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## CASE REVIEW

# Update on a rare mandibular osteolytic lesion in childhood: the buccal bifurcation cyst

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## ABSTRACT

Buccal bifurcation cyst is a rare, uncommon buccal cystic lesion associated with the permanent mandibular first molar in children just prior to tooth eruption, and is categorized by the World Health Organization as an odontogenic cyst occurring in a vital tooth, near the buccal cervical margin of the lateral aspect of a root, as a result of an inflammatory process in a periodontal pocket. The aim of this study is to present a series of three similar mandibular buccal bifurcation cyst cases, by providing clinical, radiological and histological characterization of the lesion, in order to lead clinicians through the diagnosis, treatment and follow-up process and contribute to deeper knowledge of this rare pathological entity.

## INTRODUCTION

The buccal bifurcation cyst (BCC) is a rare, uncommon buccal-located cystic lesion associated with the permanent mandibular first molar in children just prior to tooth eruption.

According to the World Health Organization (WHO), the mandibular buccal bifurcation cyst (MBBC) is categorized as an odontogenic cyst and described as a cyst occurring in a vital tooth, near the buccal cervical margin of the lateral aspect of a root as a consequence of an inflammatory process in a periodontal pocket.<sup>1</sup>

MBBC has a slightly male predominance,<sup>2</sup> is most common in 6- to 15-year-old patients and is not found in adults. Moreover, it does not occur in the maxilla and can be bilateral.<sup>3</sup>

The associated tooth has positive reaction to vitality tests and has, generally, an altered eruption pattern, with a

typical buccal tilting of the crown and lingual tipping of the roots, which, in severe cases, can perforate the lingual cortical plate. The vitality of the tooth remains positive. In literature, no cases have reported interruption of the inferior border of the mandible.

The lesion was first described by Stoneman and Worth in 1983,<sup>4</sup> as the mandibular infected buccal cyst. In literature, however, there seems to be no consensus about the terminology. In fact, the buccal bifurcation cyst (BBC) is also described as mandibular buccal bifurcation cyst (MBBC), because of its site and age specificity,<sup>5,6</sup> or mandibular infected buccal cyst.<sup>7</sup>

On the contrary, some authors prefer to use the term infected paradental cyst, due to the assumption that MBBC is a variant of the paradental cyst, the inflammation is always present and the lesion is always located next to the root of the involved tooth.<sup>8</sup> A recent review of the WHO

classification of head and neck tumours has categorized the BBC and the paradental cyst into the group of inflammatory collateral cysts.

The pathogenesis of these cysts is also still debated, but they have most likely originated from reduced enamel epithelium or from the inflammatory proliferation of epithelial cell rests of Malassez that originate from the periodontal membrane of the buccal bifurcation of the mandibular molars. The aetiology of cystic degeneration is still unknown, but inflammation is believed to be the stimulus.

The aim of this paper is to report a case series of three clinical situations of MBBC, by providing clinical, radiological and histological characterization of the lesion, in order to lead clinicians through the diagnosis, treatment and follow-up process and contribute to deeper knowledge of this rare pathological entity.

## INVESTIGATIONS

### Case report 1

Female, 8 years old, patient referred to the oral and maxillofacial surgery department of Saint Rafaël hospital of Leuven, Belgium, for a maleruption of the left lower first molar. The girl did not have any complaints, beside the fact that the 3.6 was not erupting. She did not mention any pain or tenderness to palpation.

After clinical extra- and intraoral examination, a panoramic radiography was performed. A homogeneous round radiolucent image, of about 15 mm diameter, presenting a perilesional sclerotic edge, involved the entire radicular area and the cervical region of the left lower first molar. The lesion mesiodistally extended from the germ of the second premolar to the germ of the lower second molar.

A cone beam CT was carried out, presenting the buccal localization of the lesion with an extended resorption of the vestibular cortical plate and a periosteal reaction.

Surgical treatment was performed under general anaesthesia. A mucoperiosteal intrasulcular triangular flap was prepared from the distal side of the lower left second premolar to the distal margin of the lower left second molar. The jaw bone was deperiostized, and an osteotomy was done, in order to have access to the cyst. By finding the clivage plane, the lesion was enucleated and a curettage of the residual bony cavity was performed. The residual surgical area was thoroughly rinsed with sterile 0.9% NaCl, and the flap was replaced by simple interrupted stitches suture with resorbable suture (Vicryl 3/0).

