

BOTRYOID ODONTOGENIC CYST: A RARE CASE REPORT

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ABSTRACT:

Botryoid odontogenic cyst (BOC) is considered a rare multilocular variant of the lateral periodontal cyst, usually involves the mandibular premolar-canine area, followed by the anterior region of the maxilla. A 42-year-old male patient with botryoid odontogenic cyst (BOC) was treated by surgical removal. BOC have been widely regarded as variants of the lateral periodontal cyst. The conservative enucleation of the BOC is the treatment of choice, a significant recurrence rate has been reported for BOC. An extended post-surgical follow-up is necessary for a patient who has been diagnosed with BOC.

KEYWORDS- Botryoid cyst, lateral periodontal cyst, enucleation, maxillary lesion

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INTRODUCTION:

Botryoid odontogenic cyst, essentially, is a cystic lesion with clinical and radiological features of a lateral periodontal cyst, but which shows macroscopic and microscopic features of a botryoid odontogenic cyst. Van der Waal (1992)¹ objected to the point that lesion which occurs in the lateral periodontal position, if defined as a lateral periodontal cyst, how a lesion; botryoid odontogenic cyst, extending well beyond the lateral area of the tooth should be accepted as a variant of lateral periodontal cyst.

This begs the question: is the lesion referred to as a 'botryoid odontogenic cyst' merely a botryoid variety of a lateral periodontal cyst, or is it an entity with a tendency to recur if not completely removed at operation? There is convincing evidence, that it is a variant of lateral odontogenic cyst, but the term 'botryoid odontogenic cyst' should be retained because of the tendency of this variant to recur if inadequately removed. Even small lateral periodontal cysts may be bicystic or polycystic, and with further growth, can take on a botryoid (grape-like) appearance (Altini and Shear, 1992)². As concluded by Machado de Sousa et al. (1990)³ and Ramer and Valauri (2005)⁴, it is important to separate the two lesions because the multicystic nature of the botryoid odontogenic cyst makes the lesion more expansive and increases the possibility of recurrence when surgical curettage is inadequate. It was Weathers and Waldron (1973)⁵ who first suggested the term. They reported two examples of a multilocular cystic lesion of the jaws for which they proposed the term 'botryoid odontogenic cyst' because the gross specimen resembled a cluster of grapes. Since their original description, botryoid odontogenic cysts have been widely regarded as variants of the lateral periodontal cyst and publications on the subject have not distinguished them from the multicystic lateral periodontal cysts.⁶

CASE REPORT:

A 42-year-old male patient was referred to the Department of Oral and Maxillofacial Surgery for evaluation of a swelling in the right anterior maxilla which was also significant extraorally (**Figure 1**). He had been aware of the asymptomatic swelling during the past two years. At the time of dental examination, the radiological and CT investigations revealed a well delimited, multi-locular, 3.08 x 3.05cm radiolucent lesion (**Figure 2**), between the roots of the right maxillary lateral incisor and canine. There is deviation of the teeth without root resorption.

Aspiration contents were dusky red coloured fluid with presence of cholesterol crystals and protein content above 5 gms per 100 ml. (**Figure 3**).

Under all aseptic precautions and conditions, after standard patient preparation, under nasotracheal intubation, general anesthesia was induced. Vestibular incision was taken from maxillary right central incisor to maxillary right first molar (**Figure 4**). Mucoperiosteal flap so developed exposed the expanded buccal cortical plate. Deroofing the expanded cortex allowed access to the lesion, which was an enucleated in toto. (**Figure 5**). Closure was done with 3.0 braided silk. Lesion sent for histopathologic examination. Patient is followed up to 1 year with no recurrence (**Figure 6**)



