

Nonsyndromic Bilateral Mandibular Dentigerous Cysts: Report of a Rare Case

*NONSENDROMİK BİLATERAL MANDİBULER DENTİGERÖZ KİST:
NADİR BİR VAKA RAPORU*

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Abstract

This case report was intended to present a case prediagnosed as nonsyndromic bilateral mandibular dentigerous cysts following clinical and radiographic examination and its treatment.

A 51 year-old male patient was referred by his private practitioner to Oral & Maxillofacial Surgery Department of Dental Faculty of Gazi University for the evaluation of the bilateral mandibular radiolucencies adjacent to the impacted third molar teeth which were observed during the routine orthopantomographic examination. Patient's chief complaint was the pain and swelling in the maxillary right first premolar and left canine. The medical history of the patient was unremarkable except hypercholesterolemia and hepatitis A, approximately thirty years ago. In clinical examination there was no swelling or fluctuation in the impacted left and right third molar region and the overlying mucosa presented normal features.

Orthopantomographic examination revealed the presence of bilateral mandibular cystic radiolucencies. There was a prominent displacement of the inferior alveolar nerve at both sides. Apart from these findings, periapical inflammatory diseases were identified at the apices of the maxillary right first premolar and left canine teeth.

The bilateral mandibular lesions were prediagnosed as dentigerous cysts. The cyst in the right mandible was treated by the marsupialization + enucleation technique and the cyst in the left mandible was treated by enucleation technique.

In such cases with large cystic cavities, marsupialization + enucleation technique may be the choice of treatment as it helps to preserve the supporting structures, and avoid further complications. In such cases with small cystic cavities, enucleation technique may be the choice of treatment.

Key Words: Odontogenic cysts; dentigerous cyst; third molar

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Özet

Bu makale, klinik ve radyolojik değerlendirmeler sonucunda mandibuler dişlerle ilişkili bilateral nonsendromik dentigeröz kist teşhisi konulan vaka ve tedavisini sunmayı amaçlamaktadır.

51 yaşındaki erkek hasta serbest dişhekimi tarafından rutin radyografik değerlendirme sırasında farkedilen, mandibuler gömülü üçüncü molar dişlerle ilişkili bilateral radyolüsent bölgelerin değerlendirilmesi için kliniğimize gönderilmiştir. Hastanın asıl şikayeti sağ maksiller 1. premolar ve sol kanin dişlerindeki ağrı ve şişlikti. Hastanın anamnezinde yaklaşık 30 yıl önce geçirilmiş hepatit A ve kolesterol yüksekliği hikayesi olduğu öğrenilmiştir. Klinik muayenede mandibulada her iki taraftaki gömülü üçüncü molar dişler bölgesinde herhangi bir şişlik veya fluktuasyon olmadığı ve bölgedeki mukozanın sağlıklı olduğu görülmüştür.

Panoramik radyografi incelemesinde her iki tarafta mandibuler üçüncü molar dişler çevresinde kistik radyolüseni gösteren lezyonlar ve inferior alveolar sinirde yer değişimi olduğu; sağ maksiller 1. premolar ve sol kanin dişlerin apikalinde periapikal inflamatuvar patoloji olduğu görülmüştür.

Radyografik değerlendirmeler sonucu bilateral dentigeröz kist ön tanısı konulan hastanın sağ taraftaki kistik lezyonu marsüpyalizasyon + entükleasyon ile sol taraftaki kistik lezyonu ise entükleasyon ile tedavi edilmiştir.

Büyük hacimli kistlerin tedavisinde herhangi bir komplikasyondan kaçınmak için marsüpyalizasyon + entükleasyonun, daha küçük hacimli kistlerin tedavisinde ise entükleasyonun uygun bir cerrahi teknik olduğu sonucuna varılmıştır.

Anahtar Kelimeler: Odontojenik kistler; dentigeröz kist; üçüncü molar

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A dentigerous cyst is defined as an epithelial-lined developmental cavity arising from the enamel organ due to an alteration in the reduced enamel epithelium and enclosing the crown of an unerupted tooth at the cemento-enamel junction.¹⁻⁵ After radicular cysts, dentigerous cysts are the second most common odontogenic cysts. They account for approximately

24% of all true cysts in the jaws.^{3,4,6-8} Cysts involve impacted, unerupted permanent teeth, supernumerary teeth, odontomas, and rarely may they involve deciduous teeth. They are usually seen in the second and third decades. When compared to females, the incidence is slightly higher in males. Mandible is likely to be the primarily affected site, as cysts are located in the mandible in 75% of the cases. The most frequently involved teeth are the mandibular third molars and maxillary canines.^{2-4,6,8}

