

## MULTIPLE DENTIGEROUS CYSTS WITH RADIOLOGICAL FINDINGS IN A NON-SYNDROMIC PATIENT\*

# NONSENDROMİK BİR HASTADA RADYOLOJİK BULGULARI İLE ÇOKLU DENTİGERÖZ KİST \*

Dr. Öğr. Üyesi Deniz YAMAN\*

Öğr. Gör. Dr. Gülsün AKAY\*\*

Prof. Dr. Kahraman GÜNGÖR\*\*

*Makale Kodu/Article code:* 3676 *Makale Gönderilme tarihi:* 20.05.2018

**Kabul Tarihi:** 26.06.2018 **DOI:** 10.17567/ataunidfd.437247

**Deniz Yaman:** *ORCID ID:* 0000-0002-1492-6897 **Gülsün Akay:** *ORCID ID:* 0000-0002-1767-1383 **Kahraman Güngör:** *ORCID ID:* 0000-0001-6336-4424

#### **ABSTRACT**

Dentigerous cyst is the most common odontogenic cyst of the jaws after radicular cyst and is usually observed as unilateral involvement. The multiple involvement of this cyst is rather rare if not accompanied of an underlying systemic disease or syndrome. In this report, radiographic findings and surgical treatment of a patient with non-syndromic multiple dentigerous cyst, who applied to the clinic for prosthetic treatment were presented. Panoramic radiograph revealed radiolucencies associated with the crowns of bilateral mandibular third molar teeth, right maxillary third molar, bilateral maxillary canines. In cone-beam computed tomography (CBCT), it was revealed as well-defined unilocular radiolucent areas surrounded by sclerotic border and related to the crown of the unerupted teeth. In intermittent sessions, multiple dentigerous cysts were enucleated and the associated impacted teeth were extracted under local anesthesia. According to the comprehensive research, this is the first case-report presenting the surgical treatment and revealing the radiographic findings in elderly non-syndromic patient with multiple dentigerous cyst in maxilla and mandible.

**Keywords:** Cone-beam computed tomography; dentigerous cyst; unerupted tooth

#### ÖZ

Dentigeröz kist, radikuler kistten sonra çenelerin en sık görülen odontojenik kistidir ve genellikle tek taraflı tutulum gösterir. Birden fazla dentigeröz kist olgusu, altta yatan bir sendrom veya sistemik hastalığın yokluğunda oldukça nadir görülen bir durumdur. Bu raporda, kliniğe protetik tedavi amacıyla başvuran nonsendromik çok sayıda dentigeröz kist tespit edilen hastanın radyografik bulguları ve cerrahi tedavisi sunulmuştur. Panoramik radyografide, bilateral mandibular üçüncü molar dişlerin kronları, sağ maksiller üçüncü molar, bilateral maksiller kanın dişler ile ilişkili radyolüsent alanlar tespit edilmiştir. Konik ışınlı bilgisayarlı tomografide (CBCT), iyi tanımlanmış uniloküler radyolüsent alanlar sklerotik sınırla çevrili olup, gömülü dişlerin kronu ile ilişkili olarak izlenmiştir. Aralıklı seanslarda, lokal anestezi altında çok sayıda dentigeröz kist enükle edilerek ilişkili gömülü dişler çekilmiştir. Kapsamlı araştırmaya göre sunulan vaka, maksilla ve mandibulada lokalize çok sayıda dentigeröz kist bulunan ve herhangi bir sendromu olmayan ileri yaştaki hastanın radyolojik bulgularıyla birlikte cerrahi tedavisinin sunulduğu ilk olgu sunumudur.

Anahtar kelimeler: Konik ışınlı bilgisayarlı tomografi; dentigeröz kist; gömülü dişler

**Kaynakça Bilgisi:** Yaman D, Akay G, Güngör K. Nonsendromik Bir Hastada Radyolojik Bulguları ile Çoklu Dentigeröz Kist. Atatürk Üniv Diş Hek Fak Derg 2020; 30: 122-125.

Citation Information: Yaman D, Akay G, Gungor K. Multiple Dentigerous Cysts With Radiological Findings in a Non-Syndromic Patient. J Dent Fac Atatürk Uni 2020: 30: 122-125.

### INTRODUCTION

A dentigerous cyst is an odontogenic cyst which enfolds part or all of the crown of an impacted tooth. 

1,2 It has been reported in the literature that radicular cyst is the most common cyst group followed by dentigerous cyst with a rate of approximately 24%. 

