Botryoid odontogenic cyst developing from lateral periodontal cyst: A rare case and review on pathogenesis

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Abstract

Botryoid odontogenic cyst (BOC) is considered to be a polycystic variant of the lateral periodontal cyst (LPC) as the specimen resembled a cluster of grapes. It is a non-inflammatory odontogenic cyst. The BOCs can be unicystic or multicystic. These cysts have potential to extend in the bone and become multilocular and they have a high recurrence rate. Till now, only 73 cases of BOC have been reported. The pathogenesis of BOC is still debatable. We review different pathogenesis proposed for BOC and discuss a rare case of BOC developing from lining of an abnormally large LPC which showed aggressive behaviour in terms of growth and size.

Key words: Botryoid odontogenic cyst, botryoid odontogenic cyst, lateral periodontal cyst, lateral periodontal cyst, pathogenesis of Botryoid odontogenic cyst

Introduction

Botryoid odontogenic cyst (BOC) is a rare developmental odontogenic cyst. Some authors consider it to be a variant of Lateral periodontal cyst (LPC) with higher risk of recurrence.[1] The term BOC was coined by Weathers and Waldron in 1973. To the best of our knowledge only 73 cases of BOC have been reported till now. BOC shows a striking predilection for mandible.[1-3] Mendez P *et al*. found that multi-locularity is not a diagnostic feature as 2/3rd of cases reported in literature were unilocular.[1] The pathogenesis of BOC is still debatable. Some authors consider it to be a result of compartmentalization in LPC lumen, while other group of authors says that it arises as a result of fusion of multiple independent LPCs developing in proximity. The aims of this article are to review the different pathogenesis proposed for BOC and document an atypical case of BOC developing from lining of a fast growing abnormally large LPC.

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Access this article online	
Quick Response Code:	Website: www.contempclindent.org
	DOI: 10.4103/0976-237X.103629

Case Report

A 20 year-old female reported at our college of dentistry with an extra-oral swelling of lower anterior mandible of 3-4 month duration. Clinical inspection revealed a well circumscribed swelling of 2 × 2 cm size on left lower canine-premolar region obliterating the buccal vestibule [Figure 1a]. Panoramic radiograph showed a well-corticated unilocular radiolucency between left canine and first premolar, extending beyond inter-radicular area [Figure 1b]. Displacement of adjacent roots is seen. Based on this data, the differential diagnosis of an odontogenic cyst or tumor was made.

Aspiration content showed presence of cholesterol crystals [Figures 2] and protein content above 5 gms per 100 ml. Incisional biopsy was reported to be LPC as the sections from the specimen showed a cystic lumen lined by 2-3 layers of flattened epithelial cells resembling reduced enamel epithelium. Although a diagnosis of LPC was made, this case appeared rare because of large size of the cyst extending beyond the lateral aspect of the associated premolar and also because the cyst had enlarged to the current size over a period of 3 months only, proving it to be more aggressive than usual LPCs. Based on this, enucleation was performed along with associated premolar. A gross specimen of about 1.5 cms in diameter and firm in consistency was obtained in-toto attached to lateral border of premolar [Figure 3]. Cross-sectioning of specimen revealed a cystic lumen with infoldings at places and nodular growths protruding within lumen. Histopathologic analysis reveals a cystic space lined by thin epithelial layer of 1-4 cell layer thickness. The epithelial lining is non-keratinizing and showed squamous to cuboidal to flattened cells which resemble reduced enamel epithelium. Cyst lining shows epithelial extensions of basal cells 'raining down' into connective tissue [Figure 4]. Connective tissue is fibrous, un-inflamed and shows presence of odontogenic epithelial rests. The epithelial cells in the lining show small pyknotic nuclei. Cells are separated by intercellular fluid at places.

Figure 1: Photograph of swelling on left mandible in canine-premolar area (a) intraoral view of the swelling (b) pre-operative panoramic view showing a unilocular radiolucent lesion between roots of left mandibular canine and first premolar. Displacement of lateral incisor, canine and both premolars can be seen

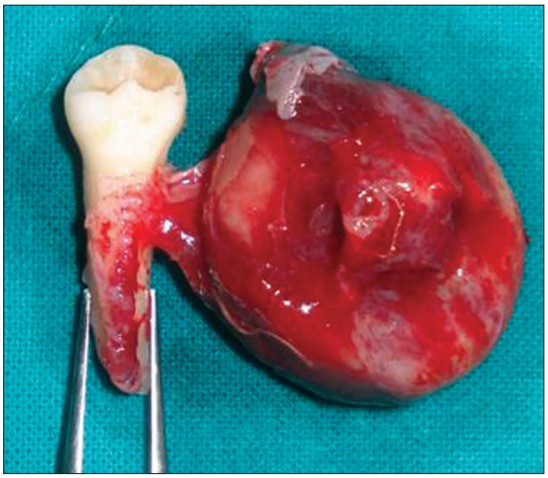

Figure 3: Photographs showing gross appearance of cyst enucleated cyst seen attached to the cervical area of the premolar

The lining shows epithelial plaques producing a mural bulge within lumen along with areas of hemorrhage. The mural growths show numerous intra-epithelial microcyst formations. The cells in plaque are fusiform while few cells are round, large and clear with pyknotic nuclei.

