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# Odontogenic keratocyst in maxillary sinus associated with an ectopically placed maxillary third molar: a rare clinical sighting

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**Abstract**

Odontogenic keratocyst is the third most common odontogenic cyst which comprises about 12% of all jaw cysts. Posterior mandible is the most commonly involved region.[2]Most commonly seen in second and third decades of life with slight male predilection. Maxillary sinus involvement by OKC is rare and <1% of cases reported in the literature till today.Diagnosis of OKC involving maxillary antrum is challenging as maxillary radiographs do not offer distinctive features due to overlapping anatomic structures. With the continuous growth of an odontogenic cyst, the cyst encroaches on the space of the sinus cause displacement of its borders which in turn may displace the developing teeth buds leading to ectopic eruption.This case report presented a 19 years old male with swelling of the left side of the face and clinical,radiographic examination,fine needle aspiration cytology suggested OKC with respected to 28 in left maxilla;which was enucleated under general anesthesia.

**Keywords:** Diagnosis, ectopic eruption, maxillary sinus, odontogenic keratocyst,

## Introduction

The odontogenic keratocyst (OKC) is a type of developmental cyst of odontogenic origin, which was first defined in 1956 by Phillipsen and it is well known for the high recurrence rate.[1] Odontogenic keratocyst is the third most common odontogenic cyst which comprises about 12% of all jaw cysts. Posterior mandible is the most

commonly involved region.[2]Most commonly seen in second and third decades of life with slight male predilection.The maxillary antrum is a part of the paranasal sinuses, lies in close proximity with developing tooth and root apices of premolar and molars. Maxillary sinus involvement by OKC is rare and <1% of cases reported in the literature till today.[3] Diagnosis of OKC involving maxillary antrum is challenging as maxillary radiographs do not offer distinctive features due to overlapping anatomic structures.[4] It is named as keratocyst because the cystic epithelium produces keratin that fills the cystic lumen with flattening of the basement membrane,palisading basal epithelial cells.[5] In this paper we present a case of odontogenic keratocyst in relation to ectopic impacted maxillary third molar which was entirely limited in the sinus cavity, without involving maxillary alveolar bone.

**Case Report**

A 19-year-old male reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of swelling on his left side of the face.(Fig-1) He noticed the swelling three months back and it increased in size gradually. Extraoral examination revealed a diffuse swelling measuring about 3*3cm. antero-posteriorly extending from lateral margin of the left alar base to the vertical line dropped from lateral canthus of eye and supero-inferiorly extending from the left infraorbital rim to the left corner of the mouth region. Intra oral examination revealed a diffuse noticeable swelling extending antero-posteriorly from the left pre-maxilla extending towards the infra-zygomatic surface region with mucobuccal fold obliteration noted with respect to 24,25,26, 27region. (Fig-2)The swelling was soft to firm in consistency and was non-tender on palpation with respect to muco-buccal fold, alveolar mucosa in relation to 24,25 26, 27 and distal to 27.Overlying mucosa appeared normal and no local raise in local temperature was noted. No pus discharge was noted. On hard-tissue examination the involved teeth were non tender on percussion and no mobility noted. Mesio-distal tipping of 27 noted with missing 28. Orthopantomogram and cone beam computed tomography revealed an ill-defined, radiolucent lesion associated with an ectopic impacted third molar placed to the posterolateral wall of left maxillary sinus with resorption of the anterior wall of maxilla. Computerised Tomography gave an impression of an expansile cystic lesion involving of left maxillary sinus.(Fig-3,4,5)On FNAC of the cystic lesion (Fig-6)revealed a thick aspirate creamy white in colour with the presence of cholesterol crystals, superficial and desquamated epithelial cells, inflammatory cells, a glucose content of 1.2mg/dl, protein content of 2.1mg/dl suggestive of keratocyst. Considering the aggressive nature, age of the patient and close proximity to vital anatomic structure, cyst enucleation along with removal of impacted 28 was done under general anesthesia,(Fig-7) and the excised specimen was sent for histopathological examination.(Fig-8)Microscopic examination showed Connective tissue lined by a thin corrugated parakeratinized stratified squamous epithelium without rete ridges and basal layer composed of hyperchromatic columnar cells with palisading arrangement.

