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Case Report

Cone Beam Computed Tomography Finding of Unusual Bilateral Dentigerous Cysts in a Nonsyndromic Patient: A Case Report

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Abstract

Introduction: A dentigerous cyst is the most commonly developed cyst of the jaw. It is most frequently associated with impacted canines and mandibular third molars. Bilateral dentigerous cysts are rare and are generally associated with developmental syndromes or systemic diseases.

Case Presentation: We report on an eight-year-old boy who presented with a complaint of bilateral malposed first mandibular molars. There was no complaint of pain and no swelling. An intraoral examination revealed buccal tipping of both the right and left first mandibular molars with slight swelling. The overlying mucosa was completely normal and there was no tenderness on palpation. This unusual case occurrence associated with the crowns of the bilateral impacted lower second molars. There are no cases reported with bilateral dentigerous cyst involving mandibular second molars. The clinical and radiographic findings, especially the cone beam computed tomography (CBCT) views, were suggestive of dentigerous cysts.

Conclusions: After the surgical removal of the cysts, the specimens were sent for histopathological evaluation. A microscopic examination suggested infected dentigerous cysts.

Keywords: Cone Beam Computed Tomography, Bilateral Dentigerous Cyst, Nonsyndromic

1. Introduction

After radicular cysts, a dentigerous cyst (DC) is the most common odontogenic cyst (1, 2), originating from a reduced enamel epithelial after crown formation (1). DCs account for approximately 24% of all true cysts in the jaw (3). This developmental cyst surrounds the crown and is attached to the neck of unerupted teeth at the cementoenamel junction (CEJ).

DCs are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth, or misalignment (4); they generally appear during tooth development in young patients. It is a common cause of radiolucency associated with the crown of an impacted tooth (5). DCs are most frequently associated with the mandibular third molars and the maxillary canines. There is no pain or discomfort unless the cyst becomes infected. The major symptom of a DC is a failure of tooth eruption (2). They usually present in the second or third decades of life, and are rarely seen during childhood (2).

DCs have a well-defined corticated margin that is associated with the crown of a tooth. Root resorption or displacement of teeth is common. Some complications of DCs include paresthesia of the inferior alveolar nerve, metaplastic and dysplastic change, displacement or obliteration of the maxillary antrum and nasal cavities by maxillary cysts, or even orbital encroachment. Some DCs may cause fractures and may result from a secondary infection. DCs are usually unilateral; the occurrence of multiple DCs is rare and is usually associated with such syndromes as mucopolysaccharidosis and cleidocranial dysplasia (1, 3, 6).

There have been 23 reported cases of bilateral DCs. In this study, we report on the unusual occurrence of nonsyndromic bilateral DCs associated with the mandibular second molar.

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2. Case Presentation

An eight-year-old boy presented to the pediatric department of the Hamadan University of Medical Sciences dental faculty with the complaint of bilateral malposed first mandibular molars. There was no complaint of pain or swelling.

An intraoral examination revealed buccal tipping of both the right and left first mandibular molars with slight swelling. The overlying mucosa was completely normal and there was no tenderness on palpation. No deviation or limitation in the mouth opening was visible.

The familial and medical histories were noncontributory and there was no abnormal finding suggesting the child had any syndrome.

For better evaluation, a panoramic radiography was obtained, which showed the right and left sides of the mandible, and the pericoronal radiolucencies with well-defined cortical borders extending from the distal aspect of the second mandibular molar to the mesial aspect of the first mandibular molar. Vertically, it extended from the alveolar crest to midbody of the mandible. There was no root resorption and no displacement of the inferior alveolar nerve canal; however, the distal displacement of the second molar bud could be seen on the right side (Figure 1).

Figure 1. A Panoramic Image Shows the Bilateral Pericoronal Radiolucencies as a Well-Defined Lesion in the Right and Left Second Mandibular Molars



The lesion on the right is bigger than the lesion on the left and extends to the mesial aspect of the first molar.

For a more accurate evaluation and better presurgical planning, cone beam computed tomography (CBCT) was performed. The CBCT showed bilateral homogenous and hypodense lesions in the distal aspect of the first molar in the mandibular axial sections. The lingual and buccal cortical plate was intact without perforation. There was no periosteal bone production (Figure 2). In the cross-sectional reconstructed views, bilateral radiolucencies caused the roots of the first molars to displace lingually, which caused a buccal tilt of their crowns. A slight swelling of the buccal

and lingual cortical plates with no perforation was seen in both sides. The lesions had no effect on the inferior alveolar canal (Figure 3).

The clinical and radiographic findings were suggestive of DCs. However, final diagnosis could only be confirmed histologically after surgical removal of the cysts. After the patient was referred to a surgeon and placed under general anesthesia, the cysts were enucleated, the unerupted permanent teeth were removed, and the specimen was sent for histopathological evaluation. Microscopic examination revealed that odontogenic epithelium surrounded the cyst. Connective tissue stroma was vascular and showed interlacing collagen fibers and fibroblasts. Severe inflammatory cell infiltration and cholesterol clefts were seen in the stroma, which suggested infected DCs.

3. Discussion

The most common type of cyst found in pericoronal locations is the DC; this cyst constitutes almost 24% of all true cysts in the jaw. However, the bilateral type of DC is extremely rare, and is usually associated with such developmental syndromes as cleidocranial dysplasia, basal cell nevus syndrome, and mucopolysaccharidosis (type VI); it may also be associated with drugs, such as cyclosporine and calcium canal blockers (1, 3, 6). Gingival hyperplasia and impaired dentition are the most common complications shared by most of these syndromes (3).

