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Inflammatory dentigerous cyst of mandibular first premolar associated with endodontically treated primary first molar: a rare case report

ABSTRACT

Aim *Inflammatory dentigerous cysts usually occur in the mixed dentition. It has been reported that inflammatory lesions from the root area of a deciduous tooth bring about the development of dentigerous cysts around the unerupted permanent tooth bud. Endodontic treatment is a common and successful procedure for periapical inflammation in children. An inflammatory dentigerous cyst can occur in conjunction with endodontically treated primary tooth.*

Case report *This article reports a case of 6 years and 6 months old boy, with a single, well-defined, unilocular, radiolucent area enclosing the first right unerupted mandibular premolar, accidentally discovered on the panoramic radiograph. The first right primary molar had received a root canal treatment18 months prior. Clinical findings combined with radiographic and microscopic examinations confirmed the diagnosis of inflammatory dentigerous cyst. As treatment, enucleation of the cyst with removal of the involved permanent tooth was chosen and a removable partial denture was supplied to the patient after surgery. The 5-year follow-up revealed good healing of the bony lesion and displaced teeth.*

Conclusion *This case presented a severe and rare complication of endodontically treated primary tooth, and the recommendation includes: (1) early diagnosis of dentigerous cysts, which is essential to prevent extensive treatment; (2) more information on the adverse side effects of an endodontic treatment should be given to patients and parents; (3) patients should be informed about the importance of follow-ups and radiographic follow-up should be routinised.*

Keywords *Enucleation; Inflammatory dentigerous cysts; Primary tooth.*

Introduction

Dentigerous cyst is a common developmental odontogenic cyst that encloses the crowns of unerupted teeth or developing teeth bud, and is attached to the cervical margins of affected teeth [Jones, Craig and Franklin, 2006; Shear and Speight, 2007]. Dentigerous cyst is the second most common odontogenic cyst, and it represents about 17.1% of all true jaw cysts [Benn and Altini, 1996]. Despite the common occurrence of dentigerous cysts, their precise origin is uncertain. Previous authors suggested two types of dentigerous cyst [Benn and Altini, 1996; Narang et al., 2012; Shibata et al., 2004; Main, 1985]: developmental and inflammatory. Developmental dentigerous cysts originate from a tooth follicle and may develop by fluid accumulation either between the reduced enamel epithelium and the enamel, or alternatively between individual layers of the reduced enamel epithelium. Inflammatory dentigerous cysts are induced by deciduous periapical inflammatory lesions that spread to the successors' follicle, causing separation of the reduced enamel epithelium from the enamel. This theory is supported by clinical and radiographic evidence of inflammation of deciduous teeth in cases with histologically confirmed dentigerous cysts associated with permanent successors [Benn and Altini, 1996; Grundy, Adkins and Savage, 1984]. Dentigerous cysts are always associated with a missing tooth or failure of tooth eruption, and may be asymptomatic until they become enlarged enough. These cysts are often found by chance when radiographs are taken for other reasons, such as miss teeth and orthodontic treatment.[Shetty and Sandler, 2004]. Dentigerous cysts often block eruption of teeth, displace teeth, become larger, destroy bone and occasionally lead to pathologic fracture [Shear and Speight, 2007].

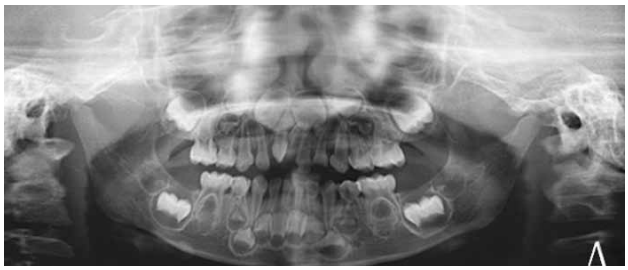

FIG. 1 Panoramic radiograph of the patient at age 3y6m.

