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Bilateral mandibular buccal bifurcation cyst: a case report emphasizing the role of imaging examination in the diagnosis

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ABSTRACT

A mandibular buccal bifurcation cyst is an inflammatory cyst that usually occurs on the buccal aspect of the permanent mandibular first molar of children. This lesion is diagnosed by an association of radiographic, clinical, and histological features. We report a bilateral case of mandibular buccal bifurcation cyst and discuss the main findings of this entity. A 7-year-old girl presented pain and delayed dental eruption in the posterior mandibular region. A cone beam computed tomography was performed and revealed hypodense lesions involving the crown and root of the mandibular first molars, with expansion of the buccal cortical and lingual tilting of the molar roots. A biopsy was carried out, and the common features of an inflammatory odontogenic cyst were histologically observed. The final diagnosis was bilateral mandibular buccal bifurcation cyst. Clinicians need to be aware of this diagnostic possibility in cases of mandibular cysts in children—especially when bilateral—to perform the correct treatment, which should not involve the extraction of the affected tooth.

Keywords
Cysts; Jaw Cysts; Odontogenic Cysts.

INTRODUCTION

Mandibular buccal bifurcation cyst (MBBC) is an inflammatory odontogenic cyst that develops on the buccal aspect of the first or second permanent mandibular molar of children.[1] Stoneman and Worth described MBBC 36 years ago as a "mandibular infected buccal cyst."[2] Since then, this lesion has also been described as juvenile paradental cyst,[3] mandibular buccal bifurcation cyst,[1,4] inflammatory lateral periodontal cyst,[5] and inflammatory paradental cyst.[6]

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The World Health Organization classifies MBBC and paradental cyst as inflammatory collateral cysts.[1] Both originate from inflammation in pericoronal tissue and seem to present the same etiology. However, a criterion has been used to distinguish each cyst: the paradental cyst is observed in the lower third molars, while the MBBC occurs in the buccal aspect of the mandibular first and second molars.[1,7] In spite of this, many cases of MBBC are found in the literature with the nomenclature of paradental cyst.[6]

MBBC is commonly misinterpreted due to the variety of names for the same lesion and the histological or clinical similarities with other entities. The aims of this paper are to report a well-documented case of a MBBC and discuss the main clinical, histological, and image findings of this lesion, in order to make clinicians aware of the most common features and treatments for this entity.

CASE REPORT

A 7-year-old female patient presented with pain and delayed mandibular molar eruption. Clinically, right and left mandibular buccal swelling of about 2–3 cm in diameter was observed. The local mucosa was more whitish than the surrounding one. The left first molar was not clinically erupted, while partial eruption of the right first molar was evident. Both regions were resistant to palpation.

Cone beam computed tomography (CBCT) was requested to establish the vestibular-lingual extension of the lesion, the relationship with the teeth, and the lamina dura maintenance. The images showed hypodense lesions involving the crowns and roots of the first mandibular molars. Basilar cortical resorption of the mandible was present on the right side. The evident expansion of the buccal cortical bone and the tilting of the molar roots toward the lingual cortex were observed (Figures 1 and 2).

These findings, as well as the relationship of the lesion with adjacent structures, were also analyzed in a three-dimensional volume rendering reconstruction (Figure 3). With the hypothesis of an odontogenic cyst, an excisional biopsy of the right side and ulectomy of the left side were performed. The histological analysis revealed cystic lesions partially lined by non-keratinized stratified epithelium, predominantly presenting a few layers of cells. Epithelial hyperplasia, exocytosis, and spongiosis were observed in some areas. The cyst wall, especially of the right side, had intense chronic subepithelial inflammation, which was mainly composed of lymphocytes and plasma cells (Figure 4). Based on the clinical, microscopic, radiographic, and CBCT images, the diagnosis was bilateral MBBC.

