

Odontogenic keratocyst arising in the maxillary sinus: A rare case report

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Abstract

Odontogenic keratocyst is an aggressive cystic lesion and a common type of tooth derived cyst due to presence of odontogenic epithelial remnants in different regions of jaw. In majority of cases, it is located in mandibular posterior region. But it can also be found in the maxilla especially in the canine region. We present a rare case of OKC in maxillary sinus which associated with ectopic third molar. Also, it can be easily confused with other lesions of maxillary sinus like sinusitis or antral polyps, which usually resemble symptomatically. There can be malignant transformation of this benign condition towards squamous cell carcinoma or ameloblastoma. So an early and accurate diagnosis of odontogenic keratocyst is a challenge for pathologists.

Keywords: Ectopic tooth, maxillary sinus, odontogenic keratocyst

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INTRODUCTION

Odontogenic keratocyst (OKC) is the third most common odontogenic cyst and comprises about 12% of all the cysts occurring in the maxillofacial region. The most significant change in the 2017 classification of developmental odontogenic cysts was that the term “keratocystic odontogenic tumor” was moved from the neoplastic category (2005) to the cyst category.^[1] The term odontogenic keratocyst was first used to describe all odontogenic cysts that contain keratin formations in the 1950s.^[2] The odontogenic keratocyst term, synonymous with primordial cyst, was used in the 1992 classification.^[3] The 2005 classification reclassified this unique lesion as a neoplasm and renamed it as “keratocystic odontogenic tumor” because of the high recurrence rate, aggressive clinical behavior, association

with nevoid basal cell carcinoma syndrome and mutations in the patched (PTCH) tumor-suppressor gene.^[4] The 2017 classification reverted back to the original and well-accepted terminology of OKC because many papers showed that the PTCH gene mutation could be found in nonneoplastic lesions, including dentigerous cysts, and furthermore, many researchers suggested that resolution of the cyst after marsupialization was not compatible with a neoplastic process.^[5] Vered *et al.* suggested that the immune-profile of sonic hedgehog (SHH)-related proteins and the SHH-induced bcl-2 oncoprotein may also be able to define the individual OKC phenotype and biologic behavior.^[6]

Involvement of the maxillary sinus by OKC is rare with <1% of cases reported in the literature.^[7] The

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maxillary sinus is a part of the paranasal sinuses, in proximity with developing tooth and root apices of premolar and molars. It is vulnerable to odontogenic infection, cyst and tumors of odontogenic origin. Diagnosis of such lesion is challenging as maxillary radiographs do not offer characteristic features due to overlapping of various structures.^[8]

In this report, we describe a case of OKC in relation to ectopic maxillary third molar in the maxillary sinus of a 15-year-old female.

CASE REPORT

A 15-year-old female reported with a complaint of pain and pus discharge with respect to the left upper back teeth region for 3 months. The pain was intermittent, dull aching, of moderate intensity and was radiating to the ear and eye. It was also associated with pus discharge with respect to the upper left second molar for 3 months. The patient visited a local doctor a week before where she was given some medications, but there was no relief from pain. Intra oral examination revealed a diffuse unnoticeable swelling on buccal aspect with obliteration of mucobuccal fold of 26, 27. The swelling was soft to firm in consistency and was tender on palpation with respect to mucobuccal fold, alveolar mucosa in relation to 26, 27 and distal to 27. Hard-tissue examination showed missing 28 and tender on percussion with respect to 26, 27. Orthopantomogram [Figure 1] revealed an ill-defined, radiolucent lesion associated with an impacted third molar displaced to the left maxillary sinus. Cone beam computed tomography [Figure 2] gave an impression of a single large destructive lesion involving lateral and posterior wall of maxillary sinus.

