

A dentigerous cyst associated with a pulpectomised primary molar: case report

A pulpectomy is a routinely performed procedure in carious primary teeth. While adverse effects are not common, these can include a dentigerous cyst. A dentigerous cyst is an odontogenic cyst that surrounds the crown of an unerupted tooth, and is caused by a fluid accumulation between the reduced enamel epithelium (REE) and the enamel surface. Residual periapical inflammation from an endodontically treated primary tooth may lead to the development of an inflammatory dentigerous cyst in the unerupted permanent successor. This case report illustrates an infected dentigerous cyst in a seven-and-a-half-year-old female child related to the mandibular left second primary molar, which had been pulpectomised two years earlier. In general, the incidence of dentigerous cysts associated with pulpectomised primary teeth is extremely low. While there is no single factor that can be attributed to cystic transformation, it is prudent that teeth receiving pulp therapy should be observed periodically, and radiographs should be taken at regular intervals.

Keywords: Dentigerous cyst, pulpectomy, zinc oxide eugenol.

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Introduction

The primary dentition plays a pivotal role in mastication, speech and aesthetics. It guides the eruption of permanent teeth. Dental caries is one of the most common causes of premature loss of primary teeth. Preservation of primary teeth should be the objective of dentists as they maintain space for the permanent teeth. Space maintainers may be required when primary teeth are extracted. Paediatric endodontic procedures have become more frequent and are generally the treatment of choice in recent times. A pulpectomy is considered a safe procedure in primary teeth, although several deleterious effects, such as the arrest of development of permanent teeth, enamel opacities of successor teeth, deflection of the permanent tooth bud, and foreign body reaction, have been reported. Another sequela to pulpectomised

primary molars is the formation of a dentigerous cyst in the periapical region of the primary molar.^{1,2} A dentigerous cyst is an odontogenic cyst that surrounds the crown of an unerupted tooth, caused by fluid accumulation between the reduced enamel epithelium (REE) and the enamel surface. Although dentigerous cysts are considered to be developmental in origin, Main has described an inflammatory variant of a dentigerous cyst that develops as a result of the intrafollicular spread of periapical inflammation from an overlying diseased primary tooth.³

Cysts in relation to pulpectomised primary molars are uncommon, and their aetiology remains unclear. This report describes one such case of an infected dentigerous cyst in relation to a pulpectomised primary molar, while also discussing the probable aetiopathology.



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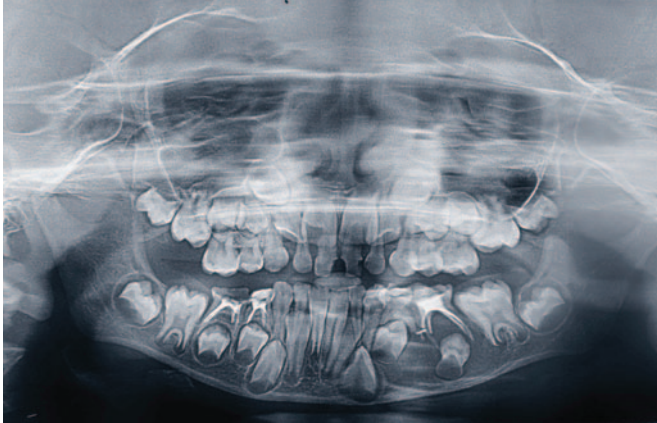


FIGURE 1: Orthopantomogram showing unilocular, well-defined, radiolucent lesion involving an unerupted mandibular left second premolar.

