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# Bilateral Mandibular Dentigerous Cysts Presenting as an Incidental Finding: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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**Conflict of interest:** None declared

**Patient:** Male, 44
**Final Diagnosis:** Bilateral dentigerous cyst
**Symptoms:** Retained anterior deciduous tooth
**Medication:** —
**Clinical Procedure:** Bilateral cyst enucleation
**Specialty:** Dentistry

**Objective:** Unknown ethiology

**Background:** Dentigerous cysts are slow-growing odontogenic cysts that usually develop unilaterally as part of a pre-existing syndrome. Non-syndromic bilateral dentigerous cysts are extremely rare, but clinicians should be aware of this condition to ensure prompt diagnosis and management and to prevent complications.

**Case Report:** A case is presented of bilateral mandibular dentigerous cysts that were discovered incidentally in a 44-year-old man who presented for extraction of a retained maxillary deciduous tooth. Histological examination of the tissue specimens following bilateral enucleation confirmed the diagnosis of bilateral dentigerous cysts.

**Conclusions:** In this case, incidental bilateral dentigerous cysts were identified and treated by enucleation. The absence of an associated syndrome should not exclude the possibility of the diagnosis of dentigerous cysts, which should be removed to prevent future complications.

**MeSH Keywords:** Dentigerous Cyst • Odontogenic Cysts • Tooth, Impacted

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## Background

Dentigerous cysts were first described by Paget in 1863, and are slow-growing odontogenic cysts of developmental origin that are most commonly associated with the crown of an impacted or unerupted third molar [1–3]. Dentigerous cysts account for 24% of all cysts affecting the jaw their prevalence has been estimated to be 1.44 cysts per 100 unerupted teeth [1]. Dentigerous cysts are often asymptomatic [4,5] and are usually incidental findings [4,5]. However, complications from dentigerous cysts include secondary infection, which causes them to enlarge and become painful [6]. Radiographically, dentigerous cysts appear as well-defined, radiolucent lesions with sclerotic borders associated with an unerupted tooth and surrounded by a cystic space exceeding 5 $mm^3$ [7].

Mandibular dentigerous cysts are usually unilateral and most often occur in the context of syndromes such as cleidocranial dysplasia and Maroteaux-Lamy syndrome [8]. Non-syndromic bilateral odontogenic cysts have only rarely been reported in the literature, with less than twenty cases reported to date [9–15]. Dentigerous cysts are most commonly treated by enucleation or marsupialization, with management selected based on criteria that include the size of the lesion, the associated tooth and its position, the relationship to adjacent teeth, and the patient's age [1].

A case is reported of bilateral mandibular dentigerous cysts in a 44-year-old man that were diagnosed incidentally during investigation before extraction of a retained maxillary deciduous tooth.

## Case Report

A 44-year-old man presented to the Oral and Maxillofacial Surgery Department for extraction of a retained anterior maxillary deciduous tooth. He had no significant past medical history. On examination, the mandibular third molars were missing. Routine radiographic examination showed bilateral, unilocular, radiolucent lesions surrounding the crowns of impacted mandibular third molars with well-defined sclerotic borders, suggestive of dentigerous cysts (Figure 1A). The right-sided lesion measured approximately 3×1.5 cm, which extending anteromedially to the distal surface of tooth #47, and from the alveolar ridge to the lower border of the mandible. The left-sided lesion was smaller than the right-sided lesion, and measured approximately 2×1 cm and involved the distal surface and apex of tooth #37 anteromedially, and extended from the alveolar ridge. The left-sided cyst had a thicker inferior border than the right-sided cyst.

Bilateral cyst enucleation with extraction of the impacted wisdom teeth and excisional biopsy were performed under general anesthesia. Bilateral surgery was performed with intraoral crest incisions distal to the second molars, 1 cm distal to the distal limit of the cyst cavity. The incisions extended medially to the mesiobuccal area of the first molar, where oblique incisions extended to the vestibule. Triangular mucoperiosteal reflections created bone windows and exposed the cysts. The impacted teeth were carefully removed with the surrounding cystic lesion, ensuring the preservation of the inferior alveolar nerves (Figure 1B).

