

# Odontogenic Keratocyst Simulating Lateral Periodontal Cyst: A Case Report

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## ABSTRACT

Odontogenic keratocyst (OKC) is a common cyst of the oral cavity which has a high recurrence rate, and it has an ability to resemble other jaw cysts. The OKC is an epithelial developmental cyst. OKC is a different entity from other odontogenic cysts that deserves special attention due to its aggressive clinical behavior and high recurrence rate. This lesion is common in the mandible, and can become quite large due to its rapid growth and its extension into the adjacent structures. WHO recommends the term keratocystic odontogenic tumor as it reflects its neoplastic nature. This lesion is commonly found in the mandible, and can become quite large due to its rapid growth and its extension into the adjacent structures. Radiographically, OKC usually appears as a well-defined radiolucency, which can either be unilocular or multilocular. The OKC is a distinct entity from other odontogenic cysts that deserves special attention due to its aggressive clinical behavior and high rate of recurrence. In 2005, WHO recommends the term keratocystic odontogenic tumor for OKC as it reflects its neoplastic nature. The parakeratinized variant are characterized by aggressive growth and tendency to recur after surgical procedures. Multiple OKC's are associated with the nevoid basal cell carcinoma syndrome. Malignant transformation of OKC has also been reported. A case of OKC in the posterior mandible, which was mimicking just like the lateral periodontal cyst is presented in this article. The clinical, radiological, and histopathological features of this cyst and its surgical management are discussed.

**Keywords:** Jaw cysts, Keratocystic odontogenic tumor, Odontogenic keratocyst, Odontogenic cysts, Periapical odontogenic keratocyst

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## INTRODUCTION

Odontogenic keratocyst (OKC) are first described by Phillipson, in 1956. Initially, OKC was known as "primordial cyst," since the origin of the lesion was thought to be from the tooth primordium. The histologic criteria necessary to diagnose OKC were given by Pindborg and Hansen in 1962. Later in 1992, the World Health Organization (WHO) histologic typing of odontogenic tumors listed OKC as the preferred terminology as the cyst contains keratinized lining. Pathogenesis of OKC is mainly related to the cell rests of the dental lamina. The dental lamina considered as the primordial epithelium, which has the ability for keratinization, proliferation, and infiltration of connective tissue during odontogenesis process, which are the characteristics of both OKC and ameloblastoma. OKC might occur along with basal cell nevus syndrome an autosomal dominant condition, which is characterized

by multiple basal cell carcinomas, OKC, skeletal abnormalities.<sup>1-3</sup>

OKC has been of special interest because of aggressive behavior, specific histopathological features, and high recurrence rate.<sup>4</sup> The WHO has designated keratocyst OKC as a keratocystic odontogenic tumor because of its neoplastic nature and 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.<sup>5</sup> OKC commonly occurs due to remnants of basal cells of the oral epithelium and odontogenic epithelial show a male predilection with male to female ratio of 1.6:1. The lesion is more common in second and third decade; keratocyst can occur in any part of the upper and lower jaw, but majority occurring in the mandible, most frequently in the angle and ramus of the mandible.<sup>1,6</sup> Radiographically, OKC present

mostly as a unilocular radiolucency with well-developed sclerotic borders. OKC, which occurs in the maxilla, are smaller in size when compared to the mandible and larger cyst expands bone.<sup>6,7</sup> Differentiating OKC from other cysts of the jaw is important as it is considered as benign neoplasms with high epithelial proliferation. Histologically, three variants were recognized initially: A parakeratinized variant, an orthokeratinized variant, and the combination of the two. "Orthokeratinized odontogenic cyst" has less aggressive clinical behavior and recurrence pattern. Numerous surgical modalities have been suggested in the management of OKC, ranging from decompression alone to simple enucleation with or curettage to resection.<sup>2,8</sup> The purpose of this paper was to report a case of OKC situated in between the roots of lower right premolars, resembling lateral periodontal cyst (LPC), and to distinguish the OKC's from other laterally positioned cysts.