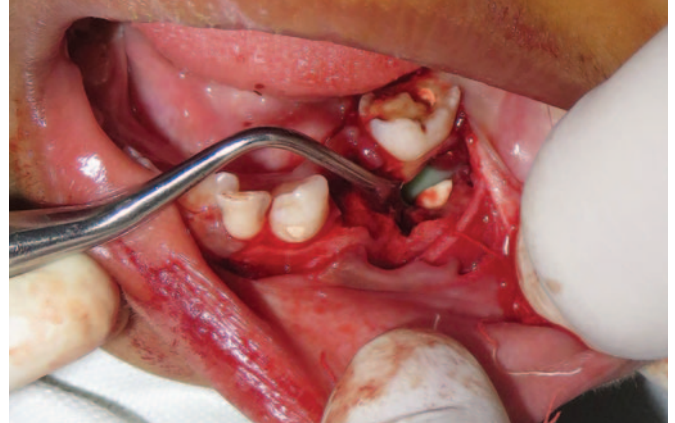


FIGURE 2: Cystic lining with exudate seen in relation to the unerupted mandibular second premolar.

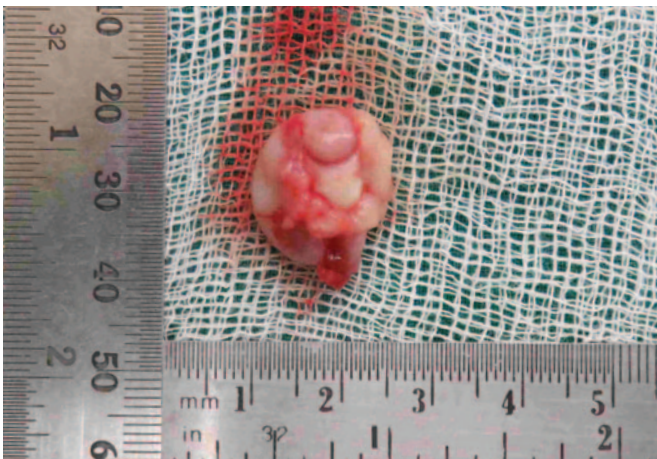


FIGURE 3: The enucleated dentigerous cyst around the neck of the second premolar.

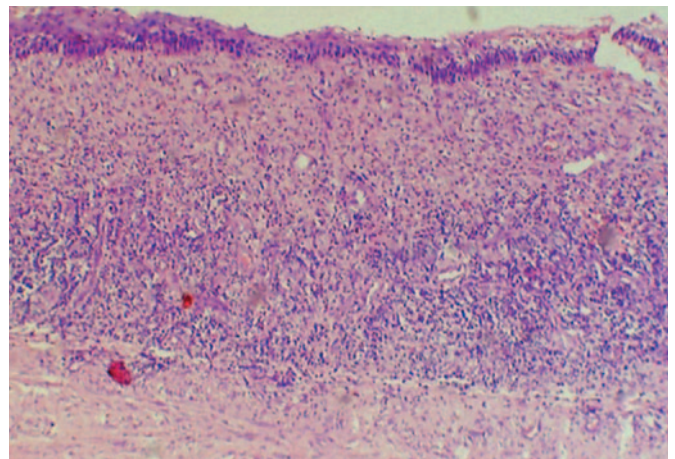


FIGURE 4: Histopathological image of the cystic lining at 10x (haematoxylin-eosin stain).

Case report

A seven-and-a-half-year-old girl reported to the Department of Paediatric and Preventive Dentistry with a chief complaint of an extra-oral swelling on the left side of the lower jaw, which gradually increased in size over three months. Previous dental records revealed that pulpectomies were performed in the mandibular right primary molars and the left second molar using zinc oxide eugenol (ZOE) as the obturating material, 20 months previously. On extra-oral examination, a firm swelling was noted on the lower left side of the face. Intra-orally, a dislodged temporary restoration with secondary caries in relation to the mandibular left second primary molar was observed. A well-defined swelling, measuring approximately 1x1.5cm², obliterating the buccal vestibule in the region of the mandibular left primary molars, was identified.

Radiographic examination

An orthopantomogram (OPG) showed a well-rounded, unilocular radiolucency with radiopaque borders below the roots of the mandibular left second primary molar, extending from the mesial margins of the mandibular left first primary molar to the mesial root of the left first permanent molar (Figure 1). A radiopaque mass was evident in the periapical region surrounding the

mandibular left second primary molar. The tooth germ of the mandibular left second premolar was observed to be displaced towards the lower border of the mandible. The mandibular right primary molars also showed radiopaque root canal filling material with no apparent radiographic changes.

