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# Odontogenic keratocyst masquerading as a dentigerous cyst in the right mandible: A case report of an unusual presentation

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**Conflicts of Interest:** Nil

**Abstract**

We encountered a case of an odontogenic keratocyst (OKC) that radiographically mimicked as dentigerous cyst and was associated with an impacted mandibular third molar. The common site of OKC is posterior mandible, with a lateral radiolucency. The attachment of the radiolucency to the coronal surface of the tooth in a mandibular molar is highly usual. The finding was pathologically confirmed after an excisional biopsy. The clinical and radiographic presentation is different than the typical description of OKC.

**Keywords:** Dentigerous cyst, Odontogenic keratocyst, Mandible, Unusual

## Introduction

Odontogenic keratocyst (OKC) is defined as 'a benign uni- or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior'. It was first described by Philipsen in 1956[(1)]. In 2005, the term OKC was replaced as KCOT by WHO (World Health Organization) based on its aggressive behaviour, recurrence, and mutations in the PTCH (Protein patched homolog) gene, but then in 2017, the term OKC was restored by WHO [(2)]. The odontogenic keratocyst is a relatively common developmental benign odontogenic cyst that represents approximately 10% of all jaw cysts[(3)].

The lesion is named keratocyst because the lining epithelium produces so much keratin that almost fills the cyst lumen. The etiopathogenesis involves many factors, one of them is, cell rests of serres (remnants of the dental lamina)[(4).]. In a recent study done by Deepashri et al in 2012, the OKC was found to be the third most common

odontogenic cyst in an Indian population accounting to 12 cases (8%) with male to female ratio of 1.4:1 and a propensity for mandibular involvement. The majority of patients were in the second and third decades of life[5].The usual presentation is in the mandible and with a lateral radiolucency, and the entity spreads mesiodistally and has minimal buccolateral expansion[6]. This paper highlights a case of OKC occurring in the posterior mandible in association with an impacted mandibular third molar, with a significant swelling in the retro molar area. The radiological investigation showed coronal attachment of the radiolucency, typical of a dentigerous cyst.

**Case Presentation**

A 44-year-old female patient named Mrs. Mahalakshmi, housewife by occupation hailing from porur reported to our department with the chief complaint of pain in her right lower back tooth region for past 2 days. Her history of presenting illness reveals history of pain associated with swelling. History of pain reveals continuous in nature, throbbing type of pain which aggravates during mastication and night time, relieves on medication. Her past medical history reveals hypothyroidism for past 20 years and under irregular medication (Tab. Thyronorm 25mcg). Patient has drug allergy for Tab. Moxikind). Intraoral examination revealed clinically missing: 28, 38, 48and also evidence of single diffuse swelling in the alveolar mucosa in relation 48 measuring approximately 1x1.5cm, extending anteroposteriorly from the distal aspect of 47 to the retromolar region, mediolaterally from the buccal vestibular region to the lingual vestibular region. The mucosa over the swelling appears pale pink in colour. The surrounding mucosa appears to be normal(Fig 1). On palpation regarding site, size, number and shape. The swelling is soft in consistency, tenderness was evident with no secondary changes. By correlating the history given by the patient and clinical findings, we arrived at a provisional diagnosis of Benign Odontogenic cyst or tumour in relation to 48 region.

**Investigations**

Patient was subjected to radiological examination where Intraoral periapical radiograph (IOPA) of 46,47,48 (Fig 2) revealed in 46, evidence of well-defined radioopacity at the distal root. Suggestive of Idiopathic Osteosclerosis or Cementoblastoma in relation to 46. In 48, long axis of 48 is perpendicular to the long axis of 47 and it is present within the alveolar bone. Suggestive of Horizontal impaction in relation to 48. Presence of radiolucency surrounding the coronal portion of 48 which is lined by corticated border. Suggestive of Dentigerous cyst in relation to horizontally impacted 48.

Panaromic radiograph (Fig 3) revealed in 48, long axis of 48 is perpendicular to long axis of 47 and it is present within the alveolar bone. Presence of radiolucency surrounding the coronal portion of tooth which is lined by corticated border. Suggestive of Dentigerous cyst irt horizontally impacted 48.

In order to determine the extent of the lesion, Cone beam Computed Tomography (CBCT) of mandible was performed. In axial section, a single well defined corticated rarefraction is noted with the impacted crown in the left mandible region with its attachment at the CEJ -CEJ. Anteroposteriorly the lesion extend from the distal aspect of the impacted tooth region to the distal root of the second molar causing the distal root resorption of the second molar. Suggestive of dentigerous cyst in relation to 48 region. Cortication of the nerve canal is intact throughout and no evidence of compression, deviation noted (Figure 4).

