CASE REPORT

Inflammatory dentigerous cyst associated with an endodontically treated primary second molar: a case report

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Abstract

Primary teeth with periapical inflammation induced by dental caries are treated through pulp therapy. Thus in pediatric dentistry, pulpectomy is considered the most common and successful treatment procedure for primary teeth with periapical inflammation. Dentigerous cyst is an epithelial lined cavity that develops from the reduced enamel epithelium related to the crown of an unerupted tooth. This report describes a case of an inflammatory dentigerous cyst associated with an endodontically treated primary second molar in a 12-year-old female patient with mixed dentition. The relation between endodontic treatment and development of dentigerous cyst is discussed.

Introduction

Primary teeth afflicted by dental caries and its sequelae can be successfully treated by endodontic therapy procedure. However, the drawbacks of the pulp treatment are the occurrence of adverse effects such as enamel defects, delay in tooth eruption, discolouration, cyst formation and damage to the permanent dentition (Alaçam, 1989; da Silva \textit{et al.}, 2002; Grundy \textit{et al.}, 1984; Lustig \textit{et al.}, 1999). A dentigerous cyst also known as follicular cyst can form in the periradicular region after pulp treatment (da Silva \textit{et al.}, 2002; Lustig \textit{et al.}, 1999) is caused by an alteration of reduced enamel epithelium which results in fluid accumulation between the epithelium and the tooth crown (da Silva \textit{et al.}, 2002; Brook and Winter, 1975). The exact histopathogenesis of dentigerous cyst is unknown, but some authors consider the occurrence of two types dentigerous cyst: 1) developmental, originating from a tooth follicle and 2) inflammatory dentigerous cyst (Benn and Altini, 1996; Lustig \textit{et al.}, 1999).

The relation between persistent and prolonged inflammation of a primary tooth and development of a dentigerous cyst involving the permanent successor has been extensively discussed (Lustig \textit{et al.}, 1999; Benn and Altini, 1996; Shear, 1994; Aguiló and Gandía, 1998). Histological evidence for this cyst suggests that inflammatory exudate induced by an infection spreads to dental follicle, causing separation of reduced enamel epithelium from the enamel leading to development of dentigerous cyst (Benn and Altini, 1996). This may be true for immature permanent teeth that found under necrotic or pulp treated primary teeth (Grundy \textit{et al.}, 1984). Many authors prefer the term “inflammatory dentigerous cyst” for the follicular cyst induced by necrotic or pulp treated primary tooth (da Silva \textit{et al.}, 2002; Benn and Altini, 1996; Aguiló and Gandía, 1998).

Many cases of dentigerous cyst arising from endodontically treated primary molars involved the permanent successor (da Silva \textit{et al.}, 2002; Benn and Altini, 1996; Lustig \textit{et al.}, 1999; Shear, 1994; Aguiló and Gandía, 1998; Asian-González \textit{et al.}, 2007).

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As long as cyst remains small, it is asymptomatic and usually only found by chance when radiographs are taken to investigate a case of missing tooth, delay in tooth eruption or for orthodontic reasons. Hence, it was proposed that “keeping an eye on the ball” is very essential to detect dentigerous cyst since it is associated with an impacted, unerupted, supernumary and even pulp treated primary teeth (Shetty and Sandler, 2004). However, it can become extremely large and is sometimes associated with cortical expansion and erosion (Lustig et al., 1999).

**Case report**

A female patient aged 12 years reported to the Department of Pedodontics and Preventive Dentistry, College of Dental Sciences, Davangere, Karnataka, India with complaint of pain and pus discharge in the right lower back tooth region since 15 days. Patient's family and medical history were non contributory. Intra oral examination revealed patient with mixed dentition and showed a stainless steel crown on 85, which was having grade III mobility and tender on both vertical and horizontal percussion. Patient past dental history revealed that root treatment had been done 3 years back for the same tooth followed by stainless steel crown placement that was confirmed by previous radiographs. Patient missed further appointments. Vestibule in relation to 44 to 47 was obliterated, red in color and was tender on palpation. Orthopantomograph examination revealed unerupted 45 surrounded by well defined, single, unilocular radiolucency of oval shaped, well demarcated by a radiopaque line, extending laterally from the distal surface of root of the mandibular right first premolar to the distal root of first permanent molar and inferiorly to lower border of the mandible (Fig. 1). Expansion of buccal cortical bone from 44 to 47 was evident on mandibular occlusal radiograph (Fig. 2). On periapical radiograph the tooth 85 exhibited the resorption of the mesial and the distal roots with radiopaque restorative material in the root canals (Fig. 3). With the above findings the case was diagnosed provisionally having a dentigerous cyst.

