Glandular odontogenic cyst: Case report and review of the literature
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Glandular odontogenic cyst (GOC) is a rare developmental cyst of the jaws. It is included in the World Health Organization (WHO) histologic typing of odontogenic tumors under the terms glandular odontogenic cyst or sialo-odontogenic cyst. The most common site of occurrence is the anterior mandible, and it occurs mostly in middle-aged people. A predilection for men is observed. Clinical findings are not specific, and an asymptomatic swelling is frequently observed. A unilocular or multilocular, well-defined radiolucency is usually seen. The microscopic features of GOC, particularly the morphology of the epithelium, strongly suggest an origin from the remains of dental lamina. GOC has an unpredictable and potentially aggressive nature, which may indicate a high tendency of recurrence. The treatment of choice is still controversial, varying from a curettage to local block excision. A long-term follow-up should be carried out. The aim of this article is to report a case of glandular odontogenic cyst that recurred four times and to emphasize the importance of long-term follow-up. The origin, epidemiology, clinical and radiographic aspects, and treatment of the GOC are also discussed. (Quintessence Int 2004;35:385–389)

Key words: botryoid odontogenic cyst, glandular odontogenic cyst, jaw cyst, lateral periodontal cyst, odontogenic cyst, sialo-odontogenic cyst

The glandular odontogenic cyst (GOC) is a developmental cyst of the jaws that was described in 1988 by Gardner et al2 as a distinct entity. Padayachee and Van Wyk3 had already described the microscopic features of these cysts but called them "sialo-odontogenic cysts." The GOC is included in the WHO histologic typing of odontogenic tumors under the terms glandular odontogenic cyst or sialo-odontogenic cyst.3

Glandular odontogenic cysts are rare, with just over 50 cases reported in the literature.4–6 Magnusson et al7 analyzed 5,800 biopsies of jaw cysts between 1977 and 1995 and observed that only seven cases fulfilled the criteria for GOC, comprising 0.012% of the total. The most common site for this cyst is the anterior mandible, but it can occur in every area of the jaws.8–15 Clinically, it appears that the GOC occurs mostly in middle-aged people11–13,15 with a mean age of 46.7 years in males and 50.0 years in females.1 The female: male ratio is 19:28,5 while other reported cases show a slight predilection for men.10,14 Recurrences have been described in 21% of the cases.

Clinical findings are not specific. An asymptomatic swelling is frequently observed. Radiographically, a unilocular or multilocular, well-defined radiolucency, with scalloped margins is usually seen.6,9,11–15,16

In the present study, a case of GOC is reported, emphasizing the potential for recurrence and the importance of long-term follow-up, as well as the origin, epidemiology, clinical and radiographic aspects, and treatment.

CASE REPORT

A 44-year-old white man was seen in the Stomatology Clinic at the Bauru School of Dentistry, University of São Paulo, complaining of pain in the mandible. He
had suffered trauma to the chin 20 years earlier and, subsequently, had undergone four surgeries to treat a lesion in the area. He was unable to provide further information regarding those surgeries, and the reports of microscopic examinations of the biopsies were not available. Intraoral examination revealed a fluctuating, purple swelling extending from the left lateral incisor to the first molar involving the buccal and lingual aspects of the alveolar ridge (Fig 1). Panoramic radiograph showed a multilocular, well-defined, osteolytic lesion with scalloped margins in the left mandible (Fig 2). A periapical radiograph obtained in 1981 had already revealed multilocular radiolucency in the area (Fig 3). An aspiration biopsy was performed, and this contained material compatible with peripheral blood; biopsy reduced the swelling in the affected area. Exploratory surgery was then carried out in which the bony cavity covering was partially removed. The surgical diagnosis was of a cavity covered by a membrane with cortical discontinuities (Fig 4). Histopathologic examination confirmed that the lesion was multilocular, and the epithelium was composed of cuboidal or columnar cells that were occasionally ciliated. The epithelial lining exhibited crypts and cyst-like spaces filled by mucous-like material. In other areas, the lesion was lined by stratified squamous epithelium with focal epithelial thickenings. The underlying connective tissue showed various small cystic spaces, lined with the same epithelium, without inflammatory cells (Figs 5 and 6). The final diagnosis was multilocular glandular odontogenic cyst. The lesion was enucleated and the osseous walls were sauceredized. Figure 7 shows the immediate postoperative of the surgical area. The material was analyzed microscopically, and the histopathologic examination confirmed the previous diagnosis.
Fig 5 Photomicrograph exhibiting cyst lined by thin, nonkeratinized epithelium and composed by connective tissue wall with microcyst. Typical thickenings are present on the epithelial lining of microcysts. (Hematoxylin and eosin stain; original magnification ×25 [left] and ×100 [right]).

Fig 6 Photomicrographs showing cystic connective tissue wall with microcyst lined by columnar and cuboidal cells presenting crypts or microcysts and focal mural and luminal thickenings. The crypts are either empty or fulfilled with eosinophilic material. (Hematoxylin and eosin stain; original magnification ×125.)

Fig 7 Immediate postoperative.

Fig 8 Healed area in the clinical control at 18 months (left). Oral rehabilitation with a partial removable denture (right).

Fig 9 Panoramic radiograph revealing bone healing, without signs of recurrence, 18 months after surgery.

