Buccal Bifurcation Cyst: Two Case Reports and a Literature Review

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Introduction: *A buccal bifurcation cyst (BBC) is an uncommon inflammatory odontogenic cyst associated with the permanent mandibular first or second molar in children.* ***Case reports***: *These reports present two cases of BBC and describes the clinical and radiographic features leading to the diagnosis and the treatment of this lesion. Two patients complained of mandibular buccal swelling around the permanent first molar. The diagnosis of BBC in both cases was based on the clinical and radiographic features. In both cases, only enucleation was performed without extracting the involved tooth. Results: There were no recurrences during follow up. All teeth remained vital and erupted normally. Conclusion: The most appropriate treatment is usually enucleation of the cyst without extraction of the associated tooth. Therefore, knowledge of the distinct features of BBC is important for diagnosis and appropriate treatment.*

Key words: Buccal bifurcation cyst, Mandibular infected buccal cyst, Cyst enucleation, Oral surgery

INTRODUCTION

A buccal bifurcation cyst (BBC) is an uncommon inflammatory odontogenic cyst associated with the permanent mandibular first or second molar in children. Stoneman and Worth were the first to report the radiographic and clinical features of these cysts and described 17 cysts, which they labelled a "mandibular infected buccal cyst-molar area."[1] In 1992, the World Health Organization (WHO) included this lesion in the category "paradental cyst" and labelled it a "mandibular infected buccal cyst."[2] Shear and Speight suggested the term "juvenile paradental cyst,"[3] whereas Pompura *et al.* called the same entity a "mandibular BBC."[4]

The diagnosis of BBC is established based on its distinct clinical and radiographic features. Although a BBC is a variation of a paradental cyst, its site and age-specific features result in its designation.[4] The typical paradental cyst occurs distal or buccal to partially erupted mandibular third molars in patients with a history of pericoronitis, whereas a BBC occurs in children aged 4 to 14 years and usually involves the mandibular first molar or, occasionally, the mandibular second molar.[1,5,6] Clinically, deep periodontal pockets are observed on the buccal aspect of the involved tooth, with buccal expansion. Some cases show an altered eruption pattern of the involved tooth with crown buccal tilting.[4-7] The symptoms appear near the time of the eruption of the molar, and the patient has slight tenderness and discomfort on the buccal aspect of the tooth. The patient often notes swelling and complains of a foul-tasting discharge. The most common radiographic finding is a radiolucent lesion at the buccal furcation of the involved tooth. The periodontal ligament space and lamina dura of the involved tooth are of normal width and density.[3,11-12] The inferior border of the mandible is intact.[4] A periosteal reaction might be seen on the buccal surface.[1] The average size of the lesion is 1.2 cm, and the lesion usually extends from the furcation to the root apices.[4]

This report presents two cases of BBC and describes the clinical and radiographic features leading to the diagnosis and the treatment of this lesion.

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Case Report

Case 1

An 8-year-old boy presented to the Department of Pediatric Dentistry of the Kyung-Pook National University, Daegu, South Korea complaining of an abrupt asymmetric enlargement of the right side of his face that started the day before. His medical history was negative for systemic conditions or allergies. The extraoral examination revealed a painful swelling on the lower right side of the face. The intraoral examination revealed an operculum over the partially erupted lower mandibular right first molar that was tender to percussion.

A panoramic radiograph showed a well-defined, semilunar, radiolucent lesion at the roots and furcation of the mandibular right first molar (Fig. 1A). A periapical radiograph showed a

radiolucent lesion distal to the mandibular right first molar (Fig. 1B). A provisional diagnosis of pericoronitis was made due to the partial eruption of the mandibular first molar. To control the infection, a 3-day course of hexamidine solution and antibiotics was prescribed at the first visit. After 3 days, the pain and swelling on the lower right side of the face had disappeared. The patient was given oral hygiene instructions. The patient did not attend the 7-day follow-up. Five months later, he returned to the clinic complaining of a palpable mass and swelling in the same area. On palpation, buccal expansion covered by normal mucosa around the involved teeth was observed.

Cone beam computed tomography (CBCT) revealed a radiolucent cystic lesion at the buccal aspect of the furcation of the partially erupted mandibular first molar (Fig. 1C, D).