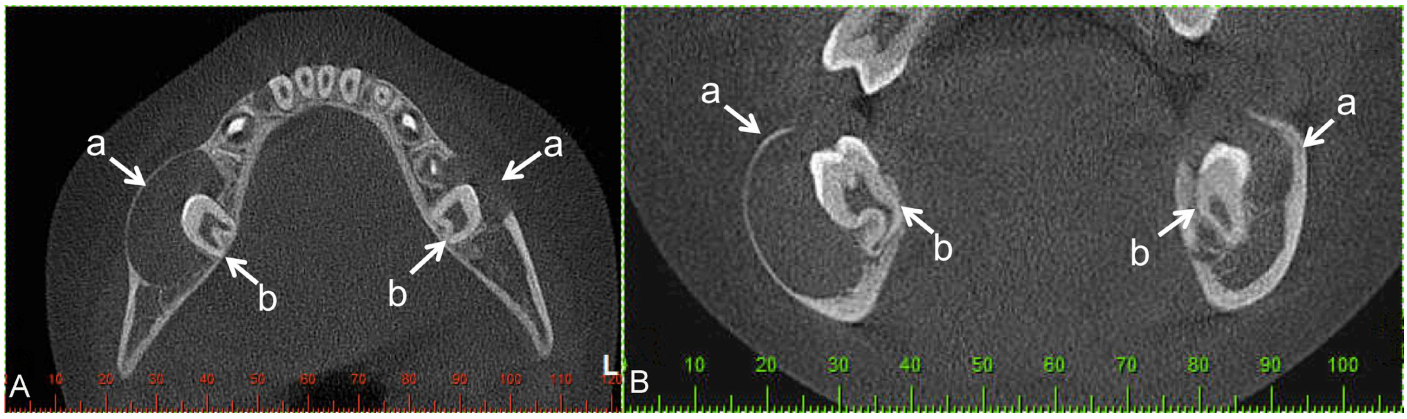

Figure 1. A – Axial view, demonstrating: (**a**) expansion and thinning of the buccal cortical bone of the posterior mandibular regions caused by the hypodense cystic lesions; (**b**) displacement of the permanent mandibular first molars to lingual side, with tilting of their coronary portions toward the buccal direction and of their apexes toward the lingual plate; L = left side. **B** – Coronal view showing the same findings described previously: (**a**) bulging of the buccal cortex; and (**b**) roots tilted toward the lingual cortex.

