

Bilateral dentigerous cysts that involve all four dental quadrants: a case report and literature review

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Abstract (J Korean Assoc Oral Maxillofac Surg 2016;42:123-126)

Dentigerous cysts are common odontogenic cysts that are associated with the crown of the tooth and typically develop from single lesions. Bilateral and multiple dentigerous cysts are very rare and occur in patients with syndromic conditions. This paper presents a case report of a 15-year-old male patient that experienced non-syndromic bilateral dentigerous cysts that simultaneously occurred in all four dental quadrants around the unerupted third molars. Clinicians should confirm the extent of cystic lesions using a panoramic view and computed tomography, and should keep the possibility of bilateral dentigerous cysts in mind as a potential diagnosis, even in a non-syndromic patient.

Key words: Dentigerous cyst, Syndrome, Third molar

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I. Introduction

Dentigerous cysts are a common type of odontogenic cyst in the jaw that arise from degenerative changes in the enamel epithelium and are accompanied by an accumulation of fluid between the crown of the tooth and the lining epithelium¹. They present as a single lesion, but bilateral or multiple lesions are usually found in patients with a developmental syndrome or systemic disease, such as Gorlin-Goltz syndrome (basal cell nevus syndrome), cleidocranial dysplasia, Maroteaux-Lamy syndrome, or mucopolysaccharidosis²⁻⁴. However, non-syndromic bilateral dentigerous cysts are very rare and we report a case of a 15-year-old male with bilateral dentigerous cysts that involved all four quadrants that had a symmetric appearance.

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II. Case Report

This case report was approved by the Institutional Review Board (HYI-15-102-1) of the Hanyang University Seoul Hospital and the patient provided informed consent. In January 2005, a 15-year-old male was referred to our clinic for asymptomatic multiple cystic lesions in both jaws, which were found by routine dental radiographs. The panoramic view revealed bilateral radiolucent cystic lesions around the unerupted mandibular third molars. In addition, well-defined radiolucent lesions were found in the superior-lateral aspects of both maxillary sinuses, which also contained the crowns of unerupted third molars. (Fig. 1. A) A computed tomography (CT) scan showed soft tissue masses obliterating both maxillary sinuses and displacing the maxillary third molars to the infraorbital area. (Fig. 1. B, 1. C) Both the patient and his parents were healthy without systemic disease or any signs of other syndromes. There were no abnormal findings in laboratory blood tests prior to the operation. Additionally, the chest radiograph was read as normal and did not show any in the clavicle and thorax areas.

The bilateral cystic lesions in the maxilla were enucleated by the Caldwell-Luc approach; then, the mandibular third molars were removed by the gingival approach under general anesthesia in February 2005. All surgical specimens were

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Fig. 1. A. Panoramic radiograph shows well-defined radiolucent cystic lesions around the bilateral unerupted third molars in both jaws. B. Computed tomography (CT) shows soft tissue masses obliterating both maxillary sinuses and displacing the maxillary third molars to the infraorbital area. C. Three-dimensional maxillo-facial shows four quadrants impacting the third molars.

Jae-Yun Jeon et al: Bilateral dentigerous cysts that involve all four dental quadrants: a case report and literature review. J Korean Assoc Oral Maxillofac Surg 2016

