

原文題目(出處)：	Non-syndromic bilateral dentigerous cysts with significant root resorption: a case report. Oral Surg 2015;8:59-62
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內文：

#### A. Clinical relevance

##### 1. Dentigerous cysts

- (1) most common developmental odontogenic cysts
- (2) associated with impacted teeth, usually third molars
- (3) bilateral dentigerous cysts are rare
- (4) usually occur in patients with a known syndrome, such as cleidocranial dysplasia

##### 2. This case :

- (1) bilateral dentigerous cysts
- (2) healthy 38-year-old female patient
- (3) aggressive biology with significant root resorption
- (4) To date only 22 cases of bilateral non-syndromic dentigerous cysts have been reported, but none with evidence of root resorption.

#### B. Introduction

##### ● Dentigerous cysts

1. most common odontogenic developmental cysts
2. associated with unerupted teeth
3. 24% of cysts in the jaw
4. Asymptomatic
5. Incidental radiographic findings
6. occasionally associated with root resorption of the responsible or neighbouring teeth
7. Bilateral or multiple dentigerous cysts are generally associated with syndromes such as cleidocranial dysplasia or conditions like mucopolysaccharidosis

##### ● Radiological

1. well-defined
2. unilocular
3. radiolucent area surrounding the crown of an unerupted tooth, most commonly a third molar,
4. continuous with its cemento-enamel junction

