Orthokeratinized Odontogenic Cyst Mimicking Mandibular Paradental Cyst: A Rare Case
Swaroop Savanur, Shitalkumar Sagari, Roopa Babannavar, Bishen Kundendu A

Abstract
The orthokeratinised odontogenic cyst is a rare epithelial developmental cyst of the jaws. This lesion resembles odontogenic keratocyst that is commonly found in the mandible, and can become quite large due to its rapid growth and its extension into the adjacent structures. Clinically the parakeratinizing odontogenic keratocyst is characterized by aggressive growth and tendency to recur after surgical treatment in contrast to orthokeratinised odontogenic keratocyst. We report a rare case with no clinical signs and symptoms other than pain in relation to left posterior mandibular region. The intraoral soft tissues adjoining to third molar are absolutely normal, whereas radiographic appearance were suggestive of Paradental cyst and periodontal cyst. The excised tissue specimen diagnosed as orthokeratinised odontogenic cyst.

Keywords: Odontogenic keratocyst; Paradental cyst; Pericoronitis; Curettage.


Introduction
Odontogenic keratocyst (OKC) is the one of the atypical developmental odontogenic cysts, which attracts many researchers due to its distinctive characteristics. OKC originates from the dental lamina remnants in the mandible and maxilla before odontogenesis is complete. It may also originate from the basal cells of overlying epithelium. OKC was first identified and described in 1876. Further Phillipsen classified it in 1956.1

Paradental cyst is defined as “a cyst occurring near to the cervical margin of the lateral aspect of the roots as a consequence of an inflammatory process in the periodontal pocket.” The incidence of the lesion reported in studies ranges from 3% to 5%, suggesting its rarity. The 1992 WHO classification included two categories of inflammatory odontogenic cyst, the radicular cyst, and the paradental (inflammatory collateral, mandibular infected buccal) cyst.2 This latter entity was defined as an inflammatory cyst arising on the lateral aspect of a vital tooth as a result of an inflammatory process in the periodontal pocket. The paradental cyst is commonly misinterpreted when associated with atypical clinical and radiographic characteristics, in turn causing diagnostic problems.

In 1962, Pindborg and Hansen3 suggested the histological criteria necessary to diagnose OKC. In recent years, World health organization4 recommended the term cystic neoplasm (now known as keratocystic odontogenic tumor (KCOT) for this lesion, as it better reflects aggressive clinical behavior, histologically high mitotic rate and association with genetic and chromosomal abnormalities.

The orthokeratinised odontogenic cyst (OOC) was first described by Schultz in 1927 as a dermoid cyst, and in 1945, it was considered by Phillipsen as an eventual type of odontogenic keratocyst. In 1981, Wright specified its clinicopathologic aspects, assuring that OOC was an individual entity, distinct from other odontogenic cysts, including the OKC.5 Hence the present article describes a rare case of orthokeratinised odontogenic cyst mimicking radiologically as a paradental cyst.

Case Report
A 40-year-old female patient who reported to the private dental hospital, with the complaint of episodes of intermittent acute pain associated with left mandibular third molar. On clinical examination, extra orally there was no facial asymmetry; intra orally there was no history of swelling and surrounding tissue in relation to erupted third molar was absolutely normal (Figure 1). There was slight tenderness on vertical percussion.

An Intraoral periapical (IOPA) radiograph revealed a radiolucent lesion in distal root of left mandibular third molar, but did not cover entire lesion, hence a panoramic radiograph
was taken. Panoramic radiograph revealed well-defined radiolucency in association with distal root of mandibular left third molar. Radiolucency was surrounded by fine, well-defined thin sclerotic border appearing to be attached distal root below cement-enamel junction and covering till the apex of root, which was almond in shape and 0.5 cm in diameter (Figure 2).

Slight widened periodontal ligament space and the lamina dura of the tooth were appreciated. No resorption or displacement of the associated tooth was seen. All the clinical and radiological features were suggestive of lateral periodontal cyst and inflammatory collateral cyst. The vitality of the tooth was confirmed with an electric pulp tester. The tooth was extracted under local anesthesia and the socket was curetted. The excised specimen was further sent for histological evaluation.

Histopathological examination of the hematoxylin & eosin stained sections showed a cystic lumen lined by orthokeratinised stratified squamous epithelium lining with surface showing flakes of keratin set in layers. The epithelial lining was about 5 to 7 layers with a prominent granular cell layer adjacent to cornified layer and basal layer showed low cuboidal morphology with lack of palisading nuclei (Figure 3 & 4). The epithelial-connective tissue junction was somewhat flat with absence of rete ridges. The connective tissue capsule was made up of dense fibrous connective tissue with minimal chronic inflammatory cell infiltration. Based on the clinical, radiographic and histopathological features, the cyst was diagnosed as orthokeratinised odontogenic cyst (OOC). Six months follow up radiograph revealed no recurrence of the lesion (Figure 5).

**Discussion**

A thorough literature survey revealed that Hofarth in 1930 had reported several cases of a jaw cyst located distal to mandibular third molars with clinical signs of pericoronitis and named it “marginal wisdom tooth cyst”. The clinical, radiological and
histological description by Hofarth\(^6\) fulfils the criteria of a Paradental cyst as defined today. However, it is Main who is credited for first describing the lesion. Various other terms used to describe the condition and its variants include Craig’s cyst, mandibular infected buccal cyst, inflammatory paradental cyst, buccal bifurcation cyst, cystis paradentalis, Hofarth’s mandibular cyst and eruption pocket cyst.\(^5\) The present case radiographically appeared as inflammatory Paradental and lateral periodontal cyst, but the periodontal condition was absolutely normal.

