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EARLY VIEW

65 **TITLE:** Bilateral dentigerous cyst treated by marsupialization: Case report

66

67 **ABSTRACT**

68

69 **Introduction**

70 Dentigerous cysts are the second most common kind of cyst lesion that develops on
71 the jaw and they have been associated to unerupted, impacted or unerupted tooth.

72 There are just a few reports on bilateral or multiple dentigerous cysts in non-
73 syndromic patients. Their traditional treatment is enucleation, and an alternative
74 treatment is rarely chosen.

75

76 **Case Report**

77 The clinical case of an 11-year-old patient with a bilateral cyst not associated with
78 any syndromes is reported hereby. He was treated by the alternative
79 marsupialization technique and with radiographic follow up for 24 months, showing
80 no recurrence.

81

82 **Conclusion**

83 The marsupialization is an excellent alternative treatment for the preservation of
84 permanent teeth.

85

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

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98

99 **INTRODUCTION**

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

106 Dentigerous cysts are the second most common odontogenic originated lesion,
107 located on the jaw in 75 % of the cases, permanent, retained, supernumerary,
108 odontomas and, exceptionally, deciduous teeth [2-4]. In most of the cases, the
109 lesions are unique however there have been reports in literature presenting cases of
110 multiple or bilateral dentigerous cysts, not associated to any kind of syndrome, being
111 extremely rare [5,6]; whereas the presence of bilateral or multiple cysts is reported
112 to be associated to syndromes like cleidocranial dysplasia, basal cell syndrome and
113 mucopolysaccharidosis [7,8]. The teeth most frequently related to this kind of lesions
114 is the mandibular third molar, the maxillary canine and the mandibular premolars
115 come second [9].

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

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

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

127 Due to their high development potential, dentigerous cysts may become extremely
128 large before being diagnosed, it is therefore important to emphasize that if there is a

129 chance to cause lesions to surrounding structures or impair the jaw (leaving it unable
130 to receive functional loads that could cause a pathological fracture), it is
131 recommendable to treat through the marsupialization surgical technique [9,12,17].

132

133 **Case Report**

134 Eleven-year-old male patient with no relevant pathological, caries lesions or
135 symptomatology background. Admitted for orthopedic treatment at the Pediatric
136 Dentistry Department of the Postgraduate Studies Division at the Dental School of
137 National Autonomous University of Mexico.

138 During the clinical intraoral and extraoral examination, there were no signs of
139 inflammation or evident impairment, two radiolucent areas can be seen in the
140 ortopantomograph by the second mandibular premolars, with a presumptive
141 diagnosis of dentigerous cysts, measuring 1.9 x 2.0 cm on their right side, and 2.8 x
142 2.2 cm on their left side, approximately (Figure1 & 2).

143 The Pathology and Pediatric Dentistry departments were consulted. The decision
144 was to carry out a conservative treatment consisting in reducing the cysts by
145 marsupialization due to their extension, in order to preserve the permanent teeth and
146 minimize the risk of a mandibular fracture derived from the extraction of the involved
147 tissue.

148 Once the treatment plan was established, under 2 % mepivacaine local anesthesia,
149 the deciduous teeth were extracted, which presented a previous pulp treatment and
150 restoration by chromium steel crowns in both quadrants. To perform the
151 marsupialization procedure through the extraction sockets, after proper hemostasis,
152 a surgical fenestration in the membrane of the cysts was created using a #2 blade.
153 The cyst membrane was sutured to the oral mucosa creating a window to maintain a
154 continuity between the cyst and the oral cavity allowing the evacuation of the cyst
155 content to relieve the intracystic pressure. The cyst cavity was packed with sterile
156 gauze to achieve hemostasis and to prevent hematoma formation. The patient was
157 advised to irrigate the cyst spaces with sterile saline three times a day for seven
158 days. After the marsupialization procedure, the patient is recalled every six months
159 during a two year period for clinical and radiographic examination reporting no

160 relevant complications in the treated sites. The eruption of the permanent teeth was
161 visible in the second appointment

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

171 By using clinical and radiographic controls six-month (Figure 4), the evolution of the
172 lesions can be seen, therefore we can show the lesions shrinking, as well as the
173 eruption of the two involved teeth, after a 24-month after treatment follow up (Figure
174 5).

175

176 **DISCUSSION**

177 Dentigerous cysts are the second most common type of cyst lesion that develops on
178 the jaw and they have been associated to unerupted teeth, mainly. Additionally,
179 there are just a few reports on bilateral or multiple dentigerous cysts in non-
180 syndromic patients. Maroteaux-Lamy syndrome is a mucopolysaccharidosis (MPS),
181 where there is a lack of N-acetyl-4-sulfatasa, among the oral clinical characteristics,
182 patients present malocclusions, gingival hiperplasia, dentigerous cysts, defective
183 mandibular condyle and retained teeth [7]. Cleidocranial dysplasia or cleidocranial
184 dysostosis, is another condition related to retained teeth and dentigerous cysts,
185 besides the possibility of odontomas and / or supernumerary teeth; although its
186 etiology remains unclear, it is associated to a mutation of chromosome 6p21,
187 affecting protein RUNX2, which acts as a transcriptional factor in the differentiation of
188 dental cells and osteoblasts, as well as tooth and bone formation [9].

