



Bilateral Dentigerous Cyst: A Case Report And Review Of Literature

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Abstract

Dentigerous cysts (DC) are the most common developmental cysts of the jaws, most frequently associated with impacted mandibular third molar teeth and impacted canines. Bilateral dentigerous cysts are rare and occur typically in association with a developmental syndrome such as cleidocranial dysplasia and Gorlin -Goltz syndrome. Conversely, their occurrence in non-syndromic patients is quite unusual. Herein, we report a case of bilateral dentigerous cysts in mandibular posterior region of non-syndromic 29-year-old female patient. Radiographically, impacted 38 and 48 and a bilateral well-defined radiolucency around the crown of the lower third molars present. Surgical excision and follow up radiograph of the patient showed a favourable osseous formation with no evidence of recurrence of the cysts. So, a comprehensive radiographic examination, especially for mandibular third molars, is essential for correct diagnosis and management of the lesions. Furthermore, systematic clinical examinations should be performed to rule out any associated syndrome.

Keywords: Dentigerous cyst, non-syndromic, Cleidocranial Dysplasia, Gorlin Goltz

Introduction

A dentigerous cyst is one that encloses the crown of an unerupted tooth by expansion of its follicle, and is attached to its neck¹. They are the second most common odontogenic cysts after radicular cysts, accounting for approximately 25% of all true cysts in the jaws². The cyst arises by the separation of the follicle from the crown of an unerupted tooth by means of an accumulation of liquid between the crown of the tooth and the reduced epithelium of the enamel. The erupting tooth exerts pressure on the impacted follicle which obstructs venous outflow. This results in a serum transudate across the capillary

wall causing elevated hydrostatic pressure and therefore pooling of fluid. This causes separation of the follicle from the crown, leading to formation of the cyst^{3,4}. The peak incidence of occurrence is in the second to fourth decades of life⁵. Most commonly affected tooth is the mandibular third molars followed by maxillary canines⁶. Pain and discomfort are not present unless it becomes secondarily infected.

Dentigerous cysts are frequently discovered on routine radiographs taken to investigate a failure of

tooth eruption, a missing tooth⁷. Radiographically they show a unilocular, radiolucent lesion characterized by well-defined sclerotic margins and associated with the crown of an unerupted tooth in the CEJ. A normal follicular space is 3-4 mm, so when the space is more than 5 mm, it is suspicious of a dentigerous cyst⁸. The dentigerous cyst most commonly presents as a solitary lesion⁸. Multiple dentigerous cysts with bilateral presentation have been associated with several conditions which involve unerupted teeth, such as mucopolysaccharidosis type VI (Maroteaux Lamy syndrome), Nevroid Basal Cell Carcinoma Syndrome (Gorlin-Goltz Syndrome) and Cleidocranial Dysplasia⁹. In the absence of these syndromes, bilateral dentigerous cyst are quite uncommon. Treatment for dentigerous cyst is either enucleation or marsupialization. As noted in Table 1, there have been 60 cases of multiple non -syndromic cysts reported in the literature from 1941 to 2023, and we only found 24 reports of non -syndromic bilateral DCs associated with mandibular third molars. Here, we report the unusual occurrence of non -syndromic bilateral DC associated with unerupted mandibular third molars.

Case Presentation:

A, 29-year-old female patient came to the department with a chief complaint of pain and swelling on lower left back region of jaw in the last 7 days. Extra oral examination revealed facial asymmetry. Diffuse swelling present on lower left side of face extending anteroposteriorly 1 cm away from left corner of mouth to ear lobe and supero inferiorly 1 cm below the ala tragus line and 1cm below the lower border of mandible (Figure1). Skin overlying swelling appeared to be normal. Intraoral examination revealed, unerupted 38 and 48. Reduced mouth opening and obliteration of left buccal vestibule.No significant swelling on right side of mandible present. The patient's medical history was not relevant and it was not associated with any syndrome.

