

Non-syndromic bilateral dentigerous cysts associated with permanent second premolars

Shiva Shirazian, Farzaneh Agha-Hosseini
Oral medicine, Faculty of Dentistry, Tehran
University of Medical Sciences, Tehran, Iran

Abstract

The dentigerous cyst is one of the most common developmental odontogenic cysts in the jaw. Occurrence of the bilateral dentigerous cyst is uncommon, and frequently associated with syndromes like basal cell nevus syndrome or cleidocranial dysplasia. There are few reports on the presence of bilateral dentigerous cyst in non-syndromic patients, and most of these are associated with first and third molars. The reported case in this paper is bilateral dentigerous cysts associated with mandibular permanent second premolars, in the absence of any signs of syndrome. To our knowledge bilateral dentigerous cysts in these locations have not been previously reported.

Introduction

Dentigerous cyst (DC) is the second most common odontogenic cyst, after radicular cyst in the jaw.¹ DC comprises 24% of all true cysts in jaws, and its prevalence in the general population is approximately 1.44 cysts for every 100 unerupted teeth.² The World Health Organization (WHO) classified the cysts developed on germs of non-erupted teeth as epithelial odontogenic cysts (follicular); these cysts have the following characteristics:

- i) they develop on non-erupted teeth;
- ii) they can manifest at any age, though the majority of cases occur during the 2nd and 3rd decades;
- iii) they are unilateral;
- iv) usually the teeth involved are the mandibular third molars and the maxillary canines;³

DCs are usually asymptomatic and have no pain. Chief patient complaint is frequently an unerupted tooth.¹

Case Report

A 10-year-old boy was referred with the chief complaint of bilateral expansion of the mandible. He had had tooth-ache 2 years previously, and referred himself to a dentist, where he had undergone a pulpectomy and filling of his

bilateral mandibular deciduous molars. The posterior areas of his mandible began to expand bilaterally about 18 months ago and this continued until 2 months ago. There was not any erythematosis, pain or suppuration. The lesion expanded slowly.

In his medical history, there was no systemic disease or sign of any syndrome. Intra-oral examination revealed bilateral bony expansion at buccal of the mandible (at right side from mesial #83 to distal #85 and at left side from mesial #74 to distal #36). There were bilateral well-defined unilocular radiolucencies at the pericoronal of the permanent second premolars (Figure 1). According to clinical and radiographic features, bilateral dentigerous cysts were considered as the first differential diagnosis of the lesions. To treat the lesion, a left side marsupialization was carried out, and 2 months later, after primary healing of the left side, the same surgery was done on the right side. The histopathologic result confirmed the dentigerous cysts at both sides. Clinical re-evaluation did not show any associated syndrome.

Discussion

As DC is usually asymptomatic, it is often found in routine radiographs. DC is solitary in most patients,^{1,2} and bilateral or multiple DCs are usually associated with syndromes such as cleidocranial dysplasia, basal cell syndrome Maroteaux-Lamy syndrome, and Hunter's syndrome.^{4,5}

The occurrence of bilateral DCs in non-syndromic patients is rare.^{1,2,4,6,7} To our knowledge, only 29 cases of bilateral DCs in non-syndromic patients have been reported (Table 1).

Most reports of bilateral dentigerous cysts are associated with third molars,^{2,6,9,12-14,16} and first molars.^{1,3,8,10,17} Choeyoungcheol reported bilateral dentigerous cysts associated with mandibular first premolar,¹⁵ and McDonnell reported a case associated with permanent mandibular second premolar and molar.¹⁸ We did not find any previous reports of bilateral permanent mandibular second premolars without any syndrome.