Aspiration of the cystic lesion revealed the presence of squames and abundant eosinophilic material, which was suggestive of keratocyst [Figure 3]. Therefore, enucleation of the cyst along with removal of 28 and antrostomy was done under general anesthesia, and the specimen was sent for histopathological examination. Microscopic examination revealed parakeratinized stratified squamous epithelium of 6–8 layers' thickness with surface corrugation. The basal cells showed nuclear hyperchromatism and palisading. 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 histopathological features, it was diagnosed as OKC of the maxillary sinus [Figures 4 and 5].

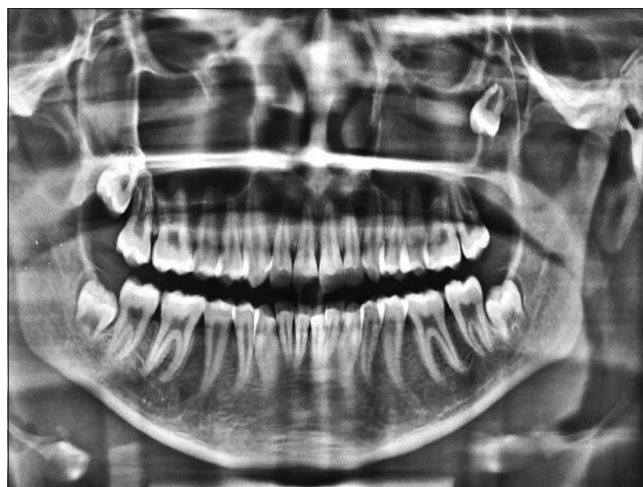


Figure 1: Orthopantomogram showing ectopically placed 28 in the lateral wall of the maxillary sinus



Figure 2: Cone-beam computed tomography showing obliteration of the maxillary sinus with impacted 28 in maxillary sinus

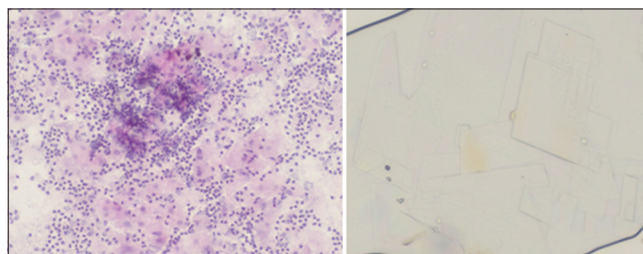


Figure 3: Fine-needle aspiration cytology shows the presence of cholesterol crystals, superficial and desquamated epithelial cells, inflammatory cells and bacterial colonies

DISCUSSION

OKC is a common developmental odontogenic cyst that accounts for 10%–12% of all jaw cysts. Toller and Browne believed it as a cyst derived from the dental lamina or its remnants and basal cells of the overlying epithelium.^[2] The origin of OKC in the maxillary sinus is controversial, presumably arising from the entrapment of odontogenic epithelium within the sinus because of the close anatomic

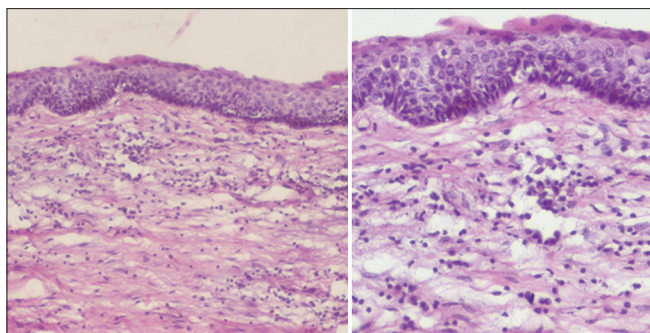


Figure 4: Connective tissue lined by a thin corrugated parakeratinized stratified squamous epithelium without rete ridges. Basal layer composed of hyperchromatic columnar cells with palisading arrangement

relationship between the dental lamina and developing antrum or the primordium of the canine and the floor of the sinus.^[9]

