Conservative Management of a Large Dentigerous Cyst in a 6-year-old Girl: A Case Report

Elif Bozdogan, DDS *Burak Cankaya, DDS, PhD*
Koray Gencay, DDS, PhD *Oya Aktoren, DDS, PhD*

ABSTRACT

Dentigerous cysts are benign odontogenic cysts that are associated with the crowns of permanent teeth. They are usually characterized as unilocular radiolucent lesions and are rarely seen during childhood. The purpose of this article was to report the case of a 6-year-old girl with a dentigerous cyst associated with the tooth buds of premolars. The therapeutic approach included extraction of the primary molar involved and marsupialization of the lesion. After 21 months of follow-up, spontaneous eruptions of the impacted premolars were noticed. In conclusion, marsupialization might be the first treatment option for conservative management of dentigerous cysts in children.

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KEYWORDS: DENTIGEROUS CYST, MARSUPIALIZATION, ERUPTION OF TEETH

Dentigerous cysts (**DCs**) are the benign odontogenic cysts associated with the crowns of permanent teeth.[1,2] They enclose the crown of nerupted, impacted permanent teeth or supernumerary teeth at the cementoenamel junction, expand by growth of the follicle, and inhibit the eruption of the cyst-associated tooth. DCs are the second most common odontogenic cysts after radicular cysts and account for approximately 24% of all epithelium-lined jaw cysts.[3,4] The teeth most often involved are the mandibular third molars, maxillary canines, and mandibular premolars.[1]

DCs are generally observed in the second and third decades of life and are rarely seen during childhood.[2,3] In most cases, they are painless and asypmtomatic and usually detected by routine radiographic examination. Swelling, sensitivity, teeth displacement, and delayed tooth eruption, however, might be seen in severe cases. Radiographically it is usually characterized as a unilocular radiolucent lesion, with well-defined sclerotic margins, enclosing the crown of an unerupted/impacted tooth.[5] Histopathologically, a DC reveals a nonkeratinized stratified squamous epithelium consisting of mucosebaceous, ciliated, and rarely sebaceous cells.[6]

The methods for treating a DC are enucleation and marsupialization, which is a conservative management recommended mostly for cases associated with displaced and unerupted teeth.[7-9]

The purpose of this report was to describe the conservative management of a dentigerous cyst that dislocated the buds of mandibular premolars in a 6-year-old girl.

CASE REPORT

A nonsyndromic, 6-year 11-month-old girl was referred to the Department of Pediatric Dentistry, Istanbul University, Istanbul, Turkey, with the complaint of painless facial swelling in her left posterior mandibular region that began 8 weeks earlier without any signs of acute or chronic infection. An intraoral examination revealed swelling in the region of the primary mandibular left second molar, which received a pulpotomy 2 years ago. Additionally, the primary mandibular first molar was extracted due to a periapical infection observed before treatment of the primary second molar (Figure 1A).

A panaromic examination showed a large, circular, well-defined uniocular area from the mandibular left canine tooth to the mandibular first molar tooth (Figure 2A).

Dr. Bozdogan is post graduate student, Dr. Gencay is professor, and Dr. Aktoren is Professor, all in the Deparment of Pedodontics, and Dr. Cankaya is research assistant at Department of Oral Surgery, all in the Faculty of Dentistry, Istanbul University, Istanbul, Turkey.

Correspond with Dr. Bozdogan at dt.ellie@hotmail.com

A computed tomography (**CT**) image was obtained due to the large size of the lesion. CT imaging showed a well-defined lesion in the mandibular left region surrounding the unerupted second premolar's crown (Figure 3A) and partially surrounding the unerupted first premolar's crown (Figure 3B). The root apices of these teeth were open and the teeth were displaced. No signs of root resorption were present in the adjacent teeth.

Clinical and radiographic examination findings displayed the lesion as a DC. The primary mandibular left second molar was extracted, and the marsupialization technique was performed in the oral surgery department clinic of Istanbul University. A biopsy tissue sample was taken from the access window of marsupialization for patho-logic evaluation; and then a silicon tube was installed to drain the intracystic pressure. Histopathological analysis showed a fibrous cystic wall with focal mononuclear inflammatory cells lined by a thin, strafied, squamous, and nonkeratinized epithelium, which confirmed the diagnosis of a DC (Figure 4).

