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http://dx.doi.org/10.17096/jiufd.58691 *ORIGINAL RESEARCH*

*EVALUATION OF KERATOCYSTIC ODONTOGENIC TUMORS USING CONE BEAM COMPUTED TOMOGRAPHY**

Keratokistik Odontojenik Tümörlerin Konik Işınlı Bilgisayarlı Tomografi ile Değerlendirilmesi

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ABSTRACT

Purpose: *The aim of this retrospective study is to determine the radiological features of keratocystic odontogenic tumors (KCOT) using cone-beam computed tomography (CBCT).* ***Materials and Methods:*** *CBCT images of 28 patients who had histopathologically-confirmed KCOT were retrospectively reviewed from the archives of the Department of Dentomaxillofacial Radiology, Gazi University Faculty of Dentistry. The location and size of KCOT, cortical expansion, cortical perforation, relation with the impacted teeth, and the impact on the mandibular canal were evaluated.* ***Results:*** *The mean age of patients at initial diagnosis was 34.5 years. Patients with an impacted tooth were significantly younger than those without an impacted tooth ($p<0.05$). Among KCOTs, 21% were localized in the maxilla whereas 79% were found in the mandible. The lesions localized in the mandible were mostly found in the retromolar-ramus region. Of these patients, 93% had cortical expansion and 75% had bone perforation. Of the 22 mandibular lesions, 20 were in close proximity of the mandibular canal and 18 (90%) of these lesions had caused destruction in the mandibular canal.* ***Conclusion:*** *KCOTs exhibit their aggressive nature in the jaw bone. CBCT is a useful radiological imaging method to examine the radiologic characteristics of KCOTs such as bone destruction and their spatial relations with the neighboring anatomic structures.*

Keywords: *Keratocystic odontogenic tumor; cone beam computed tomography; impacted tooth; inferior alveolar canal; bone destruction*

ÖZ

Amaç: *Bu retrospektif çalışmada keratokistik odontojenik tümörlerin (KKOT) konik ışınlı bilgisayarlı tomografi (KIBT) görüntüleri ile değerlendirilerek radyolojik özelliklerinin belirlenmesi amaçlandı.*
Gereç ve Yöntem: *Histopatolojik olarak KKOT tanısı konulmuş 28 hastaya ait olan ve Gazi Üniversitesi Diş Hekimliği Fakültesi Ağız, Diş ve Çene Radyolojisi Anabilim Dalı arşivinde bulunan KIBT görüntüleri değerlendirildi. KKOT'lerin konumu, boyutları, kortikal ekspansiyonun varlığı, kortikal perforasyon, gömülü dişler ile ilişkisi, mandibular kanal üzerine etkileri incelendi.*
Bulgular: *Hastaların ortalama yaşı 34.5 olarak belirlendi. Gömülü dişler ile izlenilen lezyonların bulunduğu hastaların yaş ortalaması, gömülü diş saptanmayan hastalardan anlamlı olarak daha düşüktü ($p<0.005$). KKOT'ların %21'i üst çenede, %79'u alt çenedeydi. oranında Alt çenedeki lezyonların daha sık olarak retromolar-ramus bölgesinde konumlandığı saptandı. Hastaların %93'ünde kortikal ekspansiyon, %75'inde ise kemik perforasyonu izlendi. Mandibulada lokalize olan 22 lezyonun 20'sinin mandibular kanala yakın olduğu, bunların da 18'inin (%90) mandibular kanalda kemik yıkımına neden olduğu belirlendi.*
Sonuç: *KKOT genelikle çene kemiklerinde agresif özellik gösteren lezyonlardır. KIBT, KKOT'lerin kemik yıkımı gibi radyolojik özelliklerinin ve komşu anatomik yapılar ile ilişkisinin incelenmesinde faydalı bir radyolojik görüntüleme yöntemidir.*

Anahtar kelimeler: *Keratokistik odontojenik tümör; konik ışınlı bilgisayarlı tomografi; gömük diş; inferior alveolar kanal; kemik yıkımı*