Routine blood investigations were advised which revealed normal values

Patient underwent extraction of 48 and enucleation of cyst was done. The excised specimen was sent for histopathological analysis.

Histopathological examination with H & E stain section revealed hyperplastic and acanthotic stratified squamous epithelium with focal areas showing squamoid eddies. The cyst beneath shows squamous epithelial lining with focal parakeratosis, keratin flakes and focal ulceration with sheets of lymphocytic infiltrate predominantly plasma cells amidst areas of fibrosis, hyalinisation and focal bony trabecular (Figure 5).

The overall histopathological findings were suggestive of Odontogenic Keratocyst.

**Differential Diagnosis**

The lesion was suspected primarily to be a dentigerous cyst based on the clinical and radiographic presentation of the lesion. Apart from that, Odontogenic keratocyst (OKC) and Ameloblastoma were suggested for differential diagnosis.

Based on the patient's history, clinical findings, radiological findings and histopathological findings, we arrived at a final diagnosis of Odontogenic Keratocyst (OKC) in relation to 48 region.

**Outcome and follow-up**

The patient has been asymptomatic for 3 months after the treatment. As the lesion has a high recurrence rate, so a follow-up period of atleast 5 years is necessary.

**Discussion**

The odontogenic keratocyst is a relatively common odontogenic cyst with a reported incidence of 7 to 11% among all oral developmental cysts. It is one of the most aggressive odontogenic cysts owing to its relatively high recurrence rate and its tendency to invade adjacent tissues[7].OKCs arise from the dental lamina and are characterised by a cystic space containing desquamated keratin with a uniform lining of parakeratinised squamous epithelium[8].

The reported age distribution of OKCs is considerably wide, with a peak of incidence in the third decade of life[9,10] and a slight male predominance[11]. OKCs originate in tooth-bearing regions and the mandible is more often affected than the maxilla[12].When OKCs originate from the mandible, the most common location is the angle or ramus of mandible[13]. Contrarily, the anterior region, mainly between canine and lateral incisor, and the third molar region are the most common sites of origin in the maxilla[14,15].The present follow this conventional norm regarding area specificity of the cyst.

About 50% of the patients are asymptomatic prior to seeking treatment, although there are no characteristic clinical manifestations of the keratocyst. Among the more common features are pain, soft tissue swelling and bone expansion, drainage, and various neurologic manifestations such as paresthesia of the lip[2].The aspirate from this lesion mostly contains a cheesy material suggestive of keratin. Sometimes, the aspirate may also contain a straw-colored fluid.

OKCs may be located in a per coronal position, in a periapical position or in a lateral root position. They have no relationships with any dental structures in about 30% of cases[11,16]. Large lesions, causing significant erosion of cortical plates and involvement of surrounding structures, may be seen in asymptomatic patients[17].Unlike other odontogenic lesions having similar aggressive behaviour such as ameloblastomas, OKCs infrequently cause root resorption of adjacent teeth[11].

Syndromes associated with multiple OKC'S are Nevoid basal cell carcinoma syndrome/Gorlin Goltz syndrome, Marfans syndrome, Ehler danlos syndrome and Noonans syndrome[18].

The radiological imaging techniques most commonly used in the OKCs are conventional radiography (mainly panoramic radiography), computed tomography (CT) and magnetic resonance imaging (MRI). These imaging modalities differ significantly in their technical characteristics, acquisition modalities, indications and information provided[19]. Radiographically, OKCs demonstrate a well-defined radiolucent area with smooth and often corticated margins and may be unilocular or multilocular. An unerupted tooth is seen in association with the lesion about 25 to 40% of cases[20].

There are four radiological variants of OKC. It includes replace mental, extraneous, envelopmental and collateral[21].

Histologically, OKCs has been classified as parakeratotic or orthokeratotic subtypes by some authors. The parakeratotic subtype is the most frequent (80%) and has a more aggressive clinical presentation. It has more production of keratin compared to orthokeratotic subtype[22].OKC is composed of a prominent palisade, polarized layer of cuboidal cells with some corrugations and described as having "picket fence or tombstone appearance"[23].

Treatment of OKCs includes many surgical modalities, such as enucleation, marsupialization, enucleation with use of Carnoy's solution, or cryotherapy, with a marginal or radical section[24].

