

## Botryoid Odontogenic Cyst on Mandibular Anterior and Both Body Area: a Case Report

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### Abstract

Botryoid odontogenic cyst (BOC) is considered a rare multilocular variant of the lateral periodontal cyst. In this report, a 67-year-old male visited with chief complaint of severe mobility on mandibular incisors. Multilocular radiolucent lesion was seen from the right premolar to the left premolar area, involving almost the whole mandible. Histologically, the botryoid odontogenic cyst showed focal nodular thickening of the lining epithelium. These thickening often showed swirling appearance of the cells. Cyst enucleation and bone graft on mandible anterior and both body area were performed under general anesthesia, and postoperative healing was favorable without recurrence.

**Key words:** Botryoid odontogenic cyst, Lateral periodontal cyst

### Introduction

Botryoid odontogenic cyst (BOC) has been a rare multilocular variant of the lateral periodontal cyst. It is a developmental cyst of odontogenic epithelial origin. It has been known to originate from rests of the dental lamina and represented the counterpart of the gingival cyst of the adult. The most common site of this lesion is in the periodontal space of vital tooth.<sup>1)</sup>

Botryoid odontogenic cyst was the term first employed by Weathers & Waldron for a multilocular variant of the lateral periodontal cyst.<sup>2)</sup> The BOC is apparently uncommon, and it was reported to have a tendency to recur. The BOC is more likely found in middle-aged and older adults, and the teeth more likely affected are mandibular canines and premolar area. On radiography, the cyst appears grape-like appearance.<sup>1)</sup>

This report describes a case of botryoid odontogenic cyst, which occurred from right mandibular 2nd premolar to left mandibular 1st premolar of a 67-year-old male.

### Case report

A 67-year-old male visited with chief complaint of a painless, bluish, fluctuant, swelling mass in his mandibular anterior area, and severe mobility of mandibular incisors. Radiological examination and CT scan revealed a large, multilocular radiolucent, cystic lesion with distinct radiopaque borders (Figs. 1, 2).

The lesion extended from the right mandibular 2nd premolar to left mandibular 1st premolar area. The width of the periodontal ligament space seemed normal. There was a slightly compressible swelling of the mandibular anterior area, but there were no paresthesia, tenderness, or other changes in sensation. All mandibular anterior teeth were asymptomatic, and they had neither caries nor restorations. All mandibular anterior teeth except the left 1st incisor proved to be vital on electric pulp test. At the operation, the lesion consisted of multiple and separated cystic cavities with a very thin wall. The location and radiographic appearance of the abnormality and the age of the patient suggested a differential diagnosis of botryoid odontogenic cyst, odontogenic

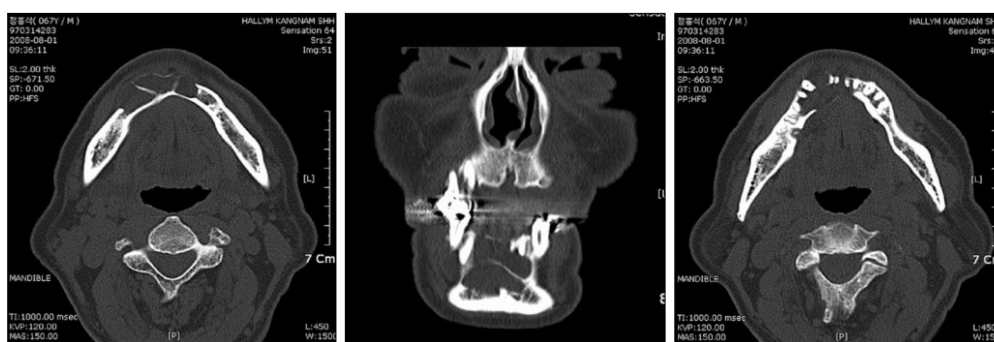
keratocyst and ameloblastoma. Finally, cyst enucleation and bone graft with xenograft (Biocera®, Osscotec, Korea) and allograft (Oragraft®, Megazen, Korea) materials.

During postoperative follow-up period, bony expansion disappeared and normal bone structure was formed on labial surface at 6 months after surgery (Fig. 3). Additionally, new bone formation occurred at operative site and there was no recurrence at 11 months after operation (Fig. 4).

The surgical specimen of the initial lesion consisted of several separate cyst cavities. Histologically, these were generally lined by one to three cell layers with thin and cuboidal epithelium. There was prominent separation of the epithelium from surrounding fibrous connective tissue wall. Botryoid odontogenic cyst showed focal nodular thickening of the lining epithelium. Focal thickening often showed swirling appearance of the cells (Figs. 5-7).