Although most dentigerous cysts are solitary, bilateral and multiple cysts may also develop. However, bilateral cysts are usually associated with a number of syndromes, including cleidocranial dysplasia and Maroteaux-Lawy syndrome.^{3,4,6,9-11} Bilateral dentigerous cysts are also suggested to be induced by the prescribed drugs. The combined effect of cyclosporine and a calcium channel blocker is reported.³ In most cases cysts are asymptomatic but they may become painful with infection, likewise seen in a case with *Serratia* infection.¹

In the absence of any syndrome, bilateral dentigerous cysts extremely rare and as far as we are concerned only 18 cases are reported until today (Table 1).^{1,3-6,8,9,12-22} In this case report, presence of nonsyndromic bilateral dentigerous cysts associ-

ated with mandibular third molar teeth is presented.

Case Report

A 51 year-old male patient was referred by his private practitioner to Oral & Maxillofacial Surgery Department of Dental Faculty of Gazi University for the evaluation of the bilateral mandibular radiolucencies adjacent to the impacted third molar teeth which were observed during the routine orthopantomographic examination. Patient's chief complaint was the pain and swelling in the maxillary right first premolar and left canine. The premolar tooth was restored with a crown, while composite restoration was present in the canine tooth. The medical history of the patient was unremarkable except hypercholesterolemia and hepatitis A, approximately thirty years ago. In clinical examination there was no swelling or fluctuation in the impacted left and right third molar region and the overlying mucosa presented normal features. The patient also had no complaints regarding these sites. It was obtained informed consent form from the patient.

Orthopantomographic examination revealed the presence of bilateral mandibular cystic radiolu-

Table 1. Review of the literature.

Authors/Year	Sex	Age (years)	Location	Treatment
Freitas, Tempest, Sicoli, Lopes-Neto, 2006 ¹²	M	14	Md. second molar and mx. third molar	Enucleation
Batra, Roychoudhury, Balakrishan, Parkash, 2004 ⁶	F	15	Md. third molars and second premolar	Enucleation
Ustuner, Fitoz, Atasoy, Erden, Akyar, 2003 ⁸	M	6	Mx. canines	Enucleation
Shah, Thuau, Beale, 2002 ⁹	M	39	Md. third molars	No treatment
De Biase, Ottolenghi, Polimeni, Benvenuto, Lubrano, Magliocca, 2001 ³	M	8	Md. first molars	Enucleation
Ko, Dover, Jordan, 1999 ⁴	M	42	Md. third molars	Enucleation
Sands, Tocchio, 1998 ¹³	F	3	Md. central incisors and first molars	Enucleation
Banderas, Gonzalez, Ramirez, Arroyo, 1996 ¹⁴	M	38	Md. third molars	Enucleation
O'Neil, Mosby, Lowe, 1989 ⁵	M	5	Md. first molars	Enucleation
Eidinger, 1989 ¹⁵	M	15	Md. first molars	Enucleation
McDonnell, 1988 ¹⁶	M	15	Md. second premolar and second molar	Enucleation
Crinzi, 1982 ¹⁷	F	15	Md. third molars	Enucleation
Swerdloff, Alexander, Ceen, Ferguson, 1980 ¹⁸	F	7	Md. first molars	Enucleation
Burton, Sheffer, 1980 ¹	F	57	Md. third molars	Enucleation
Callaghan, 1973 ¹⁹	M	38	Md. third molars	Enucleation
Stanback, 1970 ²⁰	M	9	Md. first molars	Enucleation
Henefer, 1964 ²¹	F	52	Mx. third molars	Enucleation
Myers, 1943 ²²	F	19	Md. third molars	Enucleation

M, male; F, female; Md., mandibular; Mx., maxillary

cencies. At the right side, the radiolucency surrounded the crown of the vertically impacted third molar tooth and expanded from the second molar region to the inferior border of the coronoid process. Likewise the radiolucency on the right side, an apparently smaller cystic radiolucency was observed around the horizontally impacted third molar tooth on the left side. There was a prominent displacement of the inferior alveolar nerve at both sides (Figure 1). Apart from these findings, periapical inflammatory diseases were identified at the apices of the maxillary right first premolar and left canine teeth, which, in fact, were the focus of the chief complaint of the patient. Occlusal radiograph of the maxilla did not indicate evidence of any clefting (Figure 2a).

Due to the presence of multiple dentigerous cysts, it was decided to further evaluate the patient for any possible syndromic association. For this purpose, a skeletal survey was performed to rule out the cysts in any other bone. This survey revealed no additional cysts. In a similar manner, the chest radiograph presented no specific findings (Figure 2b). Peripheral blood lymphocytes were used for the chromosomal analysis. Cytogenetic studies did not reveal any structural or numerical abnormalities, including chromosome 1qh+. Based on these analyses, no syndromic association could be made.

