

## ODONTOGENIC KERATOCYST IN ANTERIOR MAXILLA MIMICKING RADICULAR CYST: A CASE REPORT

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**ABSTRACT**

Odontogenic keratocyst is a common developmental odontogenic cyst arising from the remnants of dental lamina characterized by aggressive clinical behavior, increased potential for recurrence and its occasional association with Gorlin- Goltz syndrome. The lesion primarily occurs in early adults with increased preponderance to occur in the third molar region of the mandible. On certain occasions, odontogenic keratocyst may occur in unusual sites such as the ramus, maxillary sinus and in the anterior region. The occurrence of the lesion in the maxillary anterior region especially in relation to the periapical region of the tooth may pose diagnostic difficulties owing to its similarity to a radicular cyst. The difference in the clinical behavior and prognosis of odontogenic keratocyst from that of a radicular cyst makes it mandatory to differentiate between the two lesions. This presentation reports a periapical lesion in anterior maxilla diagnosed histopathologically as odontogenic keratocyst with relevant literature review.

**KEYWORDS :** Odontogenic keratocyst; periapical lesion; differential diagnosis; case report

**INTRODUCTION:**

Odontogenic keratocyst (OKC) is a developmental odontogenic cyst arising from the remnants of the dental lamina. The term "odontogenic keratocyst" was introduced by Philipson in 1956 because of the keratin produced by the epithelium, filling the cystic lumen<sup>[1]</sup>. OKC is known for its high recurrence rate, aggressive behavior, and its occasional association with the nevoid basal cell carcinoma syndrome. OKCs may occur at any age but the peak incidence is seen in the second and third decades of life with a gradual decline subsequently<sup>[1]</sup>. Among the jaw bones mandible is more commonly affected than the maxilla with the majority of the cysts occurring in the posterior areas mainly in third molar-ramus region followed by first molar region and sometimes in the anterior region<sup>[2]</sup>. In addition OKC has been reported in unusual locations such as TMJ, maxillary sinus, between the root apices and in the midline region<sup>[3,4]</sup>.

Lesions occurring in the midline may be evident in relation to an impacted permanent or supernumerary tooth; between the roots; in the periapical region or in rare cases away from the tooth bearing sites of the jaw. Precise diagnosis of OKC occurring in periapical region is a pre-requisite for appropriate management owing to its aggressive nature and tendency to recur. Hence, the aim of this article is to report a case of OKC present in the anterior region of maxilla clinically mimicking a periapical cyst.

**CASE REPORT:**

A 65 years old female patient reported with a chief complaint of pain and swelling in the upper front region of the jaw for 5 years. Patient gives history of trauma with 21 and 22 followed by root canal treatment for the same. On intraoral examination a single diffuse swelling measuring 3cm x 2cm in size, extending from 11 to 24 and crossing the midline was seen [Fig 1]. The swelling was tender, hard in consistency causing expansion of buccal cortical plates with no associated tooth mobility. An intra-oral periapical radiograph showed a unilocular radiolucency with a well-defined sclerotic border. Considering the provisional diagnosis as radicular cyst, surgical enucleation of the cyst with peripheral osteotomy and extraction of the associated teeth was performed. Soft tissues adhering to the capsule of the lesion in

the buccal fenestration were also removed and the specimen was submitted for histopathological investigation.



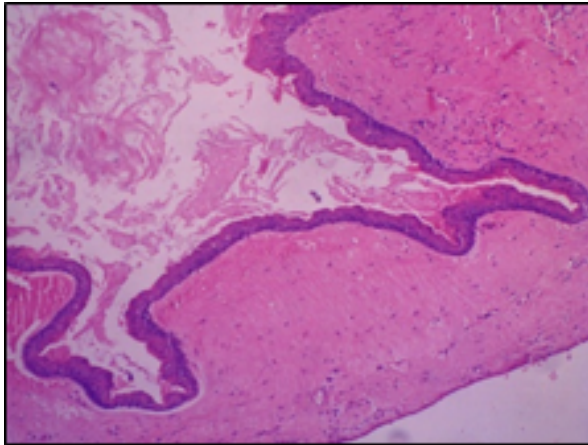
**Fig 1: Intraoral Image Showing Swelling Extending From 11 To 24**

The macroscopic appearance of the submitted tissue specimen demonstrated a soft tissue mass attached to the periapical area of maxillary incisors and canine [Fig 2].



**Fig 2: Macroscopic Appearance Of Cyst Associated With Periapical Region Of Teeth 21, 22 And 23**

Microscopic examination of the soft tissue specimen revealed a cystic lumen lined by corrugated parakeratinized stratified squamous epithelium of uniform thickness without rete ridges and filled with keratin flecks [Fig 3]. The epithelium is folded at many places and appeared to be detached from the underlying connective tissue wall. The basal cell layer showed palisading of hyperchromatic tall columnar cells giving a tombstone appearance. The connective tissue wall is made up of minimal inflammation, many engorged blood vessels and extravasated blood elements. The histological findings were suggestive of odontogenic keratocyst.