Anatomopathological analysis revealed a vestibular cyst, microscopically characterized by inflammatory infiltration of neutrophils, plasma cells, lymphocytes and eosinophils. Based on clinical, radiographic and histological examination, the diagnosis of MBBC was established.

After 2 years, a follow-up cone beam CT was carried out in order to evaluate the eruption pattern of the mandibular first molar. It was obvious that the first molar erupted in the right place, still

showing some minor buccal tilting. A slight bone defect was still noticed on the buccal area at the site of the bifurcation (Figure 1). Interestingly, the second mandibular molar was mesially tilted and was not able to erupt due to impaction at the distal edge of the first mandibular molar (Figure 1 bis).

### Case report 2

Male, 7 years old, patient referred to the department of oral and maxillofacial surgery of the National and Kapodistrian University of Athens, Greece, complaining of a slowly progressing extraoral swelling in the lower right molar area.

Intraoral examination revealed that the lower right first molar was correctly erupted and was reacting positively to vitality tests. Panoramic radiography showed the presence of a well-defined homogeneous radiolucent round area of about 13 mm diameter, involving the roots and the cervical region of the lower right first molar, with an osteosclerotic peripheral edge. The lesion extended mesiodistally from the germ of the second premolar to the germ of the second molar. Further radiographic investigation, consisting of a cone beam CT, confirmed that the lesion was localized in the buccal aspect. Resorption of the vestibular plate and a periosteal reaction were also detected. Surgical treatment consisted in the extraction of the tooth, enucleation of the lesion and curettage of the residual bone cavity.

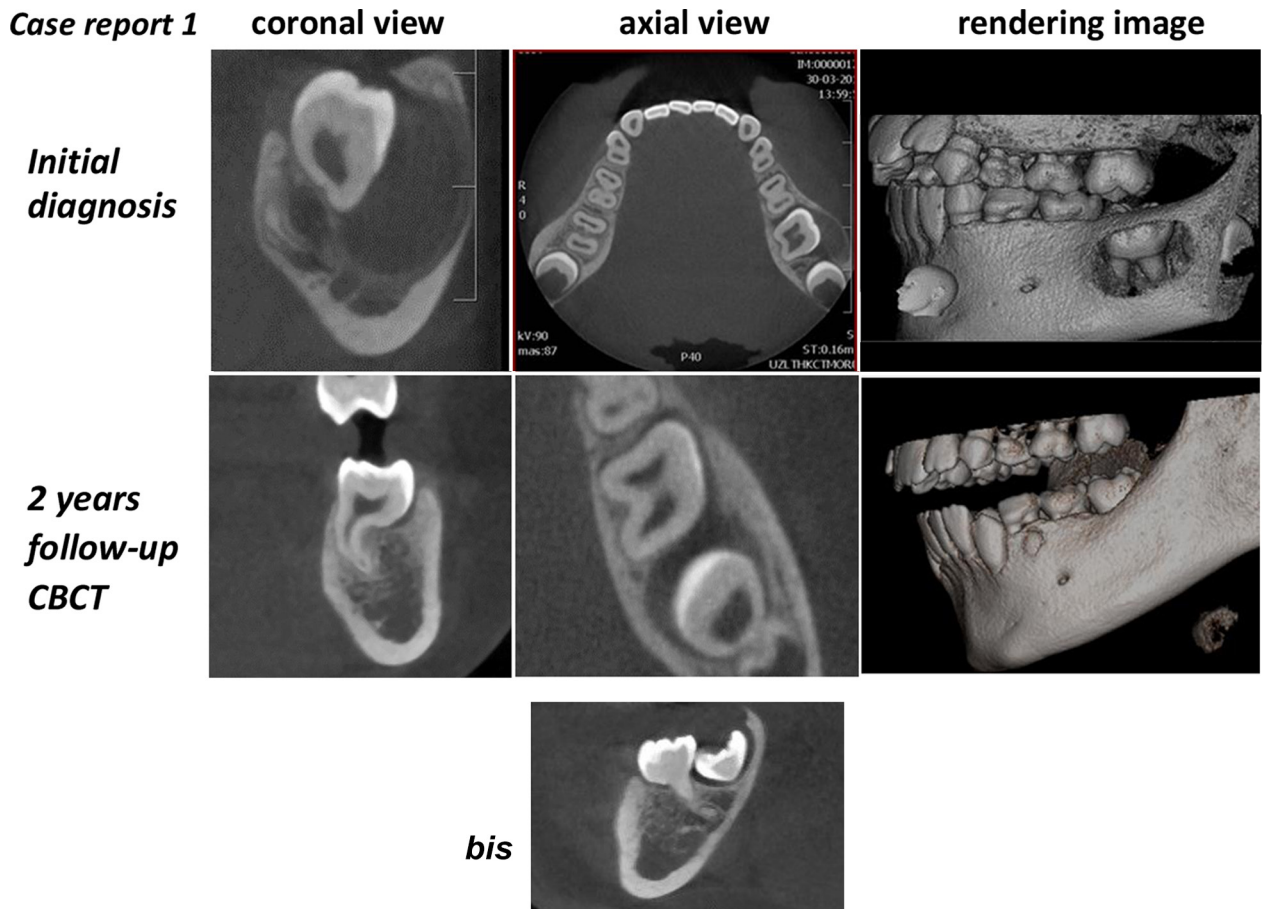
Anatomopathological examination showed a cystic cavity lined by non-keratinized squamous epithelium with elongated and interconnected rete pegs and inflammatory exocytosis. The tissue wall consisted of collagenized, vascular connective tissue with moderate to intense foci of inflammatory infiltration, mostly in subepithelial distribution, and bone spicules. Thus, the diagnostic hypothesis of MBBC was confirmed by this histological analysis (Figure 2).

