

# Multiple Dentigerous Cysts With a Complex Odontoma: An Unusual Case Report

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## Abstract

**Introduction:** Multiple dentigerous cysts are rare and mostly associated with syndromes.

**Case Presentation:** The present case report is unique, as the dentigerous cysts enclosed the lateral incisor, canine, first and second premolars of the mandible and were accompanied by a complex odontoma, instead of first mandibular molar.

**Conclusions:** Multiple unilateral nosyndromic dentigerous cysts with complex odontoma are found only rarely. The periapical inflammation of non-vital deciduous teeth and trauma may be considered ethiological factors.

**Keywords:** Dentigerous Cyst, Complex Odontoma, Teeth

## 1. Introduction

Dentigerous cysts are the second most common type of odontogenic cysts, after radicular cysts, and account for about 24% of all true cysts in the jaw. There is usually no pain or discomfort associated with the cyst, unless it becomes secondarily infected (1-3).

The teeth that are commonly affected are the mandibular third molars, maxillary canines, maxillary third molars and, rarely, the central incisors (4-6).

The cyst appears as a well-defined ovoid unilocular radiolucency with sclerotic border surrounding the crown of an unerupted tooth. Most dentigerous cysts are solitary (5). Multiple dentigerous cysts have been reported in systemic disease and syndromes such as mucopolysaccharidosis, basal cell nevus syndrome, and cleidocranial dysplasia. The occurrence of bilateral dentigerous cysts in the absence of a developmental syndrome is rare (7). As Freitas et al. and Reddy et al. have declared, there have been only 24 cases of multiple nonsyndromic cysts noted in the literature between 1943 and 2011 (3, 7).

## 2. Case Presentation

A 26-year-old woman was visited at the maxillofacial radiology department of the Isfahan School of dentistry. Clinically, some of her teeth were absent without an extraction history in the mandible's left quadrant. She had a childhood history of trauma to the mandible that resulted

in necrosis and the extraction of the mandibular right deciduous molars. During general examination, there was no relevant systemic disease or syndromes. She did not exhibit any skeletal abnormality, and she was not mentally retarded. She did not reveal pathological symptoms via neurological, skin, or ophthalmological clinical examinations. Her other teeth were vital, and she did not show signs of swelling or other clinically relevant findings. Panoramic radiograph showed five impacted teeth in the right quadrant of the mandible (lateral incisor, canine, first and second premolars, and a lesion not resembling a tooth instead of a first molar). The central incisor and second and third molars were present, and a deciduous canine was remained (Figure 1). CBCT views showed pericoronal radiolucencies around the impacted teeth, with well-defined sclerotic borders that displaced the teeth apically. Root resorption of adjacent teeth and expansion were not observed. There was a well-defined mixed radiolucent-radio opaque lesion in the position of the first molar, and CBCT densitometry of radioopacities foci of the lesion showed a density near to dentin and cancellous bone (Figure 2). 3D CBCT images showed the teeth were associated with the inferior alveolar nerve (Figure 3). The surgery was done under conscious sedation, the buccal and lingual sulcular full thickness mucoperiosteal flaps were reflected and all impacted teeth with lesions were removed, taking care not to damage inferior alveolar nerve. The flaps were repositioned and sutured. The specimens were sent for histopathologic examination, which

showed dentigerous cysts and a complex odontoma. There were several pieces of fibrotic soft tissue that were covered by thin, non-keratinized striated squamous epithelium. There were many odontogenic epithelial islands in the fibrotic tissue background. Eosinophilic hyalin structures like Rushton bodies were spread widely throughout the tissue and were more basophile because of calcification (Figure 4A - C). Microscopic examination of the lesion medial to the second molar showed some dentin tissue with tubular structure mixed with odontogenic epithelial islands and hyaline structures, which suggested a complex odontoma (Figure 4D - F). A follow-up panoramic radiograph was taken three months after surgery (Figure 5). The patient felt numbness in her lower lip after surgery, which improved during three and six months follow-ups after surgery.



**Figure 1.** Panoramic Radiograph Shows Impacted Teeth

### 3. Discussion

Dentigerous cysts are the most common developmental odontogenic cysts of the jaws. They originate from the expansion of the follicle of an unerupted tooth and attach to the cemento enamel junction. Dentigerous cysts are discovered in radiographs taken to evaluate unerupted or missing teeth. There is usually no pain or discomfort associated with the cyst, unless it becomes secondarily infected. They are generally solitary. Multiple dentigerous cysts are usually associated with Maroteaux-Lamy and cleidocranial dysplasia and are extremely rare in the absence of a syndrome or systemic disease. Reddy et al. have declared there have been only 24 cases of multiple nonsyndromic cysts noted in the literature between 1943 and 2011 (4, 7, 8), and these are mostly associated with molars and, infrequently, premolars. In the present case, multiple unilateral dentigerous cysts were found in the lateral incisor, canine and premolars. The mean age of reported cases was 22.5; in the present case, the patient was 26 years old. Patients with dentigerous cysts present impacted teeth and

