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Mandibular Botryoid Cyst with Large Extension, Treated by Decompression and Marsupialization: A Case Report

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ABSTRACT

The maxillo-mandibular complex is subject to rare pathologies, such as odontogenic and non-odontogenic cysts and tumors. The presentation can be clinically symptomatic, with localized edema, facial asymmetry, paresthesia, dental mobility, gingival discoloration and pathological fracture of alveolar bone. It can also be asymptomatic, occasionally discovered during routine radiographic examination. The lateral periodontal cyst (LPC) is a rare odontogenic cyst that usually appears on the lateral surface of vital tooth roots and can have a more aggressive presentation known as botryoid odontogenic cyst (BOC). BOC is a rare odontogenic pathology, which can be extensive causing bone destruction and tooth loss. Large cases can be treated surgically, with decompression and marsupialization, presenting good clinical progress, reduction in symptoms with bone growth potential, which associated with conventional surgical enucleation, in a pathology of smaller dimensions, has better prognosis. This study presents a case of an extensive symptomatic BOC in the mandible, discovered by routine radiographic examination, and treated by marsupialization and decompression, and discusses this pathology and the importance of proper management to avoid tissue and dental damage. The cases must be followed up for years due to the high possibility of recurrence.

Keywords: Botryoid Cyst; Lateral periodontal cyst; Osseous pathology; Odontogenic tumor.

Introduction

The lateral periodontal cyst (LPC) is a rare odontogenic cyst most commonly found in the mandible, between the roots of the canine and premolar teeth. The botryoid cyst (BOC) is a variant of LPC, most often affecting middle-aged and older adults, with a multilocular aspect generated by the spread of concentrations of epithelial remains that results in an aspect of grape cluster. Although both LPC and BOC can be managed with simple enucleation, it is worth noting that while first one has an extremely low recurrence rate, the latter can recur more easily.

Being a cyst of odontogenic origin, the LPC is considered a rare lesion in the oral cavity, with an occurrence rate of less than 1% among all odontogenic cysts¹. The diagnosis is based upon radiographic analysis where a drop-shaped radiolucency with well-defined margins is observed. LPC originates from epithelial remains and presents a thin keratin layer histologically. Although it is a rare pathology, it is not a painful lesion. It has

a slight female predilection and is more common in patients between 40 and 70 years of age. It appears laterally around the roots of vital teeth, more frequently canines and premolars of the mandible, with its most accepted origin being the remnants of dental lamina, reduced enamel epithelium and epithelial rests of Malassez. The preferred treatment plan is enucleation, always emphasizing the importance of preserving the adjacent dental element².

The BOC is a variation of the LPC, gaining its name because it resembles the shape of a bunch of grapes, morphologically³. Also considered a rare odontogenic cyst, it is radiographically identified with a multilocular appearance, due to the presence of epithelial remains in its margin. It has a higher incidence of occurrence in the mandible, having the same treatment as LPC, but requiring a more meticulous treatment plan due to its high recurrence rate⁴.

Although the causative pathogens are different, it is important

that both are treated immediately after diagnosis, avoiding sequelae such as pathological fractures, infections, local pain and tooth loss, which could cause major complications to the rehabilitation treatments⁵.

Case Report

Patient AA, 32 years old male, attended the maxillofacial surgery clinic referred by a fellow dental surgeon complaining of an intraoral mass, with a slight change of mucosa color in the region of mandibular canine and premolars on the right side, associated with mild pain and mandibular hypoesthesia in the lower right lip and mental region. The patient, otherwise healthy, was hypertensive, using medication (losartan and zolpiden), non-smoker and an occasional drinker. He did not report any local trauma or other previous surgery in the specified region.

Upon clinical examination, an expansive mandibular lesion was noted, the buccal bone plate presented slight crackling and no teeth mobility was observed (**Figure 1**). Radiographic and tomographic examination (**Figure 2 & Figure 3**) revealed an ill-defined multilocular radiolucency, with “honeycomb” aspect, adjacent to the roots of the lower right canine and premolars, without mandibular base involvement. Pulp vitality test was applied using endofrost (cold) and gutta percha (heat), revealing vital canine and non-vital premolars. The differential diagnosis was Ameloblastoma, botryoid cyst and central lesion of giant cells.



Figure 1. Intra and extra oral clinical appearance.



