Metachronous Bilateral Dentigerous Cysts Associated With Permanent First Molars

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ABSTRACT

The occurrence of bilateral dentigerous cysts (DC) has been described as being associated mostly with third molars in adult patients. Very few reports show the bilateral occurrence of dentigerous cysts associated with first molars or premolars in childhood. The purpose of this report was to describe the case of a 5-year-old boy with bilateral mandibular dentigerous cysts associated with permanent first molars. (**J Dent Child 2008;75:197-200**)

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According to the World Health Organization,[1] dentigerous cysts (DC) are developmental odontogenic cysts associated with the crowns of permanent teeth. Frequently, they do not cause symptoms or pain, but noneruption of the affected permanent tooth is a common clinical finding. A radiolucent area with well-defined sclerotic borders associated with the crown of an unerupted tooth is a typical radiographic finding.[1]

DCs are the second most common odontogenic cysts after radicular cysts and occur most frequently in the mandible during the second or third decades of life, mainly associated with impacted third molar teeth.[2] The occurrence of this lesion associated with primary teeth and permanent first molars is very rare,[3] and it is usually a single lesion. Bilateral DCs are an extremely uncommon finding in children. The purpose of this paper was to describe the case of metachronous bilateral dentigerous cysts in a nonsyndromic child patient.

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CASE DESCRIPTION

A 5-year-old boy was referred to the Stomatology Department at A.C. Camargo Cancer Hospital, Sao Paulo, Brazil, complaining of a painless lump of 25 days duration that appeared in the mandible's posterior region. His previous medical history was not contributory.

Upon extraoral exam, slight facial asymmetry was observed due to a firm, well-circumscribed nodule, which could be palpated in the left mandibular body's posterior region (Figure 1). No presence of palpable neck lymph nodes was noted. Intraoral examination revealed discoloration of

Figure 1. During clinical examination, a slight facial asymmetry was observed in the left mandibular body's posterior region.

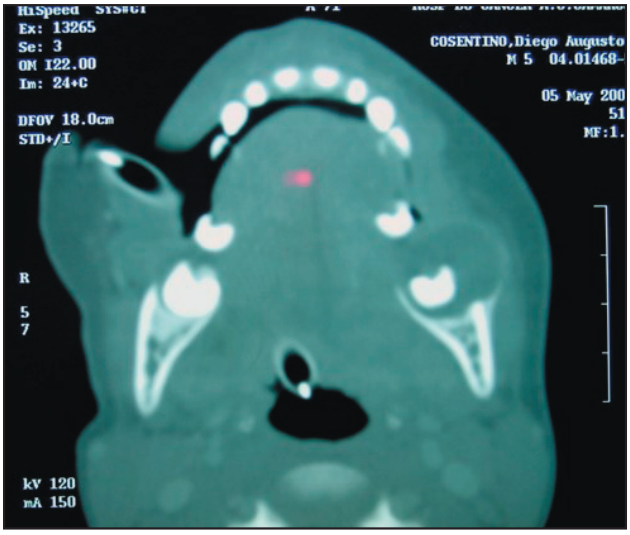

Figure 2. Computed tomography showing a rounded lesion with liquid density sited on the mandible's left side, which expanded the vestibular bone plate and adjacent soft tissues.

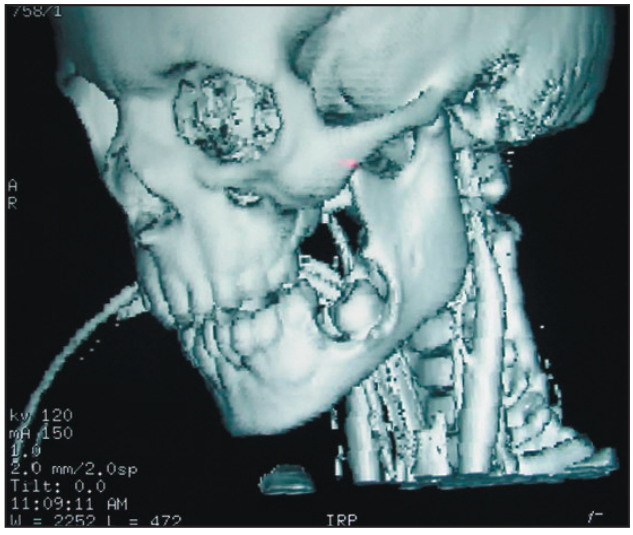

Figure 3. Three-dimensional reconstruction confirmed a well-circumscribed lesion associated with the permanent first molar's crown.

Figure 4. After 2 months, a clearly defined radiolucent lesion involving the permanent right first molar's crown was noted.

the bottom of the fold in the left posterior inferior region adjacent to the primary second molar's distal region, with noneruption of the inferior permanent first molar.

