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# Mandibular Buccal Bifurcation Cyst: Review and A Case Report

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## Abstract

Buccal bifurcation cyst (BBC) is a rare inflammatory odontogenic cyst that typically occurs at the buccal region of the first or second mandibular molars of children. It typically develops on the buccal aspect of the permanent mandibular first and second molars in younger patients (8 to 16 years old). Common clinical signs are the lack of or a delay in eruption of a mandibular first or second molar, swelling in the affected mandibular molar region, and an increase in periodontal pocket depth. Specific radiographic features include a radiolucent lesion on the buccal aspect of the tooth, tilting of the involved molar, and periosteal reaction on the buccal surface. However, the histopathologic features are inconclusive. In this report, we present a case of BBC with special emphasis on the characteristic radiographic features and differential diagnosis.

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## 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 [1-4]. According to the World Health Organization (WHO), themandibular buccal bifurcation cyst (MBBC) is categorizedas 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 [5]. The BBC and the paradental cyst are categorized into the group of inflammatory collateral cysts [6-12].

The inflamed mandibular buccal cyst was first described by Stoneman and Worth (1983) [2]. 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 [5,6] or mandibular infected buccal cyst [7]. 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 [8-10]. This entity is always associated with mandibular teeth.

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 [11-15]. The etiology of cystic degeneration is still unknown, but inflammation is believed to be the stimulus [16-18].

This paper presents a case of mandibular buccal bifurcation cyst and discusses its etiology, its clinical presentation, histogenesis and treatment modalities and its overlap in synonyms in the literature.

## Case Report

A 8 years old, patient referred to the oral and maxillofacial surgery clinic of HCF Dental Centre, Sydney, for a radiolucency around the erupting second lower molar. He was accompanied by his parents and medically fit. The boy did not have any complaints, beside the fact that the second molar was partially erupted was not erupting. He did not mention any pain or tenderness to palpation. Clinical examination revealed no swelling extra orally. There was buccal expansion in partially erupted 37 extending in the sulcus on intraoral examination. Teeth 36 and 37 were vital. Panoramic radiography revealed well-demarcated radiolucency bucco-distal of erupting 37, which was distally inclined (Figure 1). There was no abnormal periodontal pocket noted with 37. Patient was advised CBCT, which showed a well corticated, with mild expansion of buccal plate, 16/11mm radiolucency bucco-distal of 37 extending lingually (Figure 2). There was no sign of 38 formation. A clinical diagnosis of buccal bifurcation cyst was established.

He was advised surgical exploration and parents opted to be done under general anaesthesia.

Surgical treatment was performed under general anaesthesia. A triangular mucoperiosteal flap was raised buccally to access the area. The lesion was mainly seen extending buccally going around distal of 37, which was enucleated and sent for histopathological examination (Figure 3). The area was close primarily. Postoperative phase was uneventful. He was prescribed oral amoxicillin 500mg every eight hours for five days and analgesics in the form of paracetamol.

Histopathological report revealed the fibrous connective tissue wall lined in part by non-keratinising stratified squamous epithelium, which shows arcading, spongiosis and acute inflammatory cell exocytosis. The findings are consistent to inflammed buccal bifurcation cyst (Figure 4).

He came for four-month follow up with no complaints.

## Discussion

The 2017 WHO Histological Typing of Odontogenic Tumors defined the buccal bifurcation (paradental, inflammatory collateral, infected mandibular buccal bifurcation) cyst as an inflammatory odontogenic cyst arising on the lateral aspect of a vital tooth as a result of an inflammatory process in the periodontal pocket5. Lesions most often presenting a few years after the eruption of the associated tooth with slight male predominance.