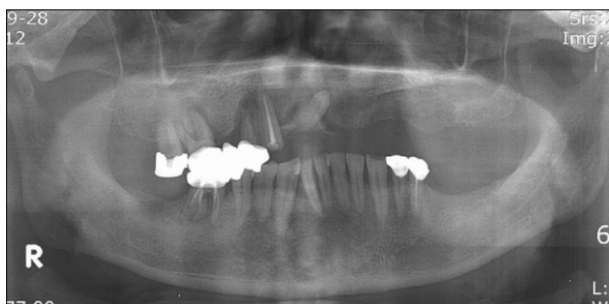
**Fig. 1.** Pre-operative panoramic view. Multilocular radiolucent lesion was seen from right mandibular 2nd premolar to left mandibular 1st premolar. There was a slight displacement of root area between right mandibular 2nd incisor and right mandibular canine. There was no root resorption and displacement of other involved teeth.



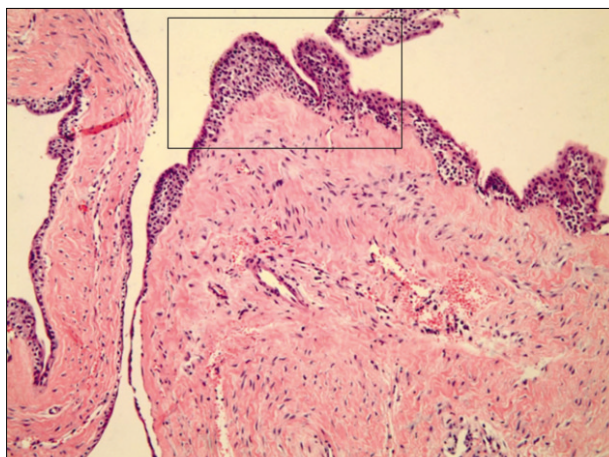
**Fig. 2.** Pre-operative axial and coronal CT scan. Multilocular radiolucent lesion was seen mandibular anterior and both body area. Bone expansion and thinning change of buccal labial cortical bone were seen on mandibular anterior area.



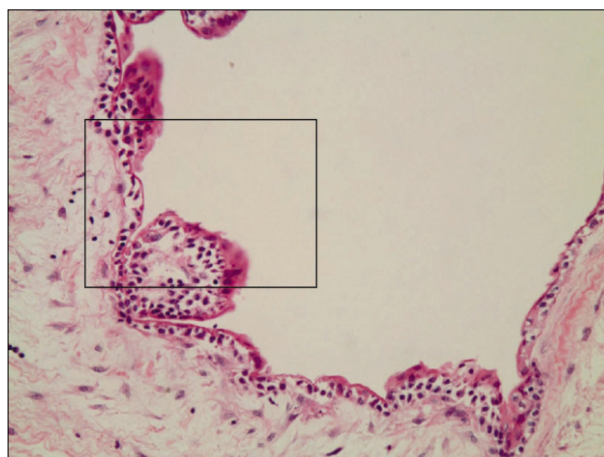
**Fig. 3.** 3-D CT scan at post-operative 6 months. New bone formation noted from right mandibular 2nd premolar to left mandibular 1st premolar. Bone expansion disappeared on labial surface and there was seen normal bone healing structure.



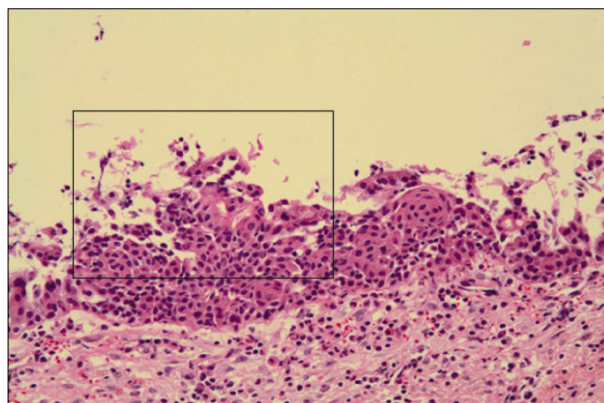
**Fig. 4.** Panoramic view at post-operative 11 months. Healing was favorable without recurrence.



**Fig. 5.** Botryoid odontogenic cyst has a thin, fibrous wall with an epithelial lining that is one to three cells' thickness in most area. This epithelium usually consists of flattened squamous cells, but sometimes the cells are cuboidal in shape (H&E stain,  $\times 100$  magnification).



**Fig. 6.** Botryoid odontogenic cyst shows focal nodular thickening of the lining epithelium, which are composed chiefly of clear cells (H&E stain,  $\times 200$  magnification).



**Fig. 7.** There was a thin epithelial lining with focal nodular thickening. These thickening often shows swirling appearance of the cells (H&E stain,  $\times 200$  magnification).

