

# CASE REPORT

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## ODONTOGENIC KERATOCYST: A CASE REPORT WITH REVIEW

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### HOW TO CITE THIS ARTICLE:

Badariramakrishna B, Archana Tangadu, Amit Byatnal, Aditi Amit Byatnal, Shrinivas C. Koppal. "Odontogenic Keratocyst: A Case Report with Review". Journal of Evidence based Medicine and Healthcare; Volume 1, Issue 9, October 31, 2014; Page: 1192-1196.

**ABSTRACT:** Odontogenic Keratocyst (OKC) may be benign cystic neoplasm, OKC may give an erroneous radiographical impression that they are dentigerous, lateral periodontal, residual, or even so called fissural cyst. It was this belief in origin of the cyst from primordial odontogenic epithelium that led for some time use as primordial cyst. The true nature OKC or currently known as keratocystic odontogenic tumor (KCOT) is still a dilemma.

**KEYWORDS:** Keratocyst, odontogenic cyst, keratocystic odontogenic tumor.

**INTRODUCTION:** Jaw cysts, especially odontogenic keratocyst (OKC) interests the dentists as it is apparently known to grow to a large size before it manifests clinically. Also, unlike other jaw cysts, it has particular tendency to recur following surgical removal. What is even more perplexing in OKC is its true nature, due to which it was renamed as keratocystic odontogenic cyst (KCOT) by Philipsen in 2005.<sup>1</sup> However, there is still an ongoing debate among oral pathologists with regard to the neoplastic nature of this lesion. The present paper presents a case of OKC and an attempt has been made to understand its nature.

**CASE REPORT:** A 32-year old male patient presented with a swelling in the left posterior region of the lower jaw of 7 months duration. History revealed that the swelling grew progressively in size with no significant aggravating or relieving factors. Past dental history and medical history was non-contributory. On clinical examination, an intraoral swelling measuring about 2x3 cm in size was observed in the left posterior region of mandible. The swelling was diffuse, asymptomatic, and bony hard on palpation with the overlying mucosa intact [Figure 1].

Orthopantomographic examination showed a well-defined radiolucency approximately 2x4 cm in size, in relation to the left ramus of the mandible associated with a mesially impacted 38 [Figure 2]. Based on the clinical and radiographic evaluation, a preliminary diagnosis of odontogenic keratocyst was made. Radio-graphically it appeared to be envelopmental type. Dentigerous cyst and a benign odontogenic tumour were considered in the differential diagnosis.

The lesion was surgically enucleated along with the disimpaction of 38, under local anesthesia [Figure 3] and submitted for histo-pathological examination. Gross examination of the specimen revealed a thin cystic associated with 38, measuring about 3x5 cm in size. The tissue was friable and was obtained in bits. The surface of the cystic lumen appeared smooth and contained white cheesy material.

Histopathological examination of the H & E stained sections showed a cystic cavity lined by Para keratinized stratified columnar epithelium, with corrugated surface [Figure 4a]. The basal layer comprised of tall columnar cells with well-defined palisading, nuclei oriented away from basement membrane and intensely basophilic.

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The superficial cells appeared polyhedral with intercellular edema at places. The epithelial connective tissue interface was flat with splitting at places, and lack of rete ridges. The capsular tissue appeared dense, with mild inflammatory infiltrate at places. Satellite cysts were also noted at places [Figure 4b].

**DISCUSSION:** Cholesteatoma was the first term ever used to describe OKC (Hauer, 1926; Kostecka, 1929).<sup>2,3</sup> The term odontogenic keratocyst was introduced by Philipsen, 1956.<sup>4</sup> Philipsen and Pindborg and Hansen<sup>5</sup> described any jaw cyst with keratin formation as keratocyst. However, it can resemble other jaw cysts in terms of clinical, radio-graphical as well as histological presentations. Hence, regarding its true nature, genetic contribution is being considered and has been a matter of debate currently.

OKC occurs in the age group of second to third decade most commonly, with slight male predominance. Mandible is the most common site of occurrence, molar-ramus area being affected frequently, as seen in the current case. Patients with OKC complain of pain, swelling or discharge, with occasional experience of paraesthesia of lower lip or teeth. But no such symptoms were noted in our case. The extent of the cyst may be even more than 40 mm, sometimes extending into the ramus or till the inferior border of the mandible. However, maxillary OKCs show rapid expansion as compared to those in mandible.<sup>6</sup>

Radio-graphically, OKC appears intraosseous, unilocular or multilocular and most commonly not associated with impacted tooth. Main in 1970<sup>7</sup> described 4 radiographic varieties of OKC, including envelop mental, replace mental, collateral and extraneous. Association with nevoid basal cell carcinoma syndrome (NBCCS) has been a major concern with regard to OKC. First reported by Gorlin and Gotz,<sup>8</sup> NBCCS has gained interest of dentists as well as general practitioners, due to the severity and aggressive nature of presentation.

NBCCS has variable expressivity, including multiple nevoid basal cell carcinomas, OKCs, congenital skeletal defects, ectopic calcifications, plantar and palmar pits, central nervous system and ocular lesions, with frontal bossing and ocular hyper telorism. Patched (PTCH) gene has been identified as NBCCS gene mapped on chromosome 9q22.3. Recurrence is the most common and worrisome matter in case of OKC, as it is very difficult to curettage the entire cyst in toto.

The epithelium being thin and friable, formation of satellite cysts in the capsular wall and epithelial lining in folding's are considered major factors leading to recurrence. However, genetic control has been considered as most important aspect that determines the recurrence. Resemblance of OKC to dentigerous cyst (DC) and orthokeratinized odontogenic cyst (OOC) causes dilemma for the dentists. An attempt by Byatnal et al<sup>9</sup> is represented in Table 1.

