Keratocystic Odontogenic Tumor Associated with an Ectopic Tooth in the Maxillary Sinus: A Rare Entity

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Abstract

The Odontogenic Keratocyst (OKC), first described by Phillipsen in 1956, has been reclassified as odontogenic neoplasm and has been renamed as Keratocystic Odontogenic Tumor (KCOT) as reported in WHO classification of head and neck tumors in 2005. KCOT is a benign intraosseous neoplasm of the jaw and its occurrence in maxilla is unusual and its appearance in maxillary sinus along with an ectopic tooth is very uncommon. This article reports an unusual case of KCOT associated with unerupted molar in the maxillary sinus of a young adult. The present case was treated surgically by gaining an access to maxillary sinus lateral wall through canine depression and enucleating the cyst containing the tooth in pieces. It was followed by soft tissue curettage and prophylactic chemical cauterization with Carnoy's solution.

Key Words: Ectopic eruption, Keratocyst, Keratocystic odontogenic tumor, M.axillary antrum

Introduction

Keratocystic Odontogenic Tumor (KCOT) is defined as a benign unicystic or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior [1-3]. KCOT occurs across a wide age range but majority of the cases (about 60%) are diagnosed in the age range of 10 to 40 years [4]. The majority of the cases involve the molar, angle and ramus areas [5].

Unusual location has also been reported, such as the anterior portion of maxilla, maxillary antrum, and maxillary third molar area [2]. A less common occurrence is a KCOT thatarises in the maxillary sinus in associationwith an ectopic tooth [6]. We hereby report a case of KCOTassociated with an ectopic tooth in the maxillary sinus.

Case report

A 32-year-old male presented with complaint of a persistent headache, present for 2 weeks. The past medical history was not contributory and no alteration was detected on extra-oral and intra-oral examination. Para nasal sinus radiographs showed a discrete opaque mass with an image of the third molar in the left maxillary sinus. The lesion seemed to be totally confined into the left maxillary sinus cavity, without nasal sinus or maxillary alveolar bone involvement (*Figure 1*).

Under general anesthesia, an access to maxillary sinus lateral wall through caninedepression was obtained. After osseous trapanation, a cystic lesion could be visualized (*Figure 2*). The cyst containing the tooth was enucleated in pieces (*Figure 3*) and soft tissue curettage and prophylactic chemical cauterization with Carnoy's solution was executed. Carnoy's solution used in the present case was composed of 3 ml of chloroform, 6 ml of absolute ethanol, 1 ml of glacial acetic acid, and 1 g of ferric chloride [7]. Patient was prescribed ivtaximax 1.5 g bd, iv flagyl 500 mg tds, and imvoveran 75 mgbd for 5 days following surgery. The histopathological examination revealed epithelial lining exhibiting parakeratotic layer, thin spinous cell layer and hyper chromatic columnar cells in the basal layer, with no rete ridges. These features confirmed the diagnosis of KCOT (*Figure 4*). The patient has been followed up for 2 years and shows no signs of recurrence.

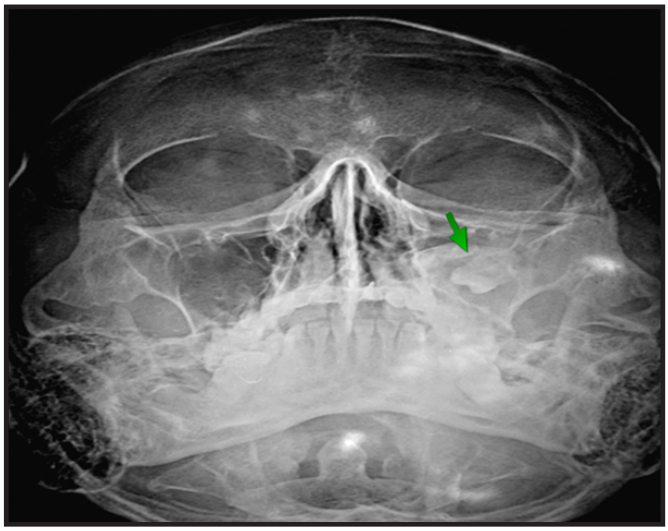

***Figure 1**. Para nasal sinus view radiograph showing unerupted tooth in the left maxillary sinus surrounded by a discrete opaque mass (arrow).*

Discussion

The Odontogenic Keratocyst(OKC) was first described by

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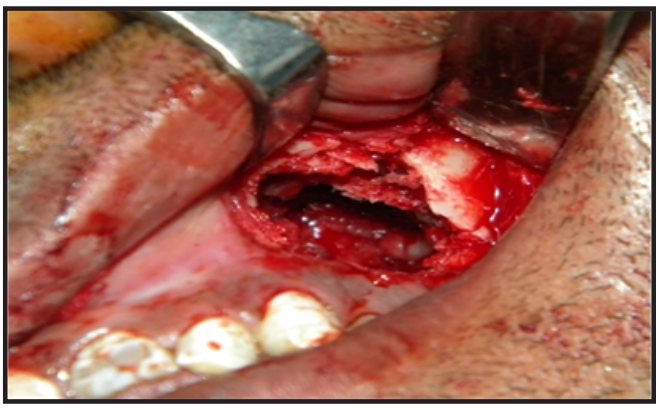

Figure 2. *Surgically exposed site of left maxillary sinus showing the cystic lesion.*