Cystic epithelium and wall interface were flat and showed detachment at focal areas. The cystic wall was fibrous with moderately dense inflammatory cell infiltrate. Focal areas were lined by pseudostratified ciliated columnar epithelium characteristic of the maxillary sinus. Based on the clinical, radiographical and histopathological features, lesion was diagnosed as OKC of the maxillary sinus.

**Discussion**

Since the odontogenic keratocyst was recognised,and later seeing its histological features defined,this lesion has

been the theme of investigation and study motivated by its tendency of recurrence and potential aggressive nature.[6] it's pathogenesis is believed to originate from dental lamina, begins as invagination of basal layer of epithelium,overlies the future maxillary and mandibular alveolar process, maintains the communication with overlying epithelium till the tooth bud formation. Develops approximately after 6 weeks of gestation and the band of epithelium that develops from anterior to posterior.[7] The controversial origin of OKC occuring in the maxillary sinus is believed to be arising from the entrapment of odontogenic epithelium within the sinus because of the close anatomic relationship between the dental lamina and developing antrum or the primordium of the canine and the floor of the sinus.[8] When the Schneiderian membrane is breached by the odontogenic pathological conditions of maxillary bone, the sinus pathology occurs. Odontogenic infections and pathology together account for 11%–12% of the total maxillary sinusitis cases.[9] Most commonly seen in second and third decades of life with slight male predilection. Maxillary cyst may involve sinus and nasal floor, premaxilla and third molar region. Swelling may present with or without pain, discharge, displacement of teeth. Enlarging cyst may lead to displacement of teeth, bucco-palatal plate expansion, missing teeth. The mode of expansion is burrowing into the bone rather than circumferential hygroscopic expansion, so small cyst is unlikely be diagnosed on routine examination. In maxilla most commonly involved side is 3rd molar followed by the cuspid.[7] Recurrence of the lesion principally seen in the first 5 years after the surgery. Recurrence rate found to vary from 0% to 62%, depending on the kind of surgical management and follow-up. The high recurrence rate of OKC is based on the great mitotic activity-8 highest among all cysts and growth potential found in epithelium. Various treatment modalities based on surgical approaches have been suggested in the literature, such as marsupialization, enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, curettage and resection.[10] The ectopic eruption of teeth aside from the mouth site is rare, although within the literature there are reports of teeth in unusual locations, like the maxillary sinus, mandibular condyle, nasal cavity, coronoid process.[11] Behind the ectopic eruption of tooth various possible etiologies have been suggested in the literature, which includes infection, trauma, developmental anomalies, and pathological conditions, such as odontogenic cysts. With respect to the latter, it is known fact that, with the continuous growth of an odontogenic cyst, the cyst encroaches on the space of the sinus cause displacement of its borders which in turn may displace the developing teeth buds.

**Conclusion**

Appearance of OKC in the maxillary sinus is rare, its radiographic image due to overlapping of the various structures may be misinterpreted. Accurate diagnosis of OKC is very important due to its aggressive nature and high rate of recurrence, so a systematic approach should be considered towards management of the lesion. Ideally a proper and accurate clinical, radiographical, histopathological examination with long term post-operative follow up are crucial elements to determine the most effective treatment outcome in order to avoid recurrence of OKC.

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Figure 1: Pre operative profile