## CASE REPORT

A 20-year-old male patient reported to the Department of Oral Medicine and Radiology with the complaint of swelling in the right lower back teeth region for past 2 months (Figure 1). Clinically revealed, on inspection there was a round diffuse swelling approximately 1 cm in diameter located on the attached gingiva in between the right premolars 44 and 45, the color of the swelling was normal (Figure 2). On palpation, the swelling was soft and fluctuant and on investigations, 44 and 45 were found to be vital on electric pulp testing. An intraoral periapical of 44, 45 region, and orthopantomograph were taken, which defined pear-shaped radiolucency in between lower right premolars along with divergence of roots (Figures 3 and 4). A mandibular occlusal radiograph was taken which revealed, irregular radiolucency in 44 and 45 region (Figure 5). Recent imaging modality, cone beam computed tomography (CBCT) of that particular site revealed the perforation of the buccal cortical plate (Figure 6). Further on aspiration a clear colored fluid was obtained (Figure 7). On the basis of clinical finding, a provisional diagnosis of LPC was given. Surgical enucleation of the lesion was performed under local anesthesia and the lesion was completely removed using a surgical curettage after which the specimen was sent for histopathological examination which revealed, a cystic epithelium showing parakeratinized stratified squamous cystic lining epithelium, which had corrugated surface and flat connective tissue interface, basal cell shows nuclear hyperchromatism arranged in palisaded pattern showing the characteristics of keratotic odontogenic cyst (Figure 8). Thereby correlating the clinical, radiological, and histopathological findings a final diagnosis of OKC was given. The patient was completely asymptomatic after 1 month follow-up postoperatively.



Figure 1: Patient profile view



Figure 2: Intraoral picture: Single round swelling in between 44 and 45 region



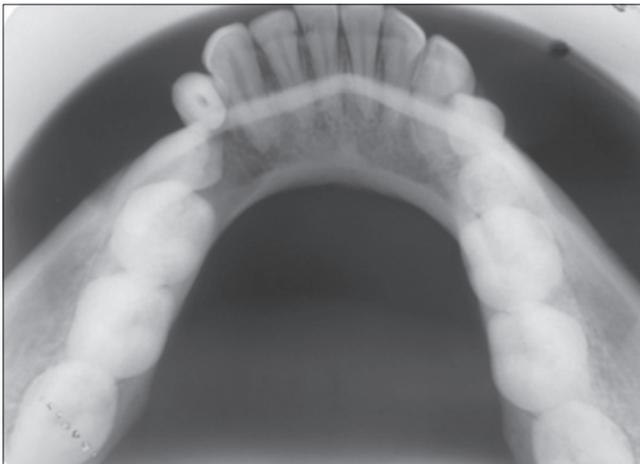
Figure 3: Intraoral periapical reveals an inverted pear-shaped radiolucency in between 44 and 45. Diverted roots of 44 and 45 are seen

## DISCUSSION

The OKC is known for its high recurrence rate, aggressive behavior, and its occasional association with the nevroid basal cell carcinoma syndrome (NBCCS).<sup>2</sup> OKC's accounts for about 7. Among 8% of all jaw cysts. More often OKC



**Figure 4:** Orthopantomograph reveals a well-defined radiolucency in between 44 and 45, which is in inverted pear-shaped appearance along with divergence roots in between 44 and 45



**Figure 5:** Mandibular topographic view reveals an ill-defined radiolucency in the 43 and 44 region extending medially from the distal aspect of 43 and laterally up to the distal aspect of 45

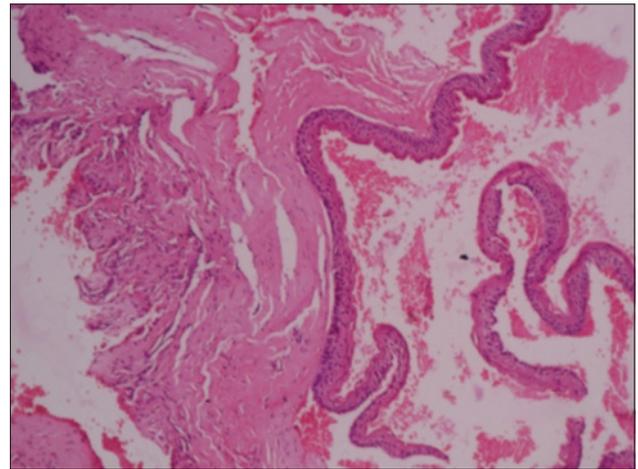


**Figure 6:** Cone beam computed tomography sagittal section: A single well-defined unilocular radiolucency in between 44 and 45

presents as a slow growing swelling in the angle or ramus of the mandible, which is painful. Patient with keratocyst may complain of pain, swelling, discharge, paresthesia of the lower lip, and in rare cases pathologic fractures. In our case, the patient was asymptomatic. The mandible is the most common site to be involved