Panoramic radiographic showed horizontally impacted 38 surrounded by a well-defined unilocular radiolucency extending from ramus involving interradicular area of 37. Also, horizontally impacted 48 with well corticated unilocular radiolucency involving 48 completely and impinging distal root of 47(Figure 2). Left submandibular lymph node was

palpable and tender. On aspiration, it revealed numerous RBCs and cholesterol crystals suggestive of infected dental cyst (Figure 3). Based on all the features, provisional diagnosis of Bilateral dentigerous cyst was given. However, large periapical cyst, odontogenic keratocyst, ameloblastoma and adenomatoid odontogenic tumor were also considered in the differential diagnosis. Routine blood examination result was within normal limits.

Bilateral cyst enucleation with extraction of the impacted teeth and excisional biopsy were performed under local anaesthesia (Figure 4&5). Bilateral enucleation 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 were extended medially to the mesiobuccal area of the first molar and oblique incisions extended to the vestibule. Bony windows were created and cysts were exposed. The impacted teeth and the surrounding cystic lesion, ensuring the preservation of the inferior alveolar nerves along with adjacent 47 and 37 were carefully removed.

The surgical specimens were then sent for final diagnosis (Figure 6). Histopathologically both the specimens were similar and showed a thin fibrous cystic wall lined by a 2-3 cell layered thick non-keratinized stratified squamous epithelium with no rete pegs. The adjacent connective tissue wall showed mild inflammatory cell infiltrate. Subepithelial layers showed parallel bundles of collagen fibres. These findings confirmed diagnosis of dentigerous cysts. One year follow up radiograph showed physiological healing with favourable bone repair without any other complications (Figure 7).

Discussion:

The dentigerous cyst is the second most common developmental odontogenic cyst in the oral cavity, with 1.44 cases reported for every 100 impacted teeth⁹. They are mainly associated with impacted mandibular third molars followed by maxillary permanent canine, mandibular premolars, maxillary third molars and rarely the maxillary premolars. Morshed stated that 1.44% of impacted teeth undergo dentigerous cyst transformation, so dentigerous cysts involving the premolars are much less. Daley et al, reported an incidence rate of 0.1-0.6%, whereas Shear found the incidence as 1.5%⁷. Dentigerous

cysts are most commonly seen in the 2nd and 3rd decades of life¹⁰. These lesions can also occur in children and adolescents and show a male predilection¹¹. In the present case report, the dentigerous cysts were associated with the mandibular third molar in a 29-year-old female.

The exact histogenesis of the dentigerous cyst is unknown. It is stated that the fluid accumulation between REE and enamel as a result of the pressure exerted by an erupting tooth when an impacted follicle obstructs the venous outflow and thereby induces rapid transudation of serum across the capillary wall. Toller concluded that the likely origin of the dentigerous cyst is the breakdown of proliferating cells of the follicle after impeded eruption. These breakdown products result in increased osmotic tension and leads to cyst formation. Bloch stated that the overlying necrotic deciduous tooth to be the origin of dentigerous cyst. Their resultant periapical inflammation will spread to involve the follicle of the unerupted permanent successor and results in dentigerous cyst formation⁷. This suggests that these exudates from the deciduous teeth might be one of the risk factors for the occurrence of dentigerous cysts⁷.

The occurrence of bilateral dentigerous cysts is extremely rare and hardly reported. They are usually associated with the Maroteaux-Lamy syndrome, (mucopolysaccharidosis type VI), Cleidocranial dysplasia, Basal cell nevus syndrome. The combined effect of cyclosporin and a calcium blocker is reported to cause bilateral dentigerous cyst¹². Cases present with pleomorphism in chromosome 1qh+ has also been reported with this condition¹³. In present case, there was no clinical evidence of syndrome.

After an extensive literature search on bilateral DCs, only 60 reported cases were identified from 1941 to 2023 (Table 1). This finding revealed the rareness of this lesion. The age range for the reported cases varies from 3 years to 57 years of age. The majority of cases were showing male predilection i.e. 42 were males and 18 were females. In all reported cases, including the present case as well, radiographic examination showed a unilocular radiolucent lesion associated with the crown of an unerupted tooth with well-defined sclerotic margins. Out of 60 cases, 24 cases have been associated with mandibular third molars, similar to our case followed by 13 cases of

maxillary first molar, 8 cases of mandibular premolars, 7 cases of maxillary canines, 4 cases of mandibular canine, mandibular second molar and maxillary third molar each.