Figure 2: Initial panoramic radiograph showing multilocular bone appearance

Due to the lesion size and proximity to neighboring teeth, the patient was oriented to perform endodontic treatment on the non-vital teeth. Consequently, an incisional biopsy and cyst decompression were scheduled. The surgical procedure was performed under local anesthesia, after local antisepsis, aspiration was performed with an 18G needle, returning a citrus liquid content, then a small vestibular incision was made to access the cystic cavity. Two fragments were obtained and stored in 10% formalin vial and sent for histopathological analysis at

the oral pathology department at the Faculty of Dentistry of the University of São Paulo. The surgical incision was then sutured and a decompression stent was introduced to keep the incision open for decompression. The patient was instructed to irrigate the cystic cavity 3x a day with 0.12% chlorhexidine solution (Periogard) to avoid obstruction and infection. (**Figure 4**). shows the results obtained after 4 weeks of decompression. A notable reduction in vestibular mass can be visualized in this image and the patient reported a major pain relief. Future cyst enucleation was decided according to radiologic evaluation of the cystic size and affected bone.

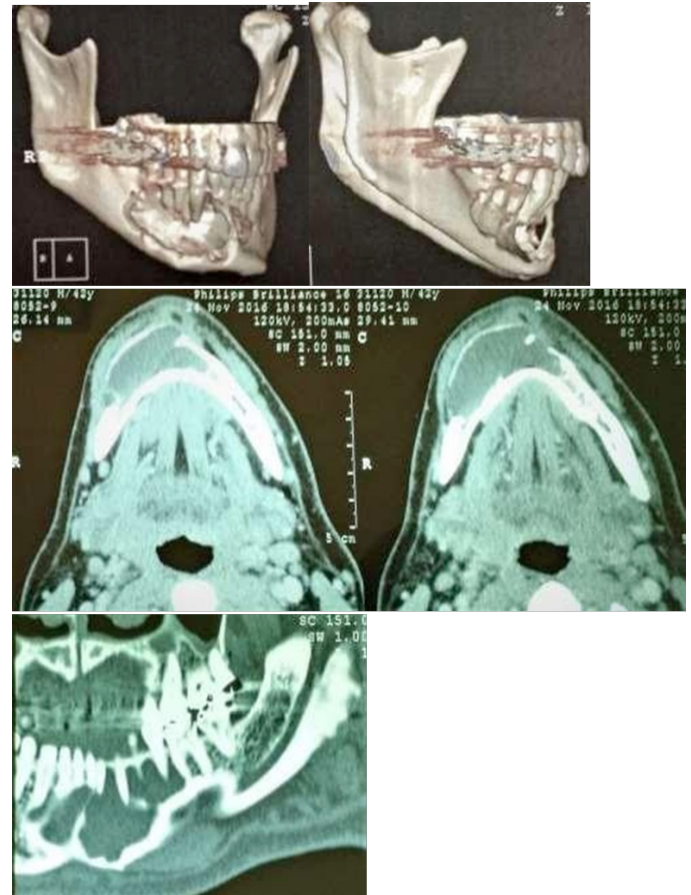


Figure 3. Initial CT scan showing bony lesion



Figure 4. cystic decompression result after 4 weeks.

The histological slides (**Figure 5**) show the presence of multiple cystic cavities of variable sizes and shapes, and thin walls lined with non-keratinized stratified squamous epithelium of variable thickness; the epithelium either consisted of a few cell layers or showed thickening generally formed by oval, sometimes

entangled, plaques. The cystic capsule is characterized by dense connective tissue that is highly collagenized, with the deposition of bundles of collagen fibers in various directions. A large number of blood vessels are noted, sometimes congested and of varying sizes, with extensive areas of interstitial hemorrhage. In the submucosa, bundles of striated skeletal muscle and trabeculae of mature bone are noted completing the analyzed histopathological picture. The result of the histopathological examination was a botryoid cyst, a variation of the lateral periodontal cyst, with characteristic stratified squamous epithelium, focal thickening with mucous cells and a multicystic aspect.

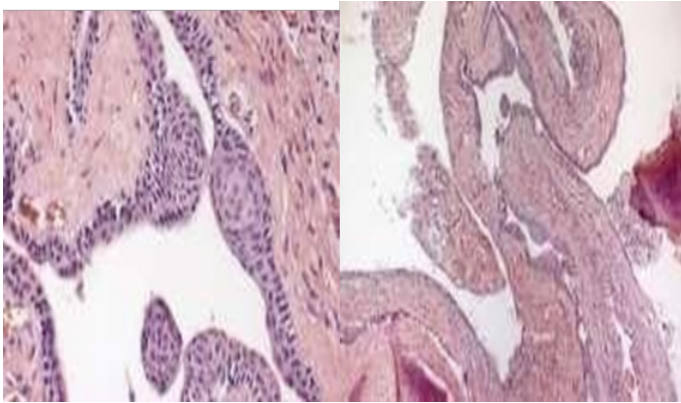


Figure 5: Histologic Slides

Local irrigations and follow-ups were maintained for 6 months, and a new radiographic examination was performed, showing notable clinical and radiographic improvement, but still showing radiolucency adjacent to neighboring teeth (**Figure 6**). At this time, it was decided to perform a new surgery for enucleation and definitive treatment as shown in (**Figure 7**).



