



Case report

Botryoid odontogenic cyst: A case report with immunohistochemical aspects

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ABSTRACT

We present a case of botryoid odontogenic cyst, affecting the anterior mandible in a 59-year-old Japanese female patient. The histochemical and immunohistochemical characteristics of the lesion are described. The nature of the cyst-lining epithelium was suggestive of an odontogenic origin as revealed by the expression of CK 10/13, CK14, and CK19, all of which have been reported to be present in the human enamel organ. An insignificant PCNA-immunoreaction was observed in the cyst-lining epithelium as compared with that of the odontogenic keratocyst. Furthermore, the appearance of diastase digestible, PAS-reactive material (consistent with glycogen) was evaluated that prefunctional cells of the dental lamina are a possible origin of the lesion.

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1. Introduction

Botryoid odontogenic cyst (BOC) is a polycystic lesion in the alveolar bone with or without proximity to a root of tooth. Since the initial report of two BOCs by Weathers and Waldron [1,2], more than 67 cases have been reported in the English literature [3]. The lesion is generally considered to represent a variant of the lateral periodontal cyst (LPC) [4–8] possibly the result of cystic degeneration and subsequent fusion of adjacent foci of dental lamina rests [5]. Although conservative enucleation of the BOC, as well as LPC, is the treatment of choice, a significant recurrence rate has been reported for BOC [9–15]. It is, therefore, of interest to document further characteristics of the cyst-lining epithelium of the lesion. In the present study, we report an additional case of BOC with histochemical and immunohistochemical characteristics of the cyst-lining epithelium.

2. Case report

A 59-year-old female patient was referred to the hospital of Meikai University School of Dentistry for evaluation of a swelling in the left anterior mandible. She had been aware of the asymptomatic swelling during the past two years. At the time of dental examination, panoramic and dental radiographs revealed a well-delimited, multi-locular radiolucent lesion, between the roots of the left mandibular lateral incisor and canine (Fig. 1). The cystic

radiolucency occupied the lateral aspects of the teeth and extended to the apical region with separation of the roots. An excisional biopsy was performed. Histopathologically, the lesion showed a multicentric cystic configuration lined by thin layer of squamous epithelium (Fig. 2a). The fibrous connective tissue wall was relatively free of inflammatory cells. The epithelium exhibited localized plaques with many clear cells containing centrally placed ovoid nuclei (Fig. 2b). The superficial layer of the epithelium showed cuboidal to columnar cells that were sometimes ciliated. These epithelial elements showed a diastase digestible PAS reaction, indicating the presence of glycogen (Fig. 3). On the basis of clinical and histopathological findings, the lesion interpreted as a BOC.

Positive immunoreactivities were obtained for CKs 10/13, 14 and 19 (Fig. 4a–c); there was expression on CK 10/13 and CK19 in the spinous and surface layers, CK14 in the basal cell layer. CK 18 weakly expressed only in the superficial layer of the epithelium (not shown). In addition to the cytokeratin profiles, immunostaining for proliferating cell nuclear antigen (PCNA) was also performed. PCNA-reactive cells were insignificant in the epithelium of BOC (Fig. 5a) as compared to that of the odontogenic keratocyst (currently termed keratocystic odontogenic tumor; 2005, WHO) (Fig. 5b) as a positive control section, in which an abundance of the immunoreactive cells could be seen.

3. Discussion

The BOC was originally described by Weathers and Waldron in [1]. The term was based on the clinical appearance that resembled a bunch of grapes. The BOC has a distinct proclivity for occurrence in the mandible anterior to the first molar [16]. Radiographically,

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Fig. 1. Radiographs showing a multilocular radiolucency between the roots of the mandibular canine and lateral incisor. The cystic lesion extends from the lateral aspects of the roots of teeth over the apical region with separation of the roots.

it is often multilocular and larger than the typical LPC, and often extends into the periapical regions of the related teeth. These two lesions share some histologic similarities; they contain characteristic thickened epithelial plaques or clear cell nests in the epithelial lining. Due to the similarity in histologic features and site of occurrence, the BOC has been considered a variant of the LPC [4–9].

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 [17]. Therefore, an extended postsurgical follow-up is recommended clinically.