1-3 According to the World Health Organization (WHO), dentigerous cysts have been classified as

developmental odontogenic cyst<sup>4</sup> and they are often located around the impacted mandibular or maxillary third molar and maxillary canine teeth.<sup>5</sup> The majority of cases are usually seen in young adults, but rarely have been reported in deciduous dentition and mixed dentition.<sup>6</sup>

Dentigerous cysts have the potential to develop and grow, but can be remain asymptomatic until



<sup>\*</sup>Bolu Abant İzzet Baysal University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Bolu, TURKEY,

<sup>\*\*</sup>Gazi University, Faculty of Dentistry, Department of Oral and Dentomaxillofacial Radiology, Ankara, TURKEY,

<sup>\*</sup> This case report was presented as a poster presentation at the 7th Scientific Congress of Oral and Maxillofacial Radiology Association, Eskisehir-Turkey 2017.

asymmery and swelling occur.<sup>1,2</sup> Therefore, radiographic imaging is important in the diagnosis and treatment planning of dentigerous cysts. In routine dental examinations, dentigerous cysts may be observed on radiographs taken due to investigate the causes of delayed tooth eruption and the presence of tooth deficiency.<sup>2,7</sup> Dentigerous cyts are seen as a unilocular radiolucent area surrounded by a sclerotic border, which is associated with the non-erupted tooth in the radiographs<sup>2</sup> whereas if infected, these cysts may have poor defined borders.<sup>5</sup>

Mostly these cysts are unilateral. It is known that bilateral and multiple dentigerous cysts are seen in Maroteaux Lamy syndrome, cleidocranial dysplasia and mucopolysaccharidosis but it is considered as a rare case in the absence of developmental syndromes or systemic diseases. <sup>2,7,8</sup>

The present report highlights the presence of multiple dentigerous cysts in the maxilla and mandible in the absence of syndrome.

#### **CASE REPORT**

A 60-year-old male patient admitted to the Clinic of Oral and Dentomaxillofacial Radiology with a chief complaint of prosthesis renewal. He was systemically healthy and extra-oral examination was within normal limits. There was no relevant past dental, medical or family history and there was no associated syndromes or systemic diseases present. Clinical examination revealed none of these abnormal findings such as expansion of the cortical bone or displacement in adjacent teeth, root resorption, dull aching pain or drainage, gingival hyperplasia. Panoramic radiograph taken for dental examination, revealed regular-shaped radiolucencies associated with the crowns of bilateral mandibular third molar teeth, right maxillary third molar, bilateral maxillary canine (Figure 1). Mandibular third molars of both side were observed inverted position.

The patient was referred for the cone-beam computed tomography (CBCT) examination to delineate the localization and structure of the lesions. The CBCT images were obtained using a Promax 3D unit (Planmeca, Helsinki, Finland), operating at 90 kVp, 9-14 mA, with a 0.16 mm voxel size, exposure time of 6 seconds and field of view of 8 cm. In CBCT, two well-defined unilocular radiolucent areas surrounded by sclerotic border related to unerupted teeth were seen in mandible. The one on the mandible right side showed diameter of approximately 21.41

mm and the left one 15.93 mm. Radiolucent lesions in the right and left mandibular posterior region were observed as adjacent to the mandibular canal (Figure 2). CBCT images revealed radiolucent areas in relation to maxillary permanent canines and right maxillary third molar. The right posterior maxilla was observed a well-defined, unilocular radiolucent lesion around the 18-number tooth crown. The size of the lesion was approximately 15x11x14 mm (supero-inferior x bucco-palatal x mesio-distal) (Figure 3). The anterior maxil lary lesion was observed as relation to the crowns of the unerupted permanent canines (Figure 4).

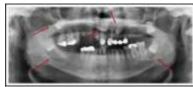


Figure 1. Panoramic radiograph showing bilateral radiolucency in mandible and right maxillary third molar, bilateral maxillary canine regions.





Figure 2. (A) Coronal section, (B) Axial section in CBCT images, demonstrating two well-defined unilocular radiolucent areas surrounded by sclerotic border related to mandibular unerupted molar teeth.

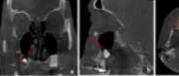


Figure 3. (A) Coronal section, (B) Sagittal section, (C) Axial section in CBCT images, showing unilocular radiolucent lesion the maxillary third molar tooth.

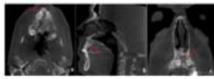


Figure 4. (A) Axial section, (B) Sagittal section, (C) Coronal section in CBCT Images, showing cystic lesion in the anterior maxilla.