Since the cysts can reach a considerable size with minimal or no symptoms, their early detection and removal are important to reduce morbidity. In our case, pain and swelling were present in the lower left back region. Therefore, it is important to perform radiological examinations when encountering unerupted teeth. A panoramic radiograph may be used for the initial examination. However, pathological analysis of the lesion is necessary for the final diagnosis. Other lesions may have the same radiological features as DCs, such as odontogenic keratocysts and unicystic ameloblastomas.

Although tooth involvement, cortical expansion, and root resorption are features mostly related to DCs, other lesions were not excluded until the results of the pathological examinations were revealed. Odontogenic keratocysts cause less expansion in the bone compared to DCs and are less likely to cause root resorption. The mean age of patients with an odontogenic keratocyst is less than that of patients with a DC; the mean size of an odontogenic keratocyst is larger than that of DCs. Furthermore, DCs are more likely to have a smooth periphery but odontogenic keratocyst mostly have a scalloped periphery¹⁴. It is not possible to differentiate unicystic ameloblastomas from DCs with clinical and radiographic examinations¹⁵.

The management of the reviewed cases of bilateral dentigerous cysts showed that 53 cases were treated by enucleation alone, 2 cases by marsupialization and 1 case with a combination of marsupialization and enucleation, 2 cases by regression, and 1 case by decompression and resection. The choice of marsupialisation depends on the size as well as the extent of the cyst, its proximity to vital structures, the risk of pathological fracture etc. Decompression of the cyst is considered in an attempt to salvage the involved teeth or adjacent delicate structures. Enucleation of the cystic lining is therefore often the preferred surgical treatment modality for dentigerous cysts, as demonstrated in the current case report.

In light of evidence suggesting the potential transformation of dentigerous cysts into more sinister lesions, such as ameloblastoma, mucoepidermoid and squamous cell carcinoma arising from the cystic lining of dentigerous cyst, it is likely that specialist intervention will consist of surgical enucleation as the definitive treatment option. However, cysts that cause major dental displacement and involve major bone loss must be decompressed. This treatment modality encourages new bone formation, as marsupialization decreases pressure within the cyst.

Conclusion:

Hence, it is important to identify patients with multiple or serial dentigerous cysts, and to investigate possible associated syndromes to prevent unnecessary complications. However, 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.

References:

1. Shafer H, Levy. Cyst and tumors of odontogenic origin in. Shafers textbook of oral pathology. 5th ed. New Delhi: Elsevier India; 2007. p. 358-61
2. Gonzalez SM, Spalding PM, Payne JB, Giannini PJ. A dentigerous cyst associated with bilaterally impacted mandibular canines in a girl: a case report. *J Med Case Rep* 2011; 5: 230
3. Zehra Yonel Bilateral Dentigerous Cysts: An Updated Literature Review and Report of a Case with Associated Root Resorption, *Dent Update* 2018; 45: 1063–1082
4. Devi P, Bhovi TV, Mehrotra V, Agarwal M. Multiple dentigerous cysts. *J Maxillofac Oral Surg* 2014; 13: 63–66
5. Speight P, Fantasia JE, Neville BW. Odontogenic and non odontogenic developmental cysts. In: World Health Organization Classification of Tumours: Pathology and Genetics of Head and Neck Tumours. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ (eds). WHO: IARC Publications, 2017: pp234–235
6. 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: 464–468
7. 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;15:91-5
8. Neville BW, Damm DD, Allen CM, Bonquot JE. Odontogenic cysts and tumors. *Oral and Maxillofacial pathology*. 2nd ed. Philadelphia. P.A.: W.B. Saunders; 2005. p. 591-2
9. Hadad H, Paolo P, JUNIOR G. Bilateral mandibular dentigerous cyst in non-syndromic patient: technical strategy and literature review. *Archives of Health Investigation*. 2020;9(2):159-63.
10. Shear M, Speight PM. Cyst of oral and maxillofacial regions. 4th ed. New Jersey: Blackwell publishers; 2007. p. 59-75.
11. Cawsons RA, Odell EW. *Cowson's essentials of oral pathology and medicine*. 8th ed. Philadelphia: Churchill livingstone, Elsevier. p. 108-9
12. Yamalik K, Bozkaya S, Erkmén E, Baris E. Nonsyndromic bilateral mandibular dentigerous cysts: report of a rare case. *Turkiye Klinikleri J Dent Sci* 2007;13:129-33
13. 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
14. Khandeparker RV, Khandeparker PV, Virginkar A, Savant K. Bilateral Maxillary Dentigerous Cysts in a Nonsyndromic Child: A Rare Presentation and Review of the Literature. *Case Rep Dent*. 2018;2018:7583082.
15. Mehdizadeh M, Hajisadeghi S, Lotfi A. Bilateral Dentigerous Cysts in a Non -Syndromic Patient: Literature Review and Report of a Case. *J Islam Dent Assoc Iran*. 2019; 31 (1):57 -63.
16. Bang KO, Yadav SR, Shenoi R, Budhraj N, Ingole P. Bilateral non-syndromic dentigerous cyst—rare case report and literature review. *Advances in Oral and Maxillofacial Surgery*. 2021 Oct 1;4:100043.