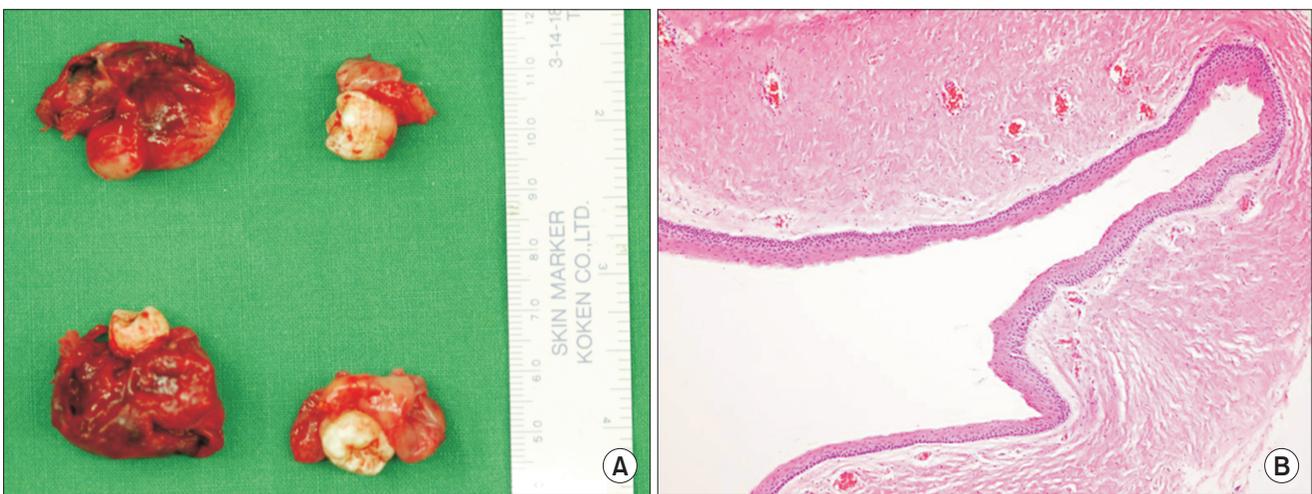


Fig. 2. A. All surgical specimens were unilocular cystic structures that contained gray-yellowish necrotic material. B. Histopathology results confirmed a typical dentigerous cyst lined by a non-keratinized squamous epithelium with mild inflammatory cellular infiltration in the connective tissue. No dysplastic changes were observed (H&E staining, $\times 40$).

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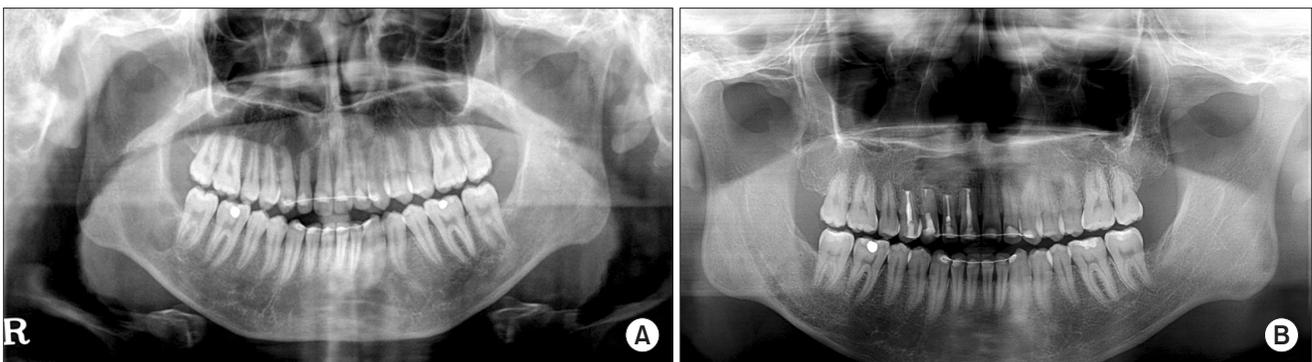


Fig. 3. A. Panoramic radiograph at 6-month follow-up. B. Panoramic radiograph at 10-year follow-up. Note the well-healed operative sites.

Jae-Yun Jeon et al: Bilateral dentigerous cysts that involve all four dental quadrants: a case report and literature review. J Korean Assoc Oral Maxillofac Surg 2016

unilocular cystic structures that contained a gray-yellowish material.(Fig. 2. A) Histopathologic analysis showed a typical dentigerous cyst lined by non-keratinized squamous

epithelium with mild inflammatory cellular infiltration in the connective tissue.(Fig. 2. B) No dysplastic changes were observed. The patient was followed periodically for 10 years

after surgery and no specific signs or symptoms related to the lesions in the operative sites were identified. (Fig. 3. A, 3. B)

III. Discussion

Dentigerous cysts are the most common type of developmental odontogenic cysts in the jaw and develop due to accumulation of fluid between the reduced enamel epithelium and the tooth crown, which leads to separation of the follicle from the crown of an unerupted tooth. They commonly arise from the mandibular third molars, maxillary canines, maxillary third molars, and mandibular second premolars¹. A search of PubMed from 1943 to 2014 revealed that only 30 cases of bilateral dentigerous cysts in non-syndromic patients were reported; 24 cases occurred in the mandible, 3 in the maxilla, and 3 in both the mandible and maxilla^{5,6}. Henefer⁷ reported a case of bilateral dentigerous cysts that simultaneously involved all four dental quadrants around the unerupted premolar and cuspids. However, in our case, the four dentigerous cysts involved un-erupted third molars and had a symmetric appearance in a non-syndromic patient.