### Case report 3

Male, 6 years old, was referred to the oral and maxillofacial surgery department of Saint Rafaël hospital of Leuven, Belgium, complaining of a painless facial swelling and small extraoral haematoma in the lower left molar area (Figure 3). Clinical extra- and intraoral examination revealed a partially erupted lower left first molar and the affected area was tender to palpation.

Radiological examination was initially done by panoramic imaging, showing the presence of a well-defined homogeneous radiolucent round area with perilesional sclerotic edge, involving the distal root and the cervical region of the lower left molar. The lesion was mesiodistally extended from the furcation of the first lower left molar to the mesial side of the second lower left molar germ. Panoramic imaging also revealed a presumption of hypomineralization of the second lower left molar germ compared to the other second molar germs. No suggestions were found for this alteration. Further radiographic examination was carried out through a cone beam CT, confirming the buccal aspect of the lesion, and showing a resorption of the vestibular cortical plate, where a periosteal reaction was noticed. The lesions were measured on the cone beam CT images and were found to be 10.82 mm in mesiodistal direction by 6.87 mm in vestibular dimension.

Figure 1. 2-year follow-up showing complete healing of the vestibular area.

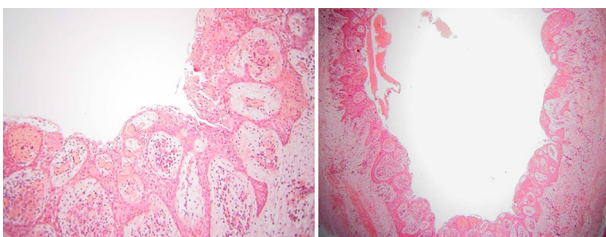


Surgical treatment was performed under general anaesthesia. The same procedure as in case 1 was maintained.

Anatomopathological examination of the cystic tissue also revealed a vestibular cyst, microscopically defined by a non-keratinized- squamous epithelium, consisting of fibrinopurulent material and granulation tissue with a mixed inflammatory infiltrate, which is in consistency with the other cases described in this paper (Figure 4).

Clinical, radiological and anatomopathological examination results confirmed the diagnosis of MBBC.

Figure 2. Histological slices of the buccal bifurcation cyst, showing a cystic cavity, lined by non-keratinized squamous epithelium with elongated and interconnected rete pegs and inflammatory exocytosis.



