Case Report
Diagnostic Dilemma: A Case Report of Odontogenic Keratocyst in Lateral Periodontal Position
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Abstract
Odontogenic keratocyst is a common cyst of the oral cavity with high recurrence rate and it has an ability to mimic other jaw cysts. Present study is a case report of a 45 year old woman presenting with an asymptomatic swelling on the gingiva between the mandibular left premolars. On clinical and radiographic examination, a provisional diagnosis of lateral periodontal cyst was given. Histopathological examination revealed odontogenic keratocyst. The lesion was successfully treated by complete enucleation. The aim of the study is to report a case of odontogenic keratocyst simulating lateral periodontal cyst. Emphasis was given to distinguish odontogenic keratocyst from other laterally positioned cysts and its cause for aggressive behavior and high recurrence rate.

Key Words: Odontogenic Keratocyst; Lateral Periodontal Cyst; Laterally Positioned Cysts; Periapical lesion; PCNA; Ki67; p53.

Introduction
Odontogenic keratocyst’s (OKCs) are epithelial developmental cysts which were first described by Phillipsen in 1956.1 According to latest World Health Organization classification, OKCs are termed as keratocystic odontogenic tumors on the basis of the tumor like characteristics of the lining epithelium and to better convey its neoplastic nature. It is a benign developmental odontogenic tumor with many distinguishing clinical and histopathological features. Among them are, a potential for locally destructive behavior, a relatively high recurrence rate and its association with nevoid basal cell carcinoma syndrome or Gorlin syndrome.2

OKCs are common in males then in females with the ratio of 1.6:1. The lesion occurs over a wide age range with a peak in the second and third decade of life. Keratocysts may occur in any part of the upper and lower jaw with majority occurring in the mandible, most commonly in the angle of the mandible and ramus. In maxillary region, there are inconsistencies regarding the predominant location of OKCs. One study shows that OKCs are divided equally between the anterior and posterior region of maxilla, some show that there are more anterior lesions than posterior lesions and other concluded that the posterior region is the predominant site.3,4

Clinical and radiographic presentation of OKCs, poses a major diagnostic dilemma as they can be located periapically, simulating periapical cyst; surrounding the crown of unerupted teeth, simulating dentigerous cyst; between the roots of teeth, simulating lateral periodontal cyst (LPC) or lateral radicular cyst; or in the maxillary midline, simulating nasopalatine duct cysts. Large unilocular OKCs can be indistinguishable from cystic ameloblastomas.5 Distinguishing OKCs from other jaw cysts is essential as it is considered as benign neoplasm and show high epithelial proliferative index.4 Hence histopathological confirmation and segregation of OKC from other jaw cysts is mandatory as OKCs have high recurrence and aggressive behavior.

The purpose of this paper is to report a case of an OKC situated between the roots of premolar teeth, simulating LPC. Emphasis was given to distinguish the OKCs from other laterally positioned cyst and reason for aggressive behavior and high recurrence rate.

Case Report
A 45 year old woman reported to the out patient department with an asymptomatic, cystic swelling located on the attached gingiva between the mandibular left first and second premolars. Duration of this lesion was one year. She had noticed that the swelling was growing slowly.
Clinical examination revealed a round swelling, almost one centimeter in diameter, fluctuant on palpation and located on the attached gingiva between the mandibular left premolars (Fig 1). The lesion was asymptomatic. The mandibular left first and second premolars were found to be vital to electric pulp testing. A radiograph of the site was taken, and it showed a round radiolucency with a radiopaque margin located between the roots of the first and second premolars (Fig 2). A clinical diagnosis of LPC of developmental origin was made. Differential diagnosis of OKC was given and cyst of inflammatory origin was ruled out as both the premolars were vital. Local anesthesia was used and full thickness mucoperiosteal flaps were elevated (Fig 3). The lesion was completely removed using a surgical curette and mucoperiosteal flap was sutured at position.

Histopathological features revealed characteristic features of OKC. Under scanner view, section showed single piece of cystic tissue, lined by epithelium overlying connective tissue stroma. Under higher magnification, epithelium appeared to be corrugated Parakeratinized stratified squamous epithelium. Epithelium was uniform in nature and ranged from four to eight cells in thickness. The basal layer composed of a palisaded row of cuboidal to columnar cells without rete ridges (Fig 4). A confirming microscopic diagnosis of OKC was made.