After marsupialization, a space maintaining appliance was applied and followed-up until the permanent teeth erupted. The silicon tube was replaced bimonthly for 6 months postoperately. The 9-month follow-up visit showed that the radiographic radiolucency was decreased, and the teeth were erupting spontaneously (Figure 2B). After the 15-month follow-up, the impacted teeth had partially erupted (Figure 2C); the clinical and radiographical findings after 21 months had shown that the first premolar reached the alveolar mucosa and the second premolar had erupted completely (Figure 1B and 2D).

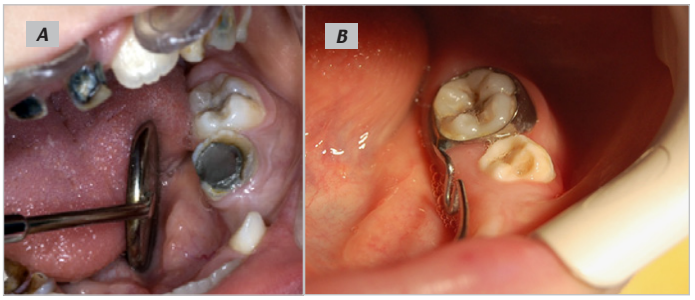

Figure 1. Intraoral photographs. (A) Intraoral view showing buccal expansion in the region of the primary mandibular left second molar, which received a pulpotomy 2 years ago. (B) Postoperative view of the second premolar, which erupted after 21 months.

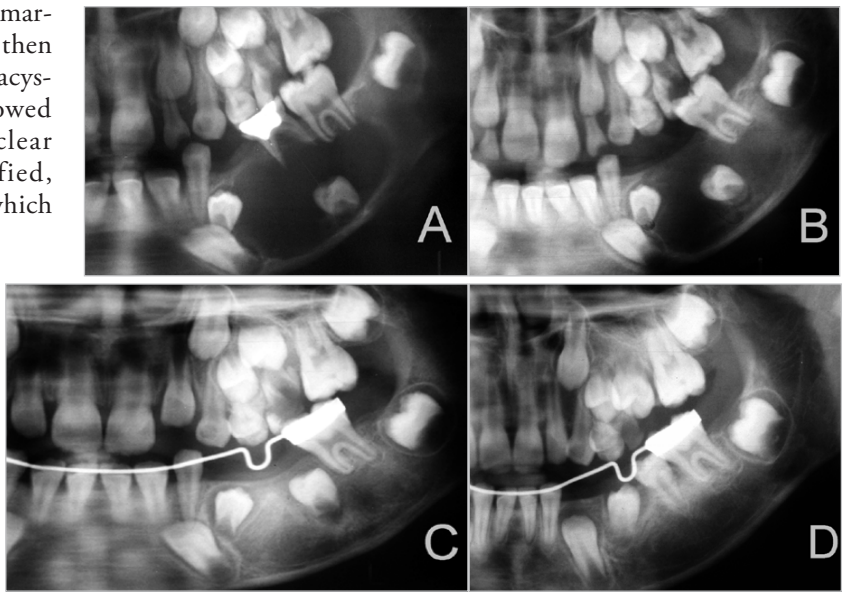

Figure 2. Panaromic radiographs. (A) Radiograph view demonstrating the dentigerous cyst related to the unerupted second premolar. (B) Postoperative radiographic view after 3 months displaying the decreased radiolucency around the second premolar. (C) Postoperative view after 6 months showing the displacement of the second premolar toward the occlusal ridge. (D) Radiographic view showing the eruption of the second premolar after 21 months.

DISCUSSION

DCs are the most common of all follicular cysts and are always associated with an impacted or unerupted tooth's crown. It may arise from the accumulation of fluid between the enamel epithelium and the crown of the permanent tooth germ or from remnants of the odontogenic epithelium.[10] It is also stated that a DC can be caused by inflammation of the periapical tissues due to necrosis or a periapical infection originating from the primary predecessor tooth, which could stimulate the developing tooth germ follicle.[11]

Benn and Altini categorized DCs as developmental and inflammatory cysts. Developmental DCs typically occur in mature teeth, usually as a result of impaction, and predominantly involve the mandibular third molars. They are generally discovered on routine radiographs in the late second or third decades of life. Inflammatory DCs could occur in immature permanent teeth as a result of inflammation from a nonvital primary tooth. These are diagnosed in the first and early part of the second decade either on routine radiographic examination or when the patient complains of a painless swelling. In this case, the patient's age and the findings reveal the cyst as an inflammatory DC; the necrosis in the primary mandibular second molar or the periapical infection in the extracted primary mandibular first molar might have stimulated the development of a DC.[12]

Patients with radicular cysts do not experience pain unless acute inflamamatory exacerbation is present. If the cyst enlarges and grows rapidly, symptoms such as buccal expansion, mild sensitivity, tooth mobility, and displacement of succedaneous teeth may occur. Koca et al., examined 35 DC patients and determined that the children's main complaint was buccal expansion in 70% and pain in 5%, while 25% had no symptoms.[13] In this case, painless facial swelling in the left posterior mandibular region was the main complaint of the patient at the referral time to the university clinic.

