Diagnosis and interdisciplinary treatment of a botryoid odontogenic cyst in the posterior mandible: Report of a case

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Botryoid odontogenic cysts (BOC) are considered to be rare polycystic variants of lateral periodontal cysts characterized by a multilocular growth pattern. The most frequent location of BOC is the mandible, predominantly the premolar-canine area, followed by the anterior region of the maxilla. The cyst shows a slight female predilection. This case report of a BOC demonstrates a treatment with initial fenestration and decompression of the cyst in order to prevent damage to adjacent structures such as the inferior alveolar nerve. The present case report emphasizes the importance of accurate clinical, radiographic, and histologic diagnostic procedures of unspecific radiolucent lesions in the jaws to establish a firm diagnosis and avoid inappropriate treatment strategies. (Quintessence Int 2014;45:233–237; doi: 10.3290/j.qi.a31211)

Key words: botryoid odontogenic cyst, cone beam computed tomography, decompression, diagnosis, therapy

The lateral periodontal cyst (LPC) is a rare developmental cyst of odontogenic origin defined as a radiolucent lesion, which develops along the lateral aspect of an erupted vital tooth. It represents nearly 0.4% of all odontogenic cysts.1 Usually, clinical symptoms are absent and (incidental) diagnosis is made by radiographic examination.

The botryoid odontogenic cyst (BOC) is considered a polycystic variant of the LPC. It was originally described in 1973 by Weathers and Waldron as an intraosseous lesion characterized by a microscopic and macroscopic multilocular growth pattern, resembling a bunch of grapes (from the Greek word botryos).2 Radiologically, the multilocularity is not necessarily visible, and many cases present as unilocular radiolucencies.3

The cysts are derived from epithelial remains of the tooth-forming organ. There are opposing views regarding the exact origin, ranging from the reduced enamel epithelium to the dental lamina or even the epithelial remnants of Malassez. In addition, it remains unclear which stimulating factor leads to the initiation of cystic growth. The polycystic appearance of BOC could be due to confluence of cysts in a multilocular LPC.4 The
present case report will demonstrate the importance of accurate clinical, radiographic, and histologic diagnostic procedures of radiolucent lesions in the jaws to initially establish a firm diagnosis and plan appropriate treatment strategies.

CASE REPORT

A 57-year-old woman was referred by her general dentist for evaluation and treatment of a cystic lesion in the right mandible. The patient had the following medical diagnoses: chronic pain syndrome, depression, secondary thrombocytosis, Hashimoto’s disease, and status post Lyme disease.

The patient had noticed gradually increasing swelling of the buccal bone in the right mandible and slight pain one week prior to referral. Her dentist had made a panoramic radiograph which identified a well-demarcated radiolucency in the right mandible (Figs 1 and 2). Anamnestically, the patient reported a surgical intervention in the region of the right mandibular first molar (tooth 46 according to FDI notation) more than a decade earlier, although the exact details remain unknown. Furthermore, the right mandibular second premolar (tooth 45) had previously been treated endodontically due to a fracture of the crown involving the pulp.

At the time of referral, the patient presented with extraoral swelling of the right mandible. Sensitivity in the area of the mental nerve was normal. Swelling of the buccal aspect of the jaw was also visible intraorally. Teeth 45 and 46 had had root canal treatment, and sensitivity testing with CO2 snow proved positive for all other teeth in the right mandible.

For further radiographic evaluation, cone beam computed tomography (CBCT; field of view, 6 × 6 cm; 3D Accuitomo XYZ Slice View Tomograph, Morita) was performed. The images showed a well-defined, multilocular, round, radiolucent structure with a sclerotic border. The apices of teeth 44 to 47 extended into the lesion, and there was no visible root resorption or divergence (Fig 3). The three-dimensional images showed an interruption in the continuity of the buccal bone wall of tooth 46. The course of the mandibular canal was not visible throughout the mesio-distal extension of the lesion.

The initial tentative and differential diagnoses were an odontogenic cyst (radicular, BOC) and an odontogenic tumor (ameloblastoma, keratocystic odontogenic tumor). To verify the diagnosis, explorative surgery including fenestration and decompression of the cystic lesion was planned. After mobilization of a buccal flap, a biopsy of the cystic epithelium was taken. Because of the size of the lesion and the non-visible course of the mandibular canal, the surgeon decided to perform initial fenestration of the cystic lesion and wait for shrinkage of the lesion by using an iodine-vaseline drain. The respective histology is shown in Fig 4.
**Figs 3a to 3c** The sagittal CBCT image (a) demonstrates that the apices of teeth 44 to 47 extended into the lesion without visible root resorption. The cortical lining of the mandible in the affected region is intact except in the region of the first molar. An interruption in the continuity of the buccal bone wall (*) can be seen on the respective coronal (b) and axial (c) CBCT images.

**Figs 4a to 4d** Histopathology (hematoxylin-eosin stain) shows a multilocular cyst (a) lined by a thin epithelium (b). Focal plaque-like thickening of the epithelium (c) is visible. The basal cell layer is formed focally by clear cells (d).