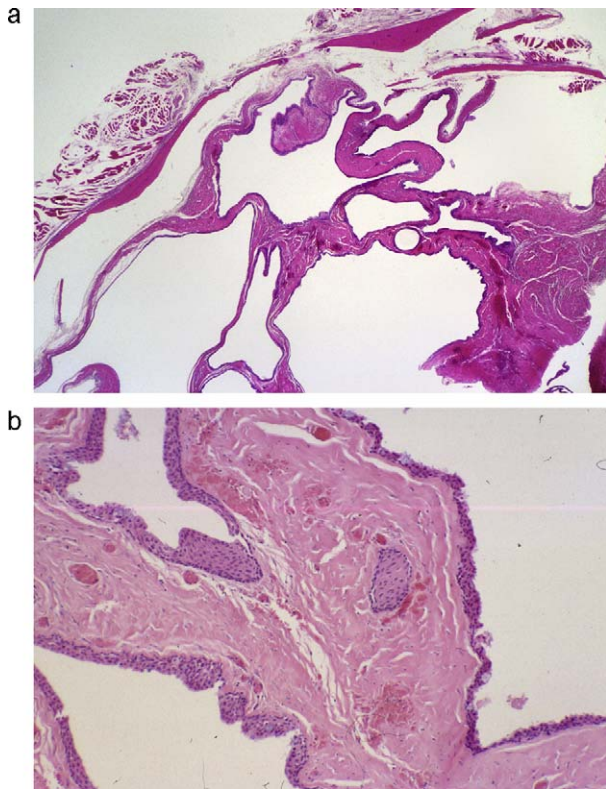


Fig. 2. Low-power photomicrograph demonstrates the polycystic nature with thin squamoid epithelial lining (a). Cyst-lined epithelium exhibiting focal plaque-like thickenings and clear cells with cuboidal to columnar cells at the superficial layer (b).

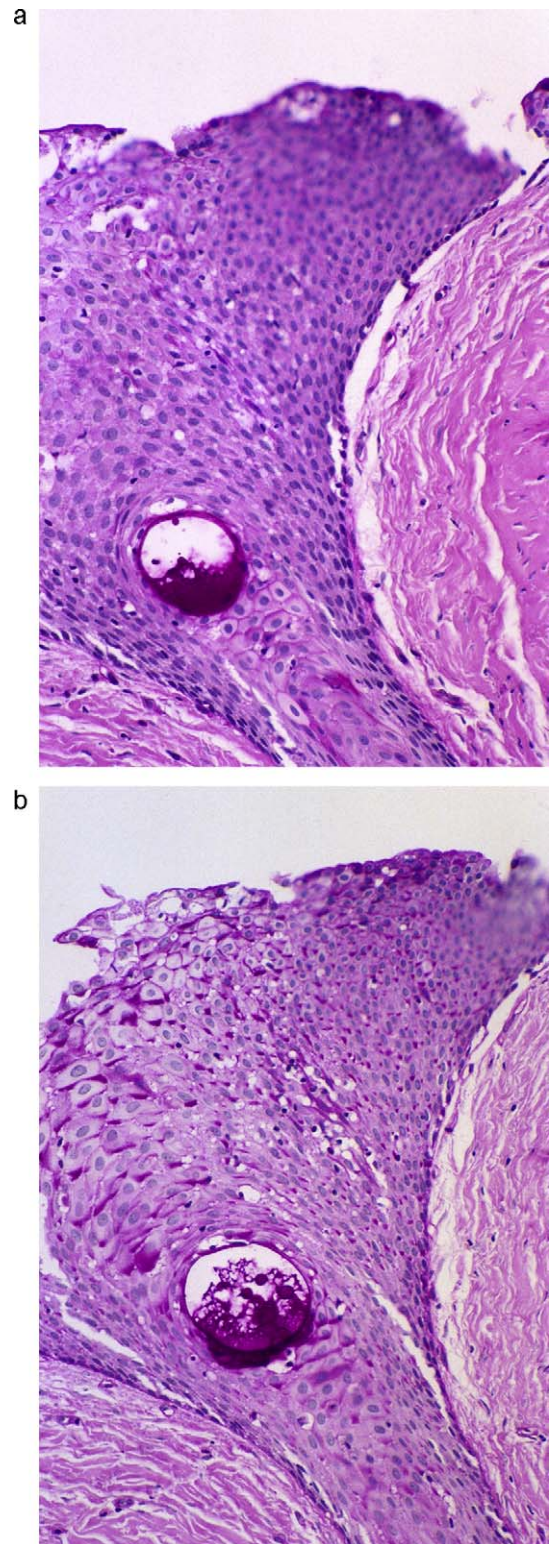


Fig. 3. PAS reactions with (a) or without (b) diastase digestion reveal the presence of glycogen in the cyst-lining epithelium.

