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ODONTOGENIC KERATOCYSTS OF MAXILLARY SINUS – A CASE REPORT

Oral Medicine & Radiology

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ABSTRACT

Odontogenic Keratocyst (OKC) is a clinico-pathologically distinct form of developmental odontogenic cyst. It is known for its aggressive biological behavior and high recurrence rate. Odontogenic keratocyst (OKC) is the third most common odontogenic cyst and comprises about 12% of all the cysts occurring in the maxillofacial region. OKC has a distinct feature to occur in body of the mandible and ramus. But it can also be found in the maxilla especially in the canine region. We present a rare case of OKC in maxillary sinus of a 35 year old female patient which is associated with ectopic third molar that can be easily confused with other lesions of maxillary sinus like sinusitis or antral polyps. Involvement of the maxillary sinus by OKC is rare with < 1% of cases reported in the literature.

KEYWORDS

Odontogenic keratocyst, Keratocystic odontogenic tumour, Odontogenic tumour, Gorlin syndrome.

INTRODUCTION

Keratocysts of the jaw are benign cystic lesions which have a potential for keratinization. Philipsen (1956) and Pindborg, Hansen in (1963) described the clinical and histologic features of this non - inflammatory cyst that uniformly formed keratin[1]. In 1971, WHO simplified the classification of jaw cysts and made the terms primordial cyst and keratocyst synonymous. This classification emphasized the predominant histological features of the cyst rather than its presumed origin from the primitive enamel organ[1]. In 2005 odontogenic keratocyst (OKC) was designated by the World Health Organization (WHO) with the term keratocystic odontogenic tumour (KCOT) as it better reflects its neoplastic nature. WHO has reclassified the lesion as a tumour based on several factors, which include

- **Behavior :** Locally destructive and highly recurrent.
- **Histopathology:** The basal layer of the KCOT budding into connective tissue with mitotic figures frequently found in the suprabasal layers.
- **Genetics:** SHH binding to PTCH inhibits growth – signal transduction thereby functioning of PTCH is lost, the proliferation-stimulating effects of SMO become predominate[2].

However, the most recent WHO classification, which was published in 2017, considered OKC as developmental odontogenic cyst again. This change was due the fact that many researchers suggest that the resolution of the cyst after marsupialization was not compatible with a neoplastic process[3,4].

Casereport

A 35 year old female reported to t h e Department of Oral Medicine and Radiology, with a complaint of pus discharge from the upper right back region of mouth since 3 months. History revealed that the discharge persisted even after the extraction of periodontally compromised 16.It was not associated with pain or any other discomfort.

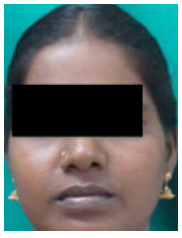


Fig – 1: Shows a diffuse swelling in the right mid face

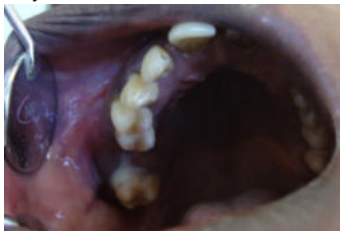


Fig – 2 Intra orally reveals pus discharge from 16

On extra-orally examination, single, diffuse, smooth surfaced swelling, measuring approximately 2×3cm in size is seen on the right mid face region. It extended medially up to nose & laterally till malar prominence, superiorly it extends to involve infra orbital margin & inferiorly up to ala tragal line. Skin over the swelling appeared normal with no secondary changes. On palpation, swelling was warm, soft and non-tender on palpation. There was no evident regional lymphadeno pathy Intra oral examination of hard tissue show congenital missing 18, with pus discharge from the socket of 16(Fig.2).