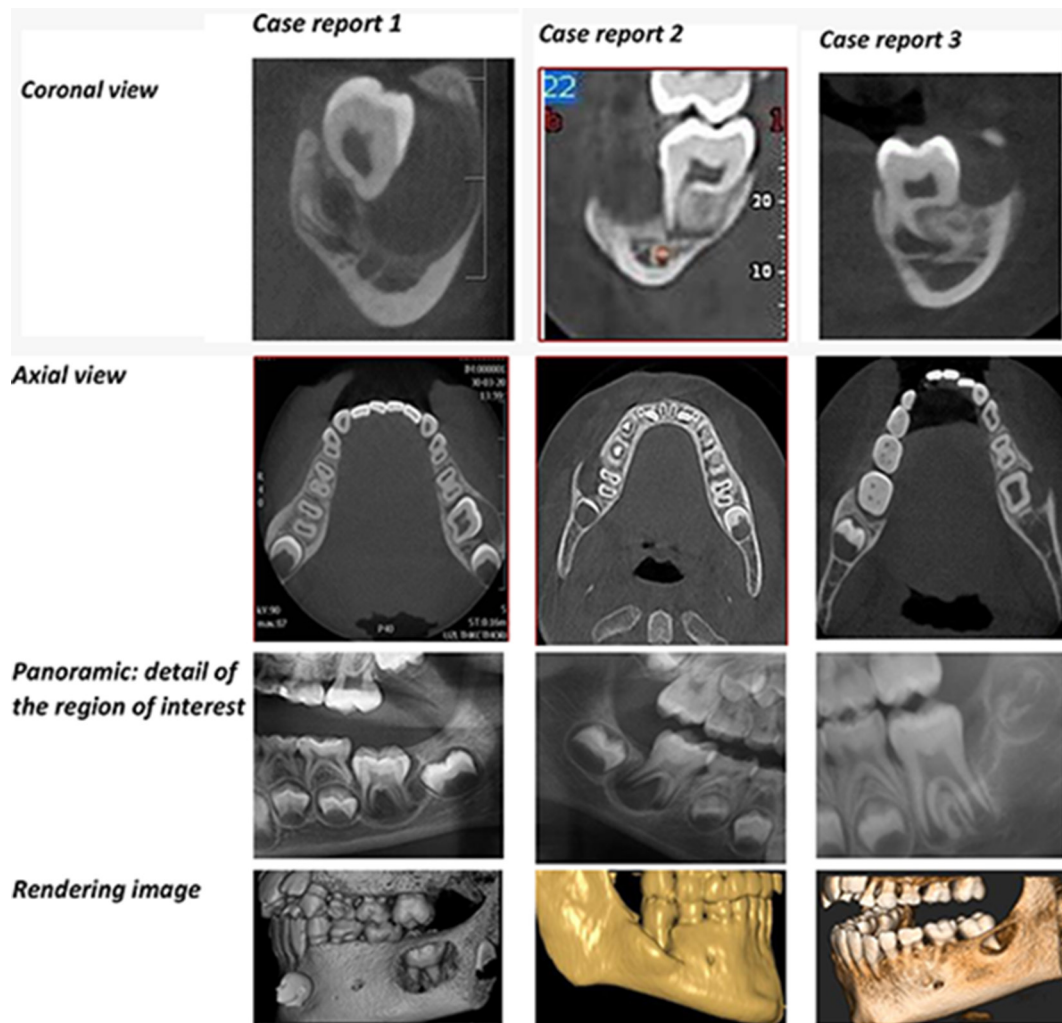
**DISCUSSION**

The reported relative frequency in percentage of MBBCs in relation to all odontogenic cysts varies between 0.9 and 4.7%.<sup>8</sup> The cases reported in this study were in line with the data in the literature; the patients were 6–8 years old and two out of three were

Figure 3. Extraoral swelling due to bone expansion. The tenderness to palpation and the redness of the extraoral tissue emphasize the inflammatory character of the MBBC.



Figure 4. The similarity of all three cases can be found in the presence of a well-defined homogeneous radiolucent round area at the vestibular side of the lower first mandibular molar. In all cases, the vestibular cortical plate was fenestrated, showing a periosteal reaction.



male. In each case, the involved tooth was a mandibular first molar. No bilateral lesions were detected.

The authors agree that the associated teeth show an altered eruption pattern, with a buccal tilting of the crown and a deep periodontal pocket on the buccal side. The vitality of the tooth always remains positive, and there can be some swelling, pain or tenderness, according to the degree of infection. No paresthesia is noticed in any known case, although the inferior alveolar nerve can be displaced lingually. Deep periodontal pockets can be measured on the buccal aspect of the tooth without gingival inflammation, and usually intraoral bony expansion, tender to palpation, is noticed.

In this study, affected teeth were vital and an abnormal eruption pattern, representing the reason for the consultation, was detected in every case. In one case, the involved molar was completely erupted. A buccal bone defect without signs of inflammation was noticed in each patient and, in two cases, intra- and extraoral swellings were present. No tenderness was appreciated

on palpation examination, except for case report 3, where the affected area was tender to palpation due to the swelling. In none of our cases, signs of alteration of sensitivity in the inferior alveolar nerve region were encountered.

