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CASE REPORT

Odontogenic keratocyst clinically mimicking an eruption cyst: report of a case

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This article describes a case of odontogenic keratocyst (OKC) in a 1 year and 7 month old girl who presented such a lesion mimicking an eruption cyst. To date, only one well-documented OKC occurring in a patient under 5 years old has been reported and it was thought to be associated with nevoid basal cell carcinoma syndrome (NBCCS). In our OKC case, the cyst was totally enucleated. No evidence of recurrence and NBCCS was found after a 4-year follow-up. The development of involving tooth in a growing child and the histogenesis of OKC are discussed in this article.
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A girl aged 1 year and 7 months was brought to the Pediatric Dental Clinic at Chang Gung Memorial Hospital in February 1998, because the left maxillary primary canine was still unerupted and the gingiva had been swelling for more than 1 week. The contralateral canine had erupted for 2 months. She had no previous dental or trauma history related to the lesion. The patient's past medical and family histories were non-contributory.

Intraoral examination disclosed the presence of a fluctuated mass of 1.0 cm in diameter on the left maxillary primary canine region (Fig. 1A). The lesion was covered by intact mucosa and was non-tender on palpation. Clinically, it mimicked an eruption cyst with regard to the child's dental age.

A multilocular radiolucent lesion was found in the left upper primary canine region (Fig. 1B). The mesial border of the lesion was the distal aspect of the left primary lateral incisor, whereas the distal and upper borders could not be defined in the periapical radiograph. The cortical bone was expanded and perforated. The left primary canine was displaced, and the root was further distally displaced by the lesion. A panoramic radiograph was taken, which showed that the solitary radiolucent lesion almost extended to the half of the left maxillary sinus. No other lesion was found in panoramic radiograph. During the taking of the panoramic radiograph, the mass was ruptured and yellowish white contents flooded out. The initial diagnosis of odontogenic keratocyst (OKC) was made. The cyst was totally enucleated and the tooth germ near the root of the left upper first primary molar was also removed.

The histological examination showed a typical OKC lined by parakeratinized stratified squamous epithelium, six to eight cell layers in thickness. The luminal surface of the lining epithelium revealed a corrugated appearance. The basal epithelial cells were cuboid to columnar, with a palisaded arrangement. The epithelium–connective tissue interface was flat, without evidence of epithelial ridges (Fig. 1C). A tooth bud in bell stage was also included in the specimen (Fig. 1D). Focal chronic inflammation and foreign body reaction, induced by the desquamated keratin, were discernible in the fibrous cystic wall.

All the primary teeth erupted. No recurrence of the OKC and no nevoid basal cell carcinoma syndrome (NBCCS) were found during the 4 years of follow-up. However, the follow-up panoramic radiograph (Fig. 1E) showed aberrant tooth germ position as a consequence of tumor and surgery.

Comments

Non-syndrome-associated OKC is usually found in the third or fourth decade of life. Occurring in a child, OKC is often related to the NBCCS. To the best of our knowledge, only one syndrome-associated OKC occurring in a child under 5 years old had been reported in the literature (1).

The girl had no family history or other obvious manifestations of the NBCCS up to the age of 5 years and 7 months. The satellite cysts, ameloblastomatoid rests, and solid islands of odontogenic epithelium, which are the most frequently found histologic pictures of syndrome-associated OKC (2), were not identified in our OKC case. However, the occurrence of NBCCS in later