Figure 1- Showing preoperative extra oral swelling

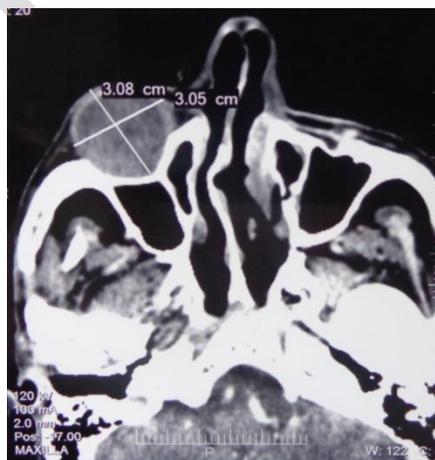


Figure 2- Showing extent of lesion in axial CT Scan



Figure 3- Contents were dusky red coloured fluid



Figure 4- Showing planned intraoral incision



Figure 5- Showing enucleation of the lesion



Figure 6- Showing postoperative picture after 12 months

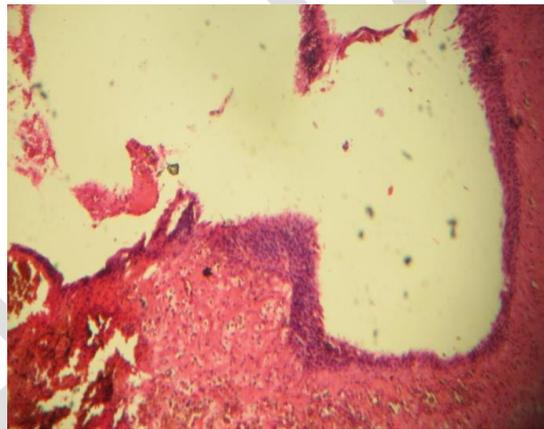


Figure 7- Showing histopathologically proven as BOC

Microscopically, the H & E stain section shows multiple cystic cavities. Two of them are smaller and located near a larger cyst and placed in a thick and dense connective tissue capsule one of these two cavities is lined by thicker darkly stained non-keratotic stratified squamous epithelial lining with one area of plaque formation. The another one is lined by single layer of epithelial cells. The larger intact cyst in this section is lined by non-keratotic thicker darkly stained stratified squamous epithelium without epithelial ridges formation. One area in the lining shows more thickening and plaque formation . The connective tissue capsule of these cyst is thick, dense, with very little but chronic inflammation and few blood vessels.

Two more areas in the sections show stratified squamous non-keratotic epithelial lining. At one place it is separated from the connective tissue capsule which is inflamed. At one area the lining is attached to the underline connective tissue capsule. The connective tissue capsule show overall chronic inflammatory response, large blood vessels with some extravassated blood. Features suggestive of Botryoid Odontogenic Cyst. **(Figure 7).**

DISCUSSION:

Lateral periodontal cyst (LPC) is an unusual cyst of odontogenic origin, most frequently found in the mandible between the roots of canines and premolars and corresponding to 0.7% to 1% of all cysts found in the jaw.⁷ Patients of BOCs frequently complain of swelling along with pain and paraesthesia. BOCs are comparatively larger cysts ranging from 4mm to 45mm size and may be unilocular or multilocular. In present case also patient reported with chief complaint of swelling and paraesthesia.⁸ The cyst frequently cannot be distinguished clinically from simple lateral periodontal cyst. Being so closely associated with the lateral aspects of the root, can easily be confused with an odontogenic keratocyst or lateral radicular cyst. An important aspect that can provide the differential diagnosis between a radicular cyst and lateral periodontal cyst (LPC) is the fact that the former is characterized by necrosis of the affected tooth, whereas the tooth remains vital in the latter and no root canal treatment is required.⁹

Botryoid odontogenic cysts typically affects adult over the age of 50years. There appears to be no significant gender distribution. Demographic, clinical, microscopic, and radiographic features concluded the uncommonness. Mandibular premolar to canine region is the main site of occurrence. With some examples of anterior extension across the midline.^{9,10} Botryoid odontogenic cyst can be considerably larger than conventional lateral periodontal cyst, and multilocularity may be clearly seen. However, histologically confirmed botryoid odontogenic cyst may appear unilocular in radiographs. Botryoid odontogenic cyst may also differ in their location from lateral periodontal cysts.^{6,9}