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Introduction

Odontogenic keratocysts (OKCs) were described for the first time in 1956 by Philipsen (1). They are developmental epithelial cysts accounting for approximately 11% of all mandibular cysts (1, 2). OKCs have the potential to demonstrate aggressive behavior. Their recurrence rates are high and they may be associated with the nevoid basal cell carcinoma syndrome. There is also ongoing debate over whether these lesions are developmental or neoplastic lesions (3). In 2005, World Health Organization (WHO) classified these lesions as keratocystic odontogenic tumors (KCOT) assuming that this classification would better reflect the neoplastic nature of these lesions. According to WHO, KCOTs are "benign uni- or multicystic, intraosseous tumors of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium" (4). The general consensus is that the OKCs arise from the dental lamina remnant in the mandible and maxilla. However, some authors have also suggested that these cysts originate from the extension of basal cells of the overlying oral epithelium (5). In recent studies, PHCT gene has been implicated in the etiology of KCOTs (6).

In case of a suspected lesion in the mandible or maxilla on computed tomography (CT), magnetic resonance imaging and nuclear medicine imaging are used as supplementary imaging techniques. CT is superior in demonstrating the degree of bone resorption, osteosclerosis, cortical bone swelling, destruction, and calcification (7). The use of conventional CT imaging systems is limited in the practice of dentistry because of high costs, large foot print, and high dose of ionizing radiation. The cone beam computed tomography (CBCT) was originally designed for the visualization of solid structures in the head–neck region. Its most significant advantage is the acquisition of images with higher diagnostic quality using sub-mm resolution and lower doses of radiation (8).

The clinical use of CBCT provides accurate information about the contents and borders of the lesions, their spatial relations with surrounding structures, and cortical expansion for which the conventional radiographic techniques are usually inadequate (9). The aim of the present study was therefore to evaluate the radiologic features of KCOTs using CBCT.

Materials and Methods

Study sample

CBCT images found in the archives of the Department of Maxillofacial Radiology of Gazi University Dental Faculty pertaining to 28 patients diagnosed with KCOT at the Department of Oral Pathology were retrospectively reviewed.

CBCT evaluation

CBCT images were acquired using Promax 3D® (Planmeca, Helsinki, Finland) with exposure settings of 90 kVp, 12 mA, and 13.8 s. These images were assessed in dimly lit and calm environment while maintaining a 50 cm distance to the 24-inch medical monitor that offers 1920 × 1080 pixel resolution and using Romexis® (Planmeca, Helsinki, Finland) computer software. On these images, the locations of the lesions in the maxilla and mandible (anterior, premolar, molar) as well as the mandible in retromolar area and ramus were determined. The lesions were classified as uni- or multilocular depending on the presence of at least one septum separating the lesion (Figure 1). The perforation and the buccal, lingual, palatal cortical expansions caused by the lesion were also evaluated (Figure 2). The largest diameters of the lesions in axial, coronal, and sagittal planes as well as the relationship between the age of the patient and the dimensions of the lesions were determined (Figure 3). Association of the lesions with impacted teeth and their relations to the root or crown region of the impacted tooth were investigated (Figure 4). In addition, lesions that cause displacement of the mandibular canal, those in the maxilla that invade or displace maxillary sinuses and nasal cavity, as well as their impact on the cortex of the mandibular canal, maxillary sinuses, and nasal cavity were examined (Figure 5).

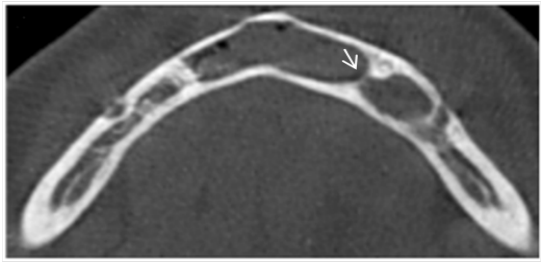

Figure 1. *Axial sequence of multilocular septated* ***(arrow)*** *lesion in the mandible.*

