

Bilateral dentigerous cyst treated by marsupialization: A case report

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ABSTRACT

Introduction: Dentigerous cysts are the second most common kind of cyst lesion that develops on the jaw and they have been associated to unerupted, impacted or unerupted tooth. There are just a few reports on bilateral or multiple dentigerous cysts in non-syndromic patients. Their traditional treatment is enucleation, and an alternative treatment is rarely chosen. **Case Report:** The clinical case of an 11-year-old patient with a bilateral cyst not associated with any syndromes is reported hereby. He was treated by the alternative marsupialization technique and with radiographic follow-up for 24 months, showing no recurrence. **Conclusion:** The marsupialization is an excellent alternative treatment for the preservation of permanent teeth.

Keywords: Bilateral dentigerous cyst, Cyst on the jaw, Dentigerous cyst, Marsupialization

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INTRODUCTION

Odontogenic cysts are a group of jaw cysts that are formed from tissues involved in odontogenesis and results because of the enlargement of the follicular space of the whole or part of the crown of an impacted or unerupted tooth and is attached to the neck of the tooth. Based on their pathogenesis, they are classified as either developmental or inflammatory lesions [1]. Their frequency is estimated to be 1.44 cysts out of 100 unerupted teeth [2].

Dentigerous cysts are the second most common odontogenic originated lesion, located on the jaw in 75% of the cases, permanent, retained, supernumerary, odontomas and, exceptionally, deciduous teeth [2–4]. In most of the cases, the lesions are unique however there have been reports in literature presenting cases of multiple or bilateral dentigerous cysts, not associated to any kind of syndrome, being extremely rare [5, 6]; whereas the presence of bilateral or multiple cysts is reported to be associated to syndromes like cleidocranial dysplasia, basal cell syndrome and mucopolysaccharidosis [7, 8]. The teeth most frequently related to this kind of lesions is

the mandibular third molar, the maxillary canine and the mandibular premolars come second [9].

Dentigerous cysts are usually asymptomatic lesions, but when incidentally infected, they can cause pain [10]. They usually cause considerable increase in volume through the cortical bone expansion and delay in tooth eruption [11, 12].

In literature, cases of spontaneous regression of dentigerous cysts have been reported, but in most of them there is a lot of controversy on the likelihood of this kind of situations [13, 14].

The aggressive potential of dentigerous cysts influences the kind of therapeutic procedure to be used [15]. Lesions with a shorter diameter, mainly in young patients or children, are removed completely in order to prevent damage to the permanent tooth and benefit its eruption while larger lesions are treated by means of marsupialization or decompression [16, 17].

Due to their high development potential, dentigerous cysts may become extremely large before being diagnosed, it is therefore important to emphasize that if there is a chance to cause lesions to surrounding structures or impair the jaw (leaving it unable to receive functional loads that could cause a pathological fracture), it is recommendable to treat through the marsupialization surgical technique [9, 12, 17].

CASE REPORT

An 11-year-old male with no relevant pathological, caries lesions or symptomatology background was presented for orthopedic treatment at the Pediatric Dentistry Department of the Postgraduate Studies Division at the Dental School of National Autonomous University of Mexico.

During the clinical intraoral and extraoral examination, there were no signs of inflammation or evident impairment, two radiolucent areas can be seen in the orthopantomograph by the second mandibular premolars, with a presumptive diagnosis of dentigerous cysts, measuring 1.9x2.0 cm on their right side, and 2.8x2.2 cm on their left side, approximately (Figures 1 and 2).

The pathology and pediatric dentistry departments were consulted. The decision was to carry out a conservative treatment consisting in reducing the cysts by marsupialization due to their extension, in order to preserve the permanent teeth and minimize the risk of a mandibular fracture derived from the extraction of the involved tissue.

Once the treatment plan was established, under 2% mepivacaine local anesthesia, the deciduous teeth were extracted, which presented a previous pulp treatment and restoration by chromium steel crowns in both quadrants. To perform the marsupialization procedure through the extraction sockets, after proper hemostasis, a surgical fenestration in the membrane of the cysts was created

using a #2 blade. The cyst membrane was sutured to the oral mucosa creating a window to maintain a continuity between the cyst and the oral cavity allowing the evacuation of the cyst content to relieve the intracystic pressure. The cyst cavity was packed with sterile gauze to achieve hemostasis and to prevent hematoma formation. The patient was advised to irrigate the cyst spaces with sterile saline three times a day for seven days. After the marsupialization procedure, the patient has recalled every six months during a two-year period for clinical and radiographic examination reporting no relevant complications in the treated sites. The eruption of the permanent teeth was visible in the second appointment.

The pathology service received fragments of non-keratinized squamous epithelial cells with edema between cells in a stroma from fibrous compact and loose connective tissue, with severe chronic diffuse infiltration, with recent bleeding areas, bacterial colonies, and bone spicules in the left region sample. According to the microscopic description of the right side sample, it was comprised by non-keratinized squamous epithelial cell strips, arch-like shaped varying in thickness and edema between cells; as well as irregular fragments of fibrous compact and loose connective tissue, richly vascularized with a severe chronic inflammatory infiltration (Figure 3).

By using clinical and radiographic controls six-month (Figure 4), the evolution of the lesions can be seen. Therefore, we can show the lesions shrinking, as well as the eruption of the two involved teeth on follow-up after a 24-month treatment (Figure 5).