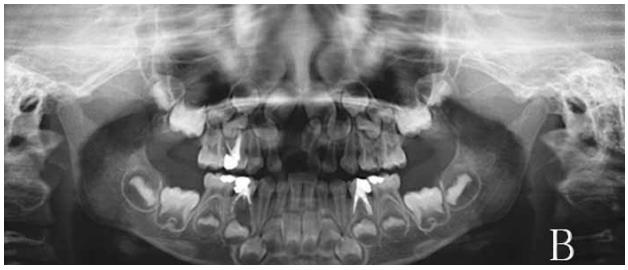

FIG. 2 Panoramic radiograph of the patient at age 5y2m.

FIG.3 Panoramic radiograph of the patient at age 6y6m.

FIG. 4 Computed tomography revealed that the dimensions of the radiolucent lesion were approximately 20.2 mm × 22.1 mm, and were not significant in the buccolingual direction.

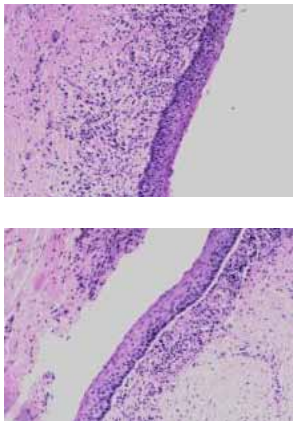

FIG. 5 Image of the histopathological findings which shows a thin fibrous wall covered with stratified squamous cells, and the wall mildly infiltrated with chronic inflammatory cells.

Case report

A 6 years and 6 months old boy presented at the Department of Orthodontics, Stomatology Clinic, Wenzhou Medical University in May 2009, with a complaint of delayed eruption of the upper incisors. He did not reporte of any tooth pain. The patient was apparently healthy on general examination and had no history of systemic diseases. According to the dental history, we found that the patient had received several endodontic treatments and had undergone extraction of a supernumerary tooth. The previous panoramic radiographs (Fig. 1, 2) showed that a supernumerary tooth was in the eruption path of the left upper incisor and that teeth 54, 74 and 84 had been endodontically treated.

The extraoral examination was unremarkable. Intraoral examination revealed a slight expansion of the buccal cortical plates at the region of teeth 44 and 45. The swelling was firm and painless on palpation. The patient was in the mixed dentition stage and only teeth 36, 31, 41 and 46 had erupted. We advised a panoramic radiograph which revealed a single, unilocular, well-delimited, radiolucent area enclosing the right mandibular first premolar bud (Fig. 3). The displacement of the permanent teeth buds near the cyst was distinct, especially the right first mandible premolar bud that had moved to the floor of the mandible. Computed tomography was performed for further evaluation of the content and the extent of the cyst. The dimensions of the radiolucent lesion were approximately 20.2 × 22.1 mm, and were not significant in the buccolingual direction (Fig. 4). On the basis of radiographic and clinical findings, a provisional diagnosis of inflammatory dentigerous cyst associated with an endodontically treated primary molar was made, however adenomatoid odontogenic tumor, keratocyst and large periapical cyst were also considered as diagnostic hypotheses. The results of routine blood and urine examinations were normal, and surgical enucleation of the cyst was performed. Extractions of teeth 84 and 85 were done under local anaesthesia and brown fluid was drained during the procedure, then, the buccal flaps were raised and the cyst was enucleated together with the retained tooth. The specimen was sent for histopathological examination, which showed a thin fibrous wall covered with stratified squamous cells, and the wall was mildly infiltrated with chronic inflammatory cells (Fig. 5). The histological examination confirmed the suspected diagnosis. Antibiotics and analgesics were prescribed. One week after surgery, the surgical wound showed good healing. A removable partial denture was used as a functional space maintainer, which also improved aesthetics and pronunciation. The patient was asked to revisit and radiographic follow-up quarterly, however, he did not come back to our department until March 2012. The patient had lost the removable partial denture three months after first wearing and paid no attention to the space in the lower dentition. Fortunately, there were no clinical or radiographic signs of recurrence

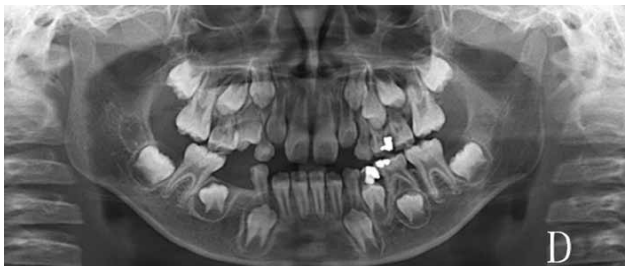

FIG. 6 Panoramic radiograph of the patient at age 9y4m.

