Botryoid odontogenic cyst: A clinicopathologic study of 10 cases

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Abstract
Botryoid odontogenic cyst is a rare multilocular variant of lateral periodontal cysts. In this study, a series of 10 cases of botryoid odontogenic cysts retrieved from the archives of the Postgraduation Program in Oral Pathology, Federal University of Rio Grande do Norte (Brazil), were reviewed for epidemiologic data, clinical presentation, radiographic and histopathologic characteristics, treatment, and recurrence.

Keywords: Botryoid odontogenic cyst; Recurrent botryoid cyst; Lateral periodontal cyst; Multicystic variant

1. Introduction
Botryoid odontogenic cyst (BOC) is a multicystic variant of lateral periodontal cysts (LPCs) and was first described by Weathers and Waldron [1] in 1973. The histogenesis of LPC and BOC remains uncertain, although a number of possible sources of odontogenic epithelium, including the rests of Malassez and reduced enamel epithelium, have been ruled out as candidates [2].

Botryoid odontogenic cyst preferentially involves the mandibular premolar-canine area, followed by the anterior region of the maxilla [3]. Adults older than 50 years are the most affected group [4]. Although most of the reported cases of BOC have shown a multilocular radiolucency, some authors demonstrated that this lesion frequently presents as a unilocular radiographic image, thus, resembling a variety of other odontogenic cysts or neoplastic conditions [5].

Botryoid cysts are characterized by multiple cystic spaces and variations in the thickness of the epithelial lining, accompanied by epithelial protrusions into the lumen [6]. Glycogen-containing clear cells may also be present, they lack mucus-producing cells, mucous pools, crypts, intraepithelial microcysts, ductlike structures, or “hobnail” cells. The same reasoning would be true for LPC, which are essentially botryoid cysts with a single space [7,8]. The present article reports 10 additional cases of BOC, and the literature is reviewed regarding clinicopathologic aspects, differential diagnosis, therapeutic management, and recurrence of this rare odontogenic cyst.

2. Case reports
Ten cases of BOC were identified in the archives of the Postgraduation Program in Oral Pathology, Federal University of Rio Grande do Norte (Brazil). The clinical and radiographic characteristics of all cases are summarized in Table 1 and are illustrated in Figs. 1 and 2. Histopathologic features are summarized in Table 2 and are illustrated in Figs. 3 to 7.

3. Discussion
Botryoid odontogenic cyst is an unusual and controversial pathologic condition. By definition, LPC is a nonkeratinized developmental cyst that occurs in alveolar bone along the lateral aspect of a vital tooth. In this respect, BOC could be defined as a “multilocular variant of LPC” [9,10].
Analysis of the 10 BOC cases studied here showed a wide range in patient age from 34 to 83 years, with a mean age of 59.8 years. A male predominance was observed: 7 (70%) patients were males and 3 (30%) were females.

With respect to the site affected, 8 (80%) cases involved the mandible and 2 (20%) the maxilla. In the mandible, BOC was found in the anterior region in 3 cases, in the posterior region in 3, and in the anteroposterior region in 2. The anterior and posterior maxillary regions were involved in one case each.

Clinically, BOC manifested as a slow-growing lesion, which was symptomatic in 7 (70%) of the 10 cases and asymptomatic in 3 (30%). This odontogenic cyst measures 1.5 to 8.0 cm in diameter (mean, 2.88 cm). The reported duration varies from 2 to 10 years (Table 1).

The initial clinical diagnosis of BOC was ameloblastoma in 4 (40%) of the 10 cases and asymptomatic in 3 (30%). This odontogenic cyst measures 1.5 to 8.0 cm in diameter (mean, 2.88 cm). The reported duration varies from 2 to 10 years (Table 1).

Lateral periodontal cyst accounts for 0.8% of all central (intraosseous) cysts of the jaw bones [11]. This lesion develops in alveolar bone along the lateral aspect of an erupted vital tooth [12]. Clinical symptoms are usually absent, and the diagnosis is made by radiographic examination [2,12]. Lateral periodontal cyst appears as a solitary, well-defined interradicular radiolucency [2].