Surgical excision

The clinical and radiographic findings were suggestive of a dentigerous cyst associated with the mandibular left second premolar. Favourably positioned premolars can erupt normally following marsupialisation of the cyst. The impacted premolar in this case was assessed to be in an unfavourable position. The proposed treatment plan was cystic enucleation of the lesion along with removal of the second premolar under local anaesthesia. A full thickness mucoperiosteal flap was reflected to expose the cyst (Figure 2). Enucleation of the cyst was performed, and the flap was approximated using 3-0 Mersilk. The cystic lining was then sent for histopathological evaluation (Figure 3).

Microscopic findings

Microscopic examination demonstrated that the cystic lumen was lined by thin non-keratinised, stratified squamous epithelium resembling REE. The

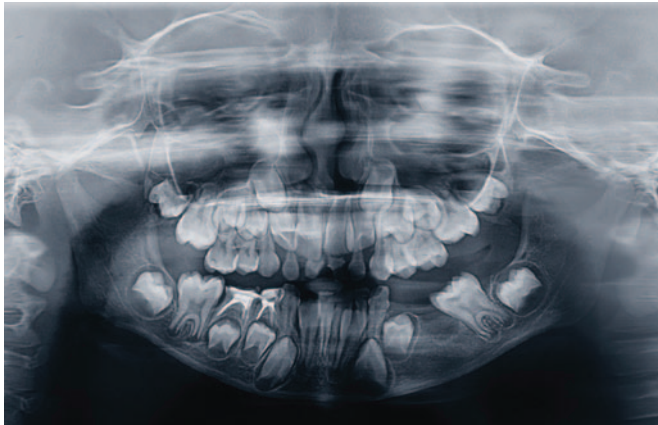


FIGURE 5: Six-month follow-up orthopantomogram.

underlying mature fibroconnective tissue showed mild, chronic inflammatory cell infiltrate. The overall histopathological features were suggestive of an infected dentigerous cyst (**Figure 4**). A comprehensive dental treatment plan for the remaining carious teeth comprising extractions, restorations and preventive treatment was planned for the patient. Unfortunately, the patient did not keep her appointments for comprehensive care but reported six months later for review, when an OPG was taken. The radiograph demonstrated satisfactory healing (**Figure 5**).

Discussion

A dentigerous cyst is the most common type of developmental odontogenic cyst, constituting around 20% of all epithelium-lined cysts. A developmental dentigerous cyst is induced by fluid accumulation either between the REE and the crown of an unerupted tooth, or between the layers of the enamel organ, and grows by the expansion of the follicle, while it is still attached to the neck of the tooth.⁴ Incidences of the development of radicular cysts following pulp therapy of primary teeth are well documented.^{1,5,6} However, the development of dentigerous cysts in relation to endodontically treated primary molars with persistent inflammation has been studied by only a few authors. Asián-González reported a case of a dentigerous cyst arising from a pulpectomised primary molar, while Nagaveni *et al.* documented a case of an inflammatory dentigerous cyst arising from a pulpectomised primary molar.^{7,8} While the pathogenesis of these inflammatory cysts is still unclear, Benn and Altini, in their study on dentigerous cysts arising as a result of an inflammatory cause, have proposed three possible mechanisms of histogenesis of these cysts. A permanent successor may erupt into a radicular cyst that has formed at the apex of a nonvital primary tooth and results in a dentigerous cyst that is extrafollicular in origin. Periapical inflammation from a nonvital primary tooth may secondarily infect a developmental dentigerous cyst, or may spread to involve the follicle of the permanent successor, resulting in dentigerous cyst formation.⁹ We propose that the chronic inflammation caused due to the extruded ZOE cement could act as a factor resulting in a dentigerous cyst. Materials used for the endodontic treatment of primary molars have also been implicated in the pathogenesis of such cystic transformations.¹⁰ Accidental periapical extrusion of root canal filling material frequently ensues during the obturation of primary teeth. Sometimes, it is beyond the control of the