#### C. Case report

##### ● General Data

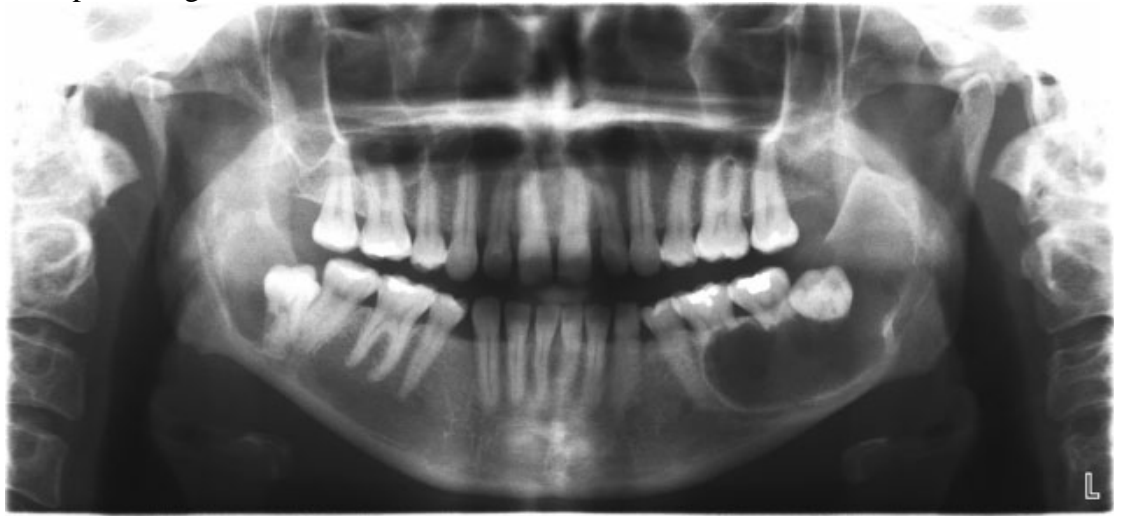
1. 38 y/o
2. Female
3. healthy

##### ● Chief complain

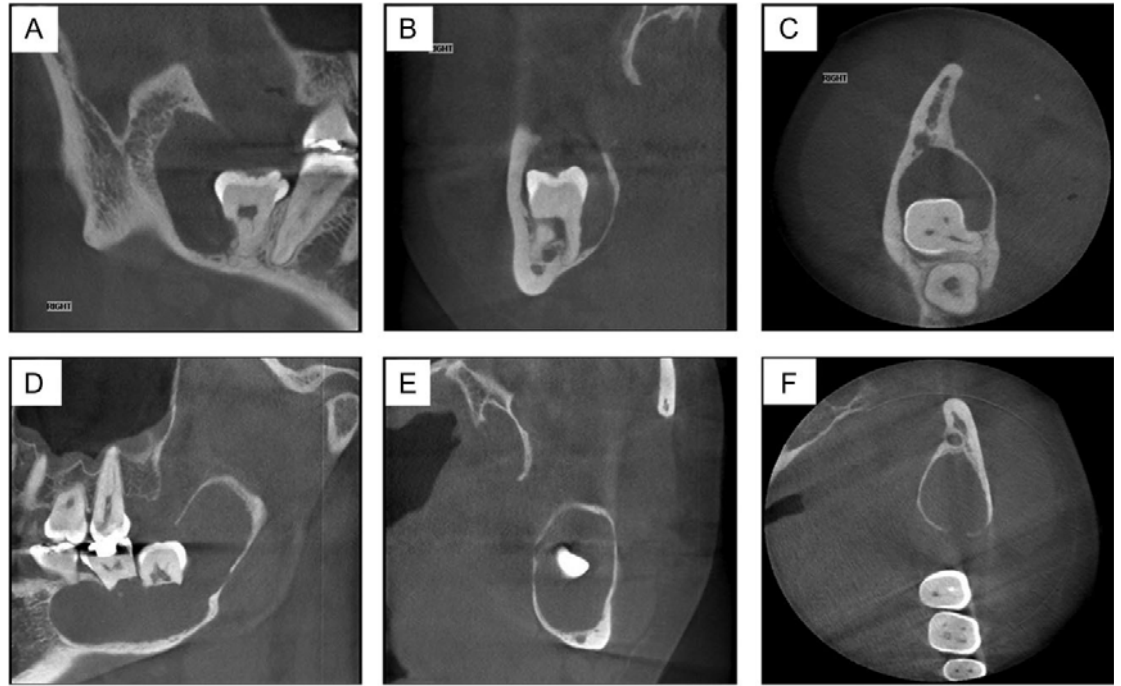
- ✓ intra-oral bone, non-tender swelling and mobility of the lower left molar teeth several weeks prior to her presentation
- ✓ no history of trauma, infection or pain

##### ● Examination

- ✓ Extraoral
  1. no visible extra-oral swelling or asymmetry
  2. mandibular contour felt regular and the lower border of the mandible was intact
  3. no paresthesia of the lower lip
- ✓ intraoral
  1. non-tender left buccal sulcus swelling
  2. firm and smooth
  3. consistent with buccal bone expansion
  4. No soft tissue expansion or dehiscence
  5. no discoloration of any teeth
  6. wisdom teeth were unerupted
  7. mobility:37 gr II; 36 gr I
  8. pulp vitality test was positive
- ✓ orthopantomogram

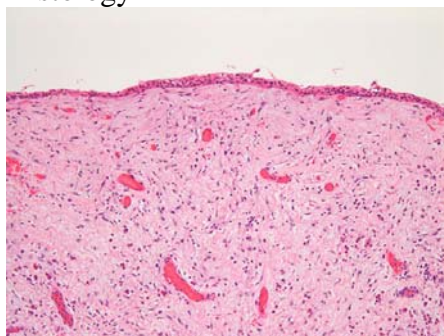


1. two large, unilocular, well-circumscribed radiolucent lesions
  2. involving the unerupted 38 and the contralateral 48
  3. from the left body of mandible into the left ramus and up to the sigmoid notch
  4. lower border of the mandible appeared intact
  5. inferior alveolar canal was displaced to the lower border
  6. extensive root resorption of LL6, LL7 and LL8
  7. similar smaller lesion was seen in relation to the impacted LR8
- ✓ cone beam computed tomography scan



- 1. aggressive biological behavior causing significant root resorption
- ✓ haematological investigations
  - 1. including alkaline phosphatase test
  - 2. normal
- ✓ myeloma screen
  - 1. negative
- ✓ no history of cutaneous lesions such as multiple naevi, basal cell carcinomas, metabolic disorders or other signs that would suggest an underlying syndrome or other systemic involvement.
- differential diagnosis
  - ✓ odontogenic keratocysts
  - ✓ ameloblastomas
  - ✓ dentigerous cysts
  - ✓ histiocytosis X
- biopsy
  - ✓ under GA
  - ✓ enucleation of both cysts
  - ✓ extraction of the involved teeth
- Twenty-four months post-operatively, the patient remains free of any recurrence

D. Histology



- variable thickness of fibrous wall lined by thin non-keratinizing stratified squamous epithelium

- occasional mural nodules composed of cholesterol clefts and associated giant cells
- the fibrous wall contained diffuse lymphoplasmacytic infiltrates
- no evidence of ameloblastoma or keratinisation

