

Botryoid odontogenic cyst: clinical and pathological analysis in relation to recurrence

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ABSTRACT

Objetives. Botryoid odontogenic cyst (BOC) is considered as an unusual multilocular variant of lateral periodontal cyst (LPC). Review of the literature indicates that this lesion has higher risk of recurrence than LPC, but objective reasons are still unknown. The aim of this study is identify main clinical and pathological variables associated with the risk of recurrence. **Study design.** A complete literature review about cases of BOC was made, from its first description up to the year 2006. A total of 67 cases of BOC were identified from year 1973 (first case) to the last publication in 2005. **Results.** 85.2% of the BOCs were located in the jaw, affecting to women (54.8% of the cases) in the fifth decade of the life. This lesion shows a well- circumscribed unilocular (60%) or multilocular (40%) radiolucency. Recurrence rate of cases with follow-up data was 32,4%. **Conclusions.** Size and multilocular patterns could be the main factors associated to recurrence in BOC.

Key words: *Recurrent botryoid odontogenic cyst, botryoid odontogenic cyst, lateral periodontal cyst.*

INTRODUCTION

Lateral periodontal cyst (LPC) represents near 0.8% of all central cyst (intraosseous) of maxillary bones (1). This lesion develops in the alveolar bone along the lateral aspect of an erupted vital tooth (2). Usually, clinical symptoms are absent and diagnosis is made by radiographic examination (2,3). Lateral periodontal cyst has been described as a solitary, well-defined interradicular radiolucency (3). Although uncommon, recurrence has sporadically been reported in the literature (3,4). LPCs have been reported chiefly in adults (range: 19-85 years) and bears a distinct proclivity for occurrence in the mandibular premolar area, followed by the anterior region of the maxilla (1). Botryoid odontogenic cyst (BOC) is considered as a variant of the LPC, with different radiographic features and higher risk of recurrence (3-15). The first reported case of a multilocular cystic lesion of the jaws resembled a cluster of grapes and called botryoid odontogenic cyst was made by Weathers and Waldron in 1973 (5). Radiographically, most are polycystic

(multilocular); but occasional unilocular cases have been encountered (6). Histopathological characteristics of BOC are similar to LPC. The recurrence rate of BOC may range between 15 and 33 %, higher than LPC (7,8). Since 1973, 67 cases of BOC have been reported in the literature. Over 62 cases with accessible information, 12 cases of recurrences of BOC (one of them with four recidivas) were reported (9). The aim of this article was a review of the literature, in order to identify clinical, radiographic or histological variables associated with the risk of recurrence and to aid practitioners for a correct treatment guidelines and appropriated control of this pathology.

MATERIAL AND METHOD

The present study reviews all the cases of BOC reported in the literature. To this effect, searches were made in PubMed® database, crossing the following key words: "botryoid odontogenic cyst", "recurrent botryoid odontogenics cyst" and "lateral periodontal cyst". A total of 67 cases of BOC

were identified since 1973 (first case) (5) up to the last case in 2005 (7). We had access to complete information about 62 cases that were the objective of our revision (Tables 1-3). The article published by Slater in 2006 (16), is a letter to editor who did not detail any clinical case. The documented variables were: age, sex, race, anatomic location, radiographic appearance, follow-up and evolution. All the reviewed cases were classified in two groups: patients with recurrence and without recurrence. In tables 1-3 the general performances of the revision are detailed.

RESULTS

Age. The patient age ranged between 23 and 85 years with a mean of 53,8 years for the reported cases (Table 1). The reported ages for the recidivant BOCs spanned 23 and 60 years with a mean of 47,7 years, whereas for not recidivant BOCs is between 26 and 78 years with a mean of 54,9 years (3,5-15).

Gender. Of 62 reviewed cases, 34 occurred in females (54,8%) and 28 in males (45,2%), showing the BOC a slight female predilection (3,5-15). No gender bias was detected in recurrent cases (Tables 2 and 3).

Race This lesion has preponderance in Caucasian patients (36 cases), and recurrent (7 cases) and non-recurrent (19 cases) variants were both more common in Caucasian race. This variable was not available in 11 cases (6,8-10,12,15).

Anatomic location. The most frequent location of BOC is the mandible (52 of 61), followed by the anterior region of the maxilla (9 of 61) (3,5-15). In one patient of 62 reviewed cases this important data was not specified (15). In the mandible, it usually occurs in the canine-premolar area (3,5-15), whereas in the maxilla it seems to shows predilection to anterior region (canine to canine) (10,15). All the reviewed recurrent cases were located in the mandible, with a single exception (Table 3).

Table 1. General characteristics of botryoid odontogenic cyst reviewed cases. M: male. F: female.