Histological examination confirmed the diagnosis of bilateral dentigerous cysts, with cystic spaces lined by cuboidal epithelium that was 2–3 layers thick and resembled a thin enamel epithelium-like lining (Figure 2A). Focally, the epithelium was

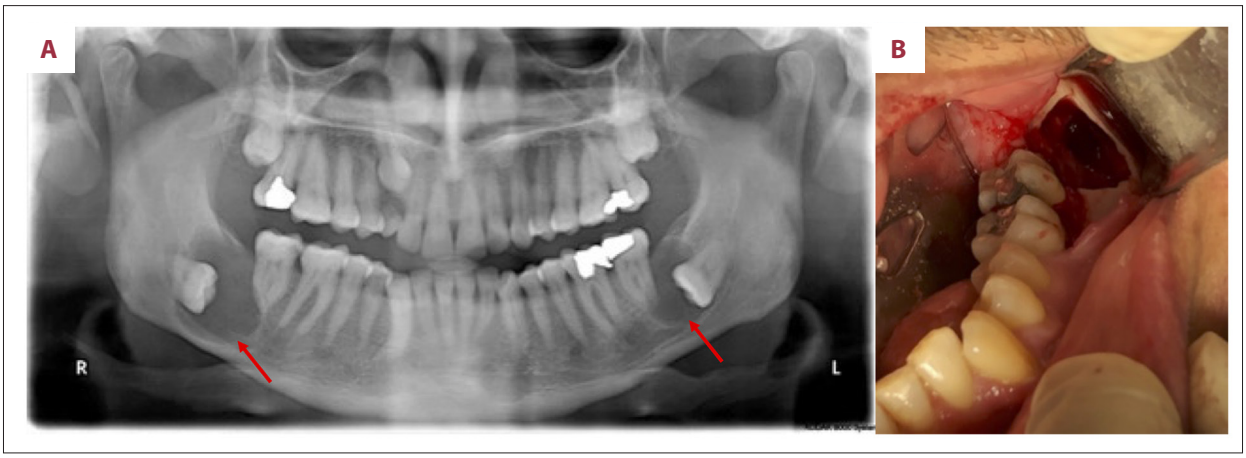


**Figure 1.** Bilateral mandibular dentigerous cysts in a 44-year-old man. (**A**) Panoramic radiograph showing bilateral radiolucent lesions surrounding the crowns of impacted mandibular third molars (red arrows). (**B**) Photograph of the cavity after removal of the left-sided cyst and the impacted tooth.



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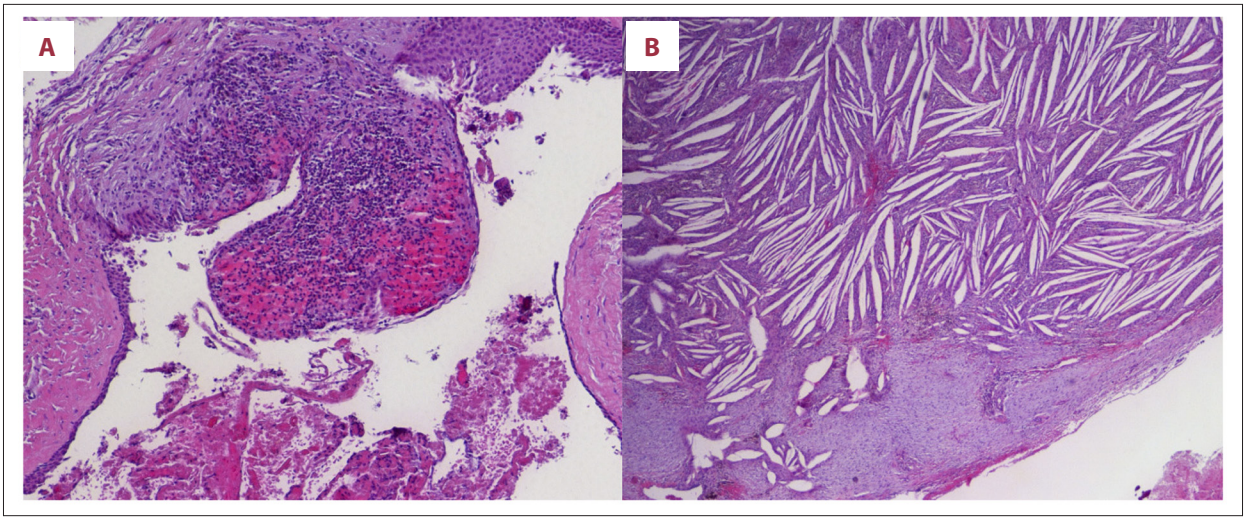