The bilateral mandibular lesions were prediagnosed as dentigerous cysts. Due to the large cystic cavity, expansive nature, apparent displacement of the impacted tooth towards the mandibular basis and the excessive weakness of the mandibu-



Figure 1. Pre-operative radiographic appearance of the bilateral mandibular radiolucencies involving the impacted the third molar teeth at both sides.

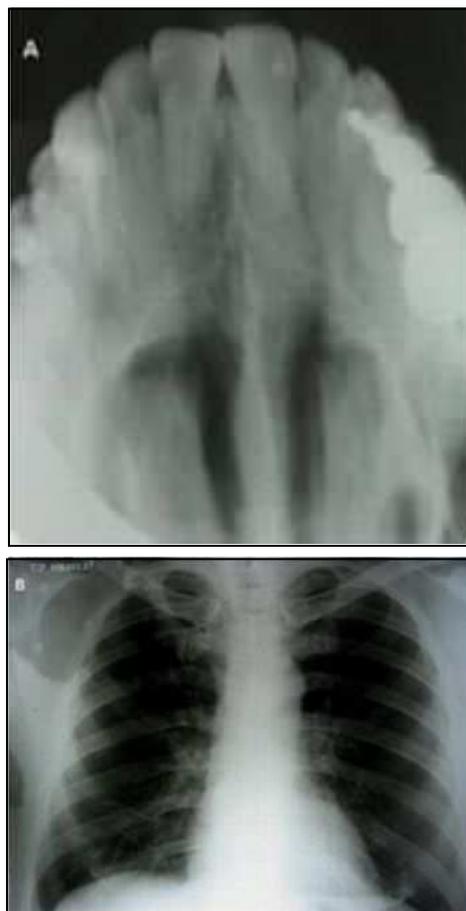


Figure 2. a: The occlusal radiograph of maxilla with no evidence of clefting. **b:** Chest radiograph showing no specific findings.

lar portion, the cyst in the right mandible was decided to be treated by the marsupialization + enucleation technique. Healing was uneventful. Two months later, the small cyst in the left mandible was enucleated and the related impacted tooth was removed (Figure 3). As the patient experienced numbness of the left lip, B complex vitamin was prescribed for 3 months.

Following the surgical treatment of the bilateral mandibular dentigerous cysts, the 2 maxillary teeth with apical lesions which lead to pain and swelling and were the focus of the patient's chief complaint were treated by apicectomy. The histopathological analysis of the operation material did not indicate any cystic features and the diagnosis of apical granuloma were achieved.

Five months after the first intervention on the right side of the mandible, it was seen that the di-



Figure 3. Panoramic radiograph of the patient two months following marsupialization on the right side and immediately after enucleation of the cyst on the left side.

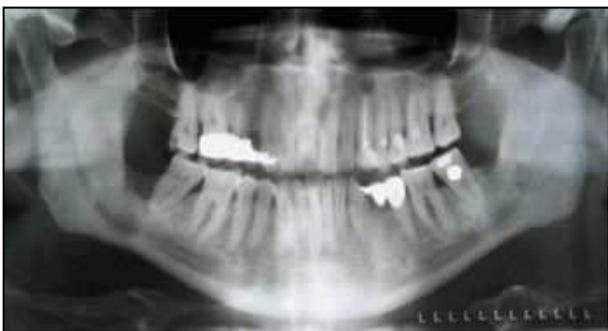


Figure 4. Post-operative radiographic appearance of the patient after one year.

mensions of the cystic cavity was reduced both clinically and radiographically. However, complete resolution of the lesion could not be achieved. Thus, enucleation was performed and the remaining cystic lesion and the tooth were removed. The patient is still under routine follow-up and will be closely monitored for long term outcome (Figure 4).

Histopathologically, both cysts were lined by non-keratinizing squamous epithelium which is 4-5 cell layers of thickness. The connective tissue walls were cellular and composed of fibrous collagen, while only some areas exhibited mononuclear inflammatory infiltration (Figure 5). Furthermore, extravasated blood, and the deposit of hemosiderin were frequently seen. The diagnosis of bilateral dentigerous cysts was achieved.