**Discussion**

In the present case, OKC was situated between the roots of premolars, mimicking LPC and cyst of inflammatory origin. However, cyst of inflammatory origin was ruled out by establishing the vitality of tooth. Segregation of the cysts occurring in the lateral periodontal position is mandatory for the accurate treatment and prognosis.
According to Shear and Pindborg, the term LPC should be reserved for cysts in the lateral periodontal position in which an inflammatory etiology has been excluded on clinical and histopathological grounds.\textsuperscript{1,5} The WHO's "Histopathological typing of odontogenic tumors" has more or less changed the LPC from a clinico radiographic entity into a histopathological one. In fact, today, the diagnosis of LPC seems to be primarily based on histopathological features.\textsuperscript{6} The OKC appearing lateral to a root is important in the differential diagnosis of LPC, because of its potential aggressive growth and tendency to recur.

Also, OKCs can be mistaken easily for inflammatory lesions, because almost half of the patients presenting with OKCs show inflammatory symptoms such as pain, swelling and drainage.\textsuperscript{7} OKCs may appear as small unilocular radiolucencies and may occur adjacent to a nonvital or endodontically treated tooth.\textsuperscript{8} Hence, radiographic and clinical characteristics of OKCs are not pathognomonic signs and may lead to a difficult diagnosis especially when this lesion is adjacent to teeth with nonvital pulp or inadequate root fillings. When an orthograde re-treatment is ineffective, a retrograde re-treatment with a biopsy is recommended\textsuperscript{9} because OKCs in an unusual location may simulate a periapical lesion.

The use of advanced diagnostic aid such as computerized tomography (CT) displayed a significant role in the assessment and follow up of OKCs. This technique is accurate in; measuring the extent of lesion, exact localization of perforation through the cortex and assessment of soft tissue involvement. Also CT has overwhelming features over conventional radiographs, which include lack of image superimposition preservation of soft tissue details, selective enlargement of areas of interest, high degree of accuracy and the possibility of three dimensional interpretation.\textsuperscript{1}

Aggressive behavior of OKCs is substantiated by immunocytochemical studies. Epithelial cells of OKC, more particularly in suprabasal layer express proteins such as PCNA, Ki67 and p53. These proteins in common are expressed in actively proliferating cells, particularly in neoplasms. Hence OKCs are considered as benign neoplasms.\textsuperscript{4} OKCs have a high recurrence rate of 62 percent.\textsuperscript{3} Reason for recurrences include: thin and friable nature of capsule due to which inadequate surgical removal of cyst; bony perforation and adherence to surrounding soft tissue structure; de novo formation of cyst i.e. remnants of dental lamina not associated with the original OKC, may transform in to OKC; OKCs with Parakeratinized layer have a much higher tendency for recurrence then orthokeratinized variant and presence of satellite cysts.\textsuperscript{7}

Histopathologically, LPC can be clearly demarcated from OKCs. LPC is characterized by thin, nonkeratinized epithelium usually one to five layer thick, resembling reduced enamel epithelium. Many of the lining cells have clear, vacuolated, glycogen-rich cytoplasm. Focal thickened plaques of proliferating lining cells often project in to the luminal areas. However, in case of OKCs the lining epithelium is highly characteristic consists of keratinized surface (parakeratinized-83% and orthokeratinized-10%) which is typically corrugated. Thickness of the epithelium is uniform with 6 to 10 layers without rete-ridges. Basal layer show prominent palisaded arrangement which often described as 'picket fence' or 'tombstone' appearance. Upper portion of the epithelium is composed of stratified squamous epithelium with high mitotic index without any clear cell formation. Epithelial plaque formation is absent in OKCs but the connective tissue wall often show small islands of epithelium.\textsuperscript{1,3,5}

For the treatment of OKCs, multiple modalities have been published, which include, decompression, simple enucleation with or without curettage and finally resection of involved jaw. With simple enucleation the recurrence rate of OKCs is very high. Hence enucleation followed by application of Carnoy’s solution and decompression followed by enucleation demonstrates less recurrence rate and are less invasive than resection.\textsuperscript{2}

**Conclusion**

This case has shown that, the clinical impression and radiographic picture are not distinctive enough to be used as the sole criterion for rendering a diagnosis of any cyst occurring on the lateral periodontal position. OKCs occurring in such a location are commonly mistaken for inflammatory...
lesions of endodontic origin and LPC. Due to high recurrence rate and aggressive behavior of OKCs all the tissues removed from lateral periodontal position, should be submitted for histopathological evaluation and a definitive diagnosis.

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**References**

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