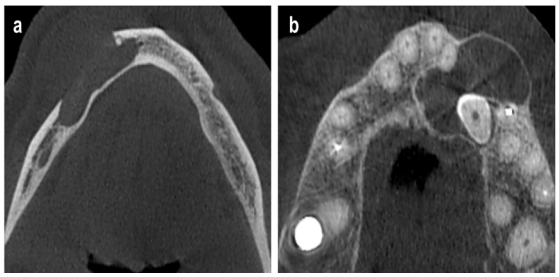

Figure 2. *Axial image of the lesion causing bone perforation in the right mandible* ***(a)****. Axial image of the lesion with an impacted tooth in the left mandible and expansion in the buccal and palatinal bones* ***(b)****.*

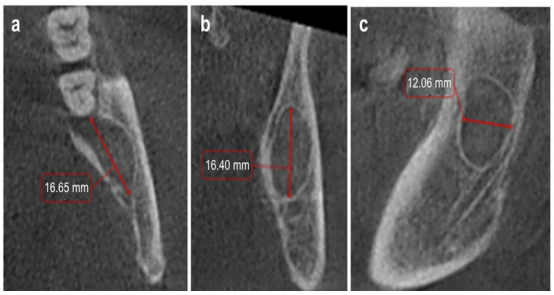

Figure 3. *Measurement of the lesion size in axial, coronal, and sagittal planes.*

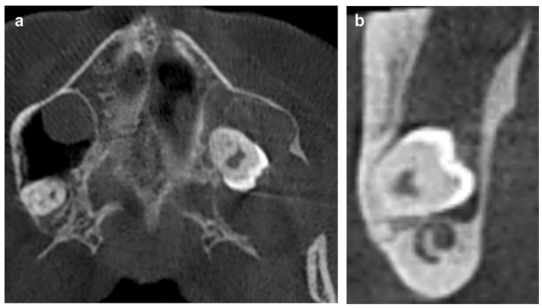

Figure 4. *The extension of the lesion to the apex of the tooth root in the left maxilla* ***(a)****. The lesion confined to the cement–enamel junction in the mandible* ***(b)****.*

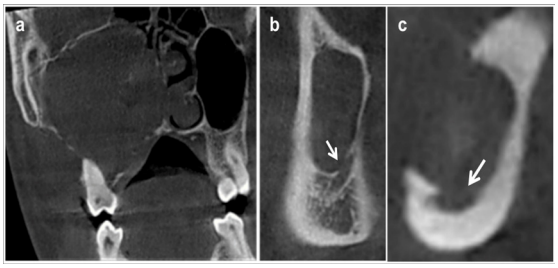

Figure 5. *Invasion of maxillary sinus and nasal cavity by a large lesion in the right maxilla* ***(a)****. Destruction of the cortex of the mandibular canal by the lesion is observed in the sagittal images* ***(****arrows in* ***b*** *and* ***c)****.*

Statistical analysis

The collected data were summarized using descriptive statistics (e.g., mean, standard deviation, range, frequency, percentage). The t-test was used to analyze the differences between continuous independent variables. Data were analyzed using the SPSS software package for Windows (Statistical Package for Social Sciences, version 15; SPSS Inc., Chicago, Illinois, USA). Confidence interval was set to 95% and *p* values less than 0.05 were considered as significant.

Results

There were 17 (61%) male and 11 (39%) were patients in the study sample. The mean age of the patients was 34.5 years (range: 17–79 years). The lesions were mostly located in the posterior region of the jaws. 6 (21%) were localized in the maxilla, and 22 (79%) were found in the mandible. In the mandible, the lesions were most commonly located in the retromolar-ramus region and less commonly in the anterior region (Table 1). Two lesions in mandible and one lesion in the maxilla were observed to cross the mid-line. (Figure 5). Among the lesions found in the mandible, 20 (91%) were adjacent to the mandibular canal, 18 (90%) caused resorption in the cortex of the mandibular canal, and 12 (60%) caused displacement in the mandibular canal. 5 out of 6 lesions localized in the maxilla invaded into the maxillary sinus and nasal cavity. Half the lesions (n=14) were unilocular whereas other half were multilocular (Table 1). The lesions caused cortical expansion in alveolar bones in, at least, one of the buccal and lingual/palatal bones in 26 cases (93%) and perforation was detected in 21 cases (75%) (Table 1).

