

# AN EXPANSILE RADIOLUCENT LESION OF MAXILLA CROSSING MIDLINE – A CASE REPORT

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## Abstract

Odontogenic Keratocyst (OKC) is a distinctive form of developmental non-inflammatory odontogenic cyst which receives special contemplation because of its unambiguous clinical behavior and histopathological features. It arises from cell rests of dental lamina and is aggressive in nature. OKC is the third most common odontogenic cyst after radicular and dentigerous cyst. Most of the studies have reported that posterior region of the mandible is the most common site, but there are less predictability regarding the prominent site of OKCs in the maxilla. Very few published literature are available with OKCs involving maxilla and crossing the midline. Here we report a case of an expansile odontogenic keratocyst of maxilla crossing the midline.

**Conclusions:** OKC is the third most common jaw cyst after the radicular and dentigerous cyst. The existing published literature shows that it has more predilection to occur in the mandibular posterior region and less likely to be in maxilla with involvement of maxillary midline and rarely crossing the midline. In spite of its large range of incidence, the lesions crossing the maxillary midline have been reported in older individual but in our case the reported individual is female and of young age group. Proper clinical examination and radiographic investigation along with biopsy is important for exact diagnosis and appropriate treatment of OKC to prevent recurrence.

**Key words:** *Odontogenic cyst, Odontogenic Keratocyst, Maxilla.*

## Introduction

Jaw cysts are very common in their occurrence due to the presence of odontogenic epithelium remnants in the jaw bone.<sup>1</sup> OKC is the third most common jaw cyst after radicular and dentigerous cyst. The term odontogenic keratocyst (OKC) was first proposed by Philipsen in 1956 to describe keratin containing jaw cysts. It is unique and important odontogenic cyst because of its aggressive behavior, high rate of recurrence, and specific histopathologic features. It accounts for about 11% - 15% of all odontogenic cysts. In 2005, the World Health Organization reclassified OKC as "keratocystic odontogenic tumor" (KCOT)<sup>2</sup> thereby enduring the same under the category of benign neoplasm of odontogenic origin and not an odontogenic cyst. It is defined as a "benign uni- or multi-cystic odontogenic intraosseous tumor, with lining of parakeratinised stratified squamous epithelium and with aggressive behavior"<sup>3</sup>.

WHO in 2017, again included OKC in the category of odontogenic cysts.

OKC is an enigmatic developmental cyst as it exhibits higher growth potential and recurrence rate due to formation of daughter cysts and thin and fragile epithelial lining. Despite of many classifications and nomenclature, the clinicians still have to face trouble in the treatment of this commonly found lesion<sup>4</sup>.

## Case Report

A 29 years old female patient reported to the department of oral medicine and radiology with the chief complaint of swelling on her right side of the face since 2 months. There was no history of pain and paraesthesia associated with the swelling. On extraoral examination a diffuse swelling was present on the right middle third of the face extending anteroposteriorly from ala of nose up to malar region and supero-inferiorly from infra-orbital margin

up-to the corner of mouth with obliteration of the nasolabial fold. The swelling was hard in consistency and non-tender on palpation (Figure 1).

## Figure Legend



Fig 1: Extraoral photo showing swelling on the right side of the face

Intra-oral examination showed a small diffuse swelling with normal overlying mucosa in relation to 13, 14, 15 region (Figure 2). On further examination posteriorly expansion of buccal cortical plate is noted in molar region. It was soft to firm in consistency in the 13, 14, 15 region while hard in molar region. The teeth 14, 15 were rotated distally. A clinical diagnosis of infected odontogenic cyst was given. On aspiration, a cheesy material was aspirated.



Fig 2: Intraoral photo showing small swelling with mild expansion of buccal cortical plate

Intraoral periapical radiograph in the region of 13,14,15 and 16,17 region showed no evidence of caries irt to the teeth. A radiolucency was present irt apical root region of 13,14,15 and 16 ,17. There was loss of lamina dura irt 13,14,15,16,17 with loss of trabeculation within the radiolucent lesion. The roots 14 and 15 were displaced distally away from each other (Figure 3).



Fig:3 IOPAR showig a radiolucent lesion with loss of lamina dura irt 11,12,21

The panoramic examination revealed a large radiolucent lesion crossing the midline measuring approximately 8 x 5 cm in dimension extending from mesial root apex region of 22 upto the distal root region of 17mediolaterally and superoinferiorly from floor of the orbit upto the alveolar crest region of 11,12,13,14,15 region. The radiolucent lesion was surrounded by thin scalloped sclerotic border (Figure 4).