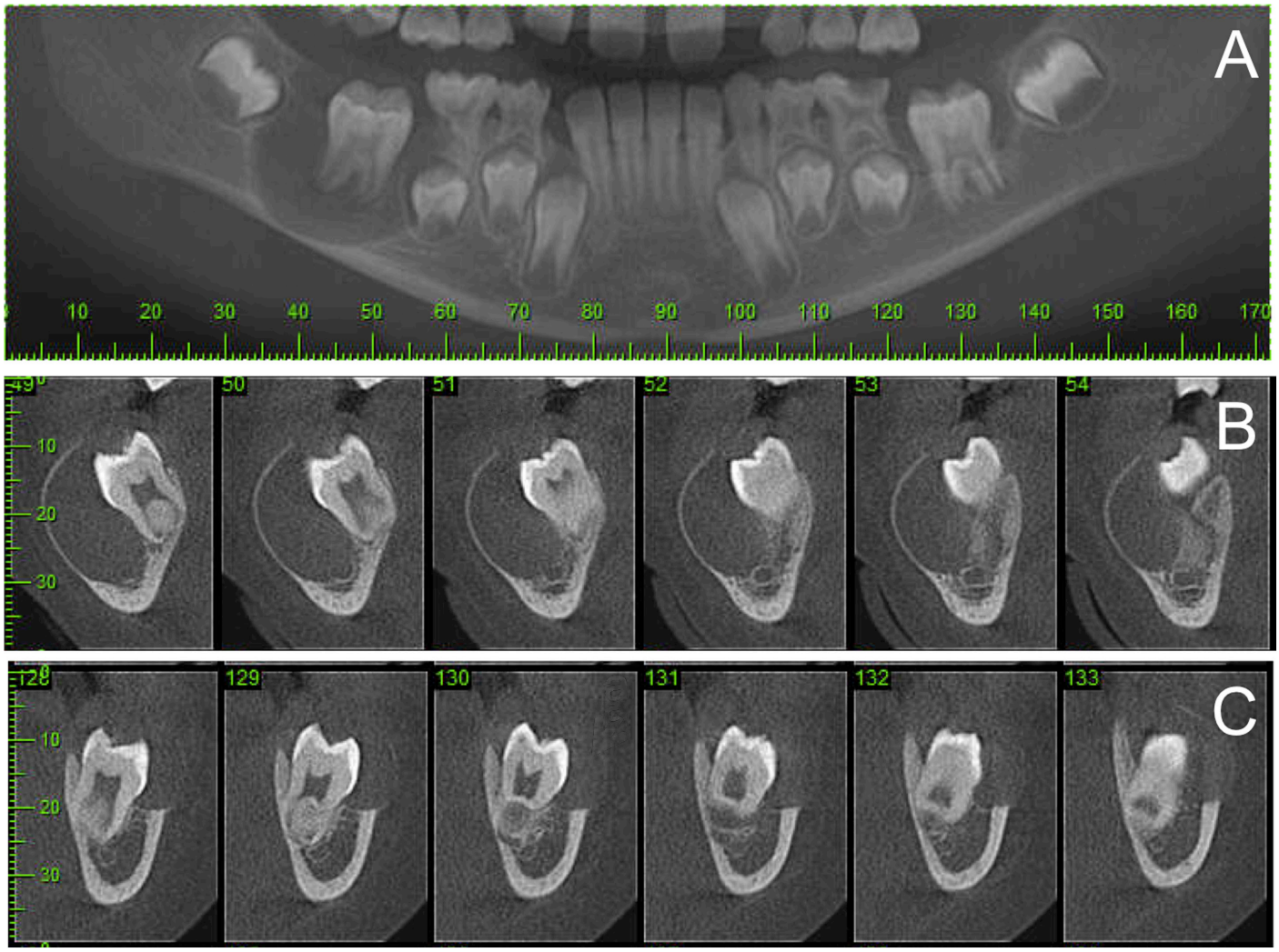

Figure 2. A – Central panoramic view in which a hypodense lesion may be observed bilaterally in the regions of permanent mandibular first molars. **B** and **C** – Transverse view of the right and left sides, respectively, in which buccal expansion of the cortical bone as well as the tilting of the teeth may be seen.

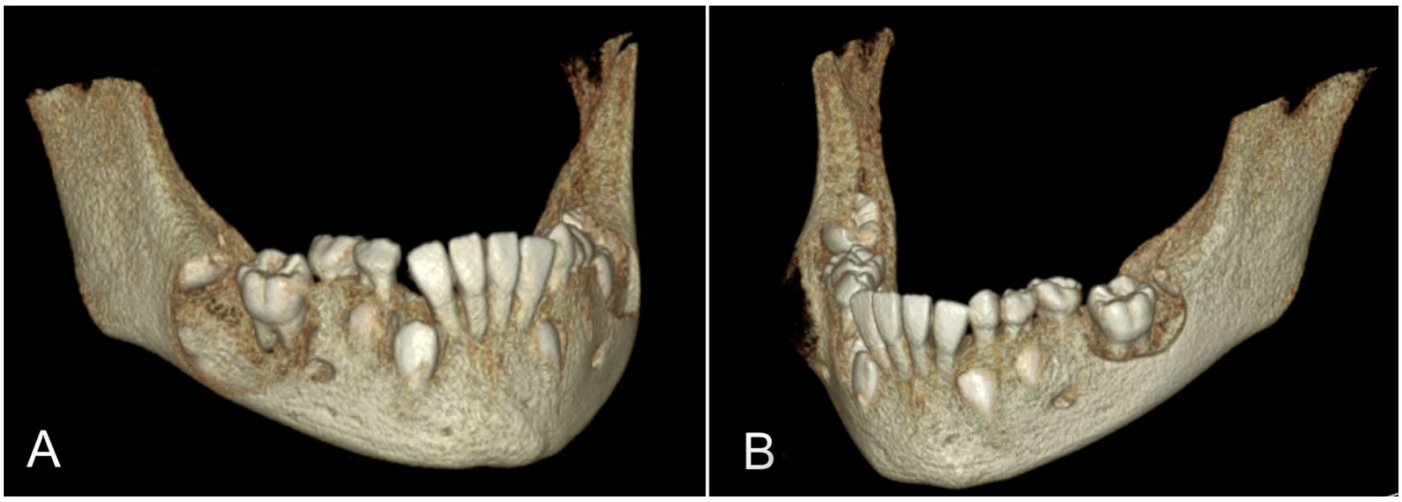

Figure 3. Three-dimensional reconstructions in bone protocol of the right (**A**) and left (**B**) mandibular regions, emphasizing the findings and the relation of the lesion to the adjacent structures.