Even though OKCs are most aggressive and recurrent form of tumors, there are few cases where OKCs have been treated by enucleation. The case discussed here also has been treated with the enucleation procedure. However, a long-term follow-up is required to establish nonrecurrence of the tumor.

The reported recurrent rate of OKCs after surgery is about 30%, with most recurrences occurring after enucleation[25]. The recurrences might be explained by different causes: incomplete removal of the epithelial cyst lining, growth of small satellite cysts left behind by conservative treatment and development of lesions localised in the adjacent region of the jaws[26].

**Conclusion**

Despite its aggressive behavior and high recurrence rate, the etiopathogenesis of the odontogenic keratocyst is not fully understood. Therefore, there is a need for further studies to better understand its characteristics for more accurate diagnosis and for the development and adoption of less aggressive therapeutic approaches that are perfectly adequate for each case in order to prevent its recurrence.

**Abbreviations**

OKC – Odontogenic keratocyst

KCOT – Keratocystic odontogenic tumor

IOPA – Intraoral periapical radiograph

OPG – Orthopantamogram

CBCT – Cone beam Computed tomography

CT – Computed Tomography

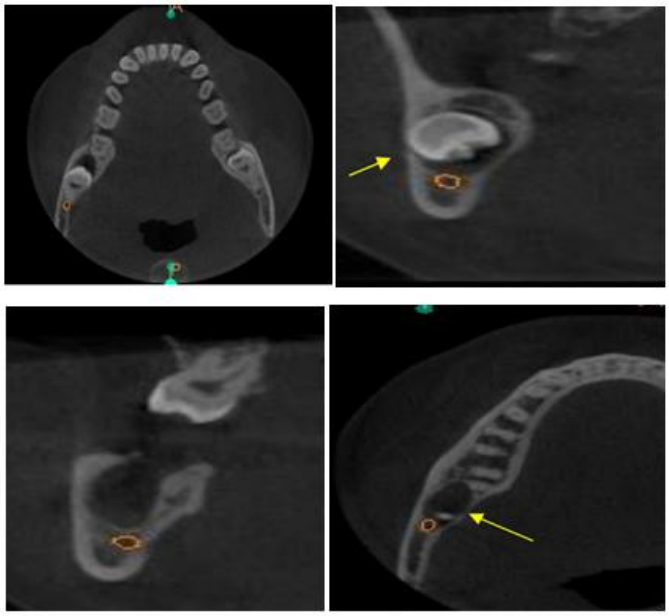
MRI – Magnetic Resonance Imaging

**Legend Figures**

Figure 1: Intraoral view of the lesion

Figure 2: IOPA of 46, 47, 48

Figure 3: Orthopantomogram showing the extent of the lesion



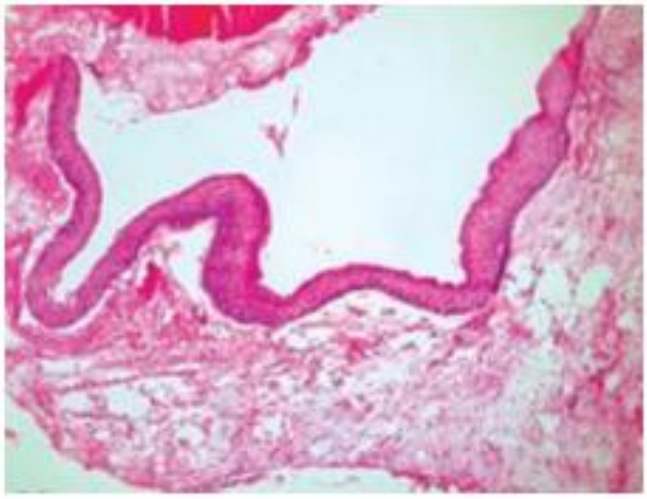


Figure 5: Odontogenic keratocyst

**References**


1. Ongole R. Textbook of Oral Medicine. Elsevier Health Sciences APAC; 2014.
2. Sivapathasundharam B, Rajendran A. Shafer's textbook of oral pathology. Elsevier Health Sciences; 2012 Jun 30.
3. Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, Preda L. Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights into imaging. 2018 Oct;9(5):883-97.
4. Augustine D, Rao RS, Lakshminarayana S, Prasad K, Patil S. Sub-epithelial hyalinization, incomplete cystic lining, and corrugated surface could be a predictor of recurrence in Odontogenic Keratocysts. Journal of Oral Biology and Craniofacial Research. 2021 Jul 1;11(3):423-9.
5. Kambalimath D.H., Kambalimath H.V., Agrawal S.M. Prevalence and distribution of odontogenic cyst in Indian population: a 10-year retrospective study. J Maxillofac Oral Surg. 2014;13(1):10–15. doi: 10.1007/s12663-012-0450-y.
6. Karikal A, Shetty T, Kudthadka A. Odontogenic Keratocyst Masquerading as a Dentigerous Cyst in the Maxilla: A Case Report of an Unusual

Presentation. European Journal of Therapeutics. 2018;24(2):121-5.