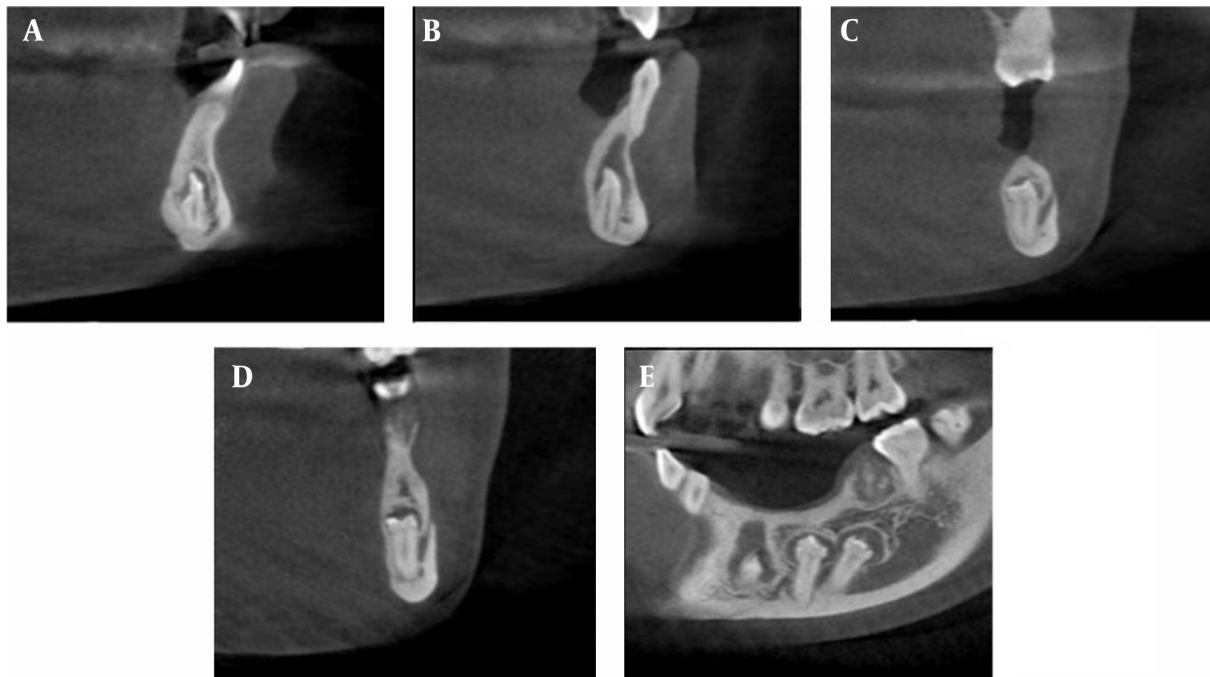
asymptomatic slow-growing swellings. In this case, the patient presented with impacted teeth, but she had not any swelling. In all reported cases, including the present case, radiographic examination showed a unilocular radiolucent lesion associated with the crown of an unerupted tooth with well-defined sclerotic margins (1, 5). In addition to the developmental origin, some authors have suggested that periapical inflammation of non-vital deciduous teeth in proximity to the follicles of unerupted permanent teeth may be a factor for triggering dentigerous cyst formation (7). The literature does not mention the role of trauma as a trigger for forming dentigerous cysts, however it may cause dentigerous cyst formation and impaction. The present case had a history of childhood trauma to the mandible.

Dentigerous cysts may be misdiagnosed as odontogenic keratocyst, but they are more common in young patients with an unerupted or impacted tooth; indeed, the mean area of the odontogenic keratocysts was larger than that of dentigerous cysts. Cortical expansion and root resorption are characteristics more associated with dentigerous cysts, but the present case did not show any of these. Gorlin-Goltz syndrome, also known as basal cell nevus syndrome, is linked with mutation in the PTCH gene (human homolog of the drosophila segment polarity gene, "patched"). It is characterized by cutaneous, skeletal, ophthalmic, and neurologic abnormalities. Partial expression of the gene may result in the occurrence of only multiple recurring OKC without any associated systemic findings (1, 2, 9, 10). With this in mind, our second differential diagnosis after multiple dentigerous cysts was multiple odontogenic keratocysts on radiographs (the type that shows partial expression of the gene), because her clinical examination did not show signs or symptoms of basal cell nevus syndrome, however histopathologic findings ignored it.