#### E. Discussion

- Dentigerous cysts
  - ✓ Clinical
    1. second most commonly occurring odontogenic cysts following radicular cysts
    2. developmental cysts
    3. enclose the crown of an unerupted tooth, most commonly third molars
    4. develops from the follicular tissues of the unerupted tooth
    5. accumulation of fluid between the reduced enamel epithelium and the associated crown
    6. mechanism that still remains unclear
    7. predilection
      - male
      - wide age range from childhood to middle age
    8. solitary, slowly progressive lesions
    9. generally asymptomatic unless they become infected
    10. present as incidental radiographic findings
  - ✓ Histological
    1. lining : thin, regular layer of non-keratinized stratified squamous epithelium, resembling the reduced enamel epithelium
    2. fibrous connective tissue capsule
    3. occasionally, cholesterol clefts are observed
  - ✓ Bilateral and multiple cysts
    1. association with several syndromes
      - cleidocranial dysplasia
      - Maroteaux–Lamy syndrome
      - mucopolysaccharidosis
    2. Multiple dentigerous cysts are thought to be extremely rare in non-syndromic patients
    3. Ochsenius et al.
      - analysed 2944 cases of odontogenic cysts and identified 546 patients with dentigerous cysts
      - 61 (11%) of which had synchronous dentigerous cysts
      - none of these patients had any metabolic or syndromic condition
      - but there is no clear evidence of how many of these cysts were bilateral or caused root resorption
- diagnostic challenge
  1. unusual bilateral simultaneous presentation of clinically and radiologically similar lesions
  2. the aggressive biological behaviour in terms of significant root resorption
- differential diagnosis
  1. bilateral odontogenic keratocysts

bilateral odontogenic keratocysts
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This case
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multilocular	X
most commonly located in the body or ramus of the mandible	O
present synchronously in syndromic cases, such as the basal cell naevus syndrome (Gorlin–Goltz syndrome)	X
locally aggressive, but not usually associated with root resorption	X
Histological (1) relatively thin fibrous capsule, which may give rise to satellite cysts (2) regular layer of stratified squamous epithelium with a well-defined basal layer and keratin production displays mitotic activity	X ;keratinisation was excluded by microscopic analysis

2. bilateral unilocular ameloblastomas

bilateral unilocular ameloblastomas	This case
most common radiolucent benign odontogenic tumour	
can cause expansion and destruction of the bone as well as various degrees of root resorption	O
encountered in young adult life as an incidental radiological finding	X
Histological (1) epithelial lined cyst (2) comprising a basal layer of columnar cells which displays areas of intraluminal or mural proliferation of ameloblastomatous tissue	X

3. odontogenic fibromyxomas

odontogenic fibromyxomas	This case
multiple radiolucent areas of varying size and bony septa, but unilocular lesions have also been described	O
often related to unerupted teeth	O
radiologically well-defined	O
microscopy demonstrates specific features, readily distinguishing odontogenic fibromyxomas from other lesions, such as dentigerous cysts stellate, fibroblast-like cells within an abundant connective tissue matrix, which contains various amounts of collagen	X

4. systemic osseolytic conditions such as histiocytosis or multiple myeloma

➤ Histiocytosis X (eosinophilic granuloma)

Histiocytosis X (eosinophilic granuloma)	This case
occurs in children or young adults	X
solitary osteolytic lesion in the mandible	O
local destructive potential and causing loosening of teeth	O
multifocal eosinophilic granuloma	X

(Hand–Schuller–Christian syndrome) is usually associated with other visceral lesions and multi-organ manifestations such as hepatosplenomegaly, lymphadenopathy and endocrine disorders	
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➤ multiple myeloma

multiple myeloma	This case
multiple osteolytic lesions that can occur in the jaw bones	O
individuals between 50 and 70 years of age	X
neoplasm composed of abnormal plasma cells producing large amounts of a single homogeneous type of immunoglobulin that can be traced in blood product electrophoresis	X ; Blood tests and paraprotein electrophoresis proved negative for multiple myeloma

F. Summary

1. The presented case displayed clinical and radiological features that were not typical enough to point confidently towards one single working diagnosis.
2. Non-syndromic bilateral dentigerous cysts are extremely uncommon
3. Commonly encountered lesions can present a diagnostic challenge and emphasizes the importance of histopathological analysis in definitive diagnosis and overall management

題號	題目
1	Which is the most common type of developmental odontogenic cyst? (A) Odontogenic Keratocyst (B) Dentigerous cyst (C) Calcifying Odontogenic cyst (D) Lateral periodontal cyst
答案(B)	出處：Oral and Maxillofacial Pathology third edition p.679
題號	題目
2	Which statement about dentigerous cyst is wrong? (A) Dentigerous cyst may occur in association with any unerupted tooth, and most often they involve maxillary canine. (B) Small dentigerous cyst are usually completely asymptomatic. (C) Dentigerous cyst encloses the crown of an unerupted tooth and is attached to the tooth at the cemento-enamel junction. (D) The prognosis for most dentigerous cyst is excellent, and recurrence seldom is noted after complete removal of the cyst.
答案(A)	出處：Oral and Maxillofacial Pathology third edition p.679-682