Complex convulations of epithelium, mural proliferations

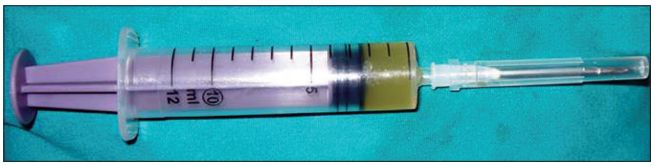

Figure 2: Photographs of the cystic aspirate yellow colored aspirate obtained

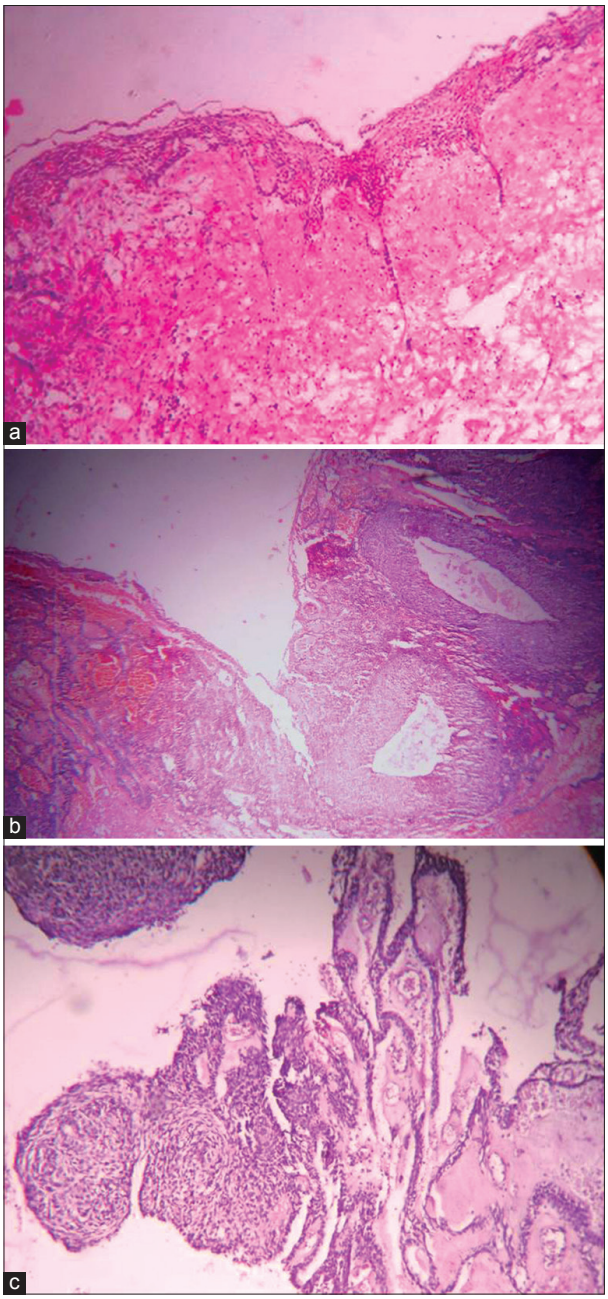

Figure 4: Photomicrographs of histologic sections of the cyst showing (a) 3-5 layered lining epithelium showing basal cell extensions "raining down" into the connective tissue capsule (×40, H and E stain) (b) complex mural proliferation and different stages of microcyst formation (×100, H and E stain) (c) PAS negative staining of intraluminal proliferations of epithelium. (×100, PAS stain)

and presence of daughter cysts within the lining shows the tendency of multi-locularity [Figure 4b]. The case shows definite botryoid odontogenic changes in lining epithelium of LPC. Hence a final diagnosis of botryoid Odontogenic cyst is made. Special staining was performed and the epithelial lining of the BOC was found to be PAS negative [Figure 4c].

The patient was reviewed after 2 months and she showed uneventful recovery.