DCs have unilocular radiolucencies in different sizes, with well-defined sclerotic borders of unerupted tooth crowns. Normal size of the follicular space is considered about 3-4 mm in radiographs, so if this space is more than 5 mm, dentists should suspect DCs.²⁴

Other pericoronal radiolucencies such as odontogenic keratocyst (OKC), ameloblastoma, odontogenic fibroma, adenomatoid odontogenic tumor (AOT), pindborg tumor, and odontoma can have similar radiographic features to DC. Radicular cyst of deciduous teeth must be considered, especially in the apex of deciduous molars, which can mimic DC of permanent premolars.^{4,8} DCs cause more bone expansion than

Correspondence: Shiva Shirazian, Department of Oral medicine, Faculty of Dentistry, Tehran University of Medical Sciences, North Kargar street, 1439955991, Tehran, Iran.
Tel. +98.2188497404 - Fax: +98.2188497400.
E-mail: shiraziash@gmail.com

Key words: bilateral dentigerous cysts, odontogenic cyst, second premolars, syndrome.

Received for publication: 6 August 2011.

Accepted for publication: 22 August 2011.

This work is licensed under a Creative Commons Attribution NonCommercial 3.0 License (CC BY-NC 3.0).

©Copyright S. Shirazian et al., 2011

Licensee PAGEPress, Italy

Clinics and Practice 2011; 1:e64

doi:10.4081/cp.2011.e64

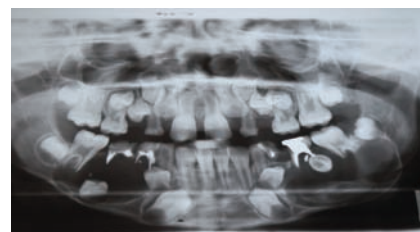


Figure 1. Panoramic radiograph showed bilateral pericoronal radiolucencies.

OKC. Microscopic characteristics of OKC are uniform thickness, palisading of basal cells with hyperchromatic nuclei, keratinized pattern with a corrugated surface, and a stratified squamous epithelium with few layers.¹⁹ Ameloblastoma and DCs can have similar clinical and radiographic features.²⁰ Pindborg tumor, AOT, and odontoma are radiolucent lesions that most often exhibit amorphous calcification within their lytic space.⁴ The varying size of multiple radiolucent areas and bony septa are the common features of odontogenic fibromixoma, but unilocular lesions also have been reported.⁸

Histopathologic evaluation is necessary for definite diagnosis.^{8,20}

Our case showed no sign of syndrome, indicating that although bilateral or multiple DCs are common in syndromic patients, detection of bilateral DCs do not necessarily indicate any syndrome.

References

1. Fregnani ER, Perez DE, de Carvalho PA, Alves FA. Metachronous bilateral dentigerous cysts associated with permanent first molars. *J Dent Child (Chic)* 2008;75:197-200.
2. Ko KS, Dover DG, Jordan RC. Bilateral dentigerous cysts-report of an unusual case and review of the literature. *J Can Dent Assoc* 1999;65:49-51.
3. De BA, Ottolenghi L, Polimeni A, et al.

Bilateral mandibular cysts associated with cyclosporine use: a case report. *Pediatr Nephrol* 2001;16:993-5.

4. Ustuner E, Fitoz S, Atasoy C, et al. Bilateral maxillary dentigerous cysts: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2003;95:632-5.
5. Norris LH, Piccoli P, Papageorge MB.

Multiple dentigerous cysts of the maxilla and mandible: report of a case. *J Oral Maxillofac Surg* 1987;45:694-7.