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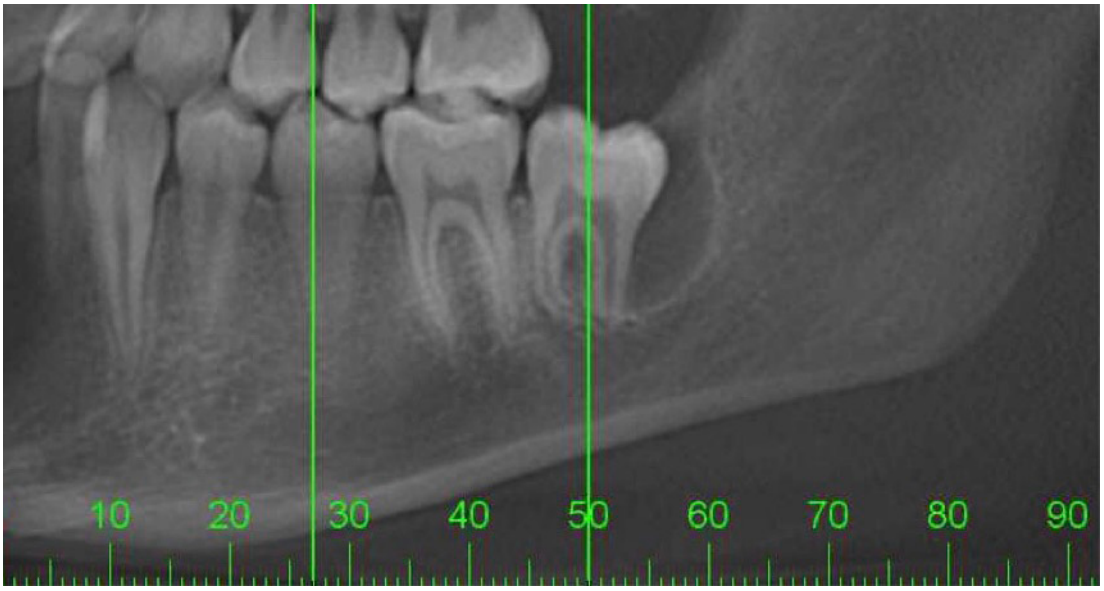


Figure 1: Sectional Orthopantomogram showing Well defined radiolucecy distal of tooth 37.

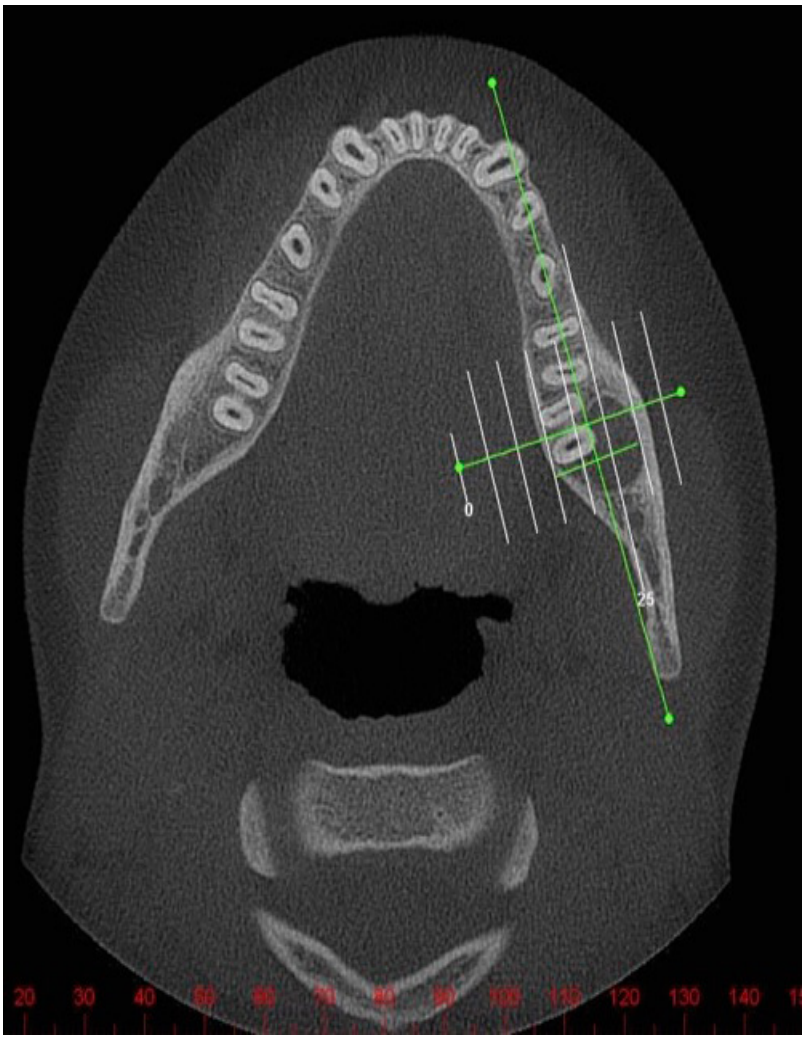


Figure 2: Axial cut of CBCT tooth 37 showing bucco-distal extension of radiolucency.