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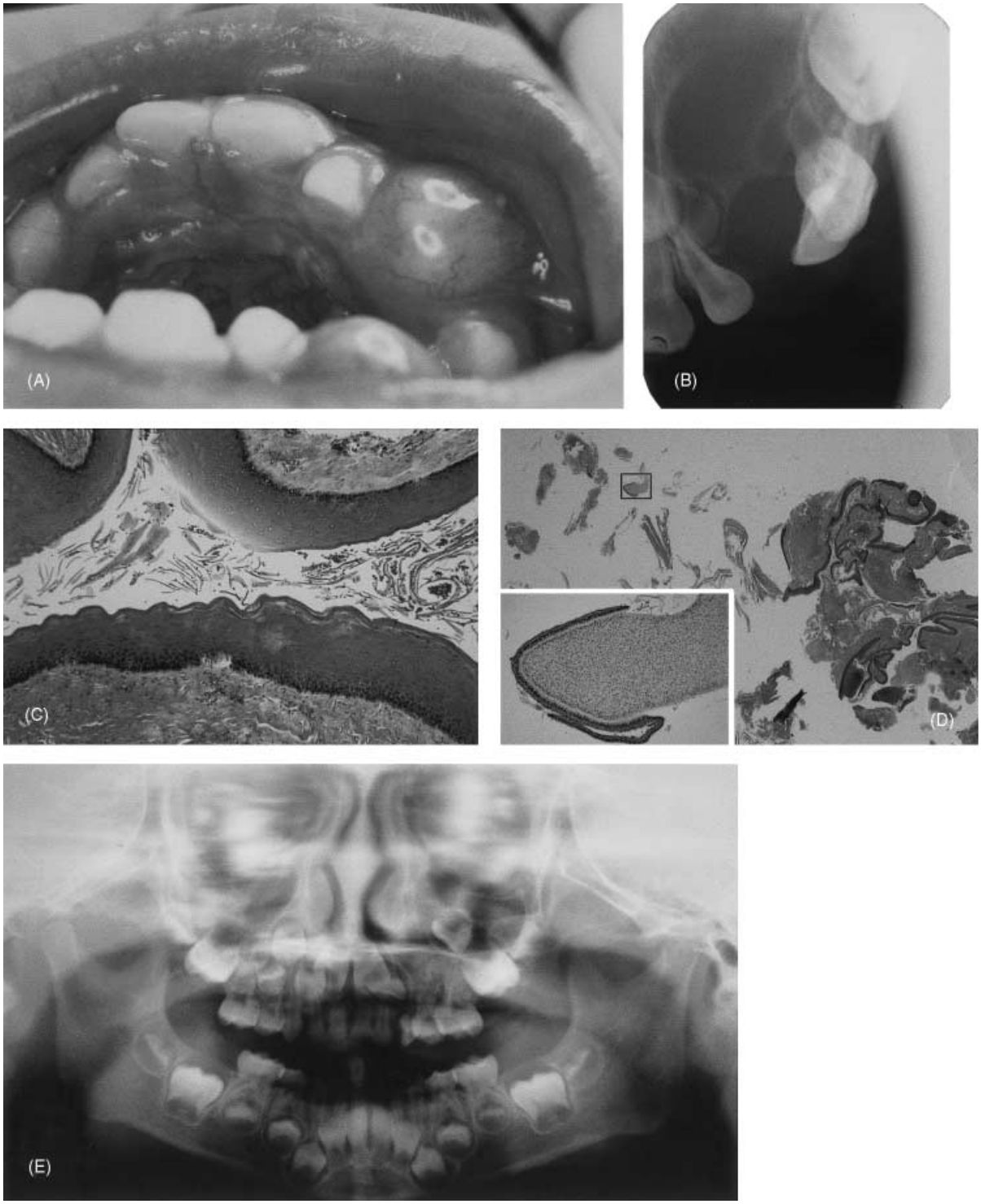

Figure 1 (A) Intraoral view of soft and fluctuated mass. The mass with 1 cm in diameter was over the maxillary left primary canine region. (B) The periapical radiography reveals a multilocular radiolucent lesion. The tooth germ removed was pointed by arrows. (C) The odontogenic keratocyst is typically lined by a parakeratinized stratified squamous epithelium. The lumen contains varying amounts of desquamated keratin. (HE 100×) (D) The tooth germ (insect) is in an extrafollicular position. (HE 4×) (E) The panoramic radiograph shows the distal eruption pathway of the upper left permanent lateral incisor, the impacted permanent left canine, the delayed development of the upper left 2nd premolar, and the missing of the upper left 1st premolar.

life should never be overlooked, because 50% of NBCCS cases have no family history, and many clinical manifestations of NBCCS remain hidden in the earlier years of life.

Delayed eruption of primary teeth is not unusual, as eruption timing and sequence in primary dentition are variable in infancy, but impaction and displacement of primary teeth may be a clue of certain pathosis if

associated with abnormal swelling or discharge. The clinical picture of this case was somehow like an eruption cyst because of the coincidence of normal eruption of the left upper primary canine. However, the multilocular radiolucency ruled out the possibility of an eruption cyst. The differential diagnosis of this case also included OKC, ameloblastoma, other odontogenic cysts and tumors, and unspecified malignancies. After the discharge of yellowish white cystic content from the ruptured cyst, a tentative diagnosis of OKC was made.

OKC is often associated with an unerupted tooth. Browne (3) has classified the OKC into follicular and extrafollicular types according to its relationship with the associated tooth. In this patient, the oral surgeon found the bell-staged tooth germ of the left upper primary canine adjacent to the cyst, and the histology of the removed specimen also showed the tooth germ in an extrafollicular position. Our finding supports the hypothesis (3–5) that a follicular OKC is extrafollicular in origin with subsequent fusion of the reduced enamel epithelium of the impacted tooth with the lining epithelium of OKC.

The excised tooth bud was presumed to be the left upper permanent 1st premolar for the following reasons. First, the timing of hard tissue formation of permanent canine starts earlier than that of the permanent 1st premolar, and is approximate to that of the permanent central incisor (6). In Fig. 1(B), we only saw an uncalcified crypt, which could be the tooth germ of the left upper first permanent premolar. Second, the excised tooth bud was in a position adjacent to the primary 1st molar. Third, the high-positioned tooth germ was obviously showing a crown with one cusp that looked like a permanent canine (Fig. 1E). In addition, both the permanent canine and the first bicuspid completed their crown formation at the age of about 6–7 years. Therefore, we can justify that the excised tooth was the left upper first premolar.

A long-term close follow-up for this patient is required in order to monitor the possibility of occurrence of NBCCS, because the sign of NBCCS may be camouflaged in very young infancy. In addition, in management of early childhood cyst or tumor, we should not only focus on the lesion itself, but also consider the surrounding developing structure.

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