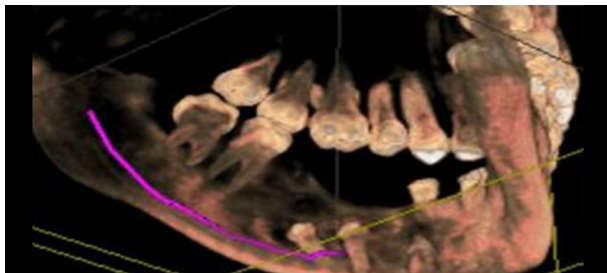
Singh Saluja et al. considered CT imaging valuable in unerupted teeth. Because CBCT provides reconstructed images of high diagnostic quality with lower exposure doses and shorter scanning time, compared to CT, CBCT was performed for this patient to give information about the lesion's extension, expansion of cortical plates, and the content and relationship of the lesion to adjacent anatomical structures, such as the inferior alveolar nerve (1, 11).

Surgical removal is suggested for dentigerous cysts because of their epithelial lining's tendency toward neoplastic transition (5, 12), so all specimen removed from a patient must be evaluated histologically.

Odontomes are hamartomatous malformations of odontogenic origin. They are asymptomatic, non-aggressive lesions that are found accidentally in radiographs. Complex odontoma are usually found in the posterior mandible and discovered before the second



**Figure 2.** A, CBCT cross sectional view of lateral incisor; B, canine; C, first premolar; D, second premolar; E, CBCT, tangential view of the impacted canine, first and second premolars and a complex odontoma. The lateral incisor cannot be seen in this cut.



**Figure 3.** 3D CBCT, buccal aspect, shows the relation of teeth to the inferior alveolar Nerve.

decade of life. The present case had a complex odontoma in the position of the first molar. The etiology of odontoma is unknown, but genetic factors and environmental causes such as trauma and infection have been proposed. This case had history of trauma, and it may be the cause for the change in the dental follicle to an odontoma (13-15).

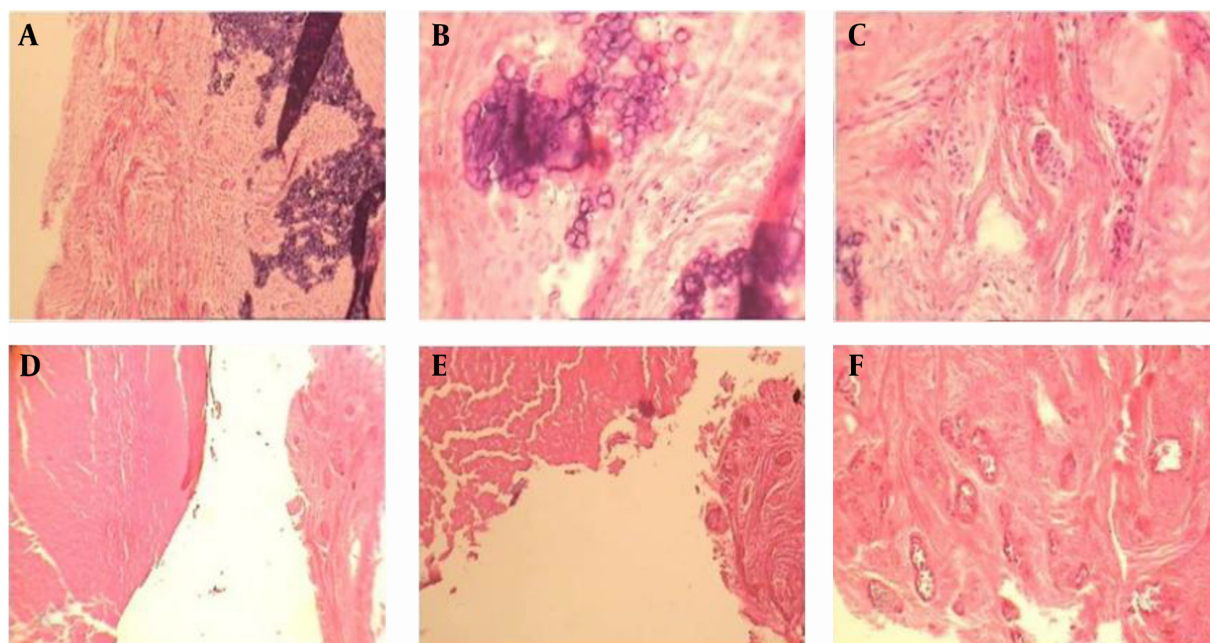
As depicted in the present case, multiple unilateral nonsyndromic dentigerous cysts with complex odontoma are rare findings. Periapical inflammation of non-vital deciduous teeth and trauma may be considered ethiological factors.