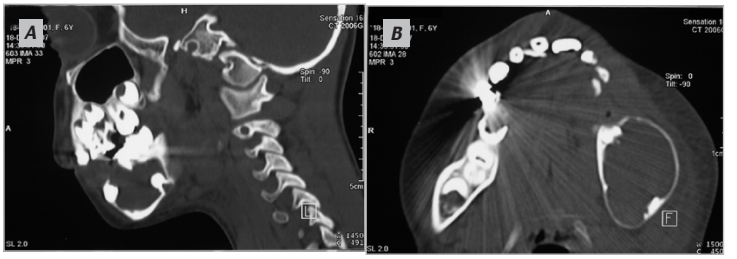

Figure 3. Computed tomography images. (A) Vertical CT image showing the well-defined lesion in the mandibular left region. (B) Axial CT image displaying the lesion surrounding the first and second premolar teeth.

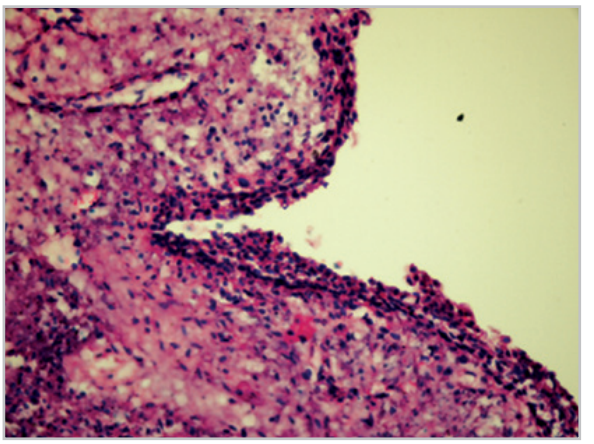

Figure 4. Histopathological view showing the cystic lesion lined by nonkeratinized squamous epithelium (original magnification 400x).

On radiographic examination, a DC can appear as a unilocular lucent cyst of varying sizes with well-defined sclorotic borders associated with the crown of an unerupted premolar.[14] Considering a normal follicular space of 3 to 4 mm, a DC can be suspected when the radiographic image of the follicular space is larger than 5 mm in diameter. In extensive cases, radiographs alone may not be sufficient to show the full extent of the lesion, and CT imaging may be necessary. CT imaging gives exact information about the lesion's size, content, and origin.[15] Koca et al., preferred panoramic radiographs for imaging procedures in all cases, while an additional CT scan was required for imaging the large lesions extending to the nasal cavity or orbital or pterygomaxillar space.[13] In this case, an additional CT scan was also required for assessing the extent of the cystic lesion, and panaromic radiographs were preferred for the periodic follow-up visits.

DCs are usually single lesions. Bilateral and multiple cysts have been reported in cases with syndromes such as basal cell nevus syndrome, mucopolysaccharidosis, and cleidocranial dysplasia. Bilateral mandibular DCs have also been reported after prolonged use of cyclosporine A and calcium channel blockers. The few reported bilateral DCs in nonsyndromic patients have been located in the maxilla and mandible.[3,14-16]

The differential diagnosis of a DC should include: radicular cysts; odontogenic keratocysts; ameloblastomas; odontogenic fibromyxoma; odontomas; and cementomas. Radicular cysts are odontogenic cysts that develop from a periapical granuloma in teeth with carious lesions associated with pulp necrosis. Odontogenic keratocysts are often multiocular and most commonly located in the mandible's body or ramus; these cysts are lined by stratified sqamous keratized epithelium. Ameloblastomas are the most common radiolucent benign odontogenic tumour that may be unilocular or multilocular. It may cause expansion and destruction of the maxilla and mandible, and histological findings may show prolifarating epithelial structures resembling "enamel organs." Odontogenic fibromyxoma usually has multiple radiolucent areas of varying size and bony septations. Odontomas are lytic lesions more often accompanied by amorphousn calcification.[1,14] Therefore, histopathologic analysis is necessary in most cases to confirm the diagnostic hypothesis. In this case report, the definite diagnosis also was based on pathologic analysis (Figure 4).