**Figs 5a to 5d** Radiologic follow-up 9 months after fenestration of the BOC using CBCT imaging. The cystic lesion exhibits reossification and shrinkage of the bony cavity in sagittal (a) and axial (b) CBCT images. The course of the mandibular canal and mental foramen are visible again** (c) and the buccal bony fenestration in the region of the first molar is still present* (d).
The patient was called in for regular postoperative check-ups and the drain was changed on a regular basis. After 9 months, another CBCT (volume 6 × 6 cm) was performed. The lesion was clearly smaller and exhibited distinctive reossification at the borders. The mandibular canal was now visible throughout; only in the region of tooth 46 was no cranial bony lining visible (Fig 5). During the 9-month period, teeth 44 and 47 had undergone root canal treatment by the referring dentist, as devitalization would have been likely after the planned enucleation of the cyst.

Eleven months after initial fenestration, the rest of the cystic epithelium was surgically enucleated and the defect was filled with iliac crest cancellous bone. To gain better access to the lingual part of the lesion, the root tips of teeth 44 to 47 were resected. During the intervention, the inferior alveolar nerve was preserved (Fig 6).

Six months after surgical removal of the cyst, a panoramic radiograph exhibited good reossification of the lesion, without signs of recurrence. Extraoral sensitivity in the area of the mental nerve was normal. One year later, another check-up including a panoramic x-ray was performed. The patient was still free of symptoms, and extraoral sensitivity in the area of the mental nerve was normal. No radiographic signs of recurrence were visible (Fig 7).

**DISCUSSION**

A BOC is considered a rare polycystic variant of a lateral periodontal cyst. The lesion is mainly seen in the mandible and shows an affinity to the molar and premolar regions. Although it can affect patients of all ages, the peak incidence is in patients aged 50 to 60 years, and there is a slight predilection for women. In most cases, a unilocular radiographic appearance is visible. Clinically, an early BOC is often asymptomatic. If not discovered early, BOC can cause continuous expansion of the involved bone that may be accompanied by swelling. Pain and paresthesia are rare symptoms. Root resorptions are an uncommon finding. Usually, affected teeth are still vital.

Histologically, BOC is characterized by multiple cysts with thin non-keratinized epithelial linings and epithelial thickenings, referred to as plaques. The epithelium is separated by connective tissue. Other authors have noted clusters of epithelial odontogenic remnants in the connective tissue wall, hyalinization of connective tissue adjacent to the basal epithelial layer, epithelial separation in the basement membrane zone, and lack of inflammation. Glycogen-containing clear cells, with cytoplasmatic glycogen, detected as periodic acid-Schiff positive may also be present.

The BOC diagnosed in this case exhibited typical clinical, radiographic, and histopathologic characteristics reported for BOCs. Recurrence rates published for
BOCs range between 15–20% and 33%.\textsuperscript{5, 6} This recurrence rate is substantially higher than for lateral periodontal cysts. This could be due to the larger size and the multilocular histologic appearance, which makes BOCs more difficult to enucleate completely. Therefore, regular clinical and radiologic follow-up visits are recommended.\textsuperscript{7, 8} In the present case, the patient will be scheduled for regular clinical and radiographic check-ups including panoramic radiographs on an annual basis.

Because of the frequent unilocular radiographic appearance, initial clinical tentative diagnosis is often not BOC, but rather a radicular cyst or an odontogenic tumor. The more expansive an osteolytic process is becoming, the more challenging is the final diagnosis based on clinical and radiographic information alone. Periapical lesions of nonendodontic origin can develop around the periapical region of teeth and can expand into large lesions, if not diagnosed early.\textsuperscript{9} There are also reports about nonendodontic lesions mimicking apical periodontitis and their subsequent misdiagnosis in the literature.\textsuperscript{10, 11} In the present case report, the final diagnosis was not clear until explorative surgery and histopathologic analysis were performed.

In a study by Santos and coworkers, the initial diagnosis was ameloblastoma in 40% of the cases, BOC in 20%, radicular cyst in 10%, residual cyst in 10%, and odontogenic keratocyst in 10%.\textsuperscript{12} BOC are distinguished from LPC by their larger size.\textsuperscript{13} A BOC frequently shows a lobulated radiographic pattern similar to that of glan- dular odontogenic cysts, and therefore the latter should be included in the differential diagnosis.\textsuperscript{14}

Patients often present with nonspecific radiographic and clinical signs and symptoms,\textsuperscript{15} or radiolucent lesions may even be incidental findings on routine radiographs. This case report underscores the importance of performing a histologic examination when a radiolucent lesion associated with teeth with vital pulps is identified during a radiographic examination.\textsuperscript{9, 16}

Once the diagnosis of BOC has been firmly established, the recommended therapy is surgical removal. As reported in the present case, surgical removal was performed only after initial fenestration, decompression, and shrinkage of the lesion with partial reossification. This procedure was chosen due to the initial extension of the cyst and the potential for damage to the inferior alveolar nerve, potential loss of the involved teeth, and fracture of the mandible following surgical removal. Furthermore, the present case report demonstrates the importance of an interdisciplinary treatment approach for unspecific expansive radiolucent lesions in the jaws including a general practitioner, an oral surgeon, and an experienced pathologist to establish a firm diagnosis and offer the appropriate treatment approach including long-term follow-up.

REFERENCES