Figure: 1- Extra oral images showing a swelling on left side of face extending below inferior border of mandible and right side shows no appreciating swelling



Figure:2- OPG showing well defined unilocular radiolucency with impacted 38 and 48



Figure:3- Aspiration showing abundant RBCs, few cholesterol crystals

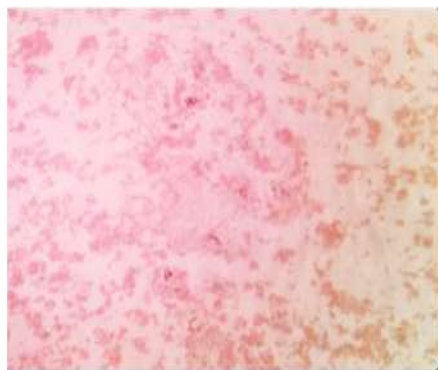


Figure:4 & 5- Surgical enucleation images of left sided cyst and impacted tooth

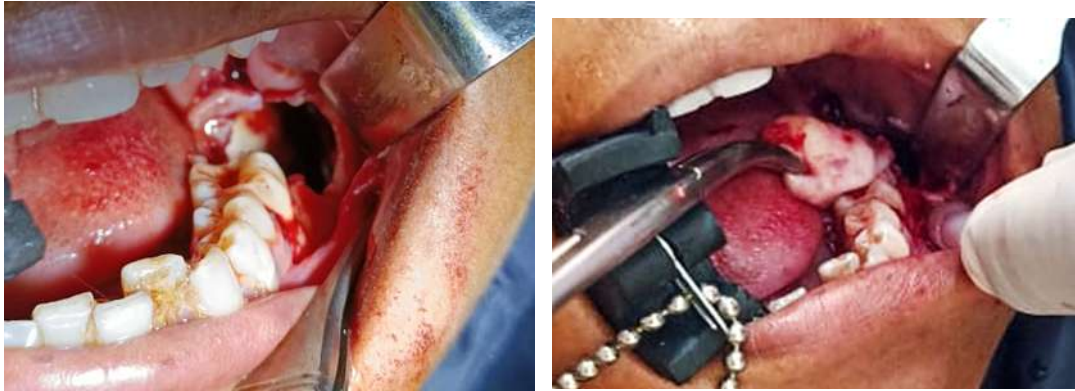


Figure:6- Surgical specimen showing extracted 38,37,47,48 and multiple tissue bits



Figure:7- Follow up(6months) OPG showing osseous formation



Table no:1 -Review of cases of bilateral dentigerous cyst in non-syndromic patients since 1941-2023

