

**Case Report / Olgu Sunusu**

**Keratocystic Odontogenic Tumor – A Diagnostic Dilemma?  
Keratokistik Odontojenik Tümör- Bir Tanı İkilemi**

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**ÖZET**

Çene lezyonları içinde keratokistik odontojenik tümör, ameloblastom ve dentigeröz kist formları en yaygın olanlardır. Odontojenik lezyonların her birine ait klasik klinik özellikler olmasın rağmen bazen histopatolojik değerlendirme olmadan klinik tanı koymada zorlukla karşılaşılabilir. Birkaç lezyonda yaş, cinsiyet ve demografik veriler üst üste binebilir. Biz, burada klinik tanının meydan okuyucu ve ilginç olan benzer tipte bir olguyu sunduk.

**Anahtar kelimeler:** Keratokistik odontojenik tümör, odontojenik keratokist

**ABSTRACT**

Among the jaw lesions, keratocystic odontogenic tumor, ameloblastoma, dentigerous cyst forms the common ones. Even though there are classical clinical features for each of the odontogenic lesions, at times, it may be difficult to arrive at a clinical diagnosis for the lesion without the histopathological examination. Age, gender and demographic data can be overlapping in few lesions. We hereby present an interesting case of similar type where in clinical diagnosis was challenging and interesting.

**Keywords:** Keratocystic Odontogenic tumor, Odontogenic Keratocyst

**INTRODUCTION:**

Keratocystic odontogenic tumor (KCOT), formerly known as the odontogenic keratocyst (OKC), is a benign unicystic or multicystic intraosseous neoplasm of odontogenic origin, which arises from remnants of dental lamina. (1) It is a benign developmental odontogenic tumor with many distinguishing clinical and histologic features. However an accurate clinical diagnosis of KCOT is a challenge.

The first description of KCOT was published in 1956 by Philipsen; the lesion attracted interest because of its specific histopathological features. KCOT occurs across a wide age range with a peak incidence in the second and third decades and a gradual decline thereafter. (2)

**CASE REPORT:**

A 26 year old male patient reported with a chief complaint of pain and swelling in right lower third of face since 1 month. Swelling was gradual in onset involving right ramus, angle and body of mandible and progressed over a period of 2 years to the present condition (Fig 1). Mild intermittent type of pain was present over right posterior teeth region, which increased on biting and chewing hard foods with a Visual Analogue Scale (VAS) of 5. Aesthetic concern was there for the patient since 1 month. There was no history of trauma, middle ear infection, exanthematous fever or paraesthesia. Family history, Dental history and Personal history were non-contributory.



Figure 1 - Facial asymmetry in right middle and lower third of face

On extra oral examination, there was a diffuse swelling measuring approximately 5 cm x 3 cm involving right ramus, angle and body of mandible

with no signs of erythema, ulceration, ecchymosis, bleeding or discharge. It was bony hard in consistency and was tender on palpation. There was no step deformity and overlying skin was pinchable. On intra oral examination, a diffuse erythematous area was present in the right buccal vestibule extending from mandibular first premolar to mandibular second molar region with clinically unerupted mandibular right third molar (Fig 2). Tenderness was present on vestibular palpation and was bony hard in consistency. However there was no obliteration of buccal vestibule and the aspiration was negative.



Figure 2 - Diffuse erythematous region in right buccal vestibule from mandibular right first premolar to mandibular second molar relation region.

Based on history and clinical examination, a provisional diagnosis of dentigerous cyst due to suspected impacted mandibular right third molar was given. Differential diagnosis of KCOT, Ameloblastoma and Calcifying Epithelial Odontogenic Tumor (CEOT) were considered. It was decided to go in for an intraoral periapical radiograph in relation to mandibular right third molar (Fig 3) which revealed a well-defined radiolucency measuring approximately 2 cm x 1 cm with sclerotic margin extending from mesial root of mandibular right first molar along the apical region of mandibular right second molar extending to the coronal part of impacted mandibular right third molar.

Orthopantomogram (OPG) (Fig 4) showed diffuse multilocular radiolucent lesion measuring approximately 7 cm x 2 cm involving right coronoid process, part of condylar process, ascending ramus, angle and body of mandible extending anteriorly 1 cm from midline and posteriorly involving the inferior border of ramus of mandible. Within the radiolucent lesion, a button shaped radioopacity indicative of horizontally impacted mandibular right third molar in buccolingual direction is seen. However origin of radiolucent lesion couldn't be

traced to the level of cementoamel junction of mandibular right third molar and as the lesion is extensive involving a major portion of right mandible; radiographic impression was in favour of KCOT, ameloblastoma. Computerized Tomographic images (Fig 5) confirmed the amount of bone destruction and extent of the lesion.



