Case Report

Keratocystic odontogenic tumor involving the maxillary sinus: A rare entity

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ABSTRACT

The keratocystic odontogenic tumor (KCOT) is a frequently encountered developmental cyst of the jaws. The occurrence of KCOT in the maxillary sinus is rare. The mucosa of the maxillary sinus is susceptible to infections, allergic diseases, and neoplasm. The anatomic position of maxillary premolar and molar teeth is in close contact with the sinus predispose to spreading of pulp and periodontal infection, odontogenic cyst, and tumors to the sinus. Diagnosis and treating KCOT in maxillary sinus is challenging as treatment has to be rendered for sinusitis because of pathology in the sinus and for KCOT. We report a case of 35-year-old female with KCOT involving the lining of the maxillary sinus and put forward hypotheses for the origin of KCOT in the maxillary sinus.

Key words: Keratocystic odontogenic tumor, maxillary sinus, odontogenic cyst

INTRODUCTION

Keratocystic odontogenic tumor (KCOT) is defined as a benign uni- or multi-cystic intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behavior.[1] It is a frequently encountered developmental cyst of importance because of its potential for aggressive clinical behavior, recurrence and its association with nevoid–basal cell carcinoma syndrome (NBCCS). KCOT is solitary unless it is associated with NBCCS.[2] Ahlfors *et al*.[3] believed that folding of the epithelial lining into the connective tissue capsule is due to active epithelial proliferation, which is responsible for aggressive nature of KCOT. Scharffetter *et al*.[4] considered both the epithelium and the connective tissue of KCOT exhibits high proliferation, and connective tissue growth was least contributing for the invasive and destructive growth of the KCOT.[4] Review of literature has shown recurrence rate of KCOT is 3–60%.[5] KCOT is commonly encountered in the mandible than the maxilla, particularly involving the premolar-molar area and ramus of the mandible.[2] Involvement of the maxillary sinus by KCOT is rare with <1% cases reported in the literature.[6] The maxillary sinus is part of the paranasal sinuses (PNSs), in proximity with developing tooth and root apices of premolar and molars. It is vulnerable for odontogenic infection, cyst and tumors of odontogenic origin.[7] Diagnosis of such lesion is challenging as maxillary radiographs does not offer characteristic features due to overlapping of various structures. We report a case of KCOT involving the maxillary sinus.

CASE REPORT

A 35-year-old female patient visited the outpatient department with a chief complaint of pain and discharge in the upper right vestibular region present for the past 1 year. The pain was mild, persistent, and radiating to the eye, forehead, and temporal region. She underwent extraction of maxillary right second premolar which was

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Access this article online

Quick Response Code:

Website: www.ccij-online.org

DOI: 10.4103/2278-0513.173256

Cite this article as: Kiresur MA, Ananthaneni A, Guduru VS, Devi RS. Keratocystic odontogenic tumor involving the maxillary sinus: A rare entity. Clin Cancer Investig J 2016;5:92-4.

then mobile 10 years back. The socket was curetted and left open for healing which was uneventful and the patient did not revisit for further follow-ups. Intraoral examination showed mild fullness of the vestibule related to maxillary first and second premolar, first molar regions with an intact mucosa. On palpation, tenderness was elicited in relation to the extracted socket and right malar region. Predicated on the clinical features, it was provisionally diagnosed as chronic sinusitis.