7. Blanchard SB. Odontogenic keratocysts: review of the literature and report of a case. Journal of periodontology. 1997 Mar;68(3):306-11.
8. Bilodeau EA, Collins BM (2017) Odontogenic cysts and neoplasms. Surg Pathol Clin 10:177–222
9. Johnson NR, Gannon OM, Savage NW, Batstone MD. Frequency of odontogenic cysts and tumors: a systematic review. J Investig Clin Dent. 2014;5:9–14. doi: 10.1111/jicd.12044
10. Chirapathomsakul D, Sastravaha P, Jansisyanont P. A review of odontogenic keratocysts and the behavior of recurrences. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006;101:5–9. doi: 10.1016/j.tripleo.2005.03.023
11. Avril L, Lombardi T, Ailianou A, et al. Radiolucent lesions of the mandible: a pattern-based approach to diagnosis. Insights Imaging. 2014;5:85–101. doi: 10.1007/s13244-013-0298-9
12. Harmon M, Arrigan M, Toner M, O'Keeffe SA. A radiological approach to benign and malignant lesions of the mandible. Clin Radiol. 2015;70:335–350. doi: 10.1016/j.crad.2014.10.011
13. Kaneda T, Minami M, Kurabayashi T. Benign odontogenic tumors of the mandible and maxilla. Neuroimaging Clin N Am. 2003;13:495–507. doi: 10.1016/S1052-5149(03)00032-7.
14. Ali M, Baughman RA. Maxillary odontogenic keratocyst: a common and serious clinical misdiagnosis. J Am Dent Assoc. 2003;134:877–883. doi: 10.14219/jada.archive.2003.0286
15. MacDonald D, Gu Y, Zhang L, Poh C. Can clinical and radiological features predict recurrence in solitary keratocystic odontogenic tumors? Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;115:263–271. doi: 10.1016/j.oooo.2012.10.018
16. Harmon M, Arrigan M, Toner M, O'Keeffe SA. A radiological approach to benign and malignant lesions of the mandible. Clin Radiol. 2015;70:335–350. doi: 10.1016/j.crad.2014.10.011
17. Eryilmaz T, Ozmen S, Findikcioglu K, Kandal S, Aral M. Odontogenic keratocyst: an unusual location and review of the literature. Ann Plast Surg. 2009;62:210–212.
18. Kargahi N, Kalantari M. Non-syndromic multiple odontogenic keratocyst: a case report. Journal of Dentistry. 2013 Sep;14(3):151.
19. Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, Preda L. Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. Insights into imaging. 2018 Oct;9(5):883-97.
20. Stoelinga PJ. Long-term follow-up on keratocysts treated according to a defined protocol. Int J Oral Maxillofac Surg. 2001;30:14–25.
21. Madhireddy MR, Prakash AJ, Mahanthi V, Chalapathi KV. Large follicular odontogenic keratocyst affecting maxillary sinus mimicking dentigerous cyst in an 8-year-old boy: a case report and review. International Journal of Clinical Pediatric Dentistry. 2018 Jul;11(4):349.
22. Kiran DN, Bali A, Anupama A. Electro-cauterization a good adjunctive therapy to treat Kcot's. Indian J Dent Sci. 2014 Oct;6(4):24–28
23. Cakur B, Miloglu O, Yolcu U, et al. Keratocystic odontogenic tumour invading the right maxillary sinus: a case report. J Oral Sci 2008;50:345–9
24. Cawson RA., Binnie WH., Speight PM., Barrett AW., Wright JN. Luca's pathology of tumors of the

oral tissues. 5th ed. London: Churchill Livingstone; 1998.

25. Johnson NR, Batstone MD, Savage NW. Management and recurrence of keratocystic odontogenic tumor: a systematic review. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013;116:e271–e276. doi: 10.1016/j.oooo.2011.12.028.
26. Mendes RA, Carvalho JF, van der Waal I. Characterization and management of the keratocystic odontogenic tumor in relation to its histopathological and biological features. Oral Oncol. 2010;46:219–225.