Discussion

Lateral periodontal cysts are rare lesions, derived from odontogenic epithelial remnants, and occurring on the lateral aspect or between the roots of vital teeth. They are lined by a thin, non-keratinising squamous or cuboidal epithelium with focal, plaque-like thickenings consisting of clear cells that may contain glycogen.[4]

Clinically LPCs are symptomless cysts most commonly arising in mandibular premolar-canine-incisor region. LPCs appeared as small round or oval well circumscribed radiolucency over radiographs ranging from 2.5 mm to 15 mm size.[5] LPCs are microscopically composed of a cystic lumen lined by non-keratinized squamous or cuboidal epithelium resembling reduced enamel epithelium. Epithelium may show intercellular fluid accumulation at places and glycogen rich clear cells. Another typical feature of LPCs is focal hyperplasia of basal cells. These plaques may extend within connective tissue or intra-luminally.

Botryoid odontogenic cyst (BOC), as reported by Weathers and Waldron, is a polycystic variant of the lateral periodontal cyst because the specimen resembled a cluster of grapes.[6] It is a non-inflammatory odontogenic cyst which has striking predilection for mandibular canine-premolar region.

Patients of BOCs frequently complain of swelling along with pain and paraesthesia. BOCs are comparatively larger cysts ranging from 4 mm to 45 mm size and may be unilocular or multilocular. In present case also patient reported with chief complaint of swelling and paraesthesia.

According to different authors BOCs may be unicystic or multicystic showing thin connective tissue septae lined by thin stratified non-keratinizing epithelium. Foci of plaque-like thickenings are more common in BOCs than LPCs. Plaques with convoluted zones are similar to those seen in Adenomatoid odontogenic tumors. Epithelial cells are seen budding off from epithelium to form multiple daughter cysts. Cells show increase in nucleocytoplasmic ratio. It is seen that these cysts have potential to extend in the bone and become multilocular.

The pathogenesis of BOC is still controversial as some authors consider it to be a result of BOC changes in LPC, while other group of authors says that it arises as a result of fusion of multiple LPCs developing in proximity.

Redman *et al*., in year 1990, postulated that BOCs may have multicentric origin.[7] They supported this theory with cases of multiple LPCs developing in close proximity. They stated that, if left untreated, these cysts may fuse to form a multicystic lesion referred to as Botryoid variety.

In year 1992 Altini and Shear stated that, with further growth LPC can take on a Botryoid (grape like) appearance.[8] He proposed 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 microcyst, develop into an irregular thin-walled multicystic structure. He classified LPC into 3 morphological types.

a. Unicystic
b. Multicystic
c. Botryoid

Same year Van der Waal stated that BOC cannot be considered a variant of LPC as it extends well beyond the lateral area of root.[9] But he did not deny that possible cells of origin for both cysts are same. In our case also, the cyst was large and extended well beyond the lateral margins of root of associated tooth [Figure 1b]

In year 1996 High *et al*. proposed the term 'Polymorphic Odontogenic Cyst' for BOC, GOC and mucoepidermoid intraosseous carcinoma due to similar recurrence rate, unusual presentation and confusing histology.[10]

Furthermore Mendez *et al*. reported, in his extensive review of 62 cases, that BOCs are predominantly unilocular and have slightly higher average age of incidence.[1] He also stated that BOCs have a striking predilection for mandibular canine-premolar area and present with symptoms like swelling, pain or paraesthesia. 12 of 37 cases followed up showed recurrence (32%).

The term BOC has been used variably in literature for a cyst which shows radiographic multi-locularity, otherwise due to multiple cystic compartments within gross specimen, and histological features similar to a LPC. Our case is in clear conjunction with the theory proposed by Altini and Shear that a BOC may develop in a LPC. Our case shows a large unilocular cystic cavity but gross specimen at places showed intra-luminal nodular growths. These nodular growths, when studied microscopically, showed basal cell hyperplasia in form of plaques, clear cells within epithelium, epithelial extensions raining down in connective tissue and numerous daughter cysts. Rest of cystic lining resembled non-keratinized epithelium of a LPC. Thus, we suggest that the term 'Botryoid Odontogenic cyst' should be used for a cyst which is essentially a variant of LPC developing either by multicentric or unicentric growth pattern, but is different in terms of aggressiveness and typical

histology with high recurrence rate.

Acknowledgment

The authors great fully acknowledge Dr. Gopa Kumar R (Professor and Head, Department of Oral medicine and radiology, Mahatma Gandhi Dental College and Hospital, Jaipur) for his able guidance on radiographic details of the case.

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How to cite this article: Arora P, Bishen KA, Gupta N, Jamdade A, Kumar GR. Botryoid odontogenic cyst developing from lateral periodontal cyst: A rare case and review on pathogenesis. Contemp Clin Dent 2012;3:326-9.

Source of Support: Nil. **Conflict of Interest:** None declared.