Eruption cyst with obvious epithelial hyperplasia in an infant

Masayuki KAGAYA1), Hisao SHIGEMATSU1), Munehisa OKADA1)
Seiji SUZUKI1), Kaoru KUSAMA2), Hideaki SAKASHITA1)

1) Second Department of Oral and Maxillofacial Surgery, Meikai University School of Dentistry
   (Chief: Prof. Hideaki SAKASHITA)
2) Department of Oral Pathology, Meikai University School of Dentistry
   (Chief: Prof. Kaoru KUSAMA)

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INTRODUCTION

The eruption cyst is a specific type of cyst associated with erupting primary or permanent teeth1). In the past, it was classified as a form of dentigerous cyst. According to the current WHO histological typing of odontogenic tumors, the eruption cyst is a separate entity and has its own morphology code designated by the International Classification of Diseases for Oncology2). There are several proposed etiologies explaining the development of an eruption cyst3,4), and some possible causative factors, such as, infection, trauma of the primary teeth, certain genetic predisposition, cyclosporine administration, etc., have been postulated5,6). Eruption cysts occur more frequently in males than in females in a 2 to 1 ratio1,7). The prevalence of the cysts is greater in the maxillary than in the mandibular. As the average age of patients with an eruption cyst has been reported to be 4.44 to 8.18 years7), eruption cysts seldom appear in newborn infants8~10). It is usually easy to make a clinical diagnosis of an eruption cyst. Even if the possibility of other lesions, such as tumors or tumor-like lesions, can not be ruled out completely according to the clinical features, it is not difficult to make a pathological diagnosis of this lesion by biopsy. In general, the eruption-cyst wall consists of a thin lining of nonkeratinized squamous cell epithelium and a certain amount of fibrous connective tissue, but no epithelial hyperplasia. The purpose of this report is to describe a rare case of eruption cyst with obvious epithelial hyperplasia and to give a brief discussion about the diagnosis and treatment.

CASE REPORT

On September 26, 2003, a 16-month-old Japanese boy was referred to our department by his dentist with a chief complaint of a swelling on his mandible. Incision and drainage of the lesion had been performed by the dentist one month before the first visit. The swelling was resolved transitorily, but recurred and increased in size in 20 days after the treatment. He had a noncontributory medical history, and there was no peculiarity in his family history. The swelling was causing some discomfort, but no feeding problems. He had a very healthy appearance. Physical examination revealed a swelling 20×15 mm in size, elastic, firm, and painless on the left first primary molar of the mandible (Fig. 1). The overlying mucosa was smooth in appearance and normal in color without any pain on palpation. Radiography showed no radiolucent lesion in the mandible (Fig.2). CT findings revealed the lesion to have the
Fig. 1 Dome-like lesion on the crest of the left posterior mandibular alveolar ridge.

Fig. 2 Radiograph showing no radiolucent lesion in the mandible.

Fig. 3 CT findings show that the lesion included the crown of the left primary first molar, but did not involve the mandibular bone.

same degree of density as the muscle, and to include the crown of the first primary molar on the mandible (Fig. 3). Clinical diagnosis of a benign tumor of the mandible was made, and excision of the dome of the lesion was performed under local anesthesia on October 7, 2003. Histopathological examination of biopsy specimen suggested an odontogenic tumor derived from the cyst wall, and so a complete excision of the lesion was recommended rather than marsupialization. On November 12, excision of the lesion with extraction of the associated first primary tooth was performed under general anesthesia. The postoperative course was uneventful. There was no evidence of recurrence 1 year after the operation.

Histopathological examination revealed that the proliferation of non-keratinizing squamous epithelium was found adjacent to the lining epithelium of the cystic lesion (Fig. 4). The intraepithelial infiltration of polymorphonuclear leukocytes and macrophages were observed in the epithelial proliferation. Immunohistochemical analysis revealed that positive reaction for cytokeratin was found in the cytoplasm of the proliferating cells, suggesting almost all of the cells were epithelial in nature (Fig. 5). On the other hand, immunoreactivity for CD-68, a marker of macrophages, was observed in the cytoplasm of scattered cells among these epithelial cells (Fig. 6). According to these findings, a final diagnosis of an eruption cyst with epithelial hyperplasia was made.

DISCUSSION

Developmental cystic anomalies diagnosed in the mouth of newborn infants include Epstein pearls, Bohn’s nodules, and eruption cysts. Epstein pearls are gingival cysts of infants that occur in the mid-palatine raphe region very near the mucosal surface. Cysts arising on the buccal and lingual aspects of the alveolar ridges of the first primary molar are called Bohn’s nodules. These cysts usually resolve themselves within a few months from birth without any surgical intervention. On the other hand, eruption cysts of newborn infants are associated with the crown of an erupting primary tooth. Therefore, this cyst may remain and increase in size until eruption of the associated tooth.

Diagnosis of these cystic lesions may be made easily, based on the characteristic clinical appearance. An eruption cyst usually appears as a bluish translucent, elevated, compressible, dome-shaped lesion of the alveolar ridge. The size is variable and dependent on the size and number of the associated teeth.
Fig. 4 HE findings reveal sheets of non-keratinized stratified squamous epithelial cells along with inflammatory infiltrates. (Original magnification × 50)

Fig. 5 A positive immunoreaction for total keratin is seen in almost of the all cells. (Original magnification × 100)

Radiographic findings on eruption cysts sometimes reveal a crescent-shaped radiolucent area above the crown of the tooth. But, in general, eruption cyst is not detected on radiological examination because this cyst usually does not involve the surrounding bone. Therefore, it is not difficult to distinguish an eruption cyst from a dentigerous cyst, an odontogenic tumor that mimics a dentigerous cyst, or an odontogenic keratocyst on a radiograph. Other possibilities to rule out in the differential diagnosis for an eruption cyst in infants may include a congenital epulis, teratoma, neuroectodermal tumor, myoblastoma, and more serious tumors.

Treatment of an eruption cyst is not performed immediately in infants. Even histopathological examination in establishing the final diagnosis is not essential. However, surgical intervention should be considered in order for the infant and the parents to lead a healthy and comfortable life, when the cyst is enlarging, symptomatic, or interfering with feeding; and more serious tumors can not be ruled out completely. Marsupialization is the surgical treatment of choice for an eruption cyst. The dome of the cyst is excised, exposing the crown of the tooth. Any malformed tooth associated with the cyst should be extracted rather than marsupialized. The excised specimen must be examined in order to make a definitive diagnosis of an eruption cyst. The prognosis of an eruption cyst is good. When a biopsy is not performed or the definitive diagnosis is not decided histopathologically from the biopsy results, an eruption cyst requires close monitoring for changes in size and appearance, because of the rare possibility of its developing into a tumor.

In the present case, the clinical diagnosis of a tumor on the mandible was made, because of the recurrence after the first treatment at the dental clinic. A biopsy was performed from the dome of the lesion. Since the histopathological findings on the biopsy specimen suggested a more serious tumor, further surgical intervention was considered. The final diagnosis of an eruption cyst with epithelial hyperplasia was made. We concluded that the obvious epithelial proliferation and thickening was led by the chronic inflammatory infiltrate, which might have been the result of occlusal trauma. Although the lumen of an eruption cyst is usually lined with a thin layer of non-keratinized stratified squamous epithelium, the presence of a chronic inflammatory infiltrate may lead to proliferation and thickening of
the epithelial lining 2). The present case emphasizes the importance of careful clinical and histopathological examinations for diagnosis of an eruption cyst with obvious epithelial hyperplasia.

REFERENCE