Figure 3 - Radiolucent lesion extending to the crown of impacted mandibular right third molar.

Even though, mild to moderate amount of buccolingual expansion was evident, anteroposterior and superoinferior extent of the lesion was more obvious and could be clearly visualized in CT images. Hence, it was finally diagnosed as KCOT. The patient was subjected to resection of the affected part, followed by reconstruction with temporomandibular joint reconstruction plate. The excised specimen was sent for histopathological evaluation which confirmed the lesion as KCOT.



Figure 4 - Diffuse multilocular radiolucent lesion involving right coronoid process, part of condylar process, ramus, angle and body of the mandible.

## DISCUSSION:

Odontogenic cysts are the most common form of the cystic lesions that affect maxillofacial region. Odontogenic keratocyst (OKC) is so called because of the ability of the cyst epithelium to produce keratin which gradually fills the cyst lumen. (3) The first to point out the neoplastic potential of OKC was Toller. (4) In 1967, Toller suggested that OKC may best be regarded as a benign neoplasm rather than conventional cyst based on its clinical behaviour.

(5) KCOT was included in the list of benign odontogenic tumors derived from odontogenic epithelium with mature fibrous stroma without odontogenic ectomesenchyme. (6) World Health Organization (WHO) defined it as a benign uni or multi cystic, intra osseous tumour of odontogenic origin, with a characteristic lining of parakeratinised stratified squamous epithelium and potential for aggressive, infiltrative behaviour. (5,7)



Figure 5 - C T image showing cortical plate destruction and extent of lesion

KCOT occurs over a wide age range with peak incidence in second and third decades. However some series of KCOT had demonstrated a bimodal age distribution with second peak in the fifth decade. (8) KCOT is found more frequently in males than in females. However distribution between genders varies from equality to a male to female ratio of 1.6:1, except in children. (9)

Tsukamoto G et al conducted a comparative study of OKC s associated with and not associated with an impacted mandibular third molar and it was found that the mean age of patients in associated group was approximately 20 years younger than that of patients in the non-associated group. (10)

KCOT tend to involve mandible much more frequently than maxilla. The frequency of mandibular involvement reported ranged from 65 per cent to 83 per cent, with about 73 per cent occurring in the third molar to the ramus region as was seen in this case report. About half of all KCOT s occur at the angle of the mandible extending for various distances into the ascending ramus and forward into the body. (11) They almost always occur within the bone, although a small number of cases of peripheral KCOT s have been reported. (5)

The radiographic findings, although highly suggestive, are not diagnostic. The radiographic findings in KCOT may simulate those of dentigerous

cyst, ameloblastoma, radicular cyst etc. Radiographically, KCOT s may appear as small, round, ovoid, well defined radiolucency, usually with corticated margins, which can be either unilocular or multilocular. (12) Frequently however these lesions may be extensive. There is involvement of an unerupted tooth in about 25 per cent to 40 per cent of cases.(12) The findings of this case report compiled with the above mentioned features as in case report the radiolucency was associated with an unerupted tooth and the radiolucency was well defined and corticated.

Such frequent association of KCOT with unerupted tooth makes it likely for the lesion to be mistaken for a dentigerous cyst.(13) A similar provisional diagnosis of dentigerous cyst was given in the present case.

KCOT tend to grow in anteroposterior direction within the medullary cavity of the bone without causing obvious bone expansion.(14) However in the present case, bony expansion was minimal. Displacement of unerupted or impacted teeth is common but root resorption is rare.(13)

The difficulty of diagnosing KCOT based upon radiographs alone has been recognized in various studies [8] as those of Haring et al who reported that in their series of 60 patients, only 23 per cent of the provisional diagnosis were correct and that most common diagnosis made was that of dentigerous cyst. (13) Proper diagnosis of KCOT is very important for initiation of appropriate therapy to prevent unnecessary complication. (15) In a similar study, Myoung H et al it was found that the radiographic impression was only in concordance with the pathologic diagnosis in 65 of 250 cases and that the most common radiographic impression was dentigerous cyst. (16)

Radiographically KCOT may be of different types. (7) Altini and Cohen studied 17 cases in which the cyst lining was typically KCOT on histological examination but which on macroscopic examination had completely surrounded the crown of the tooth and had been firmly attached to the neck. They introduced the term 'follicular primordial cyst' (follicular keratocyst) for this group of lesions. (11) Main (1970a) has referred to the variety of KCOT that embraces an adjacent unerupted tooth as 'envelopmental'. Those cysts that formed in the place of a normal tooth of the series, he called the 'replacement' variety; and those in the ascending

ramus away from the teeth he referred to as 'extraneous'.