The clinical and radiographic data suggest preliminary diagnosis of dentigerous cyst. The patient was referred to the Department of Oral Surgery. Under local anesthesia a full thickness mucoperiosteal flap was reflected in the areas of associated cysts. Each

tooth extraction and cyst enucleation were performed in separate sessions. Mucoperiosteal flaps were closed with 4-0 resorbable sutures. Antibiotic treatment with amoxicillin/clavulanic acid as an oral medication was carried out postoperatively to prevent infection. Also, nonsteroidal anti-inflammatory drug (NSAID) was started for pain management and Chlorhexidine 0.12% used as an oral rinse. The specimens were sent for histopathological evaluation and all specimens were diagnosed as dentigerous cyst. After 7 days of follow-up at each operative site, there was good healing in the surgical field without any inflammation or paresthesia. After six months in panoramic radiography image, there had been complete healing and new bone formation was observed (Figure 5). No evidence of recurrence or complications was found.

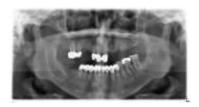


Figure 5. Panoramic radiograph after six months.

## **DISCUSSION**

Dentigerous cysts are benign odontogenic cyst and these cysts are usually associated with the crowns of impacted permanent teeth, supernumerary teeth and rarely deciduous teeth. <sup>7,9</sup> Dentigerous cytsts can be seen in all age groups, however they occur relatively rarely in the first decade of life<sup>7</sup>. In the present case patient was 60-year-old, his medical examination did not show any abnormal physical or laboratory findings suggesting any syndrome. However, dentigerous cysts associated with the impacted third molars and maxillary canines were detected.

Dentigerous cyst has a very slow growing character and is usually asymptomatic in small size unless there is any enlargement or infection. However, enlargement of the cyst and destruction of cortical bones may occur. Besides, displacement of associated teeth with delayed eruption and even pathological jaw fracture may occur. <sup>10</sup> In the present case, resorption of roots of the maxillary anterior teeth and molar teeth in both jaws was not present. Radiolucent lesions are asymptomatic and diagnosed incidentally on taken routine panoramic radiograph before prosthetic treatment.

Panoramic radiograph is primary technic for maxillofacial region evaluation. However, it provides a two-dimensional image of the lesion and does not three-dimensional relationship neighboring anatomical structures. CBCT is widely used to provide basic information about the maxillofacial region in diagnosis and surgical planning in dentistry.<sup>11</sup> Typical panoramic radiographic image of dentigerous cyst can be seen as a unilocular radioluceny with the crown of an impacted tooth surrounded by sclerotic margins. 7 CBCT can be useful in assessing expansion, cortical borders, lesion's internal structure and relationship to adjacent structures.<sup>5</sup> In present case, CBCT images were used for evaluating the lesions relation with mandibular canal, nasal fossa and adjacent teeth before surgical treatment.

Dentigerous cysts are usually solitary and the reported non-syndromic bilateral and multiple dentigerous cyst cases are quite rare. 2,7-10,12-15 According to the results of comprehensive investigations of the researchers, only 30 cases were found related to bilateral dentigerous cysts with non-syndromic patients. 12,13,15 When the studies are examined in detail, bilateral or multiple cysts can be observed in Basal cell nevus syndrome, Mucopolysaccharidosis, cleidocranialdysplasia.<sup>2,9</sup> Bilateral cysts have also been reported after a long-term of use the cyclosporine A and calcium channel blockers. <sup>16</sup> Multiple dentigerous cysts have also been reported with Klippel Feil syndrome.<sup>7</sup> In the present case, we did not find any pathology, craniofacial malformation or a defect related to the syndromes mentioned above, as a result of the clinical and radiographic examinations. In addition, there isn't present the use of systemic medication in the detailed history of the patient.

The differential diagnosis of dentigerous cyst should include hyperplastic follicle, odontogenic keratocyst, cystic ameloblastoma, and other odontogenic tumors. <sup>5,17</sup> While the size of the normal follicular space is 2 to 3 mm, a dentigerous cyst is more likely when the space is more than 5 mm. <sup>5</sup> Histological examination is essential for definitive diagnosis. <sup>8</sup>

Dentigerous cysts are benign. According to previously reported cases, there are two main surgical procedures: marsupialization and enucleation. <sup>10</sup> Treatment planning in dentigerous cyst should be made by considering factors such as size of the cyst, its relationship with anatomical structures, presence of persistent tooth, and age of the patient. All of these factors, the size of the lesion is primary determining factor in the treatment of dentigerous cyst. Surgical

enucleation is usually recommended, which may include the removal of the related tooth as well.<sup>5</sup> Removal of cyst-associated tooth and enucleation of soft tissue component is definitive treatment in many cases.<sup>17</sup> Larger lesions may be treated with marsupialization to relieve the pressure that originates from cystic fluid and to prevent damage to the involved teeth.<sup>7</sup> If untreated, cyst epithelium can rarely turn into ameloblastoma<sup>18</sup>, squamous cell carcinoma or mucoepidermoid carcinoma.<sup>5,17</sup> In our case, lesions were enucleated and cyst-associated teeth were extracted. Histopathological examination showed no metaplastic or dysplastic changes in all cystic lining.