As panoramic (Fig. 1) and periapical radiographs (Fig. 3) showed the horizontal impaction of the second premolar a decision of enucleation of the cyst along with removal of an impacted premolar was made. Patient was operated under general anesthesia. The tooth 85 was extracted (Fig. 4). The cyst was completely enucleated with removal of an impacted 45 (Fig. 4). The histopathogical examination confirmed the suspected diagnosis of inflammatory dentigerous cyst. Microscopically cyst lumen was lined by varying thickness of flattened epithelial cells resembling reduced enamel epithelium. Underlying connective tissue capsule showed chronic inflammatory cell infiltration (Fig. 5). After enucleation the cystic cavity was thoroughly irrigated and flap was closed with 3-0 silk suture. After one week recall, the sutures were removed and healing found satisfactory. Unfortunately patient missed further follow-up.

![Figure 1: Panoramic radiograph showing a single, unilocular well defined radiolucent lesion (white arrows) involving an unerupted mandibular right second premolar (yellow arrow).](image-url)
Discussion

Dentigerous cyst is an epithelial – lined developmental cavity that encloses crown of an unerupted tooth at cemento-enamel junction and thus it is also named "tooth-containing cyst" (Asian-González et al., 2007). The relation between pulp treatment, persistent and prolonged inflammation of a primary tooth and development of a dentigerous cyst involving the permanent successor has been extensively discussed (Lustig et al., 1999; Asian-González et al., 2007; Kozelj and Sotosek, 1999). The most common etiologic factor for periapical inflammation associated with primary molars is the dental caries. The inflammation at the apex of a primary tooth can cause development of follicular cyst around the permanent tooth which has been called inflammatory dentigerous cyst. It has been suggested that inflammatory exudate is induced by infection that spreads to the dental follicle, causing separation of the reduced enamel epithelium from the enamel. Another school of thought has been proposed to explain the histogenesis of the inflammatory dentigerous cyst which states that the crown of a permanent tooth may erupt into a radicular cyst of a primary predecessor resulting in the formation of an extra follicular dentigerous cyst. But this process appears to be rare because radicular cysts involving primary dentition are extremely rare. Shear (1994) has estimated that about 9% of dentigerous and 1% of radicular cysts occur in the first decade of life, while according to Donath (1985) about 4% of dentigerous and less than 1% of radicular cysts appear in this life period.

Although the pulp treatment of primary teeth is a common and successful procedure in pediatric dentistry, it seems that pulp treatment cannot prevent the development of these cysts as they have
been noted to occur both after pulp treatment (Alaçam, 1989; da Silva et al., 2002; Grundy et al., 1984; Lustig et al., 1999) and in the absence of it (Arita et al., 1989). The cystic potential of radicular lesions should be emphasized especially in primary dentition because local irritation caused by inflammation and endodontic therapy may induce adverse effects to the successor tooth follicle (da Silva et al., 2002). These conditions are very common in primary molars. Thus, a close follow up protocol should be implemented bearing in mind that pathological changes associated with an endodontically treated deciduous teeth may remain symptom – free. The materials used for endodontic treatment in deciduous teeth can provoke continued long-term local antigenic stimulation within the follicle and trigger the growth of cysts (Grundy et al., 1984; Asian-González et al., 2007; Benn and Altini, 1996). This may have occurred in this present case as previously demonstrated (Benn and Altini, 1996; da silva et al., 2002).