There is a consensus that the histologic diagnosis of KCOT is straight forward. (4) Typical histologic features of KCOT have been well characterized by Philipsen and Browne as: a thin, uniform lining of stratified squamous epithelium with a tendency to detach from the underlying connective tissue capsule; a thin corrugated surface layer of parakeratin; a spinous cell layer 4 to 8 cells in thickness, often showing intracellular oedema; a regular layer of columnar basal cells with nuclear palisading; a flat epithelial fibrous tissue junction, usually devoid of epithelial rete ridges; and a relatively thin fibrous capsule that mostly lacks inflammatory cell infiltrate. (17) The final diagnosis of KCOT requires proper clinical radiographic and histopathologic coordination. (7) The case described in this report is an epitome for the same.

#### CONCLUSION:

The difficulty of diagnosing KCOT based upon clinical features, radiographic features has been well recognized in other studies and was confirmed in the case report. However the final diagnosis must always be done based upon microscopic examination which is the gold standard for the diagnostic dilemma.

#### REFERENCES:

1. Brannon RB. The odontogenic keratocyst. A Clinicopathologic study of 312 cases. Part 1Clinical features. *Oral Surgery Oral Medicine Oral Pathology* 1976;42:54-72.
2. Pindborg JJ, Hansen J. Studies on odontogenic cyst epithelium. Clinical and roentgenological aspects of odontogenic keratocysts. *Acta Pathol Microbiol Scand* 1963;58: 283-94.
3. Prasad G, Yadav NS, Kallianpur S, Bhambal AM. Keratocystic Odontogenic Tumor- a wolf in sheep's clothing. *Annals & Essences of Dentistry* 2010;4(2):244-46.
4. Idle F, Horie N, Shimoyama T, Satio I, Tanaka A et al. Infrequent clinicopathologic features of keratocystic odontogenic tumor: A 29 year multi-institutional retrospective review. *Oral Surgery* 2009;2:1-9.
5. Madras J, Lapointe H. Keratocystic odontogenic tumor: Reclassification of the odontogenic keratocyst from cyst into tumor. *Journal of the Canadian Dental Association* 2008;74(2):165a-165h.
6. Cepeda LA, Rivera DQ, Rocha FT, Huerta L. Reclassification of odontogenic keratocyst as tumor. Impact on the odontogenic tumors prevalence. *Oral Diseases* 2010;16:185-87.
7. Hemavathy S, Roy S. Follicular odontogenic keratocyst mimicking dentigerous cyst-report of two cases. *Archives of Oral Sciences & Research*. 2011;1(2):100-3.
8. Ali M, Baughman RA. Maxillary odontogenic keratocyst. *Journal of the American Dental Association* 2003;134:877-83.
9. Oda D. Odontogenic keratocyst: The North-western USA Experience. *The Journal of Contemporary Dental Practice* 2000;1(2):1-10.

10. Tsukamoto G et al. A comparative study of odontogenic keratocysts associated with and Not associated with an impacted mandibular third molar. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;94:272-75.
11. Shear M and Speight P. *Cysts of the oral and maxillofacial regions*. 4<sup>th</sup> Edition. Singapore: Blackwell Munksgaard 2007.
12. Parikh NR. Nonsyndromic multiple odontogenic keratocysts: report of case. *Journal of Advanced Dental Research* 2010;2(1):71-74.
13. Ong ST, Siar CH. Odontogenic keratocysts in a Malaysian population: Clinical, radiological and histological considerations. *Annals of Dentistry University of Malaya* 1995;2:9-14.
14. Gang TI et al. Diagnostic ability of differential diagnosis in ameloblastoma and odontogenic keratocyst by imaging modalities and observers. *Korean Journal of Oral And Maxillofacial Radiology*. 2006;36:177-82.
15. Mozaffari E, Marmor DS, Alawi F. Odontogenic keratocyst with a misleading clinical and radiographic appearance. *Quintessence International* 2007;38(10):837-41.
16. Myoung H et al. Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. *Oral Surgery Oral Medicine Oral Pathology Oral Radiology Endodontics* 2001;91:328-33.
17. Li T J. The Odontogenic keratocyst: A Cyst or a cystic neoplasm? *Journal of Dental Research* 2011;90(2):133-42.