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MULTIPLE ODONTOGENIC KERATOCYST OF MAXILLA AND MANDIBLE:A 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 case of OKC in maxilla nad mandible which is 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, impacted third molar, maxillary sinus, odontogenic keratocyst

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

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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 non neoplastic 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]

CASE REPORT:

A 20 year old male reported with a complaint of pain and swelling in the upper anterior region of maxilla from 11 and 21 tooth region for 4 months. The pain was intermittent, dull aching, of moderate intensity. It was not associated with pus discharge. The patient visited a local doctor a week before where he 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 11,12,23 and 24 region. The swelling was soft to firm in consistency and was tender on palpation with respect to mucobuccal fold, alveolar mucosa in relation to maxillary anterior region and huge swelling in right ramus of mandible to body with bucco-lingual expansion.

Orthopantomogram [Figure 1] revealed an ill-defined, radiolucent lesion associated with lower impacted third molar and upper right and left maxillary anterior region.



FIGURE 1:Orthopentogram shows impacted lower third moral on right side and upper left and right third molar.

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Aspiration of the cystic lesion revealed the presence of cholesterol crystrals and abundant eosinophilic material, which was suggestive of keratocyst.

Therefore enucleation of the cyst followed with extraction of 48 and peripheral ostectomy followed by chemical cauterization was done and the specimen was sent for histopathological examination.



Figure 2A

FIG 2 A,B,C: Shows large swelling in the left maxillary canine premolar region. removal of the cystic lining and chemical cauterization with Carnoy's solution was carried out.



Figure 2B

Figure 2C

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FIG 3:exposure of the right mandibular body region done, cystic lining removed followed by extraction of impacted third molar and chemical cauterization done.

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.

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

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]

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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]

CONCLUSION:

OKC in the maxillary 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 maxilla. In addition, long-term follow-up must be done to detect any recurrence associated with the lesion when it occurs in the maxillary sinus.

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