Author	Nº of cases	Median Age	Gender	Location	Symtoms	Radiographic appearance	Follow up
Weathers and Waldron (5)	1	85	M	Mandible	Yes	Multilocular	Not available
Kaugars (6)	3	56,3	2 (F) 1 (M)	3 Mandible	2 Yes 1 No	3 Multilocular	1 Recurrence
Greer et al. (10)	10	46	4 (F) 6 (M)	9 Mandible 1 Maxilla	5 Yes 5 No	8 Unilocular 2 Multilocular	3 Recurrence
Phelan et al. (11)	1	23	F	Mandible	No	Multilocular	Recurrence after 3 years
Heikinheimo et al. (9)	1	51	F	Mandible	Yes	Multilocular	4 recurrence in 9 years
Redman et al. (12)	1	67	M	Mandible	No	Multilocular	Not available
Lindh et al. (13)	2	44,5	2 F	2 Mandible	2 Yes	2 Multilocular	1 recurrence 1 not available
de Sousa et al. (14)	1	54	M	Mandible	Yes	Multilocular	Recurrence after 7 years
Gurol et al. (15)	33	56,4	17 (F) 16 (M)	24 Mandible 8 Maxilla 1 not available	18 Yes 8 No 7 not available	15 Unilocular 1 Multilocular 17 not available	2 recurrence 10 no recurrence 21 not available
Carter et al. (3)	2	58	1 (F) 1 (M)	Mandible	2 No	2 multilocular	No recurrence
Ramer et al. (8)	6	56	5 (F) 1 (M)	6 Mandible	4 Yes 2 No	4 Unilocular 2 Multilocular	2 recurrence
Üçok et al. (7)	1	32	F	Mandible	Yes	Multilocular	Not available
TOTAL	62	53,8	34 (F) 28 (M)	52 Mandible 9 Maxilla 1 Not available	35 Yes 20 No 7 not available	18 Multilocular 27 Unilocular 17 not available	12 recurrence 25 no recurrence 25 not available

Table 2. General characteristics of not recurrent botryoid odontogenic cyst reviewed cases. M: male. F: female.

Author	Number of cases	Median time of follow up	Median Age	Gender	Location	Radiographic appearance
Kaugars (6)	2	0,5 years	56,5	2 (M)	2 mandible	1 multilocular 1 bilocular
Greer et al. (10)	7	1,57 years	46,4	3 (F) 4 (M)	All in mandible	6 unilocular 1 multilocular
Gurol et al. (15)	10	4,5 years	59,3	7 (F) 3 (M)	8 mandible 2 maxilla	6 unilocular 4 not available
Carter et al. (3)	2	Not available	58	1 (F) 1(M)	2 mandible	2 multilocular
Ramer et al. (8)	4	1,75 years	56,5	4 (F)	All in mandible	All unilocular
TOTAL	25	2,17 years	54,9	15 (F) 10 (M)	23 mandible 2 maxilla	16 unilocular 5 multilocular 4 not available

Table 3. General characteristics of recurrent botryoid odontogenic cyst reviewed cases. M: male. F: female.

Author	Number of recurrence	Median time of recurrence (years)	Median Age	Gender	Location	Radiographic appearance
Kaugars (6)	1	9	56	F	Mandible	Multilocular
Greer et al. (10)	3	9	45	1 (F) 2 (M)	2 mandible 1 maxilla	2 unilocular 1 multilocular
Phelan et al. (11)	1	3	23	F	Mandible	Multilocular
Heikinheimo et al. (9)	1	4 veces en 9 años	51	F	Mandible	Multilocular
Lindh et al. (13)	1	13	35	F	Mandible	Multilocular
de Sousa et al. (14)	1	7	54	M	Mandible	Multilocular
Gurol et al. (15)	2	No disponible	54,5	2 (M)	2 Mandible	1 multilocular 1 not available
Ramer et al. (8)	2	4,5	55	1 (F) 1 (M)	2 Mandible	All multilocular
TOTAL	12	7,8	47,75	6 (F) 6 (M)	11 mandible 1 maxilla	9 multilocular 2 unilocular 1 not available

Clinical presentation and recurrence. 36,4% of the reviewed cases were asymptomatic. Clinical symptoms (pain, tumefaction, paresthesia, oral fistula), were present in 63,6% of the reviewed patients (3,5-15) but this information was not available in seven of the published cases, (3,6,8,10-12,15). Only 37 reviewed cases reported follow-up information (3,6,8-15) (follow up range: 3 months–14years) (10,15). In 12 of these 37 cases recurrences were reported (32,4%) (6,8-11,13-15). Twenty-five did not show recurrence (67,6%) (3,6,8,10,15) (Tables 2 and 3). Unfortunately in 25 cases this information was not available (7,12,13,15).

Radiographic Appearance. All the reviewed cases showed a round, well circumscribed radiolucency (3,5-15). 60% (27/45) of the cases were unicystic (8,10,15), whereas the 40% (18/45) showed a multilocular image (3,5-15). No radiological information was found in 17 cases (15). Recurrent cases mainly presented multilocular images (9/11) (6,8-11,13-15) (Table 3). Only 5 of 21 patients without recurrence showed a multilocular radiographic image (3,6,10), with a mean follow-up period of two years (Table 2).
Histological characteristics. Histopathologically, no differences between recurrent and non-recurrent cases were

reported. All the studied cases presented common characteristics (3,5-15): 1.-Multiple cysts separated by connective tissue. 2.-Slight, non-keratinized squamous lining epithelium with epithelial thickenings referred to as plaques 3.-Many clear cells with cytoplasmatic glycogen, detected as periodic acid-Schiff positive. 4.-Absence of inflammatory infiltrate in subjacent connective tissue.

