikawa Y, Iwasaki H, Tokiwa N. The postoperative maxillary cyst: report of 71 cases. *J Oral Surg*. 1981; 39:191-8.
4. Yamamoto H, Takagi M: Clinicopathologic study of the postoperative maxillary cyst. *Oral Surg Oral Med Oral Pathol* 62:544, 1986.
5. Chia Li, David M. F., Killian D. M. and Sook-Bin W. Rare Mandibular Surgical Ciliated Cysts: Report of Two New Cases: *J Oral Maxillofac Surg* 72:1736-1743, 2014.
6. Sugar AW, Walker DM, Bounds GA. Surgical ciliated (postoperative maxillary) cysts following mid-face osteotomies. *Br J Oral Maxillofac Surg*. 1990;28:264-7.
7. Amin M, Witherow H, Lee R, Blenkinsopp P: Surgical ciliated cyst after maxillary orthognathic surgery: Report of a case. *J Oral Maxillofac Surg* 61:138, 2003.
8. Maruyama M, Onodera K, Ooya K. A histopathological and lectin-histochemical study of the lining epithelium in postoperative maxillary cysts. *Oral Dis*. 2002;8:241-8.