Fig. 1. (A) The panoramic radiograph shows a radiolucent well-defined semilunar lesion at the roots and furcation of the mandibular right first molar. (B) The periapical radiograph shows a radiolucent lesion distal to the furcation with cortical perforation of the mandibular right first molar. (C, D) CT shows a radiolucent cystic lesion on the buccal aspect of the tooth.

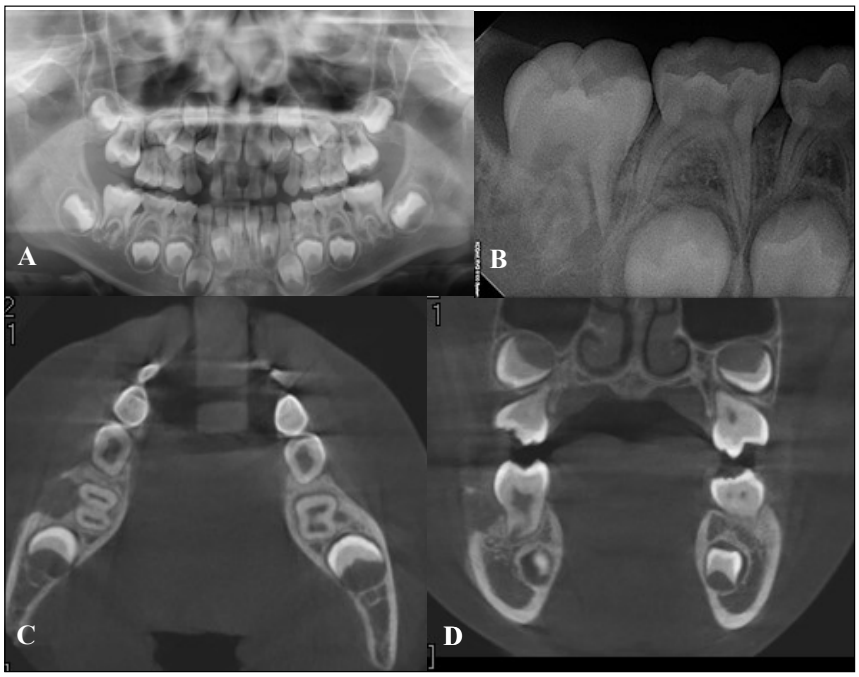
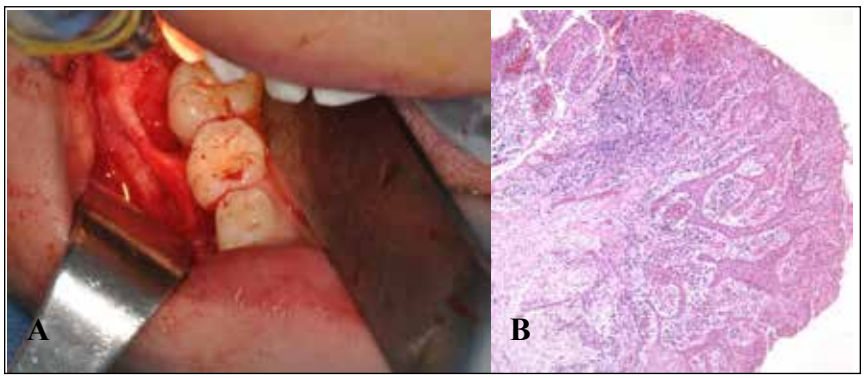


Fig. 2. (A) Operative view of the cyst enucleation. (B) A microphotograph shows the cystic lining with non-keratinized hyperplastic stratified squamous epithelium (hematoxylin-eosin stain, original magnification ×100).



We decided to perform surgical enucleation under general anesthesia without extracting the involved tooth (Fig. 2A). The surgical approach was via a full thickness flap with a gingival crevicular incision extending from #84 to the external oblique line of the mandible. The cyst was exposed and then enucleated, without extracting the involved tooth. Following irrigation with saline, the wound was closed with 4-0 Vicryl sutures.

Histologically, the cyst was lined with hyperplastic, non-keratinized, stratified squamous epithelium. The connective tissue wall showed signs of a chronic inflammatory cell infiltrate (Fig. 2B). At the 2-year follow-up, there was complete resolution of the cyst without recurrence.