The pathogenesis of the BBC remains unclear owing to the fact that the epithelial source cannot be identified. Several developmental theories have been proposed, but none has a clear outcome. Thus, the cyst can have an origin from the crevicular epithelium, the cell rests of Malassez, the reduced enamel epithelium or the dental follicle. A common certainty exists in the belief that a local inflammatory stimulus beneath the epithelial junction has an important role in epithelial proliferation. The mesiobuccal cusp is the first to break through the epithelium, and can induce communication between the pericoronal space and the oral cavity, which can generate inflammation at the epithelial attachment area.<sup>6,9</sup> This pathogenetic mechanism could justify the anatomopathological aspect of the lesion; in fact, the histological analysis of a BBC generally shows a proliferating non-keratinized, stratified squamous epithelium with an

arcading pattern. The cystic wall contains some dense, mature fibrous connective tissue, with an intense chronic inflammatory reaction presenting mononuclear and polymorphonuclear cells near the epithelium. In these cases, the cystic cavity was lined by non-keratinized squamous epithelium and the tissue wall consisted of collagenized connective tissue, presenting vascular components and inflammatory infiltration of neutrophils, plasma cells, lymphocytes and eosinophils. BBC is not universally accepted to be a distinct entity, because of the similar clinical, radiological, surgical and histological findings with other cysts. However, histologically, the lesion cannot be distinguished from a radicular cyst.<sup>10</sup>

The differential diagnosis with a lateral or radicular cyst lies in the fact that the electric pulp test of the involved tooth is negative.<sup>11</sup> There are, however, some similarities between BBCs, paradental cysts and dentigerous cysts. All of these pathologies involve the crown of the tooth. Particularly, the paradental cyst and the dentigerous cyst often occur in the third mandibular molars. Paradental cysts have an inflammatory origin, just like the BBC. Moreover, both arise on the lateral side of the tooth, and are associated with tooth eruption. Given this similar radiological characteristics of BBC and paradental cyst, we should emphasize that BBC develops more commonly on the buccal side of the first and second molar in childhood, and comes with buccal expansion of the bone. Radiographically, the lesion is well defined, radiolucent, circumscribing the roots of the affected tooth, showing an intact lamina dura. Cone beam CT is certainly useful in the delineation of the lesion. Buccal expansion, interradicular bone loss, bone cavitation and cystic content can indicate the differential diagnosis of BBC.<sup>12</sup>

In the presented cases, the histological aspect could also refer to the diagnosis of inflammatory cyst, but the integration of data concerning the very typical clinical and radiographic characteristics and especially the vitality of the affected teeth lead to confirmation of the diagnosis of bifurcation cysts. In literature, different treatment options are reported (depending on the extent of the lesion and the practising surgeon). Enucleation of the lesion without tooth extraction is the most common option and the treatment of choice.<sup>13-15</sup> In several cases, extraction of the tooth has been advocated and was found necessary; however, to date, no recurrences have been published.<sup>5,6,14,16-18</sup>

Non-surgical approach can also be established through probing the cyst and daily rinsing with saline. The marsupialization technique is most uncommon, and depends on the patient. The bifurcation cysts in this paper were enucleated without tooth extraction, except for one case, in which the involved tooth was removed by radical surgery.

In every case, after surgical treatment, clinical and radiological follow-up did not show any recurrence. Whenever not extracted, teeth could erupt normally and the vitality was kept uncompromised.

## LEARNING POINTS

1. Beware of unilateral eruption of mandibular first molars in
  - children aged between 6 and 15 years
  - extraoral swelling
  - buccal tilting of the unerupted tooth
  - vital tooth (differential diagnosis with radicular cyst)
2. Radiologic characteristics:
  - Well-demarcated radiolucency with corticated margin involving the entire radicular area and the cervical region of the lower first or second molar
  - Intact lamina dura
  - Cone beam CT:
    - buccal expansion, sometimes interradicular bone loss
    - periosteal bone reaction
    - resorption of the vestibular plate
3. Histological analysis together with data concerning age, radiological examination (cone beam CT) and vitality testing leads to the diagnosis of MBBC.
4. Best treatment option: enucleation of the cyst, no extraction if not indicated.
5. No recurrence, normal eruption.
6. Proper healing after surgical enucleation.

## INFORMED CONSENT

Written informed consent for the case to be published (including images, case history and data) was obtained from the patients for publication of this case report, including accompanying images.

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