Maxillary sinus pathology can occur when the Schneiderian membrane is breached by conditions such as the odontogenic pathology of the maxillary bone. Odontogenic infections and pathology account for 11%–12% of maxillary sinusitis cases.^[10]

It usually occurs as a single lesion, but multiple lesions are also associated with the nevoid basal cell syndrome (Gorlin–Goltz syndrome).^[11] The peak incidence is in the second and third decades of life with a gradual decline thereafter, and the frequency is higher in males than females.^[2]

OKC involves both the jaws; the mandible is more often involved than the maxilla. In mandible premolar, molar area, angle and the ramus of mandible are the most common, but in the maxilla, it is seen most commonly in the canine area, followed by third molar tuberosity and anterior maxilla. In most of the cases, it presents as a periapical lesion.^[12] Here, we present a case which has occurred in quite younger age, and the lesion was in the sinus similar to the cases reported by Silva *et al.*^[7]

The ectopic eruption of teeth in regions other than the oral cavity is rare, although there have been reports of teeth in unusual locations, such as the nasal cavity, mandibular condyle, coronoid process, and palate. One of the non-dental sites in which an ectopic tooth has been observed is the maxillary sinus. The etiology of ectopic eruption has not yet been completely clarified, but many possible etiologies have been suggested. They include trauma, infection, developmental anomalies, and pathological conditions, such as odontogenic cysts. With regard to the latter, it is known that, as the growth of an odontogenic cyst continues, the cyst encroaches on the space of the sinus and displaces its borders: it may be that the displacement

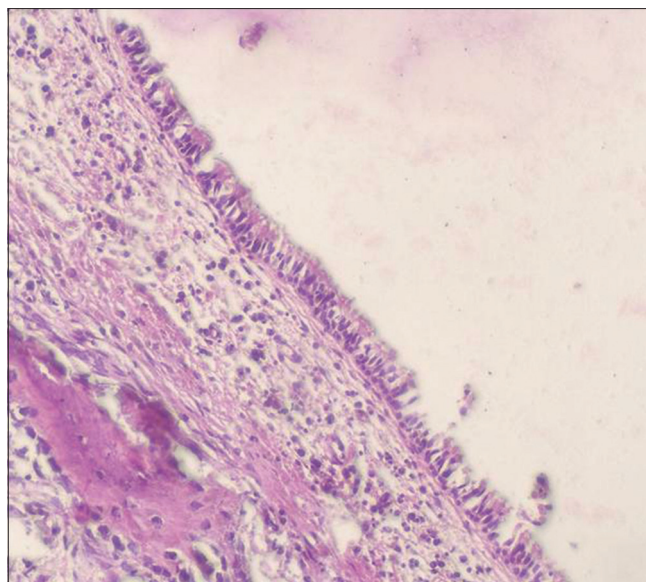


Figure 5: Focal areas of pseudostratified ciliated columnar epithelium characteristic of the maxillary sinus

of teeth buds by this expansion of a cyst results in the “ectopic” eruption of a tooth.^[13]

Radiographically, OKC appears as well-defined radiolucency which may be unilocular or multilocular.^[2] In the present case report, the radiographic examination and the computed tomography scan showed obliteration of the left maxillary sinus with ectopic third molar in it. The surgical exploration has unveiled the true pathological status with a classical histopathological picture of OKC.

Its presentation with impacted third molar tooth, location and a draining sinus made us to clinically consider the case as an infected dentigerous cyst. Hence it is important for the clinician to consider OKC in the differential diagnosis for such lesions when they occur in a younger patient.

CONCLUSION

OKC in the maxillary sinus is a rare occurrence, and it usually does not present characteristic clinical and radiographic features as its central counterpart within the jaw bone. The difference between OKC and other jaw cysts is its potential aggressive behavior and recurrence. To add to the literature, we emphasize the presence of OKC in the maxillary sinus. In addition, long-term follow-up must be done to detect any recurrence associated with the lesion when it occurs in the maxillary sinus.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and

other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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