189 Gorlin-Goltz syndrome is a genetic disorder, with tendency to develop odontogenic
190 ketarocyst tumors (OKTs). In 2003, WHO/IARC said it is important to perform
191 supplementary tests such as chest, cranial and maxillary radiographies. It is very

192 important that the dentist or pediatric dentist identifies the different impairment types
193 present, and always tries to refer to the oral pathologist in order to discard all the
194 different syndromes and / or illnesses, since there are scarce case reports not
195 associated to illnesses or syndromes in the literature – there are no more than
196 twenty cases in total [18].

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

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

218

219 **CONCLUSION**

220 It is always important to offer a conservative treatment for this kind of large cysts
221 since it does not threaten mandibular integrity with potential complications as
222 pathological fractures due to a weakened mandibular bone, additionally to obtaining
223 a progressive and better organized bone building with higher radiopacity of the lesion

224 region. That is why marsupialization is an alternative treatment for the preservation
225 of permanent teeth.

226

227 **CONFLICT OF INTEREST**

228 There was no conflict of interest

229

230 **AUTHOR'S CONTRIBUTIONS**

231 Villar Rodriguez Eunice

232 Group1 – substantial contributions to conception and design, acquisition of data.

233 Group 2 - drafting the article.

234 Group 3 - Final approval of the version to be published

235

236 Quezada Rivera Daniel M.Sc

237 Group1 - substantial contributions to conception and design, acquisition of data.

238 Group 2 - drafting the article, revising it critically for important intellectual content

239 Group 3 - Final approval of the version to be published

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241 Campos Ibarra Paola M.Sc

242 Group1 - substantial contributions to conception and design, analysis and
243 interpretation of data.

244 Group 2 – drafting the article, revising it critically for important intellectual content

245 Group 3 - Final approval of the version to be published

246 Beltran Lara Emilio M.Sc

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248 interpretation of data.

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252 Tenorio Rocha Fernando

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254 interpretation of data

255 Group 2 - drafting the article, revising it critically for important intellectual content

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307 308 **FIGURE LEGENDS**

309
310 Figure 1: Occlusal photograph of the jaw with no evident signs of lesion (A).
311 Panoramic radiograph showing two radiolucent lesions associated to teeth no. 35
312 and 45 (B).

313
314 Figure 2: Left and right Intraoral clinical photographs (A & B) showing healthy tissues
315 with no cortical expansion, inflammation or swelling.

316
317 Figure 3: Photomicrographs showing an epithelial lining on a stroma of a fibrous
318 connective tissue (A), rainbow-like epithelial pattern embedded in a highly
319 vascularized fibrous connective tissue (B).

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

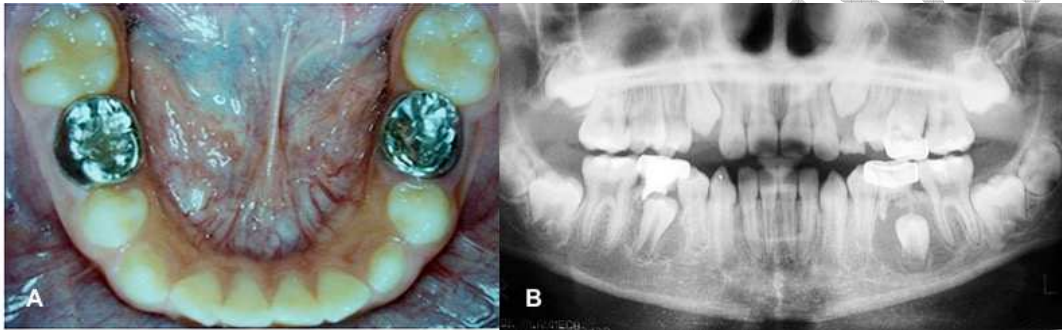
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324 Figure 5: Two year follow-up clinical (A) and radiographic images (B) showing a
325 healthy alveolar bone and normal eruption of permanent premolars.