**Figure 2.** Photomicrographs of the histology of the bilateral mandibular dentigerous cysts in a 44-year-old man. (**A**) Photomicrograph of the excision biopsy shows cystic space lined by cuboidal epithelium that was 2–3 layers thick and resembled a thin enamel epithelium-like lining. Focally, the epithelium is hyperplastic, non-keratinizing, and stratified with a lymphoplasmacytic infiltrate and thickening of the cyst wall. Hematoxylin and eosin (H&E). Magnification ×10 objective. (**B**) Photomicrograph of the excision biopsy shows cyst wall cholesterol deposition, foreign body granulomas, and hemorrhage. H&E. Magnification ×20 objective.

hyperplastic, non-keratinizing, and stratified. The cyst wall was thickened, and there was a lymphoplasmacytic inflammatory cell infiltrate adjacent to the cyst wall that contained deposits of cholesterol, foreign body granulomas, and hemorrhage (Figure 2B, 2C). There was no dysplasia and no malignancy. The histology confirmed the diagnosis of benign bilateral dentigerous cysts.

## Discussion

Dentigerous cysts are mainly associated with impacted mandibular third molars. Non-syndromic bilateral dentigerous cysts have only rarely been reported in the literature [9–11]. The patient described in this report was asymptomatic, and the bilateral dentigerous cysts were discovered incidentally. Histopathology confirmed the diagnosis and excluded the most common differential diagnoses, which include odontogenic keratocysts, which are keratin producing, and cystic ameloblastomas, which shows dysplastic features that include cell hyperchromasia and loss of basal polarity [1].

Dentigerous cysts develop around unerupted teeth following the accumulation of fluid between the enamel epithelium and enamel. The developing crown exerts pressure on the dental follicle, resulting in venous obstruction and transudation of serum through the capillary walls, which eventually separates the crown and dental follicle [4]. Although the mechanism of cyst formation is understood, the underlying etiology of dentigerous cysts remains unknown. Benn and Altini [12] proposed an inflammatory etiology for dentigerous cysts, rather than a developmental cause. They described similar histopathological features to those in the present case, with a mixture of cuboidal and stratified squamous epithelium and an associated inflammatory infiltrate [12]. However, Benn and Altini described dentigerous cysts that occurred near to deciduous teeth, which they presumed was a source of infection from adjacent caries [12]. Although the bilateral dentigerous cysts described in this case may have been present since childhood, the presence of inflammation found on histology indicated that they had occurred secondary to infection or trauma, and the bilateral presentation possibly supported that they had arisen following infection.

Bilateral dentigerous cysts can grow to a large size with minimal or no symptoms. However, early detection is important as rare complications include tooth displacement, blockage of the nasal cavity, paresthesia due to pressure on the inferior alveolar nerve or, in very rare cases, metaplastic or neoplastic change to ameloblastoma and mucoepidermoid carcinoma [10,11]. Therefore, it is important that radiographic examination of all unerupted and erupted teeth is performed, either by panoramic X-radiography or using more advanced imaging techniques such as computed tomography (CT), cone-beam computed tomography (CBCT), or magnetic resonance imaging (MRI). These more advanced imaging techniques can distinguish between the cyst and adjacent structures, which is important to reduce morbidity and plan the most appropriate surgical procedure [8].



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As in this case, dentigerous cysts are usually managed by enucleation, although large cysts may initially be marsupialized to decompress the cyst before definitive treatment. However, a wait and see approach has also been recommended as spontaneous regression is reported to occur [13,14], by cyst tissue remains *in situ* with this management approach and may be associated with complications. Complete excision of dentigerous cysts is usually curative, and the recurrence rate is low [1].

## Conclusions

A rare case is presented of incidental bilateral dentigerous cysts that were diagnosed by imaging, and treated by surgical enucleation, with confirmation of the diagnosis on histology. The histological appearances also favored an inflammatory etiology, but the cause was unknown. The absence of a concurrent syndrome does not exclude the possibility of dentigerous cysts, which should be removed to prevent future complications.

### Conflicts of interest

None.

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