## Footnote

**Authors' Contribution:** Study concept and design: Mitra Karbasi Kheir and Mahnaz Sheikhi; acquisition of data: Mitra Karbasi Kheir and Amir Hosein Moaddabi; analysis and interpretation of data: Mitra Karbasi Kheir, Mahnaz Sheikhi and Mohamad Hasan Samandari; drafting of the manuscript: Mitra Karbasi Kheir; critical revision of the manuscript for important intellectual content: Mitra Karbasi Kheir, Mahnaz Sheikhi and Mohamad Hasan Samandari; statistical analysis; administrative, technical, and material support: Mitra Karbasi Kheir, Mohamad Hasan Samandari and Amir Hosein Moaddabi; study supervision: Mahnaz Sheikhi and Mohamad Hasan Samandari.

## References

1. Saluja JS, Ramakrishnan MJ, Vinit GB, Jaiswara C. Multiple dentigerous cysts in a nonsyndromic minor patient: Report of an unusual case. *Natl J Maxillofac Surg.* 2010;1(2):168-72. doi: [10.4103/0975-5950.79223](https://doi.org/10.4103/0975-5950.79223). [PubMed: [22442592](https://pubmed.ncbi.nlm.nih.gov/22442592/)].
2. Rohilla M, Namdev R, Dutta S. Dentigerous cyst containing multiple impacted teeth: a rare case report. *J Indian Soc Pedod Prev Dent.* 2011;29(3):244-7. doi: [10.4103/0970-4388.85834](https://doi.org/10.4103/0970-4388.85834). [PubMed: [21985882](https://pubmed.ncbi.nlm.nih.gov/21985882/)].
3. Freitas DQ, Tempest LM, Sicoli E, Lopes-Neto FC. Bilateral dentigerous cysts: review of the literature and report of an unusual case. *Dentomaxillofac Radiol.* 2006;35(6):464-8. doi: [10.1259/dmfr/26194891](https://doi.org/10.1259/dmfr/26194891). [PubMed: [17082341](https://pubmed.ncbi.nlm.nih.gov/17082341/)].



**Figure 4.** A, B, C, dentigerous cysts; D, E, F, complex odontoma.



**Figure 5.** Panoramic View Three Months After Surgery

4. 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](#)].
5. Tikekar S, S Degwekar S, R Bhowate R, Kailasam S. Bilateral Dentigerous Cyst: An Unusual Case Report and Review of Literature. *J Indian Acad Oral Med Radiol.* 2010;**22**:116-8. doi: [10.5005/jip-journals-10011-1028](#).
6. Karacayli U, Gocmen-Mas N. Multiple Abnormal Unerupted Mandibular Permanent Molar Teeth: A Case Report. *Int J Morphol.* 2009;**27**(1)

doi: [10.4067/s0717-95022009000100012](#).

7. Reddy MP, Swamy KV N, Tavane PN, Malu K, Gupta PK. Non syndromic bilateral dentigerous cysts: an unusual case report. *Int J Clin Dent Sci.* 2011;**2**(3).
8. Nagaveni NB, Umashankara KV, Manjunath S. Multiple impactions in non-syndromic patient: A case report. *Arch Orofac Sci.* 2011;**6**(2):33-8.
9. Mangalore B. Unusual presentation of dentigerous cyst in a young child: a case report. *J Indian Soc Pedo Prev Dent.* 2001:21-3.
10. Kurdekar RS, Prakash J, Rana AS, Kalra P. Non-syndromic odontogenic keratocysts: A rare case report. *Natl J Maxillofac Surg.* 2013;**4**(1):90-3. doi: [10.4103/0975-5950.117874](#). [PubMed: [24163561](#)].
11. Hilgers ML, Scarfe WC, Scheetz JP, Farman AG. Accuracy of linear temporomandibular joint measurements with cone beam computed tomography and digital cephalometric radiography. *Am J Orthod Dentofacial Orthop.* 2005;**128**(6):803-11. doi: [10.1016/j.ajodo.2005.08.034](#). [PubMed: [16360924](#)].
12. Araujo JP, Kowalski LP, Rodrigues ML, de Almeida OP, Lopes Pinto CA, Alves FA. Malignant transformation of an odontogenic cyst in a period of 10 years. *Case Rep Dent.* 2014;**2014**:762969. doi: [10.1155/2014/762969](#). [PubMed: [24804126](#)].
13. Jeyaraj P, Mohan SM. Dentigerous cyst of mandible with multiple complex odontomes. *Med J Armed Forces India.* 2008;**64**(4):382-3.
14. Normak W, Reichart PA, Philipsen HP. Differential diagnosis of oral and maxillofacial lesions. Thieme Medical Publishers; 2000.
15. White SC, Pharoah MJ. Oral Radiology principles and Interpretation. St Louis: Mosby; 2004.