**Figure 7:** Aspirate revealed a clear fluid



**Figure 8:** The given H and E stained section shows parakeratinized stratified squamous cystic lining epithelium associated with connective tissue. The epithelium is thrown into folds with a corrugated surface and flat epithelial connective tissue interface. The connective tissue shows hyalinated collagen fibers with minimal inflammatory cell infiltrate predominantly of lymphocytes (×10)

than the maxilla. Frequent location for OKC is the posterior body of the mandible (90% occur posterior to the canines) and the ramus. Usually, the epicenter of the lesion is located superior to the inferior alveolar nerve canal.<sup>4,9</sup> Radiographically, OKC shows an unilocular or multilocular radiolucency with root resorption and minimal bone expansion.<sup>10</sup> In our case, the lesion presented as unilocular radiolucency displacement of roots and showed cortical expansion and perforation. The main idea of this paper was to emphasize on the varying clinical and radiological presentation of OKC. The reasons for considering LPC as a provisional diagnosis in the present case is because of the following clinical features: Cyst located in the premolar region, vitality of the associated teeth, asymptomatic diffuse swelling in the premolar region, and radiologically, it showed an single ovoid radiolucency with well-circumscribed borders in between right lower premolars any root resorption. Diagnosis of OKCs is of major diagnostic challenge both clinically and radiographically, so the differential diagnosis that can be considered are radicular cyst, which occurs laterally

as they are usually arise either from the inflammation of periodontal disease spread through a lateral foramen presented with non-vital tooth, as nasopalatine duct cyst when seen in maxillary midline, periapical cyst if located periapically, dentigerous cyst when seen surrounding the crown of unerupted tooth, calcifying odontogenic cyst, unicystic ameloblastoma presents asymptomatic swelling, more commonly in posterior region of mandible associated with root resorption of the associated teeth and bone cyst are which usually associated with the previous history of trauma and common among young adults in mandible. Radiographically, they well-delineated radiolucency with scalloping between the roots.<sup>5,6</sup> Conventional radiographic imaging such as panoramic views and periapical films are adequate to determine the location and estimate the size of an OKC advanced imaging techniques such as computerized tomography and magnetic resonance imaging can also be useful. The use of advanced diagnostic aid like CBCT, in this case, helped measuring the extent of the lesion and to identify the cortical bone perforation. LPC can be clearly differentiated from OKC histopathologically. LPC is lined by thin, nonkeratinized epithelium usually of one to five layer thick. However, OKC is usually lined by epithelium (parakeratinized -83%, orthokeratinized -10%), which is highly characteristic with 6-10 layers ridges.<sup>7,9</sup> Various surgical modalities have been proposed for treatment of OKC. They include curettage, enucleation, enucleation with cornuys solution, marsupialization, and resection. OKC have a high recurrence rate of 62%.<sup>2</sup> Percentage for malignant transformation of OKC, the high recurrence rate of OKC is due to surgical difficulties resulting in improper cyst removal, thin, and friable nature of capsule, retention of the daughter cysts.<sup>1</sup>

## CONCLUSION

This case is one of the examples that the clinical findings and the radiological pictures are not distinctive enough

to give a diagnosis of any cyst occurring in the jaws. It is very difficult to diagnose a cyst by limiting ourselves only to symptoms, age, and sex location. Hence, a proper histopathological evaluation and postoperative follow-up is mandatory.

## REFERENCES

1. Hiremath SK, Deshpande AM, Byakodi S, Magdum DB. Diagnostic dilemma: A case report of odontogenic keratocyst in lateral periodontal position. *Int J Oral Maxillofac Pathol* 2011;2:23-6.
2. Kham SA, Zacaria RK. Under diagnosis of an odontogenic keratocyst – A controversial lesion. *J Dent Med Sci* 2012;2:37-40.
3. Moeini M, Anvar SE, Bafghi RB. A case report of odontogenic keratocyst in anterior mandible position. *Am J Res Commun* 2013;1:286-91.
4. Myoung H, Hong SP, Hong SD, Lee JI, Lim CY, Chung PH, *et al.* Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;91:328-33.
5. Avinash Tejasvi ML, Balaji Babu B, Malik Afroz M. Keratocystic odontogenic tumor mimicking radicular cyst. *IJDA* 2010;2:387-90.
6. Oda D, Rivera V, Ghanee N, Kenny EA, Dawson KH. Odontogenic keratocyst: The northwestern USA experience. *J Contemp Dent Pract* 20005;1:60-74.
7. Mozaffari E, Marmor DS, Alawi F. Odontogenic keratocyst with a misleading clinical and radiologic appearance. *Quintessence Int* 2007;38:837-41.
8. Asokan GS, Jeelani S, Parthiban J, Prabhu Shankar K, Prakash CA, Shankar KA. Keratocystic odontogenic tumor – A case report and review of literature. *Int J Dent Case Rep* 2012;2:87-91.
9. Cakur B, Miloglu O, Yolcu U, Göregen M, Gürsan N. Keratocystic odontogenic tumor invading the right maxillary sinus: A case report. *J Oral Sci* 2008;50:345-9.
10. Faustino SE, Pereira MC, Rossetto AC, Oliveira DT. Recurrent peripheral odontogenic keratocyst: A case report. *Dentomaxillofac Radiol* 2008;37:412-4.

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