DISCUSSION

Dentigerous cysts are the second most common type of cyst lesion that develops on the jaw and they have been

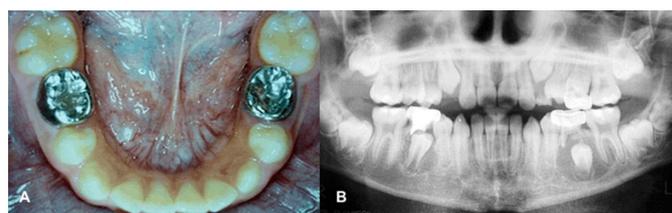


Figure 1: (A) Occlusal photograph of the jaw with no evident signs of lesion, and (B) Panoramic radiograph showing two radiolucent lesions associated to teeth no. 35 and 45.

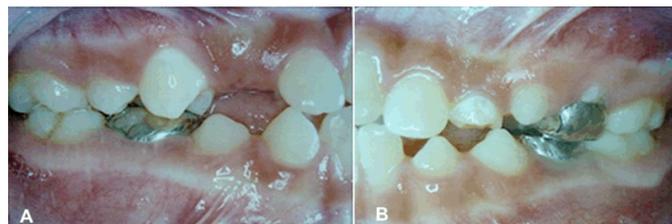


Figure 2: Left and right Intraoral clinical photographs. (A, B) Healthy tissues with no cortical expansion, inflammation, or swelling.

associated to unerupted teeth, mainly. Additionally, there are just a few reports on bilateral or multiple dentigerous cysts in non-syndromic patients. Maroteaux-Lamy syndrome is a mucopolysaccharidosis (MPS), where there is a lack of N-acetyl-4-sulfatasa, among the oral clinical characteristics, patients present malocclusions, gingival hiperplasia, dentigerous cysts, defective mandibular condyle and retained teeth [7]. Cleidocranial dysplasia or cleidocranial dysostosis, is another condition related to retained teeth and dentigerous cysts, besides the possibility of odontomas and/or supernumerary teeth; although its etiology remains unclear, it is associated to a mutation of chromosome 6p21, affecting protein RUNX2, which acts as a transcriptional factor in the differentiation of dental cells and osteoblasts, as well as tooth and bone formation [9].

Gorlin–Goltz syndrome is a genetic disorder, with tendency to develop odontogenic keratocyst tumors (OKTs). In 2003, WHO/IARC said that it is important to perform supplementary tests such as chest, cranial and maxillary radiographies. It is very important that the dentist or pediatric dentist identifies the different impairment types present, and always tries to refer to the oral pathologist in order to discard all the different

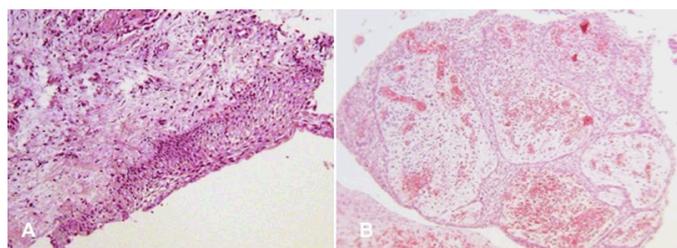


Figure 3: (A) An epithelial lining on a stroma of a fibrous connective tissue, (B) Rainbow-like epithelial pattern embedded in a highly vascularized fibrous connective tissue.



Figure 4: (A) Six-month follow-up occlusal photograph showing the eruption of permanent premolars, and (B) Panoramic radiograph showing healing of alveolar bone.

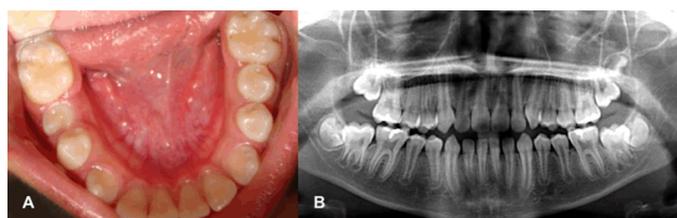


Figure 5: Two year follow-up clinical (A) Radiographic images, (B) A healthy alveolar bone and normal eruption of permanent premolars.

syndromes and/or illnesses, since there are scarce case reports not associated to illnesses or syndromes in literature; there are not more than twenty cases in total [18].

A diagnosis dentigerous cyst was given by the clinic, mainly based on radiographic imaging. A cyst can rarely be seen by plain eyes, unless it spreads to the cortical bone; and, as mentioned above, unless there is a secondary infection in which case the patient has painful symptoms. A characteristic to care for is the absence of erupting teeth, which must be verified through imaging. First, the patient will be required for periapical radiographies and orthopantomographies. A well-defined radiolucent area with a cortical around the crown of an unerupted tooth can be seen in the radiographies [9]. The diameter of the lesion is an important consideration; if it is less than 2 cm long, then it will be rather unilocular; but if the cyst lesion is not detected on time, the cyst will grow and may become multilocular, making diagnosis more difficult, even associating it to more aggressive lesions like an ameloblastoma [15].

The traditional treatment is by enucleation, carried out through an osteotomy, and the removal of the impacted tooth, as well as of the lesion. This kind of treatments often turns risky, weaken the corticals and promote fractures on the maxillary bone. Marsupialization is a less invasive treatment, although one of its disadvantages is some pathological tissue is left; remote control of the patient becomes therefore important to ensure long term success [12, 18]. We are hereby reporting a case of a bilateral dentigerous cyst in a non-syndromic patient who was treated by marsupialization of both lesions and a two-year follow up without any relapse of the lesion.

CONCLUSION

It is always important to offer a conservative treatment for this kind of large cysts since it does not threaten mandibular integrity with potential complications as pathological fractures due to a weakened mandibular bone, additionally to obtaining a progressive and better organized bone building with higher radiopacity of the lesion region. That is why marsupialization is an alternative treatment for the preservation of permanent teeth.

Author Contributions

Villar Rodriguez Eunice – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Final approval of the version to be published
Quezada Rivera Daniel – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Campos Ibarra Paola – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Beltran Lara Emilio – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Tenorio Rocha Fernando – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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