Case 2

A 9-year-old boy was referred to the Department of Pediatric Dentistry with a "radicular" cyst. The extraoral examination revealed a painful swelling on the left side of his face. The intraoral examination revealed a buccally angulated mandibular left first molar with cavitation on its occlusal surface due to enamel hypoplasia. Enamel hypoplasia was observed bilaterally. The electric pulp test of the involved tooth was positive. Panoramic and periapical radiographs showed a well-defined ovoid radiopaque line extending apically on the mandibular left first molar (Fig. 3A,B). CBCT revealed a radiolucent lesion at the buccal furcation of the mandibular left first molar, causing buccal inclination of the involved tooth (Fig. 3C,D). There was communication with the oral cavity via the alveolar crest. The cyst was enucleated under local anesthesia without extracting the involved tooth (Fig. 4A). The histological findings were the same as in Case 1 (Fig. 4B). BBC was diagnosed based on the clinical, radiographic, and histological findings. At the 6-month follow-up, the patient had recovered without recurrence.

DISCUSSION

A buccal bifurcation cyst is a rare odontogenic cyst that comprises 3.0% of all cysts of the jaw and 3.7% of odontogenic cysts.[3] Some authors suggest that the true incidence is higher, as many such lesions might be misdiagnosed.

The etiology of BBC is still unclear. It has been postulated that the tooth breaks through the oral epithelium while erupting, causing localized inflammation beneath the epithelium.[1] Another theory suggests that the cystic epithelium is derived from the epithelial cell rests of Malassez or the reduced enamel epithelium.[4,8] The fact that the mesiobuccal cusp of the first molar is the first to break through the alveolar mucosa might explain the development of the lesion on the buccal surface at about the time of tooth eruption.[1,13] Histologically, BBC is non-specific and consists of a cystic lesion lined by non-keratinized hyperplastic stratified squamous epithelium and a chronic inflammatory infiltrate in the connective tissue wall. The cyst is identical to a radicular cyst.[1,5,7] Therefore, the diagnosis is based on its distinct clinical and radiographic characteristics. CT is useful, as the axial and coronal views may show buccal expansion in the BBC. The BBC may be associated with bone loss at the furcation of the involved tooth.[14] Several studies have reported a positive electric pulp test as a diagnostic criterion for BBC.[1,5,15] The diagnosis is a lateral radicular cyst if the involved tooth is nonvital.[16]

The treatment of this cyst has changed over time. Stoneman and Worth reported successful treatment with tooth extraction and curettage of the cyst.[1] Because extraction of the mandibular molar can have a significant impact on the permanent dentition, a more conservative approach avoiding tooth extraction was subsequently adopted. Pompura *et al.* reported success after cyst enucleation without concomitant extraction in children aged 5.5 to 11 years.[4] Similar successful treatments have been reported by Shohat,[7] Thikkurissy,[12] and Vedtofte and Paetorius.[16] Levarek *et al.* performed bone grafting in three cases after cyst enucleation, proposing that this enhanced bone regeneration, provided stability, and allowed soft tissue reattachment.[17] Some authors have suggested a more conservative approach without surgery. David *et al.* suggested a non-surgical approach involving either no treatment or daily irrigation of the periodontal pocket with saline.[13] It has been speculated that microtrauma and the periodontal pocket induce a small opening into the cyst, which results in "micro-marsupialization," allowing the cyst to depressurize and heal spontaneously.[6,7,13,18] Recently, Corona-Rodriguez *et al.*[6] and Zadik *et al.*[18] reported spontaneous resolution of cases of bilateral BBCs. In our cases, both cysts were enucleated without tooth extraction.

The intraoral examination of our first patient showed an operculum over the partially erupted mandibular right first molar with swelling on the lower right side of the face. Consequently, a provisional diagnosis of pericoronitis was made, which resulted from the plaque accumulation around the involved tooth. In the second case, the patient was referred from a local dental clinic for treatment of the mandibular left first molar, with a diagnosis of a radicular cyst. It is easy to misdiagnose BBC, as it is a rare odontogenic cyst. In the first case, the involved

Fig. 3. (A, B) Panoramic and periapical radiographs show a well-defined ovoid radiopaque line extending apically on the mandibular left first molar with buccal tilting of the crown. (C, D) CT shows a radiolucent lesion in the buccal furcation area of the mandibular left first molar.