Treatment of a DC depends on the size and location of the cyst, patient age, affected dentition, and cyst's relationship with surrounding vital structures. Marsupialization or the decompression technique is indicated for children and adolescents to preserve the teeth and promote the eruption of the dislocated tooth associated with the cystic lesion. Marsupialization, by relieving intracystic pressure through the creation of an accessory cavity, maintains the impacted tooth in the cystic cavity and promotes its eruption.[6-8]

Silva et al., preferred the marsupalization technique in a 10-year-old boy with a DC surrounding his unerupted first premolars. The authors observed that, after 3 months, the radiographic radiolucency was decreased and the premolar was erupting without any other therapy; after 30 months, the premolar had erupted completely.[17] Boj et al., evaulated conservative treatment of a DC in a 6-year-old boy with an ER, Cr: YSGG laser. The authors reported that the antibacterial and anti-inflammatory properties attributed to this laser may improve the postoperative prognosis, and the erbium laser provides the advantage of a good operational field of vision as a result of its coagulation effect.[18]

In this case report, marsupialization was the treatment of choice vs enucleation of the cystic lesion since the patient was young, the lesion occupied a large mandibular area, and the cyst had led to the displacement of premolars.

The treatment planning was quite simple and atraumatic; however, follow-up care was required during the treatment period. Postoperatively, all clinical and radiographic views showed normal features, new bone formation, and spontaneous relocation and eruption of premolars in their correct position. Since immature teeth have an optimal eruption potential and children have a much greater bone regeneration capacity than adults, the prognosis of conservative surgical techniques in young children is usually successful.

Sun et al., practiced the marsupialization technique and used a removable appliance with a resin projection to decompress the cystic lesion and prevent both food debris from entering the cystic cavity and the formation of fibrous healing tissues which could impair the eruption of permanent teeth.[19] Koca et al., preferred an acrylic obturator to drain the cystic pressure and to preserve the dentition while the cystic lesions were shrinking in size. They employed conservative treatment and reported that 24% of the cases needed orthodontics to bring the unerupted tooth into the arch. In 34% of the cases, meanwhile, the permanent tooth erupted successfully into the alveolar arch without orthodontic treatment.[13]

Hayasaki et al., practiced the marsupialization technique in a 4-year, 2-month-old girl due to a DC around her mandibular right second premolar. The authors extracted a primary mandibular right second molar and applied a removable appliance with a resin projection inserted into the socket of the extracted tooth and the cystic cavity. The authors reported, however, that this tooth had to be extracted since it had not developed normally in postoperative follow-ups.[20] In this case, the marsupialization technique and a watchful waiting period for the spontaneous eruption of the unerupted tooth, without orthodontic therapy, had succesful outcomes. A silicon tube was installed to reduce the intracystic pressure and was replaced bimonthly for 6 months postoperately; after marsupialization, a space maintainer was applied and followed-up until the permanent teeth erupted.

Hyomoto et al., Fujii et al., and Yahara et al. examined groups of DC patients and divided them into 2 groups:

1. an erupted group, in which a cyst-associated tooth had erupted spontaneously after marsupialization; and
2. a nonerupted group, in which either the tooth in the cyst area had not erupted and the patient had undergone orthodontic traction or the cyst had been removed by cysteoctomy.

The authors determined that, in the erupted group, the average age was significantly younger than in the nonerupted group.[21-23] In this case report, the patient's age also was very young, and the DC-impacted tooth erupted successfully without orthodontic treatment, in spite of the cyst's enlargement.

Yahara et al. evaluated 21 patients with DCs associated with mandibular premolars after marsupialization by dental radiographs and defined the eruption either as: a half eruption (score=0.5) when the upper tip of the cusp of the impacted DC-associated tooth erupted to the line that passes the cementoenamel junction of the adjacent teeth; or as a complete eruption (score=1.0) when the DC-associated tooth reached the occlusal plane. They concluded that approximately 71% of teeth had shown half eruption in 3 months and complete eruption within 10 months.[23] In this case report, the mandibular premolar associated with the DC after marsupialization had shown half eruption within 9 months and complete eruption within 21 months due to the lesion's large size.

In conclusion, conservative surgical treatment modalities may be the first choice of treatment for dentigerous cysts in children to preserve and promote the eruption of permanent teeth.

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