Discussion

Dentigerous cysts are very common developmental cysts and they are generally solitary.^{4,12} Bilateral dentigerous cysts usually occur in asso-

ciation with a developmental syndrome or systemic disease.^{6,12} Maroteaux-Lamy syndrome (mucopolysaccharidosis, type VI)¹⁰ and cleidocranial dysplasia,¹¹ the 2 developmental conditions that are observed in young individuals with stigmata of the syndromes,^{4,6} are the two syndromes that are most frequently associated with bilateral or multiple dentigerous cysts. Bilateral dentigerous cysts have also been reported with cyclosporine and amlodipine in a patient with renal transplantation.³

As noted in Table 1,^{1,3-6,8,9,12-22} nonsyndromic bilateral dentigerous cysts cases are extremely rare and there have been only 18 cases of multiple nonsyndromic cysts reported in the literature from 1943 to 2005. Twelve of 18 cases have been associated with mandibular molar teeth, with seven^{1,4,9,14,17,19,22} of these associated with third molar teeth and five^{3,5,15,18,20} associated with first molar teeth. The present case falls in this rare category as it was not related with any syndrome. Although this fact may indicate a true rarity for this condition, there is also the possibility of bilateral dentigerous cysts to be either undiagnosed or underreported.⁶

The age range for reported cases varies widely, from 3 years to 57 years of age. The mean age of the 18 cases was 22.05 years. Twelve^{3,5,6,8,12,13,15-18,20,22} of them occurred in pa-

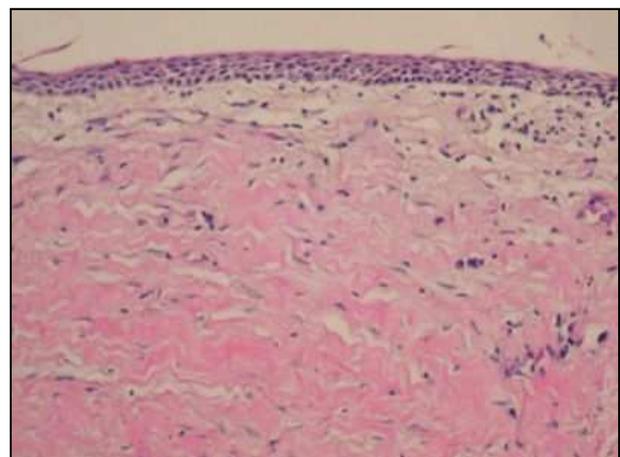


Figure 5. The cyst walls are lined by non-keratinizing squamous epithelium. The connective tissue walls were cellular and composed of fibrous collagen, while only some areas showed mononuclear inflammatory infiltration (H&E x 200).

tients under the age of 20 years and six^{1,4,9,14,19,21} of them occurred in patients upper the age of 20 years. They usually present at under the age of 20 because of the tooth eruption chronology.¹² In the present case, the patient was 51 year old.

Usually pain or discomfort is not observed with the cysts unless they become secondarily infected. Thus, dentigerous cysts are frequently identified when radiographs are taken with the purpose to analyze a failure of tooth eruption, a missing tooth or mal-alignment. Radiographic examination reveals a unilocular, radiolucent lesion that is associated with the crown of an unerupted tooth and that is characterized by well-defined sclerotic margins.^{4,6,17} A dentigerous cyst can be suspected when the follicular space is more than 5 mm, since the normal space is 3 to 4 mm.^{4,6} The present case is well in harmony with the literature in the aspect of radiological features of the lesions and also in the aspect of the asymptomatic clinical course.

The size of the lesion is the primary determinant of the choice of treatment for a dentigerous cyst. Entire removal of the smaller lesions is necessary, while larger cysts are best treated by inserting a surgical drain or by marsupialization. This enables the relief of the pressure that originates from the cystic fluid and the subsequent shrinkage of the cystic space and bone fill.^{3,5,9} As dentigerous cysts or the remnants may differentiate into ameloblastomas, squamous cell carcinomas, adenomatoid odontogenic tumors and complex odontomas, complete removal of the lesions and close monitoring of the patients are crucial.^{4,6,9} In accordance with these recommendations, in the presented case the bilateral mandibular dentigerous cysts were treated with two different approaches. Based on the possible complications and the consequences of enucleation, marsupialization + enucleation technique was preferred for the treatment of the large cyst on the right side, while the small cyst on the left side was treated by enucleation.

The present case was not related with any syndrome, systemic disease or drug usage. We consider the lack of an association with any syndrome, the asymptomatic course of development

and, if untreated, the possibility of complicated prognosis with potentially severe consequences makes this case of incidentally identified bilateral dentigerous cysts important. In such cases with large cystic cavities, marsupialization + enucleation technique may be the choice of treatment as it helps to preserve the supporting structures, avoid further complications and has superiority over enucleation procedure. We also support the suggestion that in cases with unerupted tooth, panoramic radiographs are essential for correct diagnosis.

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