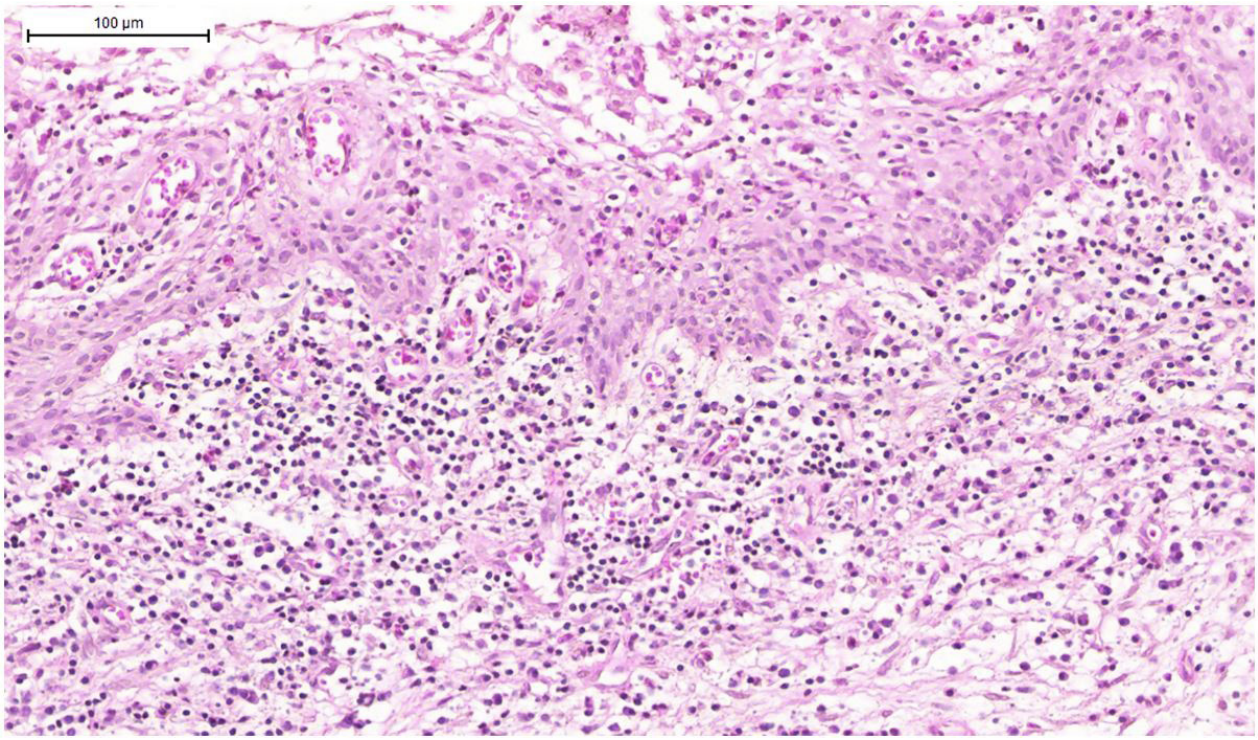

Figure 4. Photomicrograph of the lesion showing non-keratinized stratified epithelium with few layers of cells, exocytosis, and spongiosis covering the cyst wall that presented moderate chronic inflammation mainly composed of lymphocytes and plasma cells (H&E).

DISCUSSION

The etiology of MBBC is not clear, but it has been suggested that the cystic epithelium may arise from reduced enamel epithelium, cell rests of Malassez, remnants of the dental lamina or crevicular epithelium.[6] Some authors discarded the Malassez remnants origin once MBBC is not equally distributed around the root surface.[8]

Besides the origin of the cystic epithelium, the reason for the inflammatory process has also been discussed. The inflammation may be induced by food impaction in an opening pericoronal pocket (pericoronitis). The obstruction causes fluid accumulation within the blocked pocket, which leads to cystic expansion by osmosis.[9] Additionally, enamel projection into the furcation area of the tooth has been mentioned as an MBBC predisposing factor.[10,11]

Ramos et al.[12] reviewed the English language literature and found 16 manuscripts describing cases of MBBC, with a total of 56 cysts. In fact, the real epidemiology of MBBC is very difficult to evaluate, since different names are used for the same entity; for example, cases of inflammatory collateral cysts in the first or second molars have also been described as paradental cysts.[13-15]

Besides the variety of names, MBBC shares histological, clinical, and image features with other lesions.[7] The similar appearance of many cysts may lead to an incorrect diagnosis and inappropriate treatment. The careful analysis of imaging exams is an important aid in the recognition and distinction of cystic lesions. Although cysts usually present as well-defined radiolucent/hypodense lesions, different lesions may exhibit peculiar imaging features.