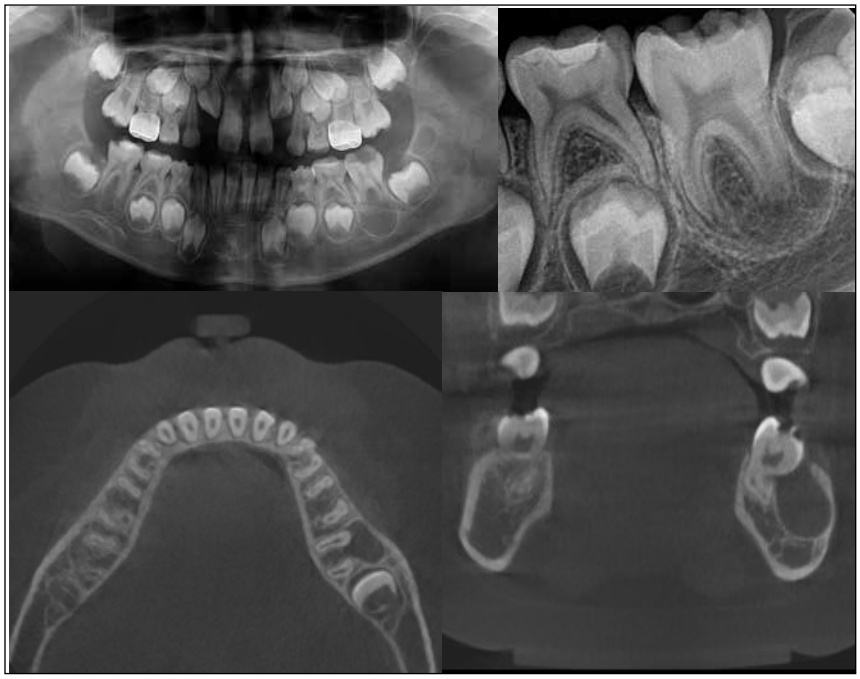
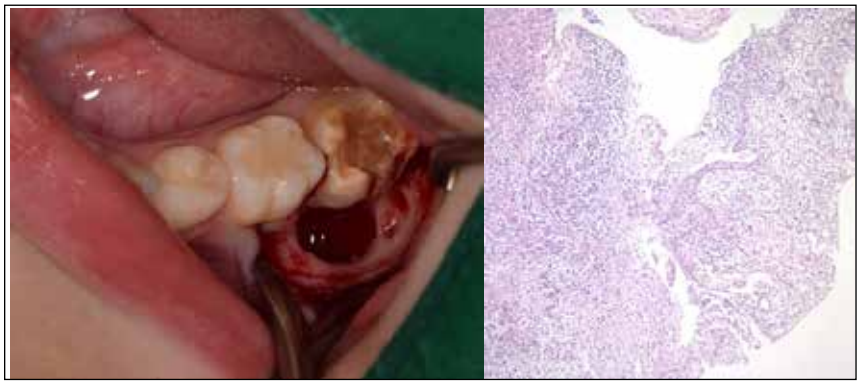


Fig. 4. (A) The operative photograph shows the cyst located at the buccal aspect of the tooth. (B) A microphotograph shows hyperplasia of the cyst lining with chronic inflammation (hematoxylin-eosin stain, original magnification ×100).



tooth was unerupted, whereas the electric pulp testing was positive in the second case. The diagnosis of BBC in both cases was based on the clinical and radiographic features. In both cases, only enucleation was performed without extracting the involved tooth, and healing was observed with radiographic evidence of cyst resolution. There were no recurrences during follow up. All teeth remained vital and erupted normally.

CONCLUSION

A buccal bifurcation cyst, which is a pediatric disease that occurs in the furcation area on the buccal aspect of an erupting permanent mandibular first or second molar, appears mainly in the first decade of life. The diagnosis is based on the clinical, radiological, and histological features. The most appropriate treatment is usually enucleation of the cyst without extraction of the associated tooth. This treatment has good long-term results according to the literature. Knowledge of the distinct features of BBC is important for diagnosis and treatment.

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