

## Keratocystic Odontogenic Tumor-A Rare Pediatric Case Report with an Impacted Premolar.

### Abstract:

Keratocystic Odontogenic Tumor which presently is also termed as Odontogenic keratocyst (OKC) this new title was given by WHO in 2005. It is a benign intraosseous cystic lesion of the gnathic bones which probably originates from the remnants of the dental lamina. We Present the case of a female child aged 12 years with aggressive KCOT and impacted maxillary second premolar

**Key-words:** KCOT, OKC, Carnoy's solution, Enucleation.

### Introduction:

According to WHO it is a benign uni or multicystic intraosseous tumor of odontogenic origin (dental lamina and its remnants) with characteristic lining of parakeratinised stratified squamous epithelium and potential for aggressive and infiltrative behavior. WHO proposed the terminology as keratocystic odontogenic tumor (KCOT) as it shows neoplastic nature. However, In 2017, the new WHO classification of Head and Neck pathology re-classified OKC back into the cystic category.[1]

This lesion features a slight preference for the male gender, second and fourth decade of life are mainly affected, involving posterior region of the mandible and ascending branch.[2]

The tumor has a predilection for the mandibular 3rd molar region and usually manifest as multilocular radiolucency with scalloped well-defined margins. Aspiration biopsies reveal keratin flakes with protein levels <4 g/100 ml.[3]

The common presence of KCOT posterior to the 3<sup>rd</sup> molar region is difficult to explain if dental lamina is believed to be the etiological derivative due to the unlikely possibility of remnants or offshoots of this dental lamina being located in the

mucosa posterior to the last molar. It is therefore probable that offshoots of the basal layer of the epithelium of the oral mucosa may also be involved in the etiology of KCOTs[4,5]

### Case Profile:

A Female child of 12 year reported to the department with the chief complain of painless hard swelling on the left side of face (no associated pain, paresthesia or discomfort) which was progressively increasing since last 4-5 months. Medical Examination revealed no systemic illness and no associated syndrome.

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Extraoral examination disclosed asymmetry on the left side, (FIGURE-1) A swelling was observed which was firm and diffused in zygomatic region. Ala of the nose was obliterated. The swelling was present medially and laterally on zygomatic arch region, superiorly over the infraorbital rim and inferiorly over 1 cm over the angle of mouth.



Figure-1: Extraoral Swelling on the left side of the face

Intraorally the hard and diffused swelling which was 4x2x2 cm in diameter in the buccal vestibule extended from distal aspect of canine to mesial aspect of second permanent molar. (FIGURE-3)



Figure-3 : Intraoral Features

### Investigations:

FNAC (Fine Needle Aspiration Cytology) showed yellowish straw-colored fluid which is a prime indication of a cystic lesion. Panoramic radiograph disclosed on the left side, well defined radiolucent lesion beneath primary second molar(maxillary), unerupted second premolar and slight resorption of first premolar. Routine laboratory parameters were normal. (Figure-2)

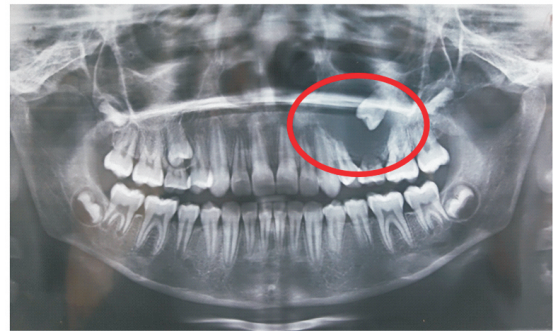


Figure-2 Pre-Operative OPG

Differential Diagnosis: Initially, the lesion was suspected to be dentigerous cyst based on clinical and radiographic presentation. Adenomatoid odontogenic tumor and keratocystic odontogenic tumor were also to be inclined to think.

### Treatment:

As the child patient was definitely positive on Frankel Behavior Rating Scale, the complete process was under local anesthesia. Trapezoidal incision(mucoperiosteal) was given extending from upper left lateral incisor to first permanent molar. Along with cystic content primary second molar and impacted second premolar was removed. After complete osteotomy a cotton pledget dipped in Carnoy's solution (1 g of ferric chloride (FeCl<sub>3</sub>) dissolved in 6 mL of absolute alcohol, 3 mL of chloroform, 1 mL of glacial acetic acid) was applied over the area for about 5 min for chemical cauterization of the tissues. Betadine irrigation was done followed by placement of G bone Modified Hydroxyapatite granules. Incision was closed by Interrupted sutures. (FIGURE-4)