Figure 2: Orthopantamogram showing well-defined radiolucency in association with distal root of mandibular left third molar

Despite of many classifications and nomenclature, unfortunately the clinicians still have to face difficulties in the diagnosis and management of Odontogenic keratocyst. OKC representing as mandibular third molar distal root cyst is very rare, so it should not be neglected to rule out even in small cystic lesions.

Further limited data exists regarding the recurrent rate of the combined Orthokeratinized and parakeratinized variants.\(^5\) In a series of 449 cases of odontogenic keratocyst now known as Keratocystic Odontogenic Tumour (KCOT) assessed by Crowley et al, 86% of cases were parakeratinized, 12.2% were orthokeratinised, and 1.6% had features demonstrating both orthokeratin and parakeratin variants.\(^5\) The recurrence rate of the parakeratinized variant in that study was 47.8%, where as that of Orthokeratinized OKCs was 2.2%.\(^5\) Subsequently, differences in cytokeratins, EMA and CEA Immunohistochemical reactivity were seen between them, hence the orthokeratinised variant of the KCOT was separated as an entity and termed “orthokeratinised odontogenic cyst” (OOC).

The orthokeratinised odontogenic cyst (OOC) was first described by Schultz in 1927 as a dermoid cyst, and in 1945, it was considered by Phillipsen as an eventual type of odontogenic keratocyst (OKC). In 1981, Wright specified its clinicopathologic aspects, assured that OOC was an individual entity, distinct from other odontogenic cysts, including the OKC.\(^9\) OOC has been reported to occur among young adults, with a male predominance. Mandible is affected twice more commonly than maxilla, with a predilection for the most posterior region. Incidentally, about 75% of OOCs are associated with impacted teeth, clinically and radiographically mimicking a dentigerous cyst.\(^9\)

The biological features of the cyst epithelium may be related to recurrences of OKCs, the mitotic index of epithelial cells lining odontogenic keratocyst is greater than in
other types of odontogenic cysts. Keratins associated with squamous differentiation or cornified epithelium (KL1, CKs 10/13 and AE1) showed pronounced staining in all but basal cells of the OOC, whereas in the parakeratinized OKCs staining was found only in the upper and surface parakeratin layers. Both EMA and CEA were consistently present in the surface parakeratin layer of the OKC but completely absent in the orthokeratinised lining. Mujio et al. studied p63 expression in parakeratinized and orthokeratinised keratocysts and concluded that parakeratinized keratocysts show more intense and diffuse expression of OKCs in compare to orthokeratinised keratocysts.13, 14

OOC is having a predilection for posterior mandible where the lining epithelium keratinizes, suggests that OOC may also originate from dental lamina and its remnants. The differences in its histological presentation and behaviour yet raise a few questions about its pathogenesis. Hence research should be carried out with more number of cases to understand the histogenesis of OOC. All the features that characterize an OOC were present in the reported case. The less aggressive nature of OOC seems to correlate with its pattern of cell proliferation and differentiation in their epithelial lining, which differ greatly from KCOT.

The differential diagnosis of the OOC includes other radiolucent lesions of the jaws, mainly odontogenic lesions such as, dentigerous cyst or paradental cyst. Odontogenic tumors such as ameloblastoma and KCOT should be included. The OOC presents similar radiographic characteristics with the ameloblastoma and the KCOT, such as its tendency to involve the mandibular angle or to appear as a multilocular radiolucency. Unlike these entities, the OOC does not cause root resorption, which is a frequent characteristic of ameloblastoma and KCOT.

**Conclusion**

The OOC has been considered as the developmental odontogenic cyst. The uniqueness of this case is the attachment of the cystic lining to the distal root of mandibular third molar, which gave an initial impression of Paradental and periodontal cyst. Hence the pathogenesis of OOC is yet to be explored, to better understand its clinical presentation and treatment plan. Numerous studies and case reports exclusive of OOC need to be compiled for validation of its pattern of cell proliferation and differentiation.

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**Author Affiliations:**
1. Dr. Swaroop Savanur, Professor and Head, Department of Orthodontics and Dentofacial Orthopedics, 2. Dr. Shitalkumar Sagari, Reader, Department of Oral and Maxillofacial Pathology, 3. Dr. Roopa Babannavar, Reader, Department of Conservative & Endodontics, Yogita Dental College and Hospital, Ratnagiri – 415709, Maharashtra State, 4. Dr. Bishen Kundendu A, Associate Professor, Department of Oral Pathology, Jodhpur Dental College, Jodhpur, Rajasthan, India.

**References**

8. Crowley TE, Kaugars GE, Gunsolley JC. Odontogenic keratocysts: a clinical and histologic comparison of the parakeratin...

Corresponding Author:
Dr Shitalkumar Sagari,
Dept. of Oral and Maxillofacial Pathology,
Yogita Dental College and Hospital,
Khed, Ratnagiri – 415709,
Maharashtra State, India.
Ph: +91 9887199662
Email: sheetalkumar9000@gmail.com

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