Fig 4: OPG showing a large radiolucent lesion with scalloped border crossing the midline

Further investigation included CT scan of the region which showed a large expansile lesion which had displaced the floor of maxillary antrum with cortical destruction (Figure 5,6,).

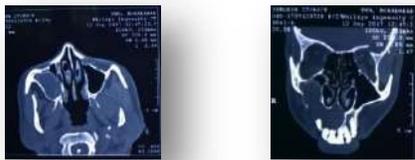


Fig 5: Axial and coronal CT showed a large expansile lesion



Fig 5: 3D CT scan of patient showing perforation of buccal cortical plate

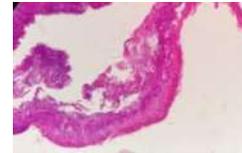


Fig 6: H & E stained section showing cystic parakeratinized epithelial lining

An incisional biopsy under local anesthesia was done and the histopathological H and E stained sections showed cystic parakeratinized epithelial lining overlying connective tissue capsule with keratin flakes and few endothelial lined blood vessels. The biopsy report confirmed the diagnosis of OKC of maxilla. Patient was advised complete resection of the lesion but she did not turn up for the treatment advised.

### Discussion

Odontogenic keratocyst (OKC) is relatively common and most aggressive developmental odontogenic cyst of the jaw .OKC has gained a special attention in the recent year because of its tendency to reccur and aggressive behavior. Many efforts have been made to classify the cyst because of its unusual and specific behavior. Previously, OKC was classified under the developmental odontogenic cysts in 1971 and 1992, in 2005 WHO reclassified and renamed it as keratocystic odontogenic tumor (KCOT).But in recent updated WHO classification of head and neck pathology 2017 KCOT was reclassified again into category of cysts<sup>5</sup>.

OKC may occur in people of any age, but it is most commonly found in the second and third decade of life and is rare in children younger than 10 years of age. OKC shows more male predisposition and tends to occur more commonly in mandible with predilection for molar ramus region. Ali M et al (2003) reviewed 393 patients of OKC, out of which 266 were in mandible and 132 were in maxilla. The most common location was third molar ramus region of mandible (137cysts) followed by maxillary canine region (54 cysts). Maxillary OKC tends to be smaller and unilocular and appear similar to inflammatory apical cyst. But there are inconsistencies regarding the prominent location of OKC in maxilla crossing the midline<sup>6, 7</sup>.In our case, a large radiolucent lesion crossing the midline was present in right maxillary region which was an unusual presentation for OKC and only few cases have been reported in literature of OKC in maxilla crossing midline.

Multiple OKC are associated with Nevoid Basal Cell Carcinoma (NBCCS) or GorlinGoltz Syndrome, characterized by multiple basal cell carcinoma,bifid ribs, calcifications of falxcerebri and frontal bossing. Radiologically, the cyst appears as well defined radiolucency, which can be unilocular or multilocular. The high rate of recurrence of OKC is mainly attributed to thin cystic lining and presence of daughter cysts or the satellite cysts .Recurrence develops usually, within 10 months to 25 years after surgical removal of the cyst<sup>8,9,10</sup>.

The treatment of OKC remains a debatable topic. Whatever the treatment modality used, the aim is to prevent recurrence. OKC is known for its high recurrence rate which lies in the range of 5 to 62.5%. The various surgical approaches include enucleation, marsupialization, decompression, curettage followed by enucleation and treatment of defect with Carnoy's solution, liquid nitrogen therapy, segmental and marginal resection as well as hemi-mandibulectomy<sup>11</sup>. The treatment of choice for small OKC is enucleation with Carnoy's solution as it results in lower recurrence rate, however, in cases of larger cyst, marsupialization is the treatment of choice.

### Conclusion

OKC is the third most common jaw cyst after the radicular and dentigerous cyst. The existing published literature shows that it has more predilection to occur in the mandibular posterior region and less likely to be in maxilla with involvement of maxillary midline and rarely crossing the midline. In spite of its large range of incidence, the lesions crossing the maxillary midline have been reported in older individual but in our case the reported individual is female and of young age group. Proper clinical examination and radiographic investigation along with biopsy is important for exact diagnosis and appropriate treatment of OKC to prevent recurrence.

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