The most common sites of dentigerous cysts are around the mandibular and maxillary third molars. The sites with high incidences of bilateral dentigerous cysts in nonsyndromic patients were similar to sites of general dentigerous cysts. The most common site was the mandibular third molar (11 cases), followed by the mandibular first molar (10 cases), maxillary third molar (2 cases), the maxillary cuspid (2 cases), mandibular first premolar (2 cases), mandibular second premolar (1 case), mandibular central incisor (1 case), and maxillary central incisor (1 case)^{5,6}.

Early detection of dentigerous cysts can be challenging because it develops as an asymptomatic lesion before the increased size causes cosmetic or dental/sinus problems. In this case, the patient also presented with asymptomatic lesions, but routine dental examinations showed extensive maxillary cysts that extended to both orbital floors. Therefore, we suggest that when clinicians encounter similar cases of unerupted third molars, routine radiographic examination may help detect early dentigerous cyst lesions and an accurate diagnosis may prevent extensive surgery or complications.

Bilateral and multiple dentigerous cysts are usually found in association with a number of syndromes such as Gorlin-Goltz syndrome (basal cell nevus syndrome), cleidocranial dysplasia, and Maroteaux-Lamy syndrome²⁻⁴. Gorlin-Goltz syndrome is a rare autosomal dominant disorder characterized by variable manifestations, including multiple basal

cell carcinomas, odontogenic keratocysts of the jaw, skeletal anomalies that include scoliosis and bifid ribs, palmar and plantar pits, calcification of the falx cerebri, and biparietal frontal bossing⁴. Cleidocranial dysplasia is a congenital disorder that causes skeletal abnormalities, i.e., delayed closures of cranial sutures and fontanelles, associated with the presence of wormian bone and clavicle aplasia. Oral manifestations include retention of deciduous teeth, multiple supernumerary teeth, delayed eruption or impaction of permanent teeth and formation of cysts around non-erupted teeth². Maroteaux-Lamy syndrome results from a defect in specific lysosomal enzymes that are required for the degradation of specific mucopolysaccharides. Oral complications include unerupted dentition, dentigerous cystic-like follicles, malocclusion and gingival hyperplasia³. Interestingly, there was one case of bilateral dentigerous cysts that was associated with a polymorphism in chromosome 1qh+⁸, and one case associated with long-term treatment with cyclosporine⁹.

In conclusion, it is important to identify patients with multiple or serial dentigerous cysts, and to investigate possible associated syndromes to prevent unnecessary complications. However, as we report in this case study, clinicians should confirm the extent of the cystic lesions using panoramic view and CT, even in a non-syndromic patient, and should keep the possibility of bilateral dentigerous cysts in mind in order to deliver accurate, early diagnoses.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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References

1. Waldron CA. Odontogenic cysts and tumors. In: Neville BW, Damm DD, Allen CM, Bouquet JE, eds. Oral and maxillofacial pathology. 2nd ed. Philadelphia: Saunders; 1990:590-3.
2. Trimble LD, West RA, McNeill RW. Cleidocranial dysplasia: comprehensive treatment of the dentofacial abnormalities. J Am Dent Assoc 1982;105:661-6.
3. Roberts MW, Barton NW, Constantopoulos G, Butler DP, Donahue AH. Occurrence of multiple dentigerous cysts in a patient with the

- Maroteaux-Lamy syndrome (mucopolysaccharidosis, type VI). *Oral Surg Oral Med Oral Pathol* 1984;58:169-75.
4. Gorlin RJ. Cysts of the jaws, oral floor and neck. In: Gorlin RJ, ed. *Thoma's oral pathology*. 6th ed. St. Louis: Mosby; 1970:470-1.
 5. Shirazian S, Agha-Hosseini F. Non-syndromic bilateral dentigerous cysts associated with permanent second premolars. *Clin Pract* 2011;1:e64.
 6. Cury SE, Cury MD, Cury SE, Pontes FS, Pontes HA, Rodini C, et al. Bilateral dentigerous cyst in a nonsyndromic patient: case report and literature review. *J Dent Child (Chic)* 2009;76:92-6.
 7. Henefer EP. Bilateral dentigerous cysts of the maxilla. Report of a case. *Oral Surg Oral Med Oral Pathol* 1964;17:296-8.
 8. Batra P, Roychoudhury A, Balakrishnan P, Parkash H. Bilateral dentigerous cyst associated with polymorphism in chromosome 1qh+. *J Clin Pediatr Dent* 2004;28:177-81.
 9. 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.