Orthopantomogram and intraoral periapical radiograph, in relation to maxillary first and second premolar, first molar showed normal bony architecture except for a small 2 mm × 2 mm ovoid radiolucent cupping bony defect with a radio opaque margin [Figure 1]. PNS view showed haziness of the right maxillary sinus. Further computed tomography (CT) scan showed a polypoidal soft tissue mass in the right maxillary sinus and a breach in the anterior wall of the sinus [Figure 2]. Based on these features, it was diagnosed as maxillary antral polyp. The patient was explained about the condition and was prepared for surgical removal of antral polyp. Under general anesthesia, access to the maxillary sinus was prepared through the canine fossa, and sinus lining along with the antral content was removed and sent for histopathological examination. Gross specimen comprised multiple bits of soft tissue of varying sizes and shapes. Microscopic examination revealed cystic wall lined by parakeratinized stratified squamous epithelium of variable cell thickness with surface corrugation. The basal cells showed nuclear hyperchromatism and palisading. Focal areas were lined by pseudostratified ciliated columnar epithelium characteristic of the maxillary sinus. The cystic epithelium was detached from connective tissue capsule [Figure 3a]. Immunohistochemical (IHC) stain with cytokeratin-17 showed strong positivity for the epithelium which confirmed the odontogenic origin of the cyst [Figure 3b]. Based on the histopathological features and IHC findings, it was diagnosed as KCOT.

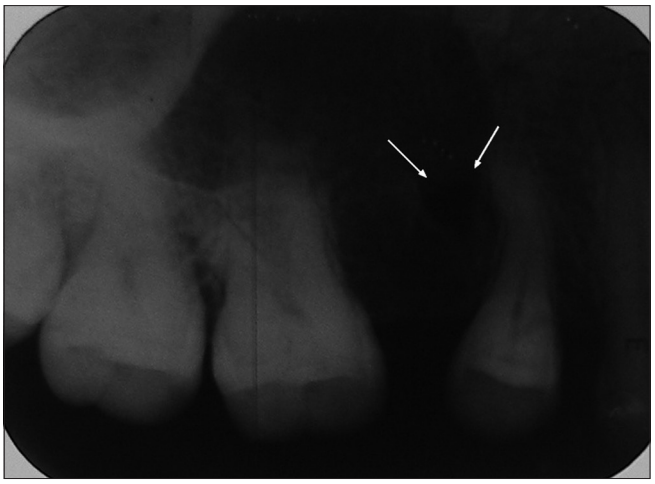

Figure 1: Intraoral periapical radiograph showing ovoid radiolucent area

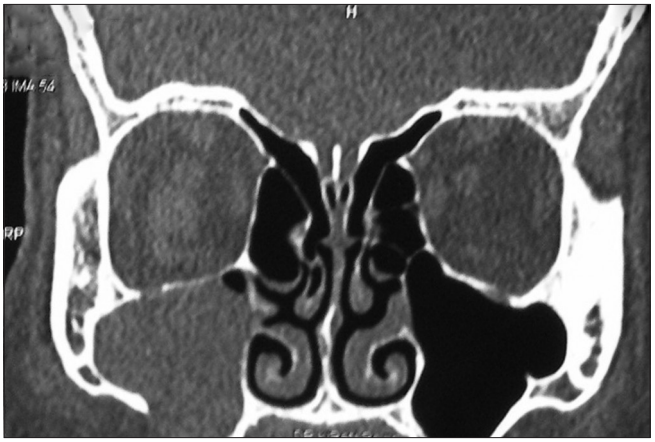

Figure 2: Computed tomography scan with haziness of right maxillary antrum and breach in the floor of sinus

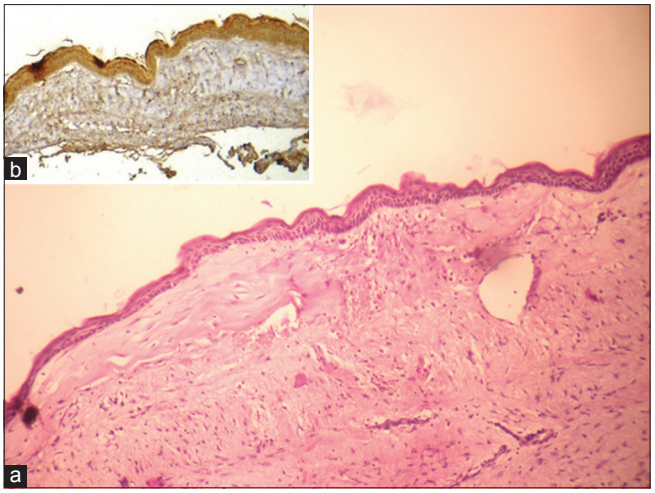

Figure 3: (a) Characteristic histologic features of KCOT (H and E, ×100). (b) Immunohistochemical stain strongly positive with cytokeratin-17