According to research by Imada et al., only 23 cases of bilateral DCs that were not related to any syndrome have been reported (3). Bilateral DCs usually present in the second or third decade of life. The age range varies from 15 - 57 years, although, similar to this case, these cysts have been seen in people at lower ages. However, extensive maxillary involvement and childhood presentation are rare (3).

DCs are usually asymptomatic without any pain or discomfort to the patient unless they become infected. The most common clinical feature is a missing tooth or teeth, although the cyst may cause a hard swelling that can result in facial asymmetry. DCs that are more than 5 mm can be discovered in a routine panoramic radiograph, but more detail can be seen with a CBCT. Some conditions, such as hyperplastic follicles, cystic ameloblastoma, ameloblastic fibroma, odontogenic keratocyst (OKC), and calcifying odontogenic cysts, may have the same radiographic features as DCs. For example, it may not be possible to differentiate a small ameloblastic fibroma or a cystic ameloblastoma from a DC if there is no internal structure. Normally, the size of the follicular space is 2 - 3 mm; if the space exceeds 5 mm, a DC is likely.

For this case, the size of the lesions ruled out a hyperplastic follicle. Differential diagnosis included OKC, al-

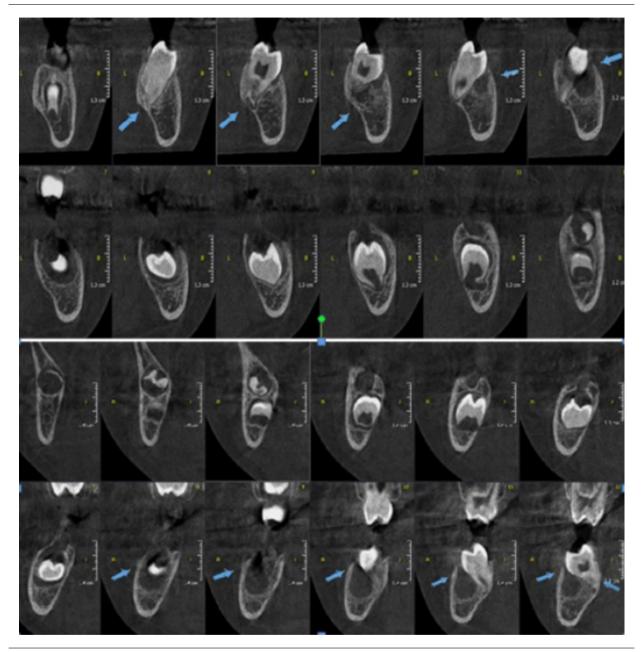
Figure 2. A CBCT Shows the Mandibular Axial Sections to Reveal the Bilateral Homogenous and Hypodense Lesions in the Distal Aspect of the First Molars

 $The \ lingual\ and\ buccal\ cortical\ plate\ is\ intact\ without\ perforation.\ There\ is\ no\ periosteal\ bone\ production.$

though it may attach further apically on the root instead of at the CEJ. DCs are more likely to have a smooth periph-

ery, while OKC is more likely to have a scalloped periphery (1). OKCs are often multilocular and located in the body or

Figure 3. The Cross-Sectional Reconstructed Views of the CBCT Show the Bilateral Radiolucencies Caused the Roots of the First Molars to Displace Lingually; the Buccal Tilt of the Crowns Can Also be Seen



A slight swelling of the buccal and lingual cortical plates with no perforation can be seen in both sides. The lesions had no effect on the inferior alveolar canal.

the ramus of the mandible (3). Radiographic diagnosis of this case included a buccal bifurcation cyst (BBC), which is occasionally bilateral. The most common location of a BBC is the first lower molar and the second molar. The diagnostic point for BBC is the tipping of an involved molar in which the root tips are pushed into the lingual cortical plate of the mandible and the crown is tipped buccally. A

panoramic or periapical radiographs would show the apparent occlusal surface of the affected tooth, but not the occlusal surface of the unaffected teeth; this is similar to our case. However, a BBC starts near the bifurcation region of the tooth and does not surround the crown as a DC does. Whereas a bilateral DC is rare, it should be considered in the differential diagnosis of bilateral pericoronal radiolu-

cencies (7).

CBCT imaging gives information about the origin size, content, cortical plates, and relationship of the lesion to the adjacent anatomical structure and makes it possible to diagnose a DC.

References

- Tamgadge A, Tamgadge S, Bhatt D, Bhalerao 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 Maxillofac Pathol.* 2011;15(1):91–5. doi: 10.4103/0973-029X.80017. [PubMed: 21731287].
- Shafer HL. Cyst and tumors of odontogenic origin in. 5 ed. 3. Shafers textbook of oral pathology; 2007. pp. 358-61.

- 3. Imada TS, Tieghi NV, Bernini GF. Unusual bilateral dentigerous cysts in a nonsyndromic patient assessed by cone beam computed tomography. *Brazil Dent Sci.* 2014;5(2):240.
- 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(1):49–51. [PubMed: 9973768].
- Shah N, Thuau H, Beale I. Spontaneous regression of bilateral dentigerous cysts associated with impacted mandibular third molars. *Br Dent J.* 2002;192(2):75-6. [PubMed: 11838012].
- Freitas DQ, Tempest LM, Sicoli E, Lopes-Neto FC. Bilateral dentigerous cysts: review of the literature and report of an unusual case. *Dentomax-illofac Radiol.* 2006;35(6):464–8. doi: 10.1259/dmfr/26194891. [PubMed: 17082341].
- 7. White S, Pharaoh M. Oral radiology principles and interpretation. 7 ed. St Louis: Mosby; 2014.