### Discussion and Conclusion

The first reported case of multilocular cystic lesion in the jaws resembled a cluster of grapes and was called botryoid odontogenic cyst.<sup>3)</sup> Since this original description, the BOC has been widely regarded as a variant of the lateral periodontal cyst and publications on the subject have not distinguished these from the smaller multicystic lateral periodontal cysts.<sup>4)</sup> Microscopically, the lesion is quite similar to the lateral periodontal cyst. Radiographically, most are monocystic; but occasional polycystic or multilocular cases have been encountered.<sup>5)</sup> The most frequent location of the BOC is the mandibular premolar area.<sup>6)</sup>

In this case, the BOC extended from the roots of the right premolar to the left premolar of the mandible. However, this multilocular radiolucency is not characteristic of the BOC. Radiographically and histologically, there are similarities between the BOC and some odontogenic cysts and odontogenic tumors such as ameloblastoma, odontogenic myxoma, glandular odontogenic cyst<sup>7)</sup> and various others in terms of appearance.

Odontogenic keratocyst (OKC) is more likely found from 10 to 40 years of age. The mandible is involved in 60 to 80% of cases, with a marked tendency to involve the posterior body and ascending ramus. The OKC tends to grow in an antero-posterior direction within the medullary cavity of the bone without

causing obvious bone expansion. This feature may be useful in differential clinical and radiographic diagnosis. The diagnosis of odontogenic keratocyst is based on the histopathologic features. The epithelial lining is 6 to 8 cells thick, with a hyperchromatic and palisaded basal cell layer. There was corrugated parakeratotic surface. The reported frequency of recurrence in various studies ranges from 5% to 62%.<sup>8)</sup>

Ameloblastoma is encountered in patients over a wide age range. A painless swelling of expansion of the jaw is the usual clinical presentation. The most typical radiographic feature is that of a multilocular radiolucent lesion. The lesion is often described as having a "soap-bubble" or "honey-combed" appearance. The follicular and plexiform patterns are the most common. Recurrence rates of 50 to 90% have been reported in various studies after curettage.<sup>9)</sup>

Odontogenic myxoma is more likely found in 25 to 30 years. Radiographically, the myxoma appears a unilocular or multilocular radiolucency that may displace or cause resorption of teeth in the area of the tumor. Histopathologically, A loose, myxomatous tumor can be seen filling the marrow spaces between the bony trabeculae. The inset shows stellate-shaped cells and fine collagen fibrils.<sup>10)</sup> Preoperative differential diagnosis can be carried out by means of incisional biopsy.

The BOC is known to be a recurrent odontogenic cyst. The recurrence rate may range from 15 to 20%.<sup>11,12)</sup> The proliferation rate of the epithelial lining has to be determined to estimate the possibility of recurrence. The risk of recurrence of the BOC is similar to that of odontogenic keratocyst.<sup>13)</sup> Histologically, the BOC has a distinctive thin, nonkeratinized epithelium which is one to five cell layers thick, and it has cystic spaces lined by squamous epithelium with thickening and clear cells.<sup>14)</sup> The BOC shows focal nodular thickening of the lining epithelium and swirling appearance. Clinicopathologic analysis of ten cases with three recurrences of BOC was reported. Eight of ten lesions were located in the mandible; the anterior mandible being the dominant site. Five of the ten lesions were unilocular, the largest measuring 4.5 × 1.2 cm. Only two of the ten botryoid odontogenic cysts were radiographically multilocular. Three lesions represented recurrences; 8,

10 and 10 years after previous surgical intervention, respectively. Perhaps the most significant information gained from this investigation of botryoid odontogenic cysts is the fact three of ten lesions recurred after initial surgical removal. It is important that practitioners take note of the fact that: 1) there may be a heightened recurrence potential for the botryoid odontogenic cyst that has gone unrecognized 2) recurrence may not occur until a decade after initial surgery; and, 3) lesions may occasionally become destructive.<sup>15)</sup>

Histologically, the surgical specimen of the 1st recurrence was covered by a hyperplastic oral epithelium, and the connective tissue between the surface epithelium and cyst epithelium contained several microcysts.<sup>16)</sup> These small cysts were lined by thin squamous epithelium and many of them were filled with keratin. It can be assumed that remnants of the thin cyst wall had been behind, resulting in multiple recurrences. In this report, histologically, there was little hyperplastic oral epithelium. Furthermore, cystic lesion was not filled with keratin. There has been no recurrence during postoperative follow-up, but further long-term check-up will be necessary in order to diagnose possible recurrences in time.

In conclusion, the BOC was commonly removed with cyst enucleation. But, size and multilocular patterns of lesion could be the main factors associated to recurrence in botryoid odontogenic cyst. The botryoid odontogenic cyst is rare but often found in cystic lesion when it is found multilocular pattern on dental X-ray screening, the clinician should include botryoid odontogenic cyst to the differential diagnosis and to optimal operation.

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