326

327 FIGURES

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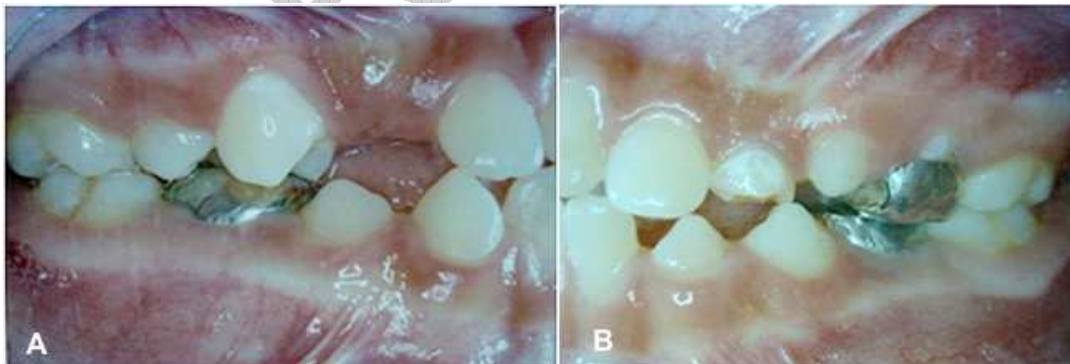


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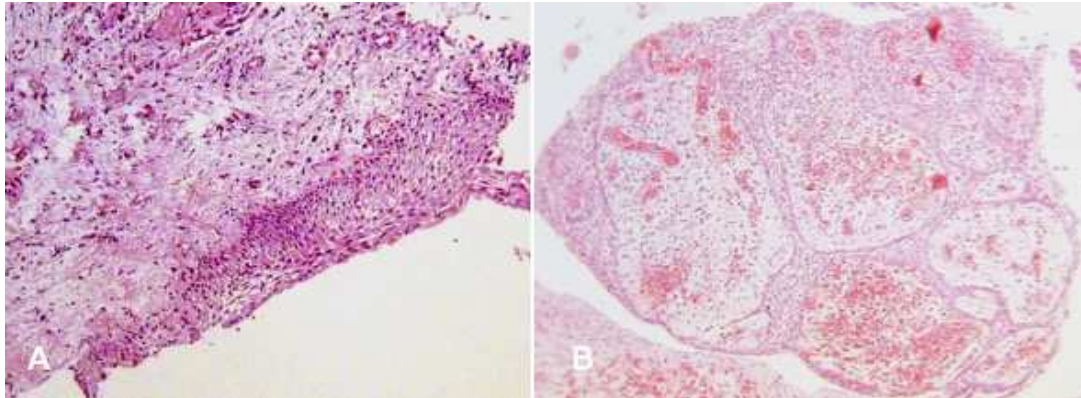


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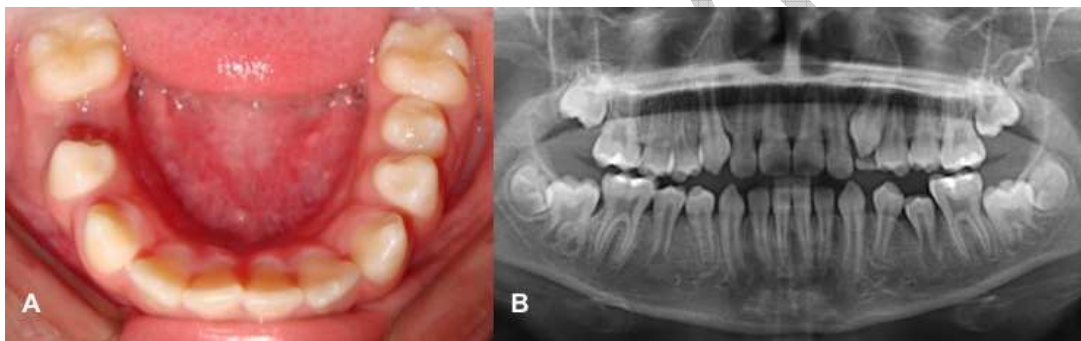


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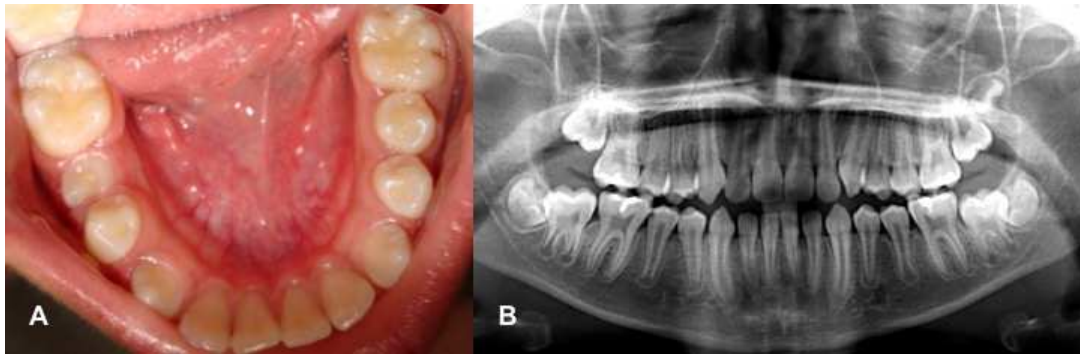
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