operator to prevent the extrusion of the material beyond the apex, especially in cases of large apical foramen and strip perforations.¹¹ Whether this extruded material aids in the healing of periapical pathoses or acts as an antigenic stimulus for the development of the dentigerous cyst remains uncertain.¹⁰ The effect of this foreign body on the periapical tissues has been extensively studied. Although described as a resorbable material, ZOE has been documented to cause deleterious effects on the permanent successor, so particular caution should be taken to prevent overfilling with ZOE in teeth with large apical foramen.¹¹ It is also known to act as an irritant for the periapical tissues, resulting in necrosis of bone and cementum.¹² When employed as an endodontic obturating material in primary teeth, a difference between the rate of resorption of ZOE cement and root structure has been noted, which leads to retention of the paste in the periapical region. The retained paste has been found to induce a foreign body reaction or an inflammatory reaction (ranging from subacute to chronic) in the dental follicle of the permanent successor.¹³ Histopathological evaluation of root canal filling materials in dogs by Silva *et al.* demonstrated an adverse tissue response such as the presence of inflammatory cells, oedema, and severely thickened periodontal ligament when ZOE comes in contact with periapical tissues.¹⁴ Initially, ZOE was the only material recommended in the clinical guidelines of the American Academy of Pediatric Dentistry (AAPD), but since 2009, based on studies recently published, the AAPD advises iodoform-based pastes as suitable alternatives to ZOE.^{15,16}

There is no definite consensus that the obturating material used in endodontic treatment forms the aetiology for such inflammatory cysts. As hypothesised by Savage, the material used in pulp therapy may cause antigenic stimulation, which could lead to the development of a cystic lesion in the periapical region.¹⁰ However, this hypothesis cannot be directly applied to the present case as the patient had a history of bilateral pulpectomies using ZOE, and the development of the dentigerous cyst was found only on the left side. Clinical and radiographic examination of the right mandibular primary molars revealed healthy tissues; the supporting tissues also appeared to be normal.

No single determinant can be attributed for the aetiopathogenesis of dentigerous cysts. Some causative factors such as infection resulting from the non-vital primary tooth, the material used for pulpectomy, and the host tissue response could all play a significant role in its pathophysiology. Thus, a relationship may exist between the pulpectomy procedure, inflammation of the periapical tissues, and the development of the dentigerous cyst involving the permanent successor. Nagaveni *et al.* and Chakraborty *et al.* have documented cases of the development of dentigerous cysts following pulpectomy of primary teeth; however, the obturating material used has not been reported.^{8,17}

Conclusion

Dentigerous cysts associated with primary teeth that have had a pulpectomy are very rare. While there is no single factor that can be held responsible for the cystic transformation, a tooth receiving pulp therapy must be observed periodically, both clinically and radiographically, to detect any pathological changes in the periapical region. Even though endodontic treatment can successfully preserve carious primary teeth, there may be complications.

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CPD questions

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CPD

1. The most common type of developmental odontogenic cyst is:

- ☐ A: Radicular cyst
- ☐ B: Dentigerous cyst
- ☐ C: Odontogenic keratocyst
- ☐ D: Residual cyst

2. The treatment for a dentigerous cyst involving an unerupted permanent tooth bud in an unfavourable position is:

- ☐ A: Enucleation of cyst with extraction of unerupted tooth
- ☐ B: Prolonged antibiotic therapy
- ☐ C: Marsupialisation
- ☐ D: Incision and drainage

3. Zinc oxide eugenol used as an obturating material in primary teeth resorbs at a rate:

- ☐ A: Slower than the physiologic root resorption
- ☐ B: Faster than the physiologic root resorption
- ☐ C: The same as that of physiologic root resorption
- ☐ D: Does not resorb.