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;35:464-8.
7. Shahrabi FS, Lotfalian M. A pigmented dentigerous cyst in a patient with multiple dentigerous cysts of the jaws: a case report. *J Contemp Dent Pract* 2007;8:85-91.
8. Cury SE, Cury MD, Cury SE, et al. Bilateral dentigerous cyst in a nonsyndromic patient: case report and literature review. *J Dent Child (Chic)* 2009;76:92-6.
9. Chew YS, Aghabeigi B. Spontaneous regression of bilateral dentigerous cysts: a case report. *Dent Update* 2008;35:63-5.
10. Maurette PE, Allais M, Morais M, Jorge J. Treatment of an bilateral mandibular dentigerous cyst by decompression in a seven-years-old child: A case report and follow up for seven years. *Rev Venez Invest Odontol* 2008;8:26-33.
11. Dinkar AD, Dawasaz AA, Shenoy S. Dentigerous cyst associated with multiple mesiodens: a case report. *J Indian Soc Pedod Prev Dent* 2007;25:56-9.
12. Garcia RG, Hernandez VE, Moreno AC, Diaz PLM, Perez JS, Campo FJR. Therapeutic approach to impacted third molar follicles. *Rev Esp Cirug Oral Maxilofac* 2005;27:80-4.
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. Shah N, Thuau H, Beale I. Spontaneous regression of bilateral dentigerous cysts associated with impacted mandibular third molars. *Br Dent J* 2002;26;192:75-6.
15. Choeyoungcheol YJ, Yeong-Chul C, Lee K. Case of bilateral dentigerous cysts treated by marsupialization. *Pediatr Dent J* 2002;29:196-8.
16. Banderas JA, Gonzalez M, Ramirez F, Arroyo A. Bilateral mucous cell containing dentigerous cysts of mandibular third molars: report of an unusual case. *Arch Med Res* 1996;27:327-9.
17. O'Neil DW, Mosby EL, Lowe JW. Bilateral mandibular dentigerous cysts in a five-year-old child: report of a case. *ASDC J Dent Child* 1989;56:382-4.
18. McDonnell DG. Bilateral dentigerous cysts. A case history. *J Ir Dent Assoc* 1988;34:63.
19. Vencio EF, Mota A, de Melo PC, as Filho AA. Odontogenic keratocyst in maxillary sinus with invasive behaviour. *J Oral Pathol Med* 2006;35:249-51.
20. Dunsche A, Babendererde O, Luttges J, Springer IN. Dentigerous cyst versus unicystic ameloblastoma-differential diagnosis in routine histology. *J Oral Pathol Med* 2003;32:486-91.

Table 1. Summary of previously reported bilateral DCs.

Author	Year	Gender	Age (ys)	Location	Treatment
Cury ⁸	2009	male	5	Permanent Man. First molars	Enucleation
Fregnani ¹	2008	male	5	Permanent Man. First molars	Marsupialization
Chew ⁹	2008	female	30	Man. Third molars	Spontaneous regression
Maurette ¹⁰	2008	male	7	Permanent Man. First molars	Decompression
Farahani ⁷	2007	Male	37	Permanent Max. Canines, permanent Man. Canines, permanent left Man. Lateral incisor	Surgical resection
Dinkar ¹¹	2007	Female	14	Max. Mesiodens	Surgical resection
Freitas ⁶	2006	male	14	Permanent left Man. Molars, right Max. Third molar	Enucleation
Garcia ¹²	2005	male	28	Man. Third molars	Extraction molars
Batra ¹³	2004	female	15	Man. Third molars	Enucleation
Ustuner ⁴	2003	male	6	Max. Canines	*
Shah ¹⁴	2002	male	39	Man. Third molars	Spontaneous regression
Choeyoungcheol ¹⁵	2002	o	o	Permanent Man. First premolars	Marsupialization
Biase ³	2001	male	8	Permanent Man. First molars	*
Ko ²	1999	male	42	Man. Third molars	Enucleation
Sands ¹	1998	female	3	Permanent Man. Central incisors and first molars	Enucleation
Carr ¹	1996	male	7	Permanent Man. First molars	Enucleation
Banderas ¹⁶	1996	male	38	Man. Third molars	*
O'Neil ¹⁷	1989	male	5	Permanent Man. First molars	Enucleation
Eidinger	1989	male	15	Permanent Man. First molars	Enucleation
MC Donnell ¹⁸	1988	male	15	Permanent Man. Second molar and second premolar	Enucleation
Norris ⁵	1987	female	7	All unerupted teeth	*
Crinzi ⁶	1982	female	15	Man. Third molars	Enucleation
Burton ⁶	1980	female	57	Man. Third molars	Enucleation
Swerdloff ¹	1980	female	7	Permanent Man. First molars	Enucleation
Callaghan ⁵	1973	male	38	Man. Third molars	Enucleation
Stanback ⁶	1970	male	9	Permanent Man. First molars	Enucleation
Henefer ⁶	1964	female	52	Max. Third molars	Enucleation
Tam ¹	1955	male	7	Permanent Man. First molars	Enucleation
Myers ²	1943	female	19	Man. Third molars	Enucleation

*not defined in the literature; °the original article in Korean