Because of the presence of mucous cells and surface columnar cells, BOC shows some microscopic similarities to the glandular odontogenic cyst (GOC) or sialo-odontogenic cyst (SOC) [18–20]. However, these lesions were felt to be best classified as a BOC. They may lie at opposite ends of a spectrum. The presence of mucous cells does not detract from an odontogenic origin, this feature has been reported in a variety of odontogenic cysts such as the dentigerous

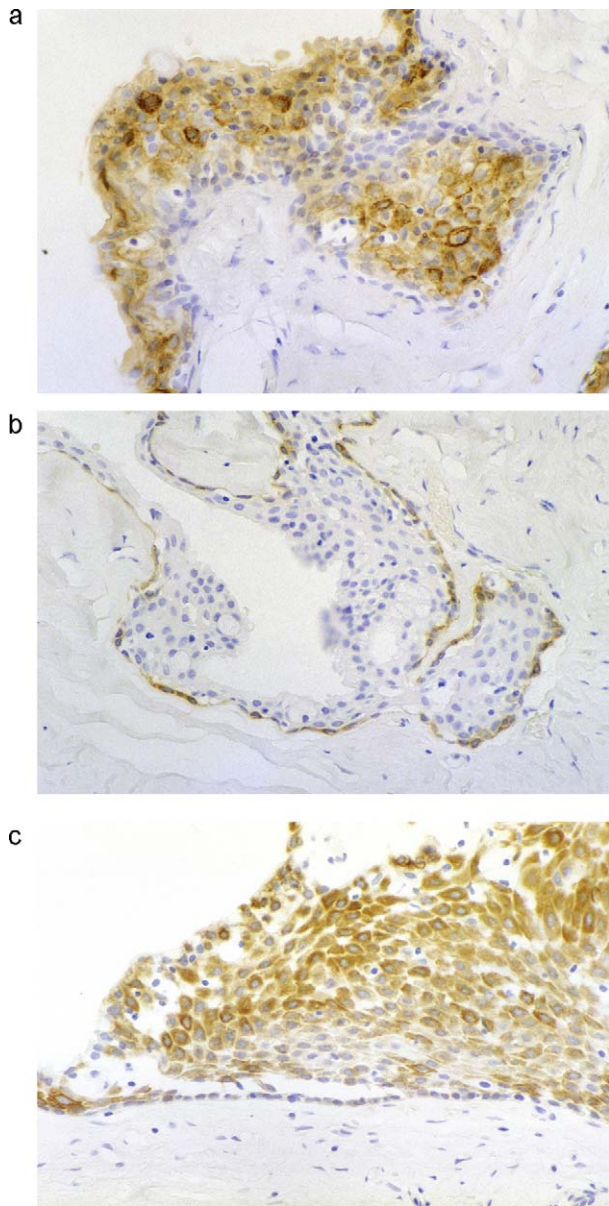


Fig. 4. CK immunoreactivities of CK 10/13 (a) and CK19 (c) show positive reactions in the spinous and surface layers, while CK14 (b) reveals a reactivity only in the basal cell layer.

cyst as a metaplastic phenomenon [21]. Furthermore, immunohistochemical studies [22,23] have suggested that the GOC is a histologic variant of BOC.

Immunohistochemical analysis on the BOC was performed by Heikinheimo et al. [13]; an odontogenic origin is supported by the presence of CK 19 immunoreactivity of the cyst-lining epithelium. These authors also observed a heterogeneously positive immunoreactions of CK 18, which has been recognized as a marker of simple ductal and glandular epithelia. They stressed the odontogenic cell of origin of BOC in which patchy distribution of CKs 13 and 16 could be found. In this regard, CKs 7, 13, 14 and 19 are present in human enamel organ [24]. Crivelini et al. [25] concluded that typical intermediate filament of odontogenic epithelium is CK 14, and that CKs 13 and 19 appear in squamous differentiation or epithelial cells near the surface epithelium. In the present study, we also observed positive staining for CKs 10/13, 14 and 19 in the respective layers of the cyst-lining epithelium. Considering these evidences, it is appar-