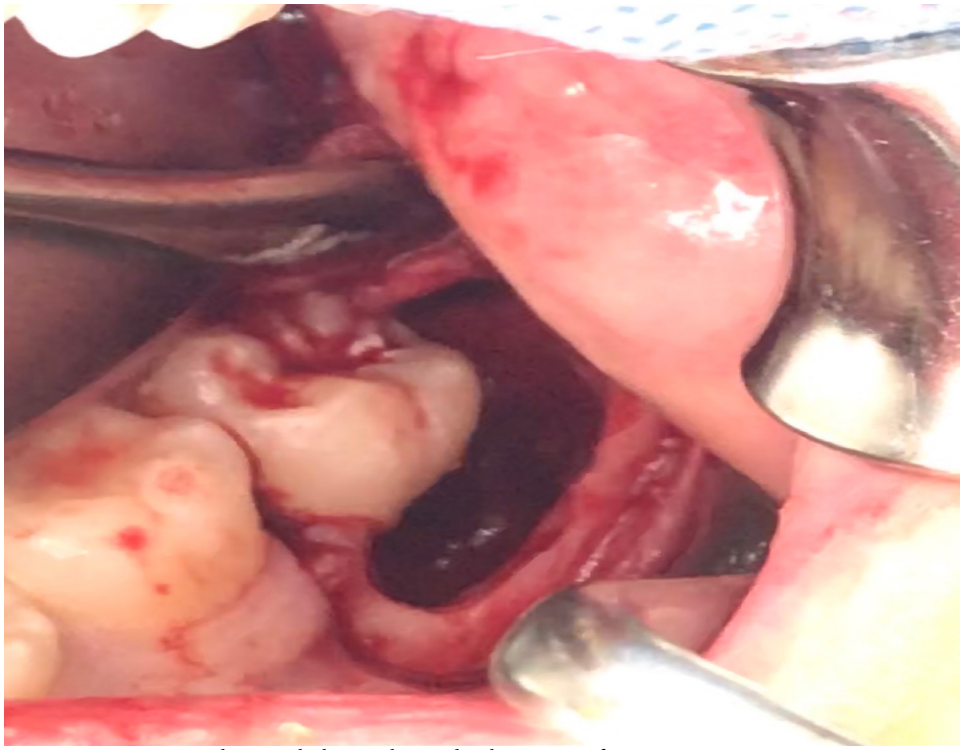

Figure 3: Intraoperative photograph showing bucco-distal extension of cyst.

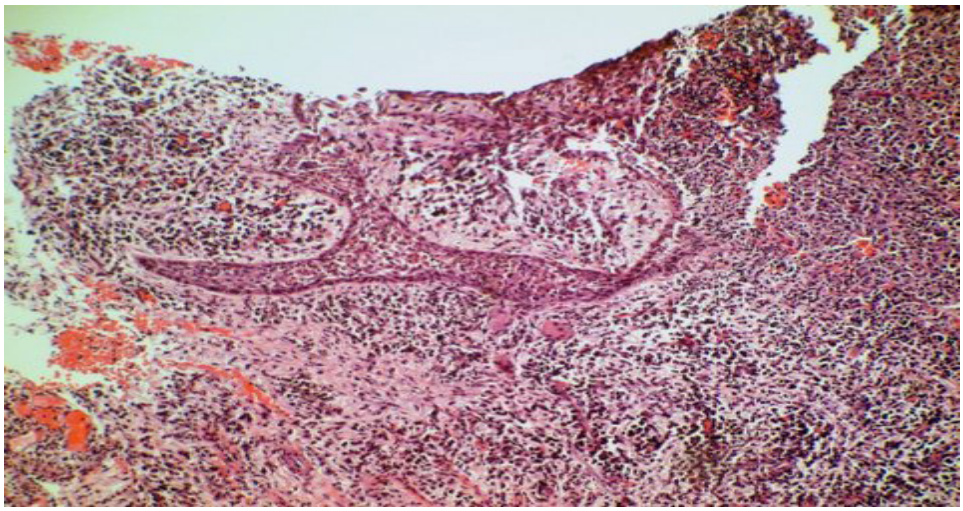

Figure 4: Photomicrograph showing fibrous connective tissue is lined in part by non-keratinising stratified squamous epithelium, which shows arcading, spongiosis and acute inflammatory cell exocytosis.