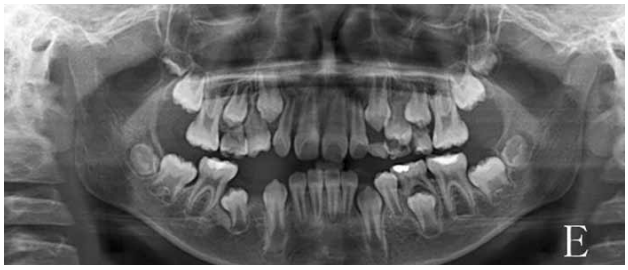

FIG. 7 Panoramic radiograph of the patient at age 10y9m.

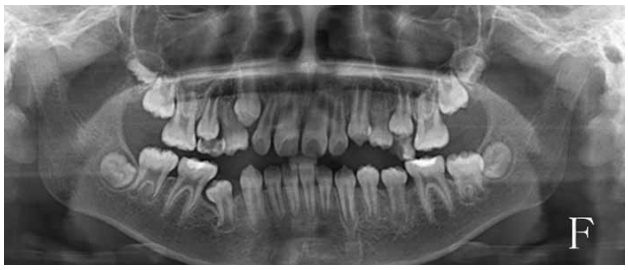

FIG. 8 Panoramic radiograph of the patient at age 11y9m.

of the lesion and the bone was reformed in the cavity (Fig. 6). Development and position of teeth 43 and 45 were normal. The right mandible molar moved forward, but the space was enough for eruption of teeth 43 and 45. Due to the crowding of maxillary dentition, a future orthodontic treatment plan may include extraction of three premolars, thus we did not ask the patient to wear a maintainer again. The patient was advised to undergo a radiographic follow-up once a year. At the 2-year follow-up, the patient presented good healing of the bone and teeth eruption (Fig. 7, 8). Orthodontic treatment will start after eruption of all permanent teeth.

Discussion

Inflammatory dentigerous cysts are usually found in mixed dentition around the first decade of life, while developmental dentigerous cysts occur in the second and third decades of life. Inflammatory dentigerous cyst develops when an apical inflammation in the primary predecessor spreads to the follicle of the unerupted immature permanent successor and stimulates cystic degeneration of the permanent tooth follicle. Several researches [Benn and Altini, 1996; Shibata et al., 2004; Main, 1985; Gondim et al., 2008] assumes that deciduous teeth with infected pulp may potentially lead to development of dentigerous cysts, as incomplete root canal treatment is a dangerous factor. A dentigerous cyst can form in the periradicular region after pulpotomy, because the prolonged inflammation may cause chronic irritation to the dental follicle of the successor tooth. [Lustig, Schwartz-Arad and Shapira, 1999; Brook and Winter, 1975]. In the literature, more than twenty cases of dentigerous cysts associated with infected primary teeth have been reported [Narang et al., 2012; Azaz and Shteyer, 1973; Shaw, Smith and Hill, 1980; Yu-Hsuan Hu et al., 2011].