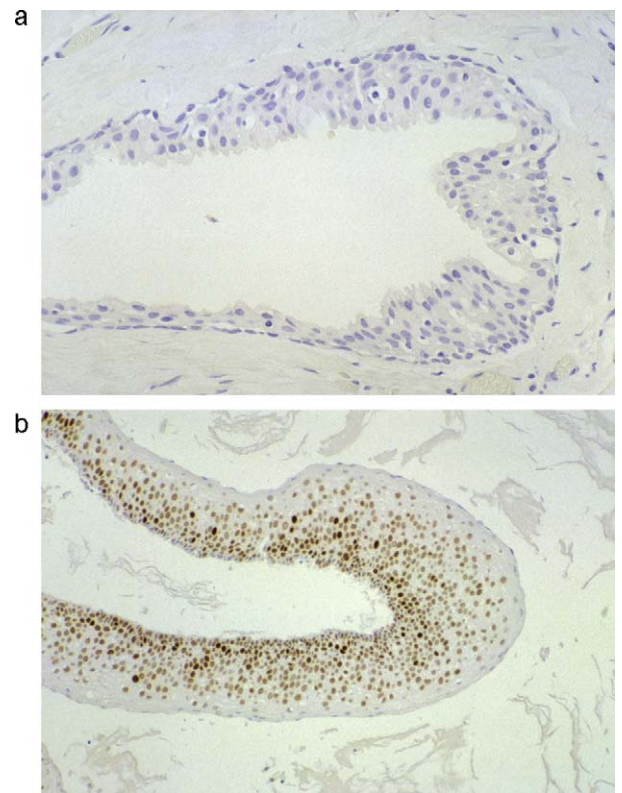


Fig. 5. PCNA immunoreactivity in the BOC (a) and OKC (b), the latter used as a positive control lesion.

ent that the epithelial cells of BOC are of odontogenic origin, as has been demonstrated by earlier investigators [5,7].

Several investigators have identified the sporadic presence of glycogen, detected as periodic acid-Schiff (PAS) positive, diastase digestible, material in the lining epithelium [16,26]. In the present study, glycogen was detected in the cyst-lining epithelium and in the epithelial plaques, but not always in the clear cells, although some of these cells were PAS positive. In this respect, Altini and Shear [7] stated that glycogen is by no means consistently demonstrable in the clear cells and is not confined to them. Redman et al. [26] observed no clear cells in their case, while the cyst-lining epithelium contained an amount of glycogen.

In a review of the English literature, no reports could be found which demonstrated the proliferative activity of the BOC. In this study, we could detect an insignificant PCNA immunoreactivity in the cyst-lining epithelium in comparison with that of odontogenic keratocyst. It can therefore be speculated that the cyst-lining epithelial cells, in which an amount of glycogen was detected, are biologically inactive. Altini and Shear [7] suggested that the reason for the limited growth potential of the LPC (and that of the BOC as well), compared with the odontogenic keratocyst is that the former arises from prefunctional cells of the dental lamina, whereas the latter presumably arises from that part of the dental lamina still possessing marked growth potential. The results of present study could be cited in support of this evidence.