An inflammatory dentigerous cyst occurs more frequently in boys than girls. They found only the mixed dentition and usually occur between 6-12 years of age and are ten times more likely to occur in the lower jaw than the upper (Kozelj and Sotosek, 1999). The prevalence of dentigerous cyst development in the mandible near the region of the second primary molars has been linked to the observations that second primary mandibular molars are associated with greater caries susceptibility and more treatment. The close physical relationship between a second primary mandibular molar and the follicle of the successor has been associated with a facilitated spread of infection in comparison with other primary teeth (Grundy et al., 1984; Lustig et al., 1999; Kozelj and Sotosek, 1999). It has been recorded that the time lapse between the pulp therapy and the detection of buccal bone expansion ranged from 5 months to 6 years (Grundy et al., 1984). Dentigerous cyst associated with pulp treated primary molar show specific clinical features such as large size, rapid growth, buccal bone expansion and displacement of succedaneous teeth (Lustig et al., 1999). Thus it can be recommended that, clinicians should investigate cases of delayed exfoliation and expansion of cortical bone in the vicinity of primary molars.

Radiographically inflammatory dentigerous cysts appear as a round, or ovoid, well demarcated unicilocular radiolucency within the corpus of the mandible. A cyst is usually associated with the roots of a non-vital primary tooth and the crown of an unerupted permanent successor with sclerotic border.

The histopathological picture of cyst in the present case showed lot of inflammation with reduced enamel epithelium. So this case was diagnosed as inflammatory dentigerous cyst. However, features of inflammation may also be found in developmental dentigerous cysts when they get infected. Therefore, based on these microscopic findings, the present case can be misdiagnosed as developmental dentigerous cyst. In this case as the cyst arise from the follicle of the successor permanent tooth due to the inflammatory reaction associated with the apex of the primary molar which in turn stimulated the proliferation of the reduced enamel epithelium of the adjacent developing premolar and based on microscopic and radiographic features, this case was diagnosed as inflammatory dentigerous cyst. Previous reports also support this phenomenon (Benn and Altini, 1996; Lustig et al., 1999; da Silva et al., 2002).

In differential diagnosis, an odontogenic keratocyst and unicystic ameloblastoma must be considered. Most frequently, they both occur in the molar region of lower jaw during the second and third decade of life, than an inflammatory dentigerous cyst which found more commonly in 6-12 years of age i.e. in mixed dentition period and in the mandibular second premolar region. The radiography does not show radiolucency associated with roots of a non-vital or pulp treated primary tooth and the crown of an unerupted permanent tooth (Kozelj and Sotosek, 1999; Holmlund et al., 1991; Ziccardi et al., 1997; Arita et al., 1989; Murakami et al., 1995; Prabhu et al., 1996; Wood et al., 1988).

Possible complications of this cyst include: permanent bone deformation, resorption of mandibular canal wall, displacement of secondary teeth, or development of an ameloblastoma or epidermoid carcinoma (Lustig et al., 1999; Holmlund et al., 1991; Ziccardi et al., 1997). Hence early diagnosis and removal of dentigerous cysts is very important to reduce morbidity.
Ziccardi et al (1997) state that, treatment modalities range from enucleation to marsupialization. It is imperative that utilization of a conservative approach to treatment of dentigerous cysts that does not involve sacrifice of the unerupted permanent tooth whenever possible. However, complete eradication of the cyst should never be compromised for saving the permanent tooth bud (Ziccardi et al., 1997; Arita et al., 1989). In cases in which the permanent tooth is severely damaged, and hopelessly displaced, complete enucleation of the cyst to include the permanent tooth bud has been recommended (Brook and Winter, 1975). In the present case, since the second premolar was mesially displaced and was unlikely to erupt on its own, enucleation with removal of the displaced premolar was elected.

**Conclusion**

Although pulp treatment is a common and successful procedure for decayed primary teeth in children, an inflammatory dentigerous cyst can occur in conjunction with endodontically treated tooth in primary dentition. Considering complications of this cyst, the following recommendations should be made mandatory: (1) a close up protocol should be implemented to investigate cases of delayed exfoliation and pulp treated primary molars; (2) more information should be given to parents regarding adverse side effects of an endodontic treatment and be instructed to follow regular recall checkups.

**References**