The mean and standard deviation of the axial, coronal, and sagittal dimensions of the lesions were 29.45 ± 10.7 mm, 31.15 ± 12.12 mm, and 20.7 ± 10.7 mm, respectively (Table 2).

Of all the lesions, 6 (30%) were observed to cause root resorption and 20 lesions were adjacent to the teeth. The lesions were found to be related with impacted teeth in 11 patients (39%). The lesions most commonly co-occurred with impacted third molar teeth (n=8, 73%) followed by the canine teeth (n=2, 18%). All lesions that co-occurred with an impacted tooth caused displacement of the impacted tooth (Figure 4). All lesions that co-occurred within the maxilla were found to have extended to the root

apex; however, lesions localized in the mandible were confined to the cement–enamel junction of the impacted tooth (Figure 4) (Table 3). There was no significant relationship between the age of the patients and dimensions of the lesions or between the age and multilocular occurrence of the lesions. Patients with impacted teeth were significantly younger than others (mean age, 23.2 ± 7.96 years and 41.7 ± 15.3 years, respectively; $p<0.05$).

Table 1. *The frequency distribution of the location, unilocular/multilocular appearence, cortical expansion and bone perforation of keratocystic odontogenic tumors in the study sample.*

		Mandible (n=22)	Maxilla (n=6)	Total (n=28)
Location	Anterior-premolar	2 (9%)	2 (40%)	4
	Anterior-premolar-molar	2 (9%)	4 (60%)	6
	Molar-retromolar	1 (4.5%)	-	1
	Molar-retromolar-ramus	11(50%)	-	11
	Retromolar-ramus	4 (18.5%)	-	4
	Premolar-molar-retromolar-ramus	1 (4.5%)	-	1
	Ramus	1 (4.5%)	-	1
Loculation	Unilocular	9 (41%)	5 (83%)	14 (50%)
	Multilocular	13 (59%)	1 (17%)	14 (50 %)
Cortical	Expansion	20 (91%)	6 (100%)	26 (93%)
	Perforation	16 (73%)	5 (83%)	21 (75%)

Table 2. *Dimensions of keratocystic odontogenic tumors in axial, coronal, and sagittal planes.*

	Mean±Standard deviation	Min–Max
Axial	29.45±10.7 mm	12.17–55.7 mm
Coronal	31.15±12.12 mm	12.4–58 mm
Sagittal	20.7±10.7 mm	10.7–54.32 mm

Table 3. *Evaluation of the lesions co-occurring with an impacted tooth in terms of age, positional change in the relevant tooth, and extension of the lesion to the apex of the tooth root.*

Impacted tooth number	Age (years)	Change in position	Extension of the lesion to the apex of the tooth root
23	45	Yes	Yes
13	17	Yes	Yes
48	28	Yes	No
48	20	Yes	No
48	17	Yes	No
48	17	Yes	No
44	24	Yes	No
48	21	Yes	No
28	23	Yes	Yes
48	23	Yes	No
48	21	Yes	No

Discussion

In the present series, KCOT lesions affect the mandible more frequently than the maxilla as previously reported (10). Mandibular lesions with the impacted teeth, which were confined to the cement–enamel junction, suggest that these lesions tend to display scalloped growth pattern superiorly and anteriorly. Unilocular, multilocular, or multiple well-circumscribed radiolucent lesions surrounded by a thin radiopaque border with a smooth or loculated periphery are the typical radiographic features of KCOTs (11). In previous studies, these lesions were reported most commonly as unilocular and, less commonly, as multilocular lesions (12, 13). However, one lesion that appeared as unilocular in panoramic view was later reported to be multilocular in CBCT images (9). In the present study, we observed equal number of unilocular and multilocular lesions. We considered that previous studies have failed to demonstrate septa of the lesions because of the limitations of the conventional methods in delineating these lesions. Multilocular structure of the lesion, size, and presence of soft tissue invasion are important features in determining surgical technique (14). Multilocular lesions were reported to show recurrence more frequently than unilocular lesions (13). It was also suggested that KCOTs begin as