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References

- [1] Weathers DR, Waldron CA. Unusual multilocular cysts of the jaws (botryoid odontogenic cysts). *Oral Surg Oral Med Oral Pathol* 1973;36:235–41.
- [2] Waldron CA. Odontogenic cysts and tumors. In: Neville BW, Damm DD, Allen CM, Bouquot JE, editors. *Oral and maxillofacial pathology*. Philadelphia: WB Saunders; 1995. p. 493–538.
- [3] Mendez P, Junquera L, Gallego L, Baladron J. Botryoid odontogenic cyst: clinical and pathological analysis in relation to recurrence. *Med Oral Pathol Cir Bucal* 2007;12:E594–8.
- [4] Fantasia JE. Lateral periodontal cyst. An analysis of forty-six cases. *Oral Surg Oral Med Oral Pathol* 1979;48:237–43.
- [5] Wysocki GP, Brannon RB, Gardner DG, Sapp P. Histogenesis of the lateral periodontal cyst and the gingival cyst of the adult. *Oral Surg Oral Med Oral Pathol* 1980;50:327–34.
- [6] Cohen DA, Neville BW, Damm DD, White DK. The lateral periodontal cyst. A report of 37 cases. *J Periodontol* 1984;55:230–4.
- [7] Altini M, Shear M. The lateral periodontal cyst: an update. *J Oral Pathol Med* 1992;21:245–50.
- [8] Carter LC, Carney YL, Perez-Pudlewski D. Lateral periodontal cyst. Multifactorial analysis of a previously unreported series. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;81:210–6.
- [9] Lynch DP, Madden CR. The botryoid odontogenic cyst. Report of a case and review of the literature. *J Periodontol* 1985;56:163–7.
- [10] Kaugars GE. Botryoid odontogenic cyst. *Oral Surg Oral Med Oral Pathol* 1986;62:555–9.
- [11] Greer Jr RO, Johnson M. Botryoid odontogenic cyst: clinicopathologic analysis of ten cases with three recurrences. *J Oral Maxillofac Surg* 1988;46:574–9.
- [12] Phelan JA, Kritchman D, Fusco-Ramer M, Freedman PD, Lumerman H. Recurrent botryoid odontogenic cyst (lateral periodontal cyst). *Oral Surg Oral Med Oral Pathol* 1988;66:345–8.
- [13] Heikinheimo K, Happonen RP, Forssell K, Kuusilehto A, Virtanen I. A botryoid odontogenic cyst with multiple recurrences. *Int J Oral Maxillofac Surg* 1989;18:10–3.
- [14] de Sousa SO, Campos AC, Santiago JL, Jaeger RG, de Araújo VC. Botryoid odontogenic cyst: report of a case with clinical and histogenetic considerations. *Br J Oral Maxillofac Surg* 1990;28:275–6.
- [15] High AS, Main DM, Khoo SP, Pedlar J, Hume WJ. The polymorphous odontogenic cyst. *J Oral Pathol Med* 1996;25:25–31.
- [16] Gurol M, Burkes Jr EJ, Jacoway J. Botryoid odontogenic cyst: analysis of 33 cases. *J Periodontol* 1995;66:1069–73.
- [17] Chi AC, Neville BW, Kliger BJ. A multilocular radiolucency. *AADA* 2007;138:1102–3.
- [18] Padayachee A, Van Wyk CW. Two cystic lesions with features of both the botryoid odontogenic cyst and the central mucopidermoid tumor: sialo-odontogenic cyst? *J Oral Pathol* 1987;16:499–504.
- [19] Gardner DG, Kessler HP, Morency R, Schaffner DL. The glandular odontogenic cyst: an apparent entity. *J Oral Pathol* 1988;17:359–66.
- [20] Farina VH, Brandao AAH, Almeida JD, Cabral LAG. Clinical and histologic features of botryoid odontogenic cyst: a case report. *J Med Case Rep* 2010;4:260.
- [21] Sciubba JJ, Fantasia JE, Kahn LB. Odontogenic cysts. In: Rosai J, Sobin LH, editors. *Atlas of tumor pathology, tumors and cysts of the jaw*. Washington, DC: AFIP; 2001. p. 15–49.
- [22] Semba I, Kitano M, Mimura T, Sonoda S, Miyawaki A. Glandular odontogenic cyst: analysis of cytokeratin expression and clinicopathological features. *J Oral Pathol Med* 1994;23:377–82.
- [23] Koppang HS, Johannessen S, Haugen LK, Haanaes HR, Solheim T, Donath K. Glandular odontogenic cyst (sialo-odontogenic cyst): report of two cases and literature review of 45 previously reported cases. *J Oral Pathol Med* 1998;27:455–62.
- [24] Domingues MG, Jaeger MM, Araújo VC, Araújo NS. Expression of cytokeratins in human enamel organ. *Eur J Oral Sci* 2000;108:43–7.
- [25] Crivelini MM, de Araújo VC, de Sousa SO, de Araújo NS. Cytokeratins in epithelia of odontogenic neoplasms. *Oral Dis* 2003;9:1–6.
- [26] Redman RS, Whitestone BW, Winne CE, Hudec MW, Patterson RH. Botryoid odontogenic cyst. Report of a case with histologic evidence of multicentric origin. *Int J Oral Maxillofac Surg* 1990;19:144–6.