Author	Year	Gender	Age	Location	Treatment
Boyko	1941	M	*	Mandibular third molars	Enucleation
Myers	1943	F	19	Mandibular third molars	Enucleation
Tam	1955	M	7	Mandibular first molars	Enucleation
Henefer	1964	F	52	Maxillary third molars	Enucleation
Stanback	1970	M	9	Mandibular first molars	Enucleation
Callaghan	1973	M	38	Mandibular third molars	Enucleation
Burton	1980	F	57	Mandibular third molars	Enucleation
Swerdloff	1980	F	7	Mandibular first molars	Enucleation
Crinzi	1982	F	15	Mandibular third molars	Enucleation
McDonnell	1988	M	15	Mandibular second premolars and second molars	Enucleation
Eidinger	1989	M	15	Mandibular first molars	Enucleation
ONeil	1989	M	5	Mandibular first molars	Enucleation
Banderas	1996	M	38	Mandibular third molars	Enucleation
Carr	1996	M	7	Permanent mandibular first molars	Enucleation
Sands	1998	F	3	Mandibular central incisors and first molars	Enucleation
Ko	1999	M	42	Mandibular third molars	Enucleation
De Biase	2001	M	8	Mandibular first molars	Enucleation
Shah	2002	M	39	Mandibular third molars	Spontaneous regression
Ustuner	2003	M	6	Maxillary canines	Enucleation
Batra	2004	F	15	Mandibular third molars and second premolars	Enucleation
Frietas	2006	M	13	Maxillary third molars and mandibular second molars	Enucleation
Yamalik	2007	M	51	Mandibular third molars	Enucleation
Shahrabi	2007	M	37	Maxillary and mandibular canines	Surgical resection
Fregnani	2008	M	5	Permanent mandibular first molars	Marsupialization
Chew	2008	F	30	Mandibular third molars	Spontaneous regression
Maurette	2008	M	7	Permanent mandibular first molars	Decompression
Cury	2009	M	5	Permanent mandibular first molars	Enucleation
Saluja	2010	M	22	Maxillary premolars and mandibular premolars	Enucleation

Krishna	2010	F	12	Maxillary canines and unilateral mandibular lateral incisor	Enucleation
Tikekar	2010	M	11	Second premolars	Enucleation
Prasad	2010	F	12	Maxillary canines	Enucleation
Grewal	2011	M	11	Maxillary canines	Enucleation
Shirazian	2011	M	10	Mandibular second premolars	Marsupialization
Tamgadge	2011	M	10	Maxillary premolars	Enucleation
Reddy	2011	F	11	Mandibular first premolars	Enucleation
Ishihara	2012	M	13	Mandibular premolars	Enucleation
Aher	2013	M	24	Maxillary and mandibular third molars	Enucleation
Byantal	2013	M	13	Maxillary mesiodens	Enucleation
Imada	2014	F	42	Mandibular third molars	Enucleation
Subash	2014	M	14	Maxillary and mandibular second molars	Enucleation
Morais	2014	M	15	Mandibular third molars	Enucleation
Naik	2014	M	25	Mandibular third molars	Enucleation
Vassiliou	2015	F	38	Mandibular third molars	Enucleation
Kaushik	2015	M	*	Maxillary and mandibular canines	Enucleation
Sanjay	2015	F	24	Mandibular canines	Enucleation
Devi	2015	M	17	Maxillary central and lateral incisors and mandibular canines and first premolars	Enucleation
Jeon	2016	M	15	Maxillary and mandibular third molars	Enucleation
Esmaelizadeh	2016	M	8	Mandibular second molars	Enucleation
Iyengar	2017	M	7	Mandibular first molars	Enucleation
Shruthi	2017	F	15	Mandibular third molars	Enucleation
Khandeparker	2018	M	10	Maxillary second premolars and canines	Enucleation
Gogula	2018	M	40	Mandibular third molars	Enucleation
Mehdzadeh	2018	M	28	Mandibular third molars	Enucleation after Marsupialization
Vasiapphan	2018	M	27	Mandibular third molars	Enucleation
Kishore M	2018	F	19	Md molars	Enucleation
AlKhudair	2019	M	19	Mx molars	Enucleation
Pant	2019	M	10	Mx canines	Enucleation
Sarwpriya S	2019	F	27	Mx third molars	Enucleation

Amal M	2019	M	44	Md third molars	Enucleation
Kshiteej B	2021	F	55	Md third molars	Enucleation
Monika K	2023	F	29	Mandibular third molars	Enucleation