Dentigerous Cyst Associated with a Formocresol Pulpotomized Deciduous Molar

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Abstract

This report presents a case of dentigerous cyst associated with a formocresol pulpotomized deciduous molar detected during routine examination. Dentigerous cyst is an epithelial-lined developmental cavity that encloses the crown of an unerupted tooth at the cementoenamel junction. The present case describes a 9-year-old girl sent to the dental clinic by her dentist, who had accidentally discovered in the panoramic radiograph a single, unicocular, well-defined, radiolucent area enclosing the second left unerupted mandibular premolar. The second left primary molar had been pulpotomized 2 years before and buccal swelling without redness occurred near the tooth, evidencing bone expansion. Surgical treatment was carried out, the tooth was extracted, and a cystectomy was performed under local anesthesia in the dental office. The histological study confirmed the suspected diagnosis of dentigerous cyst. The relation between pulpotomy and dentigerous cysts is discussed. (J Endod 2007;33:488–492)

Key Words

Dentigerous cyst, primary molar, pulpotomy, tooth impacted

Pulpotomy is considered a safe procedure, although side effects such as malposition, delay in eruption, enamel defects, discoloration, cyst formation, and damage to the permanent dentition have been reported (1–3). A dentigerous cyst can form in the periapical region after pulpotomy (4, 5) caused by an alteration of the reduced enamel epithelium (after completion of amelogenesis), which results in fluid accumulation between the epithelium and the tooth crown (6). The persistent and prolonged inflammation may cause chronic irritation to the dental sac of the unerupted tooth, which in turn leads to the development of a dentigerous cyst (7). Histological evidence for this process has been found (8), concluding that inflammatory exudate is induced by infection that spreads to the dental follicle, causing separation of the reduced enamel epithelium from the enamel. This would be true for immature permanent teeth found under necrotic or pulp-treated primary teeth (9). Inflammation and prolonged irritation are apparent causes of, and the trigger for, proliferation of the epithelial rests (10). This explains why cysts are found in conjunction with necrotic or pulp-treated teeth in the primary dentition (9–11). Periapical cysts described in association with pulpotomized primary molars seem to show specific clinical features: large size, rapid growth, buccal expansion, and displacement of succedaneous teeth (5, 11). Grundy et al. (9) recorded the time lapse between the pulpotomy and the detection of buccal bone expansion; it ranged from 5 months to 3 years, the average being 20 months.

A dentigerous cyst is an epithelial-lined developmental cavity that encloses the crown of an unerupted tooth at the cementoenamel junction, and thus it is also named “tooth-containing cyst.” The cyst arises from the separation of the follicle from the crown of an unerupted tooth and, although it may involve any tooth, the mandibular third molars are the most commonly affected, followed by maxillary canine, mandibular premolars, supernumerary teeth, and, rarely, the central incisor (12).

Dentigerous cysts are the second most commonly occurring odontogenic cysts after radicular cysts, accounting for roughly 24% of all true cysts in the jaws, representing the most common intrabony lesion of jaws in children. These cysts are frequently discovered when radiographs are taken to investigate a failure of tooth eruption, a missing tooth, or misalignment (13).

The cyst usually presents as a single lesion. However, bilateral and multiple cysts can occur in association with syndromes, including delayed dental eruption, such as cleidocranial dysplasia, nevus basocelular syndrome, and Hunter’s disease, and also in association with alterations of the PTCH gene (14), the superexpression of the P53 gene (15), and treatments with cyclosporins.

Epidemiological studies carried out by Mourghed (16) found a total prevalence of 0.8% for a dentigerous cyst and 3.6% in patients with an unerupted tooth.

Although cysts related to primary molars are frequent (10), they tend to be overlooked because they usually resolve after removal or exfoliation of the primary teeth. For the same reasons, biopsies are seldom taken. The only cases reported have been those in which the cyst became enlarged before detection and in which severe symptoms and side effects were already present (5, 10). The prevalence in the mandible and the relationship of cyst development to the second primary molars have been stressed (7, 10, 17). Two possible explanations are that the mandibular molars are associated with greater susceptibility to caries and more treatment and that a mandibular second primary molar is more closely associated with its successor’s follicle (5); such associ-
ation can more easily facilitate the spread of inflammation in comparison with other primary teeth (17).

This study reports a case of dentigerous cyst associated with formocresol pulpotomy detected during routine examination in a patient with transitional dentition.