Figure 6: Radiographic appearance at 6 months post-op. There is an evident radiographic sign of bone formation in the peripheral areas.



Figure 7: Enucleation surgery. A vestibular bone defect is noted in the decompression stent area where irrigation was performed. Cystic extension decreased, with defined margins. A small releasing incision was made to permit the complete closure of the defect.

The patient was medicated with antibiotics (cephalexin), NSAIDs and analgesics (ketoprofen and paracetamol), with periodic follow-ups until complete recovery after surgery, which was uneventful. The patient evolved with complete remission of symptoms and returned to his normal routine. He is under regular monitoring, remaining asymptomatic and without radiographic signs of recurrence after 4 years of control, as shown in **Figure 8**.



Figure 8: Radiographic examination at 4 years follow-up shows no signs of recurrence.

Discussion

The BOC is a rare and relatively new entity, first described by Weathers and Waldron in 1973⁶. It has been defined as a multilocular variant of LPC, which in turn is defined as a non-keratinized developmental cyst of the alveolar bone occurring on the lateral aspect of vital teeth⁷. However, LPC and the BOC are not considered to be of the same entity by other authors, and the latter is defined as a multicystic odontogenic lesion with histological characteristics of LPC or cystic lesion similar to LPC⁸⁻⁹. BOC can occur at any age, but more usually in individuals above 50 years of age, with a slight female predilection¹⁰⁻¹¹. The most frequent location is the mandible, followed by the anterior region of the maxilla, appearing as a multilocular radiolucency that can grow into considerable sizes with consequent swelling, pain, paresthesia and other signs¹¹⁻¹². The differential diagnosis for multilocular radiolucencies include odontogenic keratocyst, multicystic ameloblastoma, odontogenic myxoma, ameloblastic fibroma, central odontogenic fibroma and intraosseous mucoepidermoid carcinoma¹¹⁻¹³. An incisional biopsy should be performed to confirm the diagnosis¹². BOC shows a high recurrence rate (21.7%), when compared to LPC (2.4%)¹⁴, its recurrence rate resembles that of glandular odontogenic cysts and odontogenic keratocysts¹⁵⁻¹⁶. The high recurrence rate can be attributed to the multicystic nature of the cyst, making it difficult to achieve complete excision, and increases the risk of future recurrence due to the presence of cystic epithelial remnants after excision¹⁷. In a rare presentation, a case with 4 recurrences within 9 years was described in 1989¹⁸. Histologically, both LPC and BOC have numerous possible origins, including cells of rests of dental lamina, epithelial rests of Malassez and reduced enamel epithelium¹⁹. Some authors suggested that BOC arises from changes in several adjacent cell rests especially from the post functional cells of dental lamina¹⁰, being a polycystic variant of LPC developing by cystic transformation of multiple islands of dental lamina rests⁴⁻¹⁹.

Treatment of choice for multicystic BOCs is meticulous surgical excision, with curettage of all cystic epithelium.

Conservative enucleation is not recommended due to the high recurrence rate¹¹. Hence, long-term follow-up is highly indicated, with radiographic examination²⁰. It is worthy to emphasize on the importance of pathological categorization and diagnosis due to different treatment options among different cystic variants. LPC, for instance, can be treated with simple enucleation with a very low recurrence rate, BOC requires cystic decompression followed by surgical excision, whereas ameloblastoma treatment ranges from enucleation to en bloc marginal resection²¹.

Conclusion

BOC is a more aggressive variant of LPC, which presents an extensive lesion clinically and radiographically that, if left untreated, may lead to tooth loss, pathological fractures and neurosensory disorders. Initial marsupialization and decompression can be indicated in cases of large cystic lesions, gradually decreasing the cystic size and alleviating the clinical symptoms, allowing for future surgical excision with less morbidity and excellent results.

Ethical Disclosures

Protection of human and animal subjects

The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of Data

The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to Privacy and Informed Consent

The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

Conflicts of Interest

The authors declare they have no conflicts of interest.

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