DISCUSSION

BOC is an unusual and controversial pathology. By definition, LPC is a non keratinized development cyst occurring in the alveolar bone along the lateral aspect of a vital tooth (17). The so-called BOC could be defined as a "multilocular variant of LPC" (17). Nevertheless, other authors (2,18) do not consider LPC and BOC the same entity, and define BOC as a "multicystic odontogenic lesion with histological characteristics of lateral periodontal cyst", or "cystic lesion similar to lateral periodontal cyst". Otherwise, High et al. (19), proposed the term "polymorphic odontogenic cyst". This term may include BOC and other lesions as glandular odontogenic cyst and mucoepidermoid intraosseous carcinoma. All of them would have similar recurrence rates, unusual presentation and confuse histological pattern. In fact, some reported cases identified as BOC have been considered radiographic and histopathologically as glandular odontogenic cysts (16). This discussed question emphasize the relevance of the careful histopathological diagnosis on this lesions (16,19-21).

Clinical and histopathological differences between LPC and BOC may be considered a controversial question. Usually this lesion presents a common site of occurrence in most of the published cases (1-15,22). Mean age of published BOCs is 53,8 years (3,5-15) (rate: 23-85 years) (5,11), in agreement with previously established reports (8,15) and for LPC is 46 years (rate: 14-78 years) (3). Also, mean age of non-recurrent BOCs was slightly superior (more of 7 years) than recurrent BOCs. According to this point, BOCs appear in older patients than LPCs. Otherwise, recurrent BOCs appear in younger patients than non-recurrent BOCs. Radiographically, 81,1% of recurrent BOCs showed a multilocular radiolucency (6,8-11,13-15), as opposed to 23,8% of non-recurrent BOC (3,6,10). Therefore, a multilocular cyst diagnosed as BOC increases the risk of recurrence. Radiographic images of LPC show a well circumscribed, round or ovoid radiolucency, with sclerotic margins (1,2). Most of cases present small size (rate: 2,5 to 15 mm) (3,4,22) and are usually located between apex and the cervical margin of a tooth (1,2). Otherwise, Greer et al. (10) reported series of BOCs between 5 mm and 45 mm size. Therefore, the lesion size could represent a radiological difference between LPC and BOC, although this variable was not considered in our study. Moreover, mean size of recurrent BOCs described in the literature is 31 mm (10), as opposed to non-recurrent cases: 9,6 mm (10). Although more studies are needed, joint to the multilocular pattern, lesion size could be another important factor in relation to BOC recurrence.

LPCs are usually asymptomatic (3). Of the reviewed BOCs,

63,6% induced symptoms, maybe because of its greater size. Therefore, BOC must not be considered as LPC variant, but a multicystic lesion, of faster growth and similar histology to the LPC. This idea has been reported by van der Wall (18) in 1992 but not clinically confirmed.

Microscopically the main difference between LPC and BOC is the multilocular histological aspect of BOC. Complete enucleation would be more difficult in these cysts. (14). The few number of BOC documented cases difficult the known of the main epidemiology data. Gurol et al. (15) described a series of 33 cases of BOCs that, curiously, represents half of the total of cases published of this pathology in 34 years. Histopathologic differential diagnosis between paradental cyst and infected mandibular buccal cyst may be difficult, and variables like age of presentation, anatomic localization, radiographic aspect or involved teeth should be considered. As well, histological differences between LPC and BOC may not conclude the diagnosis (16), and clinical or radiological variables are usually needed.

Recurrence rate published for BOC range between 15-20% (7) and 33% (8). In a series of 33 cases, Gurol et al. (15) reported only 12 cases with a detailed follow-up and two of them recur in a non-defined time after the first surgery. This author reported a recurrence rate of 16,6% only including the followed-up cases. In our revision, 12 recurrences of 37 cases of BOC with follow up information (6,8-11,13-15) could be found, so recurrence rate is 32,4%. We think that real recurrence percentage of BOC could be higher, because many cases were not controlled, or followed for short periods. Heikinheimo et al. (9) described a case with four recurrences in a 9 years period.

In conclusion, BOCs represent an unusual pathology, in relation to vital teeth of mandibular canine-premolar area. Size and multilocular patterns could be the main factors associated to recurrence. High recurrence rate detected in this review, suggests the need of a more aggressive treatment than usually applied (enucleation and curettage). Systematic use of Carnoy solution could diminish recurrence percentages, as it happens with queratocysts, but new studies are necessary to deeply understand the effectiveness of this treatment.

Otherwise, BOQs have been reported in older patients than LPCs, and clinical symptoms are usually present at the time of diagnosis. Besides, BOQs presents larger size and higher risk of recurrence than LPCs and may show a multilocular radiographic pattern.

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