Use of keratin profiling has been done by various researchers to understand the similarities and dissimilarities between the 3 cysts. Based on the table presented, it is clear that OKC or KCOT has a more aggressive nature than DC and OOC. However, the origin of OKC still remains unanswered, whether it originates from dental lamina or reduced enamel. Surgical enucleation, curettage, enblock resection, hemimandibulectomy are the modes of treatment, depending on the size and extent of the lesion. However, post-operative follow-up is a must, to check for recurrences.<sup>6</sup> The present case was treated by surgical enucleation, as it is considered the first line of treatment. The patient is under regular follow-up since last three months, with currently no signs of recurrence.

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**CONCLUSION:** OKC, better known as KCOT is an aggressive lesion, due to its characteristic to recur more commonly. Dental lamina is considered to be the origin of OKC, however, still being questioned. Treatment most commonly considered is surgical excision. Currently, other modes being carried out include use of liquid nitrogen cry-therapy, decompression and marsupialization, with less chances of recurrences post-operatively. Complications of OKC have to be kept in mind as there are many OKCs that have transformed in squamous cell carcinoma and basal cell carcinoma, commonly. To conclude, OKC might be commonly encountered in clinical practice, but understanding its true nature and its prognosis is of utmost importance while treating it.

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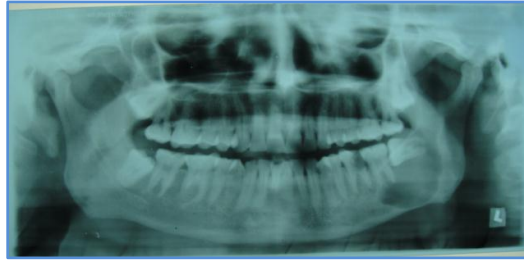
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**Fig. 1**

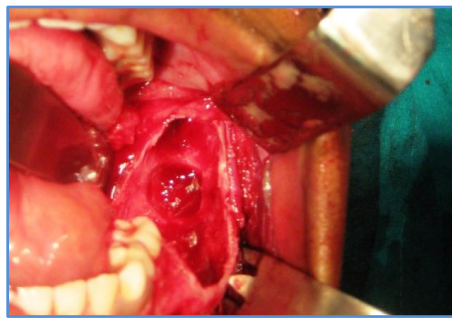
**Fig. 1:** Intraoral examination showing a swelling measuring about 2x3 cm in size

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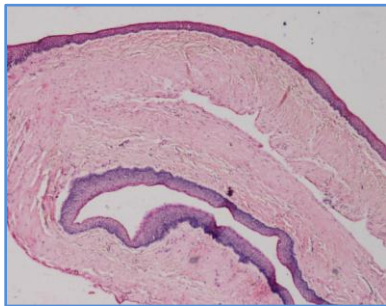
**Fig. 2**

**Figure 2:** Ortho-pantomo graph showing a well-defined radiolucency approximately 2x4cm in size, in relation to the left ramus of the mandible associated with a mesially impacted 38.

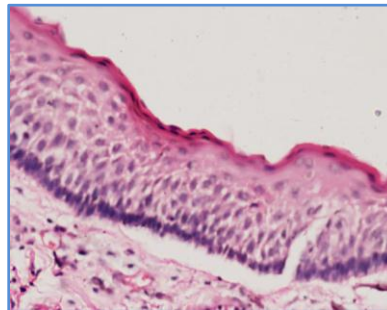


**Fig. 3**

**Figure 3:** Surgical enucleation of the cyst with disimpaction of 38.



**Fig. 4a**



**Fig. 4b**

**Figure 4a:** H&E stained section showing a cystic cavity lined by parakeratinized stratified columnar epithelium, with corrugated surface

**Figure 4b:** H&E stained section showing tall columnar basal cells with nuclear palisading.

|      | <b>OKC</b> | <b>DC</b> | <b>OOC</b> | <b>Study</b>       |
|------|------------|-----------|------------|--------------------|
| CK13 | +          | +         | +          | da Silva MJ et al. |
|      | +          | *         | +          | Meara JG et al.    |
|      | +++        | *         | +++        | Koizumi Y et al.   |

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|          |        |      |     |                  |
|----------|--------|------|-----|------------------|
| CK17     | +++    | +    | +++ | Meara JG et al.  |
|          | +      | *    | +   | Koizumi Y et al. |
| CK18     | -      | +    | -   | Meara JG et al.  |
|          | -      | *    | -   | Koizumi Y et al. |
| CK10     | ++     | *    | ++  | da Silva et al   |
|          | ++     | *    | +++ | Koizumi Y et al. |
|          | ++     | +    | ++  | Stoll et al      |
| CK7      | -      | *    | -   | Koizumi Y et al. |
| CK19     | + / ++ | *    | -   | Koizumi Y et al. |
|          | -      | ++   | *   | Stoll et al      |
| Ki67 (%) | 10.7   | *    | 8.6 | Koizumi Y et al. |
|          | 52.1   | 14.7 | *   | Stoll et al      |
| PCNA     | +++    | +    | +   | Thompson et al   |
| P63      | +++    | *    | +   | Dong et al       |

**Table 1: Expression profile of cytokeratin and labeling index of proliferative markers in OKC, DC and OOC**

CK – Cytokeratin; DC = Dentigerous cyst; OOC = Orthokeratinized odontogenic cyst; KCOT = Keratocystic odontogenic tumor.

- = Negative; + = Mild expression; ++ = Moderate expression; +++ = strong expression; \* = Not performed in the study.

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Date of Submission: 08/10/2014.  
Date of Peer Review: 09/10/2014.  
Date of Acceptance: 21/10/2014.  
Date of Publishing: 23/10/2014.