Compared to previously reported cases, this case was much more valuable and rare. This case report provides enough information on whether inflammation observed in dentigerous cyst is a primary/secondary phenomenon. As we know, deciduous periapical inflammatory lesions could spread to the successors' follicle, leading to the formation of inflammatory dentigerous cyst, while if a developmental dentigerous cyst develops a secondary infection in this case it can be defined as infected dentigerous cyst. However, it is difficult to distinguish the two entities according to the histopathologic features, if proper dental history is not collected. Most of the previous cases only showed infected primary teeth, radiographic evidences and histopathologic features, however these evidences could not prove that the dentigerous cysts developed after the inflammation. The evidences should include a radiograph showing that there was no dentigerous cyst before the deciduous periapical inflammatory lesions occurred. In this case, a panoramic radiograph was taken for a supernumerary tooth when the patient was 3 years and 6 months old, showing no abnormalities of the right lower first premolar compared to the other side. The second panoramic radiograph was taken for another supernumerary tooth when he was 5 years and 2 months old: 4 months after root canal treatment of the right lower primary first molar, the cyst could not be detected.

One year 4 months later, the patient came to the Department of Orthodontics, to find out the reason for delayed eruption of the upper incisors. The panoramic radiograph revealed a large radiolucent area below the right lower primary molar. This information combined with clinical and microscopic examinations confirmed the diagnosis of inflammatory dentigerous cyst.

This case was a big challenge for us. What should dentists do to manage the risk of inflammatory dentigerous cyst after root canal treatment for infected primary teeth? Figure 1B shows that a radiolucent lesion between teeth 44 and 84, which means that the periapical inflammatory lesions of tooth 84 had not disappeared 4 months after root canal treatment. This might be the main reason for the rapid expansion of the cyst. If we discover the dentigerous cyst when it is small, the treatment would be a simple and effective extraction of the primary tooth

above the dentigerous cyst. This case should remind dentists that young patients with treated root canal should undergo a follow-up every 6 months to careful monitoring the successors' follicle. This is essential and helpful for patients.

The treatment options of a dentigerous cyst should be based on the size of the cyst, age of patient, location of the cyst, dentition affected, and relationship with the surrounding structures [Motamedi and Talesh, 2005]. In this case we had two treatment options: complete enucleation and marsupialisation. Decompression or marsupialization has been advocated by several authors for treating dentigerous cyst in children [Martinez-Pérez and Varela-Morales, 2001; Berdén, Koch and Ullbro, 2010]. Children have a much greater capacity to regenerate bone than adults, and teeth with open apices have a large eruptive potential. This conservative technique creates an accessory cavity which helps relieving intracystic pressure and accelerates healing of the cystic lesion. However, the patient should be followed-up until the complete eruption of permanent teeth in their designated location and sometimes the unerupted teeth needs orthodontic correction. The disadvantage of marsupialisation is the risk of pathologic tissue left *in situ*, and the fact that patients should frequently come to hospital for revisit. In cases in which the permanent tooth is severely damaged, complete enucleation of the cyst including the permanent tooth bud has been recommended [Wood et al., 1988]. Many authors chosen enucleation of the cyst with removal of the involved tooth [Motamedi and Talesh, 2005; Desai et al., 2005; Muthray et al., 2006] which was our treatment choice in this case. This surgical method can avoid relapse of dentigerous cyst. In the present case, the premolar was unlikely to erupt on its own and considered the risk of developmental deformity of tooth bud, enucleation with removal of the displaced tooth was chosen. Regular follow-ups are still necessary to evaluate healing of the cyst and the development of displaced teeth. A removable partial denture was used as a functional space maintainer and a future orthodontic treatment plan should be considered.

Conclusion

Primary teeth with periapical inflammation caused by dental caries are commonly treated through pulp therapy which is considered the most common and successful treatment procedure. However, inflammatory dentigerous cysts can occur in conjunction with endontically treated primary teeth. Considering complications of a dentigerous cyst, early diagnosis of dentigerous cysts is essential to prevent extensive treatment; more information of adverse side effects of an endodontic treatment should be given to patients and parents; patients should be instructed to follow regular visits and radiographic follow-ups should be routinised.

Consent

Written informed consent was obtained from the patient's legal guardian(s) for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

We declare that we have no competing interests.

Author Contributions

Contributed materials and designed the study: Feiou Lin. Performed the experiment: Feiou Lin and Linjie Yao. Wrote and revised the paper: Linjie Yao, Xingqiao Xu. Data collection: Manman Ren, Dengfeng Liu and Zhenyu Ni.

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