Non-syndromic bilateral dentigerous cysts associated with permanent second premolars

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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. 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. The World Health Organization (WHO) classified the cysts developed on germs of non-erupted teeth as epithelial odontogenic cysts (folicular); 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;
v) DCs are usually asymptomatic and have no pain. Chief patient complaint is frequently an unerupted tooth.

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-syndrome patients is rare.1,2,4,5 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,2,4,9,12-14,16 and first molars.1,3,11,17 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 crows. 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.2,4,18

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.5,6 DCs cause more bone expansion than

References

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3. De BA, Ottolenghi L, Polimeni A, et al. 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 sepata are the common features of odontogenic fibromixoma, but unilocular lesions also have been reported.8 Histopathologic evaluation is necessary for definite diagnosis.2,20

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.

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

Received for publication: 6 August 2011. Accepted for publication: 22 August 2011.

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Table 1. Summary of previously reported bilateral DCs.

| Author          | Year | Gender | Age (ys) | Location | Treatment               |
|-----------------|------|--------|----------|----------|-------------------------|
| Marquette10      | 2008 | male   | 7        | Permanent | Decompression           |
| Banderas16      | 1999 | male   | 38       | Man. Third molars | *                        |
| Farahani2       | 2007 | Male   | 37       | Permanent Max. Canines, permanent left Man. Lateral incisor | Surgical resection       |
| Freitas6        | 2006 | male   | 14       | Permanent left Man. Molars, right Max. Third molar | Marsupialization         |
| Garcia12        | 2005 | male   | 28       | Man. Third molars | Extraction molars        |
| Batra13         | 2004 | female | 15       | Man. Third molars | Enucleation              |
| Maurette2       | 2008 | male   | 7        | Permanent Man. First molars | Enucleation              |
| Dinkar11        | 2007 | Female | 14       | Max. Mesiodens | Surgical resection       |
| Carr3           | 1996 | male   | 7        | Permanent Man. First molars | Enucleation              |
| Bandera16       | 1996 | male   | 38       | Man. Third molars | *                        |
| O’Neil17        | 1989 | male   | 5        | Permanent Man. First molars | Enucleation              |
| Eidinger        | 1989 | male   | 15       | Permanent Man. First molars | Enucleation              |
| MC Donnell18    | 1988 | male   | 15       | Permanent Man. Second molar and second premolar | Enucleation              |
| Norris1         | 1987 | female | 7        | All unerupted teeth | *                        |
| Under1        | 1982 | female | 15       | Man. Third molars | Enucleation              |
| Burton4         | 1980 | female | 57       | Man. Third molars | Enucleation              |
| Swerdloff1      | 1980 | female | 7        | Permanent Man. First molars | Enucleation              |
| Callaghan6      | 1973 | male   | 38       | Man. Third molars | Enucleation              |
| Stanback6       | 1970 | male   | 9        | Permanent Man. First molars | Enucleation              |
| Henefer2        | 1964 | female | 52       | Max. Third molars | Enucleation              |
| Tam1            | 1955 | male   | 7        | Permanent Man. First molars | Enucleation              |
| Myers2          | 1943 | female | 19       | Man. Third molars | Enucleation              |

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