**Fig 3: Photomicrograph Showing A Keratinized Epithelium Of Uniform Thickness With Surface Corrugation (H And E X100)**

#### DISCUSSION:

Odontogenic keratocyst is a distinctive form of developmental odontogenic cyst that deserve a special consideration because of its specific histopathologic features and clinical behavior and association with nevoid basal cell carcinoma syndrome<sup>[5]</sup>. While the posterior mandible is the most common site of presentation followed by maxillary posterior region; lesions occurring in unusual sites have been reported in literature. Such lesions may clinically resemble other cysts such as dentigerous cyst, periapical cyst, lateral periodontal cyst and nasopalatine duct cyst thus posing difficulties in diagnosis and subsequent treatment. Based on the anatomic site, Main (1970) had described four radiographic variants of odontogenic keratocyst namely envelopmental, primordial, collateral and extraneous types<sup>[6]</sup>. The aggressive behavior and high rate of recurrence warrants an appropriate diagnosis of OKC especially if it occurs in less common clinical sites such as the periapical region of the tooth.

Several literature reports have described an occurrence of OKC in periapical region of the teeth although the description of such cases in anterior maxillary periapical region is inadequate. Kamali and Lim (2017) reported a case of odontogenic keratocyst mimicking a radicular cyst in relation to periapical region of maxillary central incisor. The lesion was treated by enucleation and peripheral osteotomy and six-month follow-up was uneventful<sup>[7]</sup>. Jafaripozve et al (2013) reported a case of odontogenic keratocyst in the periapical region of anterior maxillary teeth and associated with endodontically treated maxillary lateral incisor. The lesion was treated with excision of the lesion, curettage and preservation of involved teeth with uneventful wound healing<sup>[8]</sup>.

Clinical cases of OKC occurring in the periapical lesion of the mandibular and maxillary posterior teeth was reported by Dasgupta et al (2015) and Gowhar et al (2016)<sup>[9,10]</sup>. The surgical treatment consisted of decortication and curettage of soft tissue mass followed by histopathological confirmation of OKC<sup>[9]</sup>.

#### CONCLUSION:

The occurrence of OKC in the periapical region of anterior maxilla may lead to its interpretation as a radicular cyst. An adequate knowledge of atypical presentations of odontogenic keratocyst is necessary to avoid misdiagnosis. Further, the aggressive behavior, high rate of recurrence and neoplastic transformation of odontogenic keratocyst makes it vital for their accurate diagnosis and further management.

#### REFERENCES:

1. Shear M, Speight P (2008). Cysts of the oral and maxillofacial region. 4th ed. New York, United States: Wiley.
2. Chkoura A, Chbicheb S, El Wady W (2008). Keratocystic odontogenic tumour: a case report and review of the literature. *The Internet Journal of Dental Science*; 6:1-6
3. Ali M, Baughman RA (2003). Maxillary odontogenic keratocyst: A common and serious clinical misdiagnosis. *J Am Dent Assoc*; 134:877-83.
4. Yadav S, Verma A, Sheorain A, Sharma A (2013). An unusual case presentation of follicular odontogenic keratocyst with an impacted mesiodens. *J Craniofac Surg*; 24:e300-2.
5. June HB, Young HK, Mun JC, Bong WP (2013). Expansile keratocystic odontogenic tumor in the maxilla: immunohistochemical studies and review of literature. *J Korean Assoc Oral Maxillofac Surg*; 39:182-7.
6. Shewale VN, Shewale K, Taqui S (2011). Collateral variant of keratocystic odontogenic tumour. *Asian Journal of Oral Health & Allied Sciences*; 1:156-9.
7. Kamali U, Lim D (2017). Keratocystic odontogenic tumour of anterior maxilla mimicking radicular cyst: a case report. *Int J Oral Maxillofac Surg*; 46:306-7
8. Jafaripozve S, Allameh M, Khorasgani MA, Jafaripozve N (2013). Keratocystic odontogenic tumor in the anterior of the maxilla: A case report and literature review. *J Oral Maxillofac Radiol*; 1:90-2.
9. Dasgupta S, Rai S, Misra D, Panjwani S, Singh N (2015). Diagnostic dilemma: Radicular cyst or keratocystic odontogenic tumor? *SRMJ Res Dent Sci*; 6:203-5.
10. Gowhar O, Singh NN, Ain TS, Sultan S (2016). Keratocystic odontogenic tumor of maxilla mimicking radicular cyst. *IJSS Case Reports & Reviews*; 3:1-3.