unilocular lesions and gradually become multilocular (15). Macdonald-Jankowski and Li (16) reported that more multilocular lesions could be found in older patients compared to unilocular lesions. However, in contrast to these studies, the present study found no correlation between the age and unilocular or multilocular appearence of the lesions. Dammer *et al.*(17) suggested that small keratocysts near the alveolar process with a maximum diameter of 1 cm should be treated by simple excision; however, large keratocysts near the base of the skull, which have invaded soft tissue, should be treated by radical excision. All lesions in the present study were above 1 cm in diameter in all 3 orthogonal (sagittal, axial, and coronal) planes. In another study in which the CBCT has been used, it has been reported that the diameter of KCOT lesions varied between 1.3 cm and 7.1 cm (9). Another characteristic feature of KCOTs is that 25%–40% of the lesions co-occur with an impacted tooth (18). This rate was 39% in the present study, and impacted tooth was most commonly observed in young patients in their second decade. Buccolingual expansion was reported in approximately two-third of the patients (19). CBCT images allowed us to delineate the margins of the lesions in all 3 orthogonal planes (axial, sagittal, and coronal). The present study determined that approximately 93% of the lesions caused buccolingual or buccopalatinal expansion. An important characteristic of KCOT is its trend to grow along the major axis of the jaws (12). Occasionally, KCOTs may erode the buccal plate or mandibular lingual cortex; therefore, on radiographs, a radiolucent shadow is frequently seen in the mandible (20). However, bone perforation and erosions caused by the lesions in CBCT images on various planes avoid false positive or false negative interpretations and allow more accurate investigation. The present study determined cortical perforation in at least one of the buccal, lingual, and palatinal bones in 75% of the cases. KCOT lesions are most commonly diagnosed in the third decade of life (19, 21). The mean age in the present study was 34.5 years. Consistent with the current findings, previous studies have reported that KCOTs affect males more frequently than females (21, 22). Lesions co-occurring with unerupted teeth are usually encountered in younger patients (16).

KCOTs are often incidentally detected on routine panoramic radiographs (23). The main symptom is swelling, although more than three-fourth of the lesions are detected incidentally (19). KCOTs can cause displacement and resorption of teeth, and the inferior alveolar nerve canal may also be displaced (12). These lesions were reported to rarely cause tooth root resorption (24). This rate was reported to be 8% – 41% (15, 16). In addition, we found that lesions occurring in dentate regions have caused root resorption in 30% of the cases. The lesions caused displacement of the mandibular canal in 60% of the cases. Sagittal images acquired using CBCT showed disruption in the continuity of the mandibular canal cortex caused by KCOTs. To our knowledge, previous studies did not address the impact of KCOTs on the cortex of mandibular canal. KCOTs can invaginate and occupy the entire maxillary antrum (12). In the present study, lesions localized in the molar region of the maxilla almost completely occupied the maxillary sinus. Panoramic radiography is a useful imaging method widely used in the practice of dentistry offering low cost and low dose of radiation and extensive imaging area. This method, however, has some inherent problems. Panoramic radiography provides only two-dimensional images and fails to demonstrate distortion in the horizontal plane, magnification in the vertical plane, and true relationships. The accuracy of the image largely depends on the operator and varies greatly with patient positioning (25). CBCT images allow visualization of the lesions and anatomic structures in orthogonal planes without distortion and magnification (26).

Conclusion

High rate of cortical expansion, mandibular canal displacement, and bone perforation associated with KCOTs indicate their aggressive nature. In preoperative assessment, CBCT with low doses of radiation and high resolution provides valuable information about KCOTs.

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Conflict of interest
None declared

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