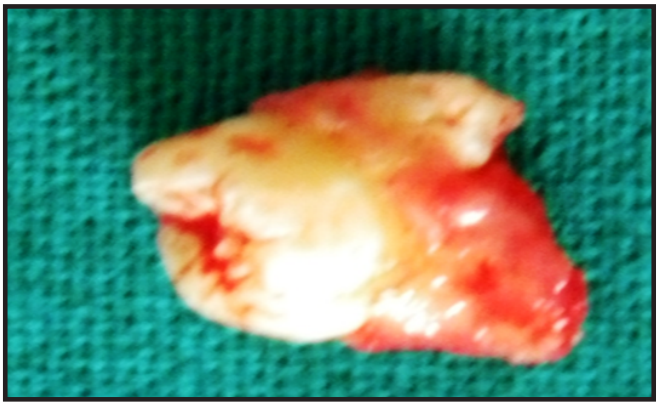

Figure 3. *Picture showing removed tooth with fragmented cystic lesion.*

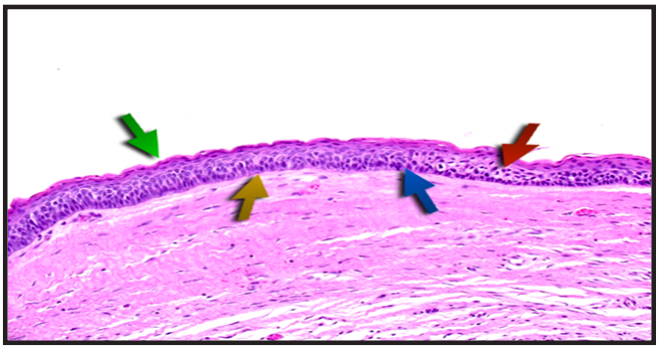

Figure 4. *Photomicrograph of the fragmented cyst showing epithelial lining exhibiting parakeratotic layer (green arrow), thin spinous cell layer (red arrow) and hyperchromatic columnar.*

Phillipsen in 1956, who used the term to describe jaw cysts exhibiting keratinization of their epithelial linings [1]. OKC has now metamorphosized as Keratocystic Odontogenic Tumor (KCOT) as reported in WHO classification of head and neck tumors in 2005 [2]. KCOT may be found in patients who range in age from infancy to old age, but about 60% of cases are diagnosed in people between 10 and 40 years of age [4]. There is a slight male predilection. In our case, the patient was in his fourth decade of life and was a male. The mandible is involved in majority of the cases, withless than 1% cases occurring in the maxilla with sinus involvement [4,8]. KCOT when involving the maxilla sinus must be carefully assessed because the orbital damage and the spread of associated infections could lead to local and systemic compromise to the patient.

Abnormal tissue interactions during tooth development may potentially result in ectopic tooth development and eruption. Ectopic eruption of a tooth into a region other than the oral cavity is rare although there have been reports of tooth in the nasal septum, mandibular condyle, coronoid process and the palate [9-12]. Occasionally, the tooth may erupt into the maxillary antrum [13]. One interesting aspect in our presented case is that the KCOT was completely restricted in the maxillary sinus without alveolar bone or erupted tooth association, while the previously cases reported large KCOTs had probably developed in the maxillary bone and expanded to the sinus [14].

Histologically KCOT have been classified as parakeratotic and orthokeratotic [1]. These types refer to the histologic characteristics of the lining and the type of keratin produced. The parakeratotic variant exhibits flattened parakeratotic epithelial cells toward the luminal surface; and the basal epithelial layer is composed of a palisaded layer of cuboidal or columnar epithelial cells, which are often hyperchromatic. However, the orthokeratotic variant shows an orthokeratotic surface, and do not demonstrate a hyperchromatic and palisaded basal layer [4]. The parakeratotic type is more frequent (80%) and has a more aggressive clinical presentation than the orthokeratotic variant [2]. The lesion reported in this case is a parakeratotic KCOT. Furthermore, the epithelial lining is composed of a uniform layer of six to eight cells thick stratified squamous epithelium, with a hyperchromatic and palisaded basal cell layer. Detachment of portions of the cyst-lining epithelium from the fibrous wall is commonly observed [3].

KCOT has been a theme of investigation and study motivated by its tendency of recurrence and potential aggressive nature. WHO in 2005 has reclassified OKC as KCOTbased on its growth capacity and development characteristics related to the mutation of a tumor suppressor gene, PTCH (patched gene), found in sporadic as well as in keratocysts associated with basal cell nevus syndrome [2].

The recurrence of OKC, which is usual, is thought to be based on great mitotic activity and growth potential found in epithelium, furthermore other sources of recurrences such as remnants of dental lamina and epithelial islands have also been proposed [15]. These findings lead to recommendation of surgical approach for its treatment which may include marsupialization, enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, curettage and resection [16,17]. The present case was treated with enucleation and chemical cauterization, and on follow up no recurrence was found, which was expected, because cyst was not adhering to the bone.

Conclusion

To conclude, KCOT is relatively rare in maxillary antrum along with ectopic maxillary molar. Thus it is suggested to include KCOT in the differential diagnosis of maxillary sinus radiolucency to avoid misdiagnosis and to decrease the chances of recurrence. Ideally a biopsy specimen examination and accurate clinical, radiographic, trans-surgical observation along with follow up are essential to avoid recurrence.

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