DISCUSSION

Mikucliz in 1876 reported the first lesion, Philipsen in 1956 coined the term odontogenic keratocyst.[2] In 2005, the World Health Organization renamed it as KCOT.[1] Prevalence of KCOT in jaws varies from 3.3% to 11%.[5] Toller and Browne believed it as a cyst derived from the dental lamina or its remnant and basal cells of the overlying epithelium. It has distinctive histologic features which include the epithelial lining of 5–8 cell thickness that show a basal layer of palisaded cells and corrugated parakeratinized surface.[2]

Origin of KCOT in the maxillary sinus is controversial, presumably 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] In the present case, we arrived at four plausible

modes of origin. First, it must have originated *de novo* from entrapped odontogenic epithelium as described above. Second, history of the patient revealed the extraction of a mobile tooth, the etiology for mobility was not explored. It could have been an existing KCOT which resulted in mobility. After extraction, the lesion constrained to a 2 mm × 2 mm defect, which grew into the maxillary sinus. Third, it would have started as a peripheral KCOT which could have intruded the maxillary bone, which facilitates easy invasion compared to the mandible. This possible factor was included because a breach in the anterior wall of the maxillary sinus was observed under CT scan. Fourth, as the patient underwent extraction, the trauma could have triggered the cell rests and resulted in cyst formation.

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 10–12% of maxillary sinusitis cases.[7] Less than 1% of KCOT cases occur in the maxilla involving the sinus.[8] The peak age of incidence is the second and third decade of life with studies showing bimodal age distribution with other peak in the fifth decade or later. The mean age of patients with NBCCS is lower than the mean age of patients with single sporadic cyst. KCOT can involve both the jaws, the mandible has been far more often involved than the maxilla. Most of the mandibular KCOT are common in premolar, molar area, and extends up to the angle and the ramus of mandible.[5] In maxilla, KCOT is seen in the canine area, followed by third molar-tuberosity and anterior maxilla and in most of the cases presenting as a periapical lesion.[9] Here, putting aside the age, which was continent with the literature, the lesion was in the sinus similar to the cases reported by Silva *et al*.[6]

KCOT appears as well-defined radiolucency which may be unilocular or multilocular. KCOT has an infiltrative behavior. Hence, little expansion is observed.[2] In the present case report, the radiographic examination and the CT scan showed a characteristic picture of antral polyp. Ali and Baughman observed that of 54 submitted samples for histopathological examination, the clinical diagnoses were periapical cyst or granuloma (31.5%), lateral periodontal cyst (25.9%), and globulomaxillary cyst (27.8%).[9] In such cases, the histopathological diagnosis can be considered as an important diagnostic aid. In this present case, clinical and radiographic picture was implicative of maxillary sinusitis. Had not the patient accepted for surgical intervention, she would have treated for sinusitis with a nonresponsive outcome. The surgical exploration has unveiled the true pathological status with a classical histopathological picture of KCOT.

Management of sinus disease requires appropriate antibiotics and removal of the underlying pathology. Treatment of KCOT includes excision of the overlying attached mucosa in conjunction with cyst enucleation and treatment of the bony defect with the Carnoy solution.[10] Recurrence has not been reported in the literature for KCOT involving the maxillary sinus; the patient was followed up for 1 year with no sign of recurrence.

CONCLUSION

KCOT in maxillary sinus is a rare occurrence, and it does not present characteristic clinical and radiographic features as its central counterpart within the jaw bone. The difference between KCOT and other jaw cyst is its potential aggressive behavior and recurrence. Due to lack of extensive literature, it is difficult to predict the behavior and recurrence of KCOT in maxillary sinus with appropriate treatment modalities. To add to the literature, we emphasis the presence of KCOT in the maxillary sinus, which needs an aggressive treatment modality and long-term follow-up when compared to maxillary sinusitis.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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