Diagnostic work up included plain film radiographic view (OPG and waters view) and advanced imaging modality. (CT scan with axial and coronal section)

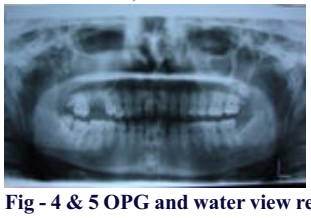
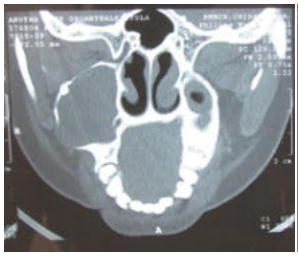


Fig - 4 & 5 OPG and water view revealed impacted third molar in right maxillary sinus



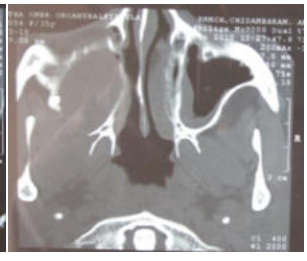


Fig -5 & 6: Axial & coronal section of CT reveals a hyperdense mass in maxillary antrum.

A well-defined expansile radio-lucent lesion was appreciable in panoramic view, waters view and computer tomographic images. On CT axial view a radiopaque mass measuring approximately 3× 2 cms in diameter with 1712 HU resembling tooth like structure is seen in right maxillary antrum. Evidence of buccal cortical expansion with radiolucent mass enchroching into the right sinus was better appreciated in the axial view. Breech along the postero-lateral wall of the maxillary sinus with deviation of nasal septum and effusion of nasal conchae was appreciated CT.

Surgical enucleation of the lesion along with impacted tooth was done under general anesthesia and tissue was submitted for histopathological examination. Histopathologial diagnosis of KCOT was made.

DISCUSSION

OKC represents 10% to 12% of odontogenic developmental cysts[5].

WHO has defined OKC in 2005 as a benign uni or multicystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium with high potential for infiltration and recurrence[5,6,7].

However, the most recent classification, which was published in 2017, considered it as a developmental odontogenic cyst again reverting back to the original and well-accepted terminology of OKC as many papers showed that the PTCH gene mutation could be found in non-neoplastic lesions, including dentigerous cysts, and furthermore. Many researchers have suggested that resolution of the cyst after marsupialization was not compatible with a neoplastic process[3,4,8]

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. 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 case[7,8]

OKC of maxilla have diagnostic difficulties owing to lack of specific clinical and radiographic characteristics. They are less common in maxilla than mandible with only 31.3% in maxilla. But when they do occur, they are more common in the canine region which was the case in our patient also. OKC has been shown to have a bimodal age distribution with first peak in the second and third decades and the second peak in the fifth decade with male predominance. It is said that the lesions in the second peak are more common in maxilla. . The literature suggest that less than 1% of all case of OKC occur in maxilla and exhibit sinus involvement.[9,10]

Multiple OKC are reported to be associated with nevoid basal cell carcinoma syndrome (NBCCS) or Gorlin-Goltz syndrome. Distinctive clinical feature of NBCCS are nevoid basal cell carcinomas, bifid ribs, calcification of the falx cerebri, frontal bossing, multiple epidermoid cysts and medulloblastoma[7,8].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[11,7,8].

Histopathological examination will show cystic lining which will be composed of uniform layer of stratified Squamous epithelium. The luminal surface shows flattened parakeratotic epithelial cells exhibiting wavy appearance. The basal epithelial layer is composed of palisadad layer of coloumnar cells. Similar finding was seen in the present case[6]. Incomplete removal of the cyst lining, growth of a new OKC from satellite cysts or odontogenic rests left behind after surgery and development of a new OKC in an adjacent area are few of the proposed mechanism of recurrence reported in literature[7,12].

To minimize invasiveness and recurrence, the most effective treatment option appears to be enucleation of the OKC and subsequent application of Carnoy's solution. Alternatively, marsupialization followed by cystectomy is likewise effective, as this treatment does not result in a significantly higher rate of recurrence than enucleation plus Carnoy's solution[7,13] . In the present case, surgical enucleation with Carnoys solution of the cyst was done under general anesthesia. Impacted tooth was removed along with the lesion[14,15].

Most recurrences take place within 5–7 years after treatment. Literature review suggests that recurrence rate is relatively low with aggressive treatment, whereas more conservative methods tend to result in more recurrent cases[15].

CONCLUSION

OKC occurring in maxillary sinus is an uncommon clinical presentation. Inherent limitation of OPG in interpreting maxillary lesion warrant CT evaluation. The aggressive nature of OKC needs an aggressive treatment strategy. The odontogenic keratocyst have high rate of recurrence up to 62.5%.

The 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. 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.

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