They typically affect the permanent mandibular first and second molar teeth. However, the first molar is involved more frequently than second molar. Bilateral lesions have been reported in 26% of all BBC cases [3,8,13,21,24,26,27,28,35,37,40-42].

To date approximately 73 cases of buccal bifurcation cysts have been reported within the literature through various case reports and cases series (Table 1). All described buccal bifurcation cysts have been reported to occur in association with either the mandibular first or mandibular second permanent molars [19-26]. The typical age of onset is between 4 and 14 years, as this time frame corresponds to the eruption of each of these respective teeth [27-31]. The true cause for the inflammatory response leading to cyst formation remains unclear. Some have postulated that cusp perforation through the oral mucosa during tooth eruption may represent a potential mechanism for formation [6,7]. Alternative explanations have also historically included the presence of enamel projections extending from the cement-enamel junction to the furcation area of the tooth. However, this theory remains questionable as most reported cases do not present with this feature [28,29]. Vedtofte and Praetorius [13] suggested the use of the descriptive term 'Inflammatory paradental cyst', because of its inflammatory origin and also due to its location at the side of the tooth. The use of inflammatory paradental cyst term has some limitation and overlap with lateral periodontal cyst, which is

of developmental origin. Instead author feels the mandibular buccal bifurcation cyst can be similar to the entity so called as juvenile inflammatory paradental cyst [32-37]. This fits in similar to presentations, age of occurrence, histological features and treatment modalities.

The histogenesis of this cyst has been widely discussed and three possible origins are generally accepted: crevicular epithelium, the cell rests of Malassez and the reduced enamel epithelium. Craig [12] has suggested that either the cell rests of Malassez or the reduced enamel epithelium might provide the cell of origin. He favored the latter source because in his study, the rest of Malassez always appeared inactive and that if the Malassez rests were responsible, the lesion should be equally distributed around the root surface. His serial sections indicated that the development of paradental cyst may follow hyperplasia and cystic change in reduced enamel epithelium. He suggested that the presence of an extension of reduced enamel epithelium over the enamel projections might be the source and could explain the frequent buccal location of the cyst. Ackermann et al. [16] favored origin from reduced enamel epithelium but suggested that cyst formation occurs as a result of unilateral expansion of the dental follicle secondary to inflammatory destruction of periodontium and the alveolar bone.

## Clinically, Buccal Bifurcation Cysts Present with a Classic Array of Findings

1. Involvement of partially erupted vital first or second molar,
2. Buccal expansion with soft tissue swelling,
3. Delayed or altered eruption of the involved tooth, and
4. An increase in periodontal pocket depth in the affected area.

## Radiographic Findings are also Classically Present

1. A radiolucent lesion located on the buccal aspect of the affected tooth,
2. Distal inclination of the involved molar with the root apices pointing toward the mandibular lingual cortex,
3. An intact periodontal ligament space and lamina dura, extension of the radiolucent lesion buccodistal of the mandible without alteration in the osseous anatomy of the cortex, and
4. A periosteal reaction on the buccal surface of the mandible (varying from a single layer to an onion-skin appearance).