The lesion is similar to the lateral periodontal cyst but exhibits some differences. The lesion is multicystic with thin fibrous connective tissue septa. The cyst cavities are of varying size, with the smaller ones tending to be orientated towards the larger ones. The cyst cavities are sometimes

lined by a thin non-keratinised epithelium comprising, for the most part, 1–2 layers of flat cells but occasionally are lined in some areas by a somewhat thicker stratified squamous epithelium. In many of the cysts there are foci of plaque-like thickenings, most of them consisting of flat fusiform cells. Weathers and Waldron suggested that these plaques may possibly be the source of new cyst locules. Interspersed clear, glycogen containing cells are unusual both in the lining epithelium and in the plaques. Clusters of epithelial odontogenic rests, largely consisting of clear cells, may be seen in the surrounding connective tissue. A discrete hyalinized zone may be seen immediately subjacent to the basal epithelial layer. This zone may promote separation of the epithelium from the capsule . There is an increase in the nucleo-cytoplasmic ratio in much of the epithelium with resultant crowding of the cells, the nuclei of which are pyknotic.^{6,9,10}

Altini and Shear(1992)² described in some detail,, a hypothesis of how a unicystic lateral periodontal cyst may progress to a multicystic, yet encapsulated lesion, and then by progressive enlargement of the many microcysts, develop into an irregular thin-walled multicystic structure, which is identified now as a botryoid odontogenic cyst. It is not unusual to see epithelial cells from one or more plaques apparently budding off from the mother cyst, later apparently becoming detached and sometimes breaking down centrally to form a daughter cyst. There is also microscopic evidence that basal cells of the mother cyst may rain down into the wall. Unless removed intact, the lesion seems to have the potential to extend in the bone and become multilocular.^{2,4,6}

Heikinheimo et al.¹¹ have shown that botryoid odontogenic cyst epithelium strongly expresses cytokeratin 18, which is absent from gingival epithelium but present in some dentigerous cysts and focally in ameloblastomas. Cytokeratin 19, which is also a major component of odontogenic epithelium, was also expressed by all cells of the botryoid odontogenic cyst epithelium. Heikinheimo et al.¹¹ were, therefore, convinced that as their location also suggests, these cysts were odontogenic.^{9,11} Botryoid odotogenic cysts may thus arise from remnants of the dental lamina, from remnants of the reduced enamel epithelium, or from the root sheaths of Hertwig. Proliferation and cystic change in several epithelium rests may give rise to the polycystic character of botryoid odontogenic cysts.^{9,11,12}

The determinants of the propensity for botryoid odontogenic cysts are also obscure, but it may not always be possible to remove every lobule intact. Extension joint to the main body of the lesion by a thin stalk, and with discrete microcysts in the cyst wall, rests of odontogenic epithelium may also be seen in the connective tissue. Any of these rests, if left after excision, might proliferate to form additional cysts. The delay of many years typically seen before recurrence may suggest that proliferation of the parent odontogenic epithelium has formed a new lesion, but the fact that recurrences have been in the site of the original cyst indicates that it is the tissue of the latter is responsible.^{6,8,9}

Feature that distinguishes BOC from lateral periodontal cyst is the larger size of the former. The importance of the differentiation between lateral periodontal cyst 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.³

Botryoid odontogenic cysts have an undoubted propensity for recurrence. Because of surgical difficulties in removing botryoid odontogenic cyst entirely, they should be excised rather than enucleated. Wide excision with sacrifice of teeth does not seem to be justified. Instead a prolonged radiographic follow-up can be maintained. Any recurrence can then be controlled by a further limited excision before the cyst reaches a large size.^{4,6,9}

CONCLUSION:

This case shows a presentation of an uncommon lesion, Botryoid odontogenic cysts, emphasizes the importance of microscopic examination of the lesions, particularly when these are associated with teeth having normal responding vital pulps. The significance of separating the BOC from LPC is based on the size and gross appearance of the former; the BOC is more expansive than the LPC because of its multicentric nature. The higher recurrence rate of BOC is not because of the cell growth activity, but because of difficulty in complete surgical removal of a multilocular lesion. Therefore, an extended postsurgical follow-up is recommended clinically.

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