Panoramic radiography evidenced a unilocular lesion on the mandible's left side, with clear limits that dislocated the permanent second molar germ, reabsorbed the primary second molar's distal root, and involved the nonerupted permanent first molar's entire crown, measuring 2 cm in diameter. Computed tomography revealed a rounded lesion with liquid density sited on the mandible's left side, expanding the vestibular bone plate and adjacent soft tissues (Figure 2). Three-dimensional reconstruction using the cuts from computed tomography confirmed a bounded lesion associated with the permanent mandibular first molar's crown (Figure 3). During nitrous-oxide sedation to perform the computed tomography, aspiration punction was performed and 3 ml of liquid was withdrawn. According to clinical and image features, a DC was considered as the main diagnostic hypothesis. Thus, an incisional biopsy of the lesion and marsupialization (decompression) were performed simultaneously to allow eruption of the involved tooth. Microscopically, the diagnosis of DC was confirmed.

After 2 months of clinical and radiographic follow-up, the patient complained of a swelling on the right side of his face. On clinical examination, the presence of a nodule located in the right mandibular body's posterior region was observed. Radiographically, a well-circumscribed radiolucent lesion 2 cm in extent was noted involving the permanent right first molar's crown (Figure 4). On the left side, there was still a residual lesion associated with the permanent first molar's crown, similar to that observed on the right side. Thus, under general anesthesia, the patient was submitted to enucleation of the bilateral cystic lesions, maintaining the involved permanent teeth. Histopathologically in both lesions, cystic cavities lined by cubic epithelium were observed, confirming the diagnosis of bilateral DCs. Clinical re-evaluation showed no associated syndromes.

After 15 days, it was clinically possible to note the initial eruption of both first molars. After 24 months, complete eruption could be observed, without clinical and radiographic signs of recurrence (Figure 5).

DISCUSSION

DCs usually occur singly, and are mainly associated with impacted permanent mandibular third molar teeth. Moreover, they occur mainly in patients during the second and third decades of life.[1] Their bilateral presentation is considered rare and can be sometimes associated with syndromes, such as cleidocranial dysplasia and Maroteaux-Lamy syndrome.[4] De Biase et al[5] reported a case of bilateral mandibular cysts in an 8-year-old boy treated with cyclosporin (**CsA**) after

Figure 5. Clinical and radiographic follow-up showing complete eruption of the mandibular first molars without signs of recurrence.

renal transplantation. The genesis of mandibular cysts might be associated with the combined post-transplantation use of CsA and a calcium channel blocker. The occurrence of DCs in childhood can also be considered uncommon.[6] In the present case, the patient was 5 years old, and there were no associated syndromes.

Most previous articles reporting bilateral DCs were cases associated with impacted third molars. To our knowledge, there are only about 24 cases of bilateral dentigerous cysts reported in the dental literature. Very few reports showed the occurrence in other sites, such as central incisors, second premolar, and first molars.[3,7-12] Nevertheless, only 8 cases have been shown to be associated with mandibular first molars, as in the present case. Table 1 summarizes the data of bilateral mandibular first molars reported in the dental literature.

It is plausible that bilateral dentigerous cysts are either under-recognized or under-reported, particularly in the third molar area, because not all pericoronal tissues associated with these impacted teeth are sent for histopathological analysis. In our opinion, as the noneruption of first molar teeth is not a common finding, most patients would probably ask their dentist for diagnosis and treatment. This may reflect the true rarity of this condition associated with permanent first molars.

An interesting aspect observed in the present case was how fast the second cyst developed on the right side. When the image exams were obtained for evaluation of the cyst on the left side (first lesion), it was not visible—being detectable only after 2 months, when the patient complained of a swelling in the region. Although the second lesion had developed in only 2 months and the presence of bilateral DCs was rare, the radiographic findings were very suggestive of a DC. Thus, the hypothesis of diagnosis for the second lesion was also DC.

There are 2 common surgical treatments for this condition: (1) marsupialization; and (2) enucleation. Marsupialization is indicated in larger dentigerous cysts, allows lesion decompression, and decreases the cystic cavity. Sometimes, later enucleation can be performed with a less extensive surgical procedure. Marsupialization and enucleation were the treatments of choice for our patient, which agrees with other authors.[17,18] There are few reports in the dental literature reporting spontaneous regression of dentigerous cysts, although they were not confirmed histopathologically.[19,20]

In summary, bilateral dentigerous cysts affecting the permanent first molars are extremely rare and can arise at different times (metachronous). In addition, the presence of bilateral dentigerous cysts does not mean that a patient is syndromic. Correct investigation should be conducted, however, because this possibility does exist.

Table 1. Summary of Previously Reported Bilateral Mandibular First Molars

Authors	Year	Gender	Race*	Age (ys)	Treatment
De Biase et al[5]	2001	Male	C	8	Enucleation
Sands and Tocchio[13]	1998	Female	N/A	3	Enucleation
Carr et al[7]	1996	Male	N/A	7	Enucleation
O'Neil et al[6]	1989	Male	Bl	5	Enucleation
Eidinger[14]	1989	Male	C	15	Enucleation
Swerdloff et al[15]	1980	Female	C	7	Enucleation
Stanback[10]	1970	Male	N/A	9	Enucleation
Tam JC[16]	1955	Male	N/A	7	Enucleation

**** Bl=black; C=Caucasian; N/A=not available.***

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