One feature that distinguishes BOC from LPC is the larger size of the former [13] because LPC has a limited growth potential. The importance of the differentiation between LPC and BOC is due to the histologic multilocular aspect of the latter that renders this lesion more expansive, thereby, increasing the possibility of recurrence because its complete surgical removal is more difficult [13,14].

In the present study, radiographic data were available for 9 of the 10 patients with BOC. Multilocular radiolucencies were detected in 6 cases (Fig. 1) and unilocular radiolucencies in 3. The differential diagnosis for a multilocular radiolucency within the jaws includes numerous conditions. Odontogenic keratocyst and ameloblastoma are the most common, with both lesions exhibiting a preference for the posterior mandible. Central giant cell granuloma also frequently affects the posterior mandible, although this tumor tends to involve anterior portions of the jaws. Less common conditions that can be included in the differential diagnosis are odontogenic myxoma, ameloblastic fibroma,
central odontogenic fibroma, and intraosseous mucoepidermoid carcinoma. A variety of other rare conditions, such as odontogenic tumors, odontogenic cysts, and other benign lesions, may also appear as a multilocular radiolucency. Because this radiographic pattern is not specific, a biopsy and histopathologic examination are required to reach a definitive diagnosis [15].

Macroscopic examination of case 6 revealed the presence of a main cystic cavity and other smaller cysts. The cyst measured 1.5 × 1.0 cm in diameter (Fig. 2).

Follow-up data were available for all 10 cases of BOC. Recurrence was observed in 4 cases, with the same patients undergoing several surgical procedures.

The main histopathologic findings of the 10 cases of BOC (Table 2) were a multicystic histologic appearance (Fig. 3) in all 10 (100%), subepithelial hyalinization (Fig. 4) in 7 (70%), squamous cuboidal epithelium consisting 2 or 3 layers in 10 (100%), focal epithelial plaques (Fig. 5) in 10 (100%), microcysts (Fig. 6) in 4 (40%), clear vacuolated cells (Fig. 7) in 3 (30%), and absence of inflammation in 8 (80%).

**Table 2**

<table>
<thead>
<tr>
<th>Histopathologic findings</th>
<th>No. of cases</th>
<th>%</th>
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<tbody>
<tr>
<td>Multicystic histologic appearance</td>
<td>10</td>
<td>100</td>
</tr>
<tr>
<td>Thin, nonkeratinized squamous or cuboidal epithelium lining</td>
<td>10</td>
<td>100</td>
</tr>
<tr>
<td>Focal epithelial plaques</td>
<td>10</td>
<td>100</td>
</tr>
<tr>
<td>Clear vacuolated cells</td>
<td>3</td>
<td>30</td>
</tr>
<tr>
<td>Microcysts</td>
<td>4</td>
<td>40</td>
</tr>
<tr>
<td>Hyalinization juxtaepithelial</td>
<td>7</td>
<td>70</td>
</tr>
<tr>
<td>Absence of inflammation</td>
<td>8</td>
<td>80</td>
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Fig. 3. Botryoid odontogenic cyst containing multiple cystic spaces (asterisks) in case 3 (40×).

Fig. 4. Botryoid odontogenic cyst showing juxtaepithelial hyalinization (arrow) in case 4 (400×).

Fig. 5. Botryoid odontogenic cyst showing whirlpool-like thickening (arrow) of the epithelial layer in case 3 (400×).

4. Conclusion

In conclusion, BOC is a rare multicystic variant of LPC, which is commonly observed in adults older than 50 years. A high recurrence rate (40%) was observed in the present study, with size and multilocular pattern probably being the main factors associated with recurrence. All cases presented multiple cysts, squamous cuboidal epithelium consisting of 2 or 3 layers, and focal epithelial plaques. Long-term follow-up of patients diagnosed with BOC is necessary.
References