**Case Reports**

A 9-year-old girl presented to the dental clinic sent by her dentist, who had accidentally discovered a lesion in the panoramic radiograph. Her medical and dental history was uneventful. She had suffered a dental injury. The four upper incisors were splinted with composite resin and did not show symptoms or signs of pulp involvement. Upper temporary molars have not yet exfoliated. A Chrom-nickel crown (3M, TM) was evident on tooth #75, pulpotomized 2 years before as related by the patient’s mother. Local buccal swelling without redness occurred near the tooth, evidencing bone expansion, although the gingiva and alveolar mucosa appeared normal (Fig. 1). A panoramic radiograph showed a mandibular radiolucent lesion (Fig. 2). A single, unilocular, well-defined, radiolucent area enclosed the second left mandibular premolar. A dentigerous cyst was initially diagnosed. To better study the radiolucent lesion, taking into account its proximity to the mentonian nerve, a mandibular TAC (Dentascan) was requested (13, 18) (Fig. 3). Thus, the great size of the lesion could be appraised not only in the mesiodistal aspect, but also in the buccolingual one. The swelling, the buccal cortical break, and the bone expansion were obvious. These features indicated a benign lesion.

The patient was informed about the nature of her lesion, a dentigerous cyst, and the low possibility (6% according to WHO) (19) of developing an ameloblastoma. The patient’s parents and her dentist asked for the total excision of the cyst. Before intervention the following medication was prescribed and supplied to the patient: (1) hydroxyzine dichlorhydrate (Atarax): 25 mg the night before the intervention and other 25 mg dose 1 hour before. (2) Amoxycillin/Clavulamic acid 500/125 mg (Augmentin): one dose the night before, continuing for 6 days after the intervention every 8 hours. (3) Ibuprofen (Dalsy): a 200 mg dose the night before and another one 1–2 hours before the intervention, continuing it with the same guideline as that of the recommended antibiotic.

With the suspected diagnosis of a dentigerous cyst associated with a retained premolar, surgical treatment was carried out, the tooth was extracted, and a cystectomy was performed under local anesthesia in the dental office. The buccal flaps were raised and the cysts enucleated together with the retained tooth (Fig. 4). The histological study confirmed the suspected diagnosis (Fig. 5). Microscopically, a dense fibrous connective tissue cyst wall was lined by remnants of dysplastic keratinizing stratified squamous odontogenic epithelium. The lining cells demonstrated a degree of nuclear and cellular pleomorphism and nuclear hyperchromatism. In focal areas, islands and strands of cells invaded the cyst wall. These invasive cords were composed of keratinizing squamous cells with occasional mitotic activity. The cords were separated by collagenous fibrous connective tissue and focal areas showed chronic inflammatory cell infiltrates. No evidence of nuclear palisading or reverse polarity of cells was noted within the islands. Healing was uneventful and, 1 week after the operation, the surgical sites showed good healing. After 3 months, there had been complete healing. There was also no evidence of recurrence of the cysts (Fig. 6).
The maxillofacial territory is the location of a great variety of cysts and neoplasias—whose identification can be very difficult. The most important of these lesions are maxillary cysts (12, 16). Cysts are pathological cavities with a defined wall of connective tissue and an epithelial carpet. Cysts, which are filled with a liquid, semiliquid, or gaseous content, grow centrifugally, spread out slowly, and are infiltrative (20).

The exact dentigerous cyst histological origin is unknown. Classically they have been defined as cysts of development of the dental follicle. Authors relate them to traumatic pathology or inflammatory processes. Thus, Shetty and Sandler (21) propose “having the eye in the grain,” when talking about pulpotomized deciduous teeth. The histology of the dentigerous cyst consists of dense fibrous connective tissue cyst wall lined by keratinizing, stratified squamous odontogenic epithelium.

Among the complications, secondary local infections, such as osteitis and osteomielitis, disturb the inferior alveolar nerve and/or lingual nerve (22), although remote infections such as mediastinitis (23) are possible. These circumstances, together with the possibility of ameloblastic transformation, support the surgical decision adopted in this case.

With respect to the neoplastic transformation, most of the odontogenic cysts show hyperplastic changes in the squamous epithelium, but they are caused by the chronic inflammation. The epithelial proliferations observed in the cyst can adopt different forms because they grow inside the separated connective wall of the cyst epithelial layer or grow by invading the connective wall of the cyst (24, 25). The presence of blood vessels in the stroma close to the epithelial proliferations aid in identifying the hyperplastic epithelial growth in the cyst’s wall arising from inflammatory changes from that of ameloblastoma, in which these blood vessels are not observed (26). Piatelli et al. (27) carried out an immunohistochemical evaluation of Ki-67 in dentigerous cysts, unicystic ameloblastomas, and ameloblastomas arising in dentigerous cysts and did not find statistically significant differences between the unicystic ameloblastoma and ameloblastoma arising from the dentigerous cyst. The authors concluded that these immunohistochemical data support the concept that an ameloblastoma arising from a dentigerous cyst has a biological behavior similar to that of the unicystic ameloblastoma and should be considered as merely a histologic variant.

The relationship of cyst development to the mandibular second primary molars curiously affected or pulpotomized has been stressed (5, 7, 10). There are two possible explanations: (1) that the mandibular molars have a greater susceptibility to caries and, consequently, are more frequently treated; and (2) that a mandibular second primary molar is more closely associated with its successor’s follicle; such association can more easily facilitate the spread of inflammation in comparison with other primary teeth (17).