Figure 2: Pre operative intraoral picture


Figure 3: Pre operative orthopantomogram

Figure 4: CT Coronal View

Figure 5: CT Axial View

Figure.6-Intra operative picture

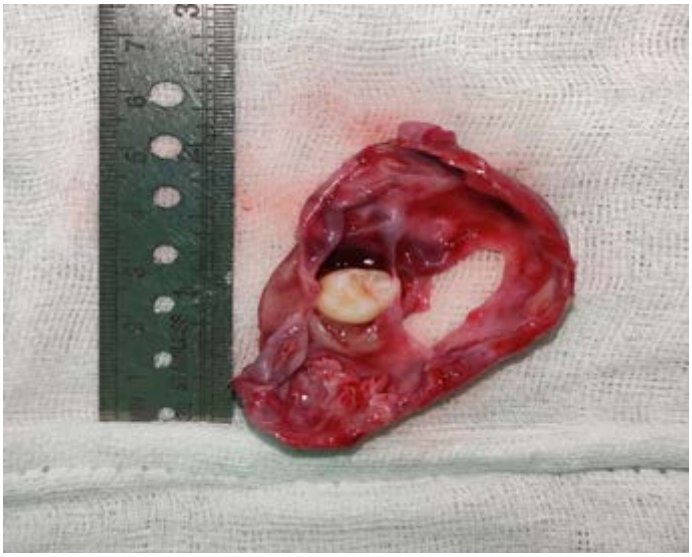

Figure 7: Excised specimen

Figure 8: Cystic content

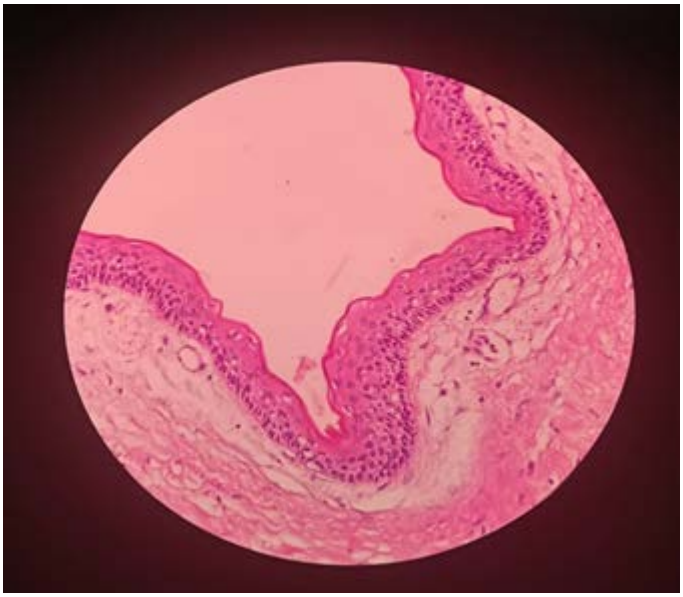

Figure 9: Connective tissue lined by a thin corrugated para-keratinized stratified squamous epithelium without rete ridges and basal layer composed of hyperchromatic columnar cells with palisading arrangement

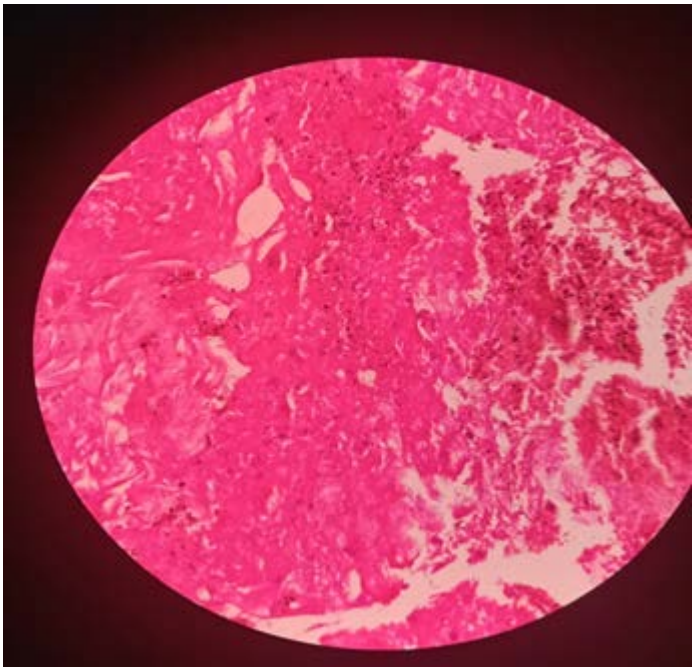

Figure 10: presence of cholesterol crystals, superficial and desquamated epithelial cells, inflammatory cells