| Year | Author | Number of cases | Treatment | Follow-up |
|---|---|---|---|---|
| 1970 | Stanback [35] | 1 Bilateral | Marsupialization | 2 years |
| 1979 | Fantasia [36] | 3 | No Data | No Data |
| 1980 | Swerdloff [37] | 2 (1 Bilateral) | Enucleation | 6 months |
| 1983 | Stoneman and Worth [19] | 17 | Enucleation/tooth extraction | No Data |
| 1985 | Trask et al. [38] | 1 (1 Bilateral) | Enucleation/tooth extraction | No Data |
| 1989 | Vedtolfe and Praetorius [13] | 5 (2 Bilateral) | Enucleation | 1–6 years |
| 1989 | Camarda et al. [39] | 2 | Enucleation | 5 years |
| 1990 | Packota et al. [40] | 5 (1Bilateral) | Enucleation | 6 months |
| 1992 | Bohay et al. [41] | 2 (1Bilateral) | Enucleation | 8 months |
| 1995 | Martinez-Conde et al. [27] | 2 (1Bilateral) | Enucleation/tooth extraction | No Data |
| 1998 | David et al. [21] | 3 (3 Bilateral) | No procedure/Irrigation with saline and hydrogen peroxide in 1 case | 1–2 years |
| 2003 | Shohat [24] | 5 (2 Bilateral) | Enucleation/tooth extraction | 2–3 years |
| 2007 | Gallego [42] | 1 Bilateral | Enucleation left side/no procedure right side | 1 year |
| 2009 | Iatrou [33] | 4 | Enucleation/tooth extraction | No Data |
| 2010 | Borgonovo et al [28] | 2 | Enucleation | 1 year |
| 2011 | Corona-Rodriguez et al. [8] | 1 Bilateral | Enucleation left side/no procedure right side | 6 months |
| 2011 | Chrcanovic et al [11] | 1 | Enucleation | No Data |
| 2011 | Zadik Y et al [4] | 1 | Self Resolution | No Data |
| 2011 | Lizio G et al [32] | 5 | Marsupialization | 2.8 years |
| 2012 | Ramos L et al [26] | 1Bilateral | Curettage | 1 year |
| 2012 | Borgonovo et al [3] | 1 Bilateral | Ennucleation | 15 months |
| 2013 | Borgonovo et al [29] | 1 | Ennucleation | 10 months |
| 2013 | Isser A et al [44] | 2 | Enucleation | 18 & 2 months |
| 2014 | Borgonovo et al [25] | 1 | Ennucleation | 11 months |
| 2014 | Friedrich et al [6] | 1 | Ennucleation | No Data |
| 2018 | De Grauwe A et al [20] | 3 | Enucleation with 0ne extraction | 2 years |

Table 1: Patients reported with buccal bifurcation cyst.

## Histologically

1. Buccal bifurcation cysts demonstrate an epithelial lining composed of nonkeratinized stratified squamous epithelium with focal areas of hyperplasia.
2. There is often a dense inflammatory infiltrate involving both the connective tissue wall and the lining epithelium of the cyst, which is analogous to the histologic findings seen in other types of inflammatory odontogenic cysts (ie, radicular cysts, residual periapical cysts, lateral radicular cysts).
3. The diagnosis of the BBC cannot be made from the histopathologic features, because these are nonspecific; therefore, the diagnosis of the MBBC should be made according to its distinctive clinical and radiographic characteristics.

## Treatment

The treatment of the BBC has evolved over time and has been controversial. Stoneman and Worth [2] reported successful treatment using tooth extraction and curettage of the lesion, as well as with endodontic treatment of the tooth and curettage of the lesion. Enucleation and extraction were reported by Martinez-Condeet al. [27]. Others have reported complete success after enucleation of the cyst without tooth extraction [20,25,37,38]. Recently, a more conservative approach has been suggested, in which the lesion has resolved without surgery, by either periodontal probing or daily irrigation of the buccal pocket with saline [4,21]. The latter investigators [21] speculated that periodontal probing induces a small opening into the cyst that results in "micro marsupialization," allowing the cyst to depressurize and heal spontaneously.

These non surgical regimens have been tried only in a limited number of cases, and therefore cyst enucleation without tooth extraction remains the current treatment modality of choice. Prognosis for buccal bifurcation cysts after enucleation is excellent. In the largest prospective series on outcomes for buccal bifurcation cyst, no recurrences were noted after cyst enucleation and maintenance of the involved teeth.

The case presented is a good example of buccal bifurcation cyst and treated successfully with ennucleation.

## Conclusion

Buccal bifurcation cyst is a rare, seen in children with erupting first and second lower molars. It is categorized as of inflammatory origin by World Health Organisation most likely arising from inflamed periodontal pocket of erupting molars. Ennucleation is the treatment of choice with removal of tooth. Recurrence is unheard of and tooth erupts normally.

## Competing Interests

The author declare that they have no competing interests.

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