In the periradicular region of pulpotomized deciduous molars, besides the dentigerous cyst, a glandular odontogenic cyst (28) or a periradicular cyst (4) can develop. Periradicular cysts in the primary dentition are located around the roots and in the interradicular area; this is in contrast to the periapical location seen in permanent teeth (5). Inflammation and prolonged irritation are apparent causes of, and the trigger for, proliferation of the epithelial rests. This explains why cysts are found in conjunction with necrotic or pulp-treated teeth in the primary dentition (5). Periradicular cysts that have been described in association with pulpotomized primary molars seem to show specific clinical features: large size, rapid growth, buccal expansion, and displacement of succedaneous teeth (9). Symptoms are similar for both periradicular and dentigerous cysts, but they differ by stage of inflammation (chronic or acute). Some features of chronic inflammation are buccal bone expansion, facial swelling, and sinus tract formation.
Several differences can be found between the radiological features of periradicular cysts and dentigerous cysts: (1) periradicular cysts are frequently unilocular (2, 9), although occasionally they are multilocular (29); (2) they are well-defined radiolucencies, located both periradicularly and interradicularly; and (3) the pericoronal space of the underlying permanent tooth appears normal, usually with an intervening total or partially distinct cortical layer of bone (30). On the contrary, dentigerous cysts: (1) generally have no distinct boundaries between the roots of the primary tooth and the crown of the underlying succedaneous tooth; (2) a single, unilocular, well-defined, radiolucent area encloses each kind of cyst (8, 17); and (3) a radiolucent area that embraces the permanent tooth bud, ill-defined lesion borders, and arrest of the root development are pathognomonic signs of damaged tooth germs (6, 7). As shown in occlusal radiographs, there are no differences between the two types of cysts with respect to size, buccal bone expansion, or displacement of adjacent permanent teeth (17). Shaw et al. (17) noted that distinguishing between dentigerous and radicular cysts on histologic grounds is difficult but that a histologic examination may determine whether the lesion is chronic or acute in nature.

Because dentigerous cysts can attain considerable size with minimal or no symptoms, early detection and removal of the cysts is important to reduce morbidity. Moreover, frequently they presented without pain and are discovered during investigation of asymptomatic slow-growing swellings (31). It is therefore important to perform radiographic examination of all unerupted teeth. Moreover, an alteration in the pattern of permanent tooth eruption under pulpotomized teeth (1, 32) was previously reported. Whereas bitewing and periapical radiographs are typically performed in the routine examination of patients with a healthy dentition, this series of radiographs may occasionally fail to delineate the full extent of a lesion, if present. A panoramic radiograph supplemented with skull series or more advanced imaging such as tomography may permit a better delineation of the extent of the lesion and its relationship to adjacent anatomical structures.

Treatment choices include extraction of the primary tooth and preservation of the underlying permanent tooth (5). Some practitioners prefer marsupialization only (8, 9, 17), whereas others perform marsupialization and also pack the space with medicated gauze to prevent early wound closure (5, 17). Neither treatment modality requires complete removal of the cyst wall. When there is no threat of damage to the permanent tooth bud, complete curettage of periradicular cyst has been performed (9, 17). In cases in which the permanent tooth is severely damaged, complete enucleation of the cyst to include the permanent tooth bud has been recommended (6, 7, 33). Enucleation was the goal if it did not jeopardize the permanent tooth germ; otherwise, marsupialization was elected. Additional packing of the cystic cavity was unnecessary (5). In the case presented, because the premolar was unlikely to erupt on its own, enucleation with removal of the displaced tooth was favored.

The present case shows the spontaneous evolution of a follicle of a retained premolar associated with a pulpotomized deciduous tooth into a dentigerous cyst. The presumed diagnosis was made because of a radiolucent lesion in the orthopantomography. The posterior histological study confirmed the diagnosis. This case, together with other published cyst cases related to a pulpotomized root–treated deciduous tooth, allow us to recommend that failures of pulp therapy, such as periradicular rarefaction, root resorption, and enlarged follicular space, should be treated by extraction; follow-up should be maintained until normal eruption of the succedaneous tooth. Parents should receive more information regarding adverse side effects of pulpotomy and be instructed on how to recognize significant signs and symptoms.

Conclusions

A dentigerous cyst can occur in conjunction with necrotic or pulp-treated teeth in the primary dentition. The time lapse between the pulp therapy and the detection of buccal bone expansion ranged from 5 months to 3 years. Taking into account that ameloblastic transformation can occur, practitioners should investigate cases of delayed exfoliation and expansion of cortical bone in the vicinity of primary molars, particularly if they are pulpotomized.

References

Case Report/Clinical Techniques