MBBC presents as a U-shaped radiolucent lesion that overlaps the roots. The space of the periodontal ligament and lamina dura remains unchanged. Tilted apexes toward the lingual cortical bone, the prominence of lingual cusps (due to inclination), an intact lower border of the mandible, expansion of the buccal cortical bone, a periosteal reaction, and displacement of adjacent non-erupted teeth also have been commonly observed.[1,4,16] Considering all these image features, the achievement of a three-dimensional analysis—as in the present case—is an important tool.

Clinically, MBBC usually presents an increase in the probing depth on the buccal gingiva. Other features, such as pain, swelling, and localized abscess, also may be seen. The clinical and radiographic characteristics of the present case are consistent with the previously published literature.[16,17]

Histologically, MBBC is lined by non-keratinized stratified epithelium with a fibrous cyst wall presenting intense chronic inflammation.[6,7,12,18,19] However, the diagnosis of this cyst cannot be ascertained only with the histopathological characteristics, if they are not specific. These features are common to all inflammatory cysts, which is a fact that strongly emphasizes the importance of the correct imaging interpretation.[6,7]

The dentigerous cyst, the lateral radicular cyst, and the periodontal pocket have been mentioned as possible differential diagnoses of MBBC.[7] The circumferential dentigerous cyst, which is characterized by the complete circumscription of the tooth,[20] may be a differential diagnosis of MBBC.[12] The cemento-enamel junction connects the cyst to the tooth, and it may occur in the first or second molars,[21] as MBBC, interfering with their eruption. Moreover, the histologic exam of a dentigerous cyst may show the presence of inflammation, hyperplasia, and leukocyte exocytosis,[7,22] similar to that observed in MBBC. In these cases, computed tomography may be an important aid, because through the different sections it is possible to observe the buccal expansion and the lingual tilting of the apexes, which are typical features of the MBBC.

A lateral radicular cyst is an inflammatory radicular lesion that shares histological features with other inflammatory cysts, including MBBC. However, a lateral radicular cyst is associated with a non-vital tooth, and a simple pulp vitality test may discard this hypothesis. A "paradental cyst" mimicking a periodontal pocket was described by Pelka et al.[14] in a tooth showing a probing depth of 15 mm on the buccal aspect. However, this probing depth is not expected in a recently erupted molar of a child.[14] In this kind of situation, the hypotheses of MBBC should be considered.

Bilateral cases of MBBC, as in this report, have been observed less frequently than unilateral cases.[6,12] It is important to emphasize that in the current report, one cyst was related to a non-erupted tooth. In a retrospective study, Pompura et al.[17] analyzed 44 MBBC, of which 6 were associated with non-erupted teeth. In 25% of the cases, a second subclinical contralateral MBBC was observed in the radiographic examination and confirmed at the time of surgery. Thus, when there is a hypothesis of MBBC, the clinicians must carefully evaluate the contralateral side.

The treatment of MBBC has changed significantly over the years. Initially, it was treated with enucleation and extraction of the involved tooth.[2,23] Currently, the treatment of choice has been enucleation without the extraction of the involved tooth. Recently, Levarek et al.[24] successfully used a bone graft as an adjuvant treatment after the enucleation and curettage of the cyst. Conservative treatment characterized by daily irrigation of the buccal pocket with a saline solution until the complete resolution of the lesion was reported,[16] as well as cases of self-resolution.[16,18,25] The potential self-resolving nature of MBBC has been suggested as this lesion is not observed in adults or in the maxilla.[17] A previous study conjectured that periodontal probing may induce a small opening in the cyst, causing a condition of "micromarsupialization," which permits depressurizing and cyst healing.[16]

In summary, the diagnosis of MBBC should be ascertained based on the correlation of clinical, imaging, and histopathological exams. Clinicians must recognize this entity and be aware of this diagnostic possibility in lesions occurring in the mandibular molars (especially bilateral) of children. The CBCT aspects were very useful in this case due to the vestibular and lingual inclinations of the teeth.

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The authors retain an informed consent signed by the patient's mother authorizing the publication of the clinical data and images. The manuscript is in accordance with the Institutional Ethics Committee.

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