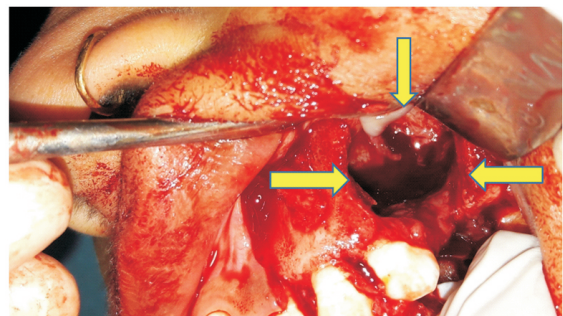


Figure-4 : Lesion with Impacted Second Premolar

### Discussion:

Keratocystic odontogenic tumor (KCOT) or also termed as OKC is one of the rare odontogenic cysts originating from the dental lamina remnants before odontogenesis is complete in maxilla and mandible. With high recurrence, OKC is locally

destructive. Due to lack of Specific clinical and radiographic characteristic, KCOT of maxilla is difficult to diagnose. With only 31.3% cases reported in maxilla, mandible is more commonly affected. Bimodal age distribution is mostly observed with first peak in 2<sup>nd</sup> and 3<sup>rd</sup> decade and second peak in 5<sup>th</sup> decade or later.

Microscopically the given H & E-stained tissue section exhibits fibrous connective tissue capsule with attached epithelial lining. There was also separation of epithelium from connective tissue at few areas. Epithelium shows columnar basal cells with reverse polarized nuclei at few areas and flat cells at few areas with keratinized surface layers. Surface of the epithelial lining was predominantly parakeratinized. There were areas with thin lining of epithelium. Epithelial connective tissue interface was corrugated and proliferated at many areas. One end of the section shows dense infiltration of chronic inflammatory cells with proliferation of cystic lining. (FIGURE-5)

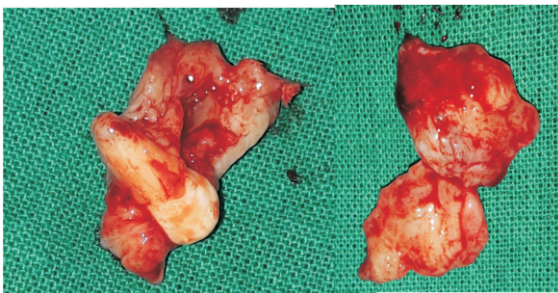


Figure-5 : Lesion was sent for Histopathology

Connective tissue was fibrocellular in nature with sub-inflammatory cell component at few areas.

The recurrence of these lesions in children has not been well documented; however, Hyun et al.<sup>6</sup> reported a case of recurrent OKC in a 7-year-old male patient who presented a lesion in the right mandibular body. The initial treatment recommended by these authors was marsupialization, followed by curettage, and only after relapse of the lesion a complete enucleation was performed. In general, recurrences are not commonly described in pediatric non-syndromic patients.

Therefore, it must be emphasized that the stage of development and eruption of the tooth need to be considered for the therapeutic planning of pediatric patients<sup>7</sup>. Infants with permanent, non-erupted teeth need a more conservative surgical treatment whenever it is possible.

Third molar followed by maxillary canine are the two most common impacted teeth and dentigerous cyst is the most common cyst associated with these impacted teeth. According to the case we presented, in this young female, other than dentigerous cyst, KCOT was primarily considered since it has features similar to both dentigerous cyst and Adenomatoid Odontogenic Tumor.

Significant healing was observed in the patient when post-operative OPG was recorded, (FIGURE-6) who has been asymptomatic for 4 months after the operation. As the lesion has a high recurrence rate, so a follow-up period of at least 5 years is necessary.

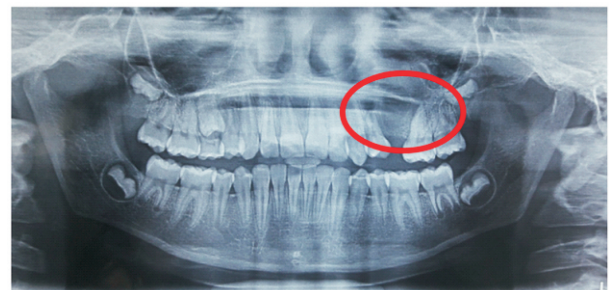


Figure-6: Post Operative OPG after 4 months of follow up

### Conclusion:

Enucleation with Carnoy's solution and Peripheral Osteotomy has combined effect of Carnoy's solution and peripheral osteotomy. The cyst is first enucleated followed by rinsing the defect with saline and then packing it with a gauze soaked with Carnoy's solution and leave it for 3 minutes. Then cystic lumen is re-rinsed with saline to see cystic wall remains, which would be dark brown colored and fixated, thus allowing complete removal of remains. After that peripheral osteotomy is performed and overlying attached mucosa is excised. Finally, defect is filled with Vaseline-iodoform gauze.

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