## **CONCLUSION**

Multiple dentigerous cyst not accompanied with syndrome is uncommon. In case of multiple dentigerous cyst, a through clinical and systematic examination should be done to exclude any associated syndrome. Dentigerous cysts are asymptomatic and they may cause bone defects. Therefore, it is important to perform radiographic examination including panoramic radiography with cone-beam computed tomography to determine the most appropriate surgical treatment approach by defining the cyst dimension and its relation to the anatomic landmarks.

## **REFERENCES**

- Soames JV, Southam JC. Oral Pathology. 2<sup>nd</sup> ed. Oxford; Oxford University Press: 1993. p. 74-6.
- 2. Ko KS, Dover DG, Jordan RC. Bilateral dentigerous cysts: report of an unusual case and review of the literature. J Can Dent Assoc 1999; 65:49–5.
- 3. Tamgadge A, Tamgadge, S, Bhatt D, Bhalera, S, Pereira T, Padhye M. Bilateral dentigerous cyst in a non-syndromic patient: report of an unusual case with review of the literature. J Oral and Maxillofac Pathol 2011; 15:91-5.
- Nunez-Urrutia S, Figueiredo R, Gay-Escoda C. Retrospective clinicopathological study of 418 odontogenic cysts. Med Oral Patol Oral Cir Bucal 2010; 5:e767-73.
- 5. White SC, Pharoah MJ. Oral radiology. Principles and interpretation. 6<sup>th</sup>ed. St Louis; CV Mosby:2009. p. 346-50.
- Demirtaş N, Mihmanli A, Bayer S, Özalp Ş. Dentigerous cysts in the mixed dentition: report of three cases. J Dent Fac Atatürk Uni 2016; 14:23-6.
- 7. Devi P, Thimmarasa VB, Mehrotra V, Agarwal M. Multiple dentigerous cysts: a case report and review. J Maxillofac Oral Surg 2015; 14:47-51.

- 8. Freitas DQ, Tempest LM, Sicoli E, Lopes Neto FC. Bilateral dentigerous cyst: Review of literature and report of unusual case. Dentomaxillofac Radiol 2006; 35:464-8.
- Ustuner, E, Fitoz S, Atasoy C, Erden I, Akyar, S. Bilateral maxillary dentigerous cysts: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003; 95:632-5.
- 10. Imada TSN, Neto VT, Bernini GF et. al. Unusual bilateral dentigerous cysts in a nonsyndromic patient assessed by cone beam computed tomography. Contemp Clin Dent 2014; 5:240-2.
- 11. Deana NF, Alves N. Cone beam ct in diagnosis and surgical planning of dentigerous cyst. Case Rep Dent. 2017; 2017:7956041.
- 12. Jeon JY, Park CJ, Cho SH, Hwang KG. Bilateral dentigerous cysts that involve all four dental quadrants: a case report and literature review. J Korean Assoc Oral Maxillofac Surg 2016; 42:123-6.
- Dhupar A, Yadav S, Dhupar V, Mittal HC, Malik S, Rana P. Bi-maxillary dentigerous cyst in a nonsyndromic child - review of literature with a case presentation. J Stomatol Oral Maxillofac Surg 2017; 118:45-8.
- 14. Shahrabi Farahani S, Lotfalian M. A pigmented dentigerous cyst in a patient with multiple dentigerous cysts of the jaws: a case report. J Contemp Dent Pract 2007; 8:85-91.
- Khandeparker RV, Khandeparker PV, Virginkar A, Savant K. Bilateral Maxillary Dentigerous Cysts in a Nonsyndromic Child: A Rare Presentation and Review of the Literature. Case Rep Dent 2018; 15:2018:7583082.
- De Biase A, Ottolenghi L, Polimeni A, Benvenuto A, Lubrano R, Magliocca FM. Bilateral mandibular cysts associated with cyclosporine use: A case report. Pediatr Nephrol 2001; 16:993-5.
- 17. Regezi J, Sciubba J, Jordan R. Oral pathology: Clinical Pathologic Correlations. 5<sup>th</sup> ed. Philadelphia; Saunders Company:2008. p. 246-8.
- 18. Kondamari SK, Taneeru S, Guttikonda VR, Masabattula GK. Ameloblastoma arising in the wall of dentigerous cyst: Report of a rare entity. J Oral Maxillofac Pathol 2018; 22:7-10.

## Yazışma Adresi

Dr. Öğr. Üyesi Deniz YAMAN Bolu Abant İzzet Baysal University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Bolu, TURKEY Tel: (+90) (0374) 253 45 01

e-mail: denizyaman89@hotmail.com