Analgesic, anti-inflammatory, and antibiotic medications were prescribed to the patient. A follow-up of 18 months showed a well-healed area, covered by noninflammatory mucosa exhibiting normal features. A partial removable denture was installed, and the patient was rehabilitated (Fig 8). The panoramic radiograph revealed the surgical area to be totally healed, without signs of recurrence (Fig 9).

DISCUSSION

The clinical aspects of the present case are consistent with those reported for GOC in the literature. There is a slight predilection for men, and the condition occurs mostly in middle-aged patients. The anterior mandible is the most common site of occurrence for this cyst, but in the current case report, the lesion...
extended to the left first molar. Radiographically, a multilocular radiolucency is usually seen, although unilateral lesions have been described. Larger cysts may produce swelling, as was found in the present case. The pain reported by the patient was probably due to stretching and pressure on the neurovascular bundles, since inflammation is a rare finding. The GOC has an intraosseous location, and it bears no direct relationship to the periodontal ligament, as does the lateral periodontal cyst (LPC). It usually occurs apically in relation to the teeth, but may show interdental extensions.

The preoperative aspiration of a colorless, low-viscosity fluid content may be a helpful clinical indication of a GOC. In the present case, the aspirated fluid was compatible with peripheral blood, perhaps a result of the previous surgeries.

The microscopic characteristics of GOC, particularly the morphology of the epithelium, strongly suggest an odontogenic epithelial origin, especially from the remains of dental lamina. The thin, cuboidal epithelium in some areas resembles the reduced enamel organ epithelium. Furthermore, mural or luminal plaques of eosinophilic and fusiform or rounded cells are seen in other lesions with accepted odontogenic origin, such as lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC), gingival cyst of adults, and occasionally in denterogenous cyst. These areas of epithelium thickening may be comparable to the proliferative changes seen on epithelial cells of dental lamina. They may be indicative of an active process of proliferation of the odontogenic epithelium in their genesis. Immunohistochemical studies strongly support the odontogenic nature of GOC.

The identification of osteodentin and the negative reaction for epithelial membrane antigen (EMA) in the area of glandular structures suggest that these features are not of true glandular origin and support the concept of odontogenic differentiation in the GOC. The cystic epithelium reacted positively to antibodies directed against cytokeratins 7, 13, 14, and 19 and negatively to cytokeratins 8 and 18.

The GOC shares some histopathologic features with LPC, such as plaquelike epithelial thickenings. This might explain why some authors believe the GOC to be a clinical-microscopic variant of the LPC. However, the LPC is a less aggressive lesion with limited growth potential and low recurrence. Some clinical (location, multilocularity, and age of occurrence) and microscopic (multicystic aspect and plaquelike epithelial thickenings) features of the GOC are shared with BOC. These characteristics might indicate a common origin for these lesions. It is possible that both lesions are merely variants of the same entity. However, based on clinical, radiographic, behavioral, and histopathologic features, some authors believe that GOC, LPC, and BOC should be considered different entities.

Although the presence of intraepithelial mucous-containing cystic spaces is a characteristic shared with other odontogenic cysts, the GOC has microscopic features and biologic behavior distinct enough to be regarded as an entity.

The GOC has been linked to the mucoepidermoid carcinoma. According to Magnusson et al, the central mucoepidermoid carcinoma, especially the low-grade variant, is regarded as the most important histopathologic differential diagnosis from GOC. It has been speculated that GOC may represent the most benign end of the spectrum of central mucoepidermoid carcinoma.

However, Gardner et al censured the term sialo-odontogenic cyst because an origin in salivary glands could not be established. The term was used due to the fact that the mucous pools are often lined by eosinophilic cuboidal cells, resulting in some resemblance to salivary gland ducts. A second reason for the use of the term sialo-odontogenic is the presence of mucous cells and pools of mucin in the epithelial lining. However, these features can be found in various odontogenic cysts and do not imply a salivary gland origin.

Some slow-growing lesions of the jaws should be included in the differential diagnosis of the GOC, such as cysts (periapical inflammatory cyst, LPC, BOC, residual cyst, dentigerous cyst, aneurismatic osseous cyst, odontogenic keratocyst); myxoma; central lesion of giant cells; fibrous dysplasia; ameloblastoma; and central mucoepidermoid carcinoma.

GOC has an unpredictable and potentially aggressive nature, suggested by its osseous extensions, penetration of cortical bones, locally invasive growth and high recurrence rate following conservative treatment. The current case supports this conclusion. The four surgeries undergone by the patient in the previous 20 years, all without resolution, indicate the high tendency of GOC to recur. Figure 4 shows extensions of the lesion, perforating the cortical buccal tissue. Hussain et al found a high recurrence rate of 55% and a long recurrence interval, with an average of 4.9 years. The mechanism of recurrence may be partly related to the thinness of the cyst wall and to the presence of microcysts, making complete removal at surgery difficult.

In the 47 reported cases until 1998, Koppan et al observed a recurrence rate of 21%. This number increases to 50% when the BOC is considered and decreases to 0% to 30%, when the LPC is analyzed.

The treatment of choice is still controversial because of the few reported cases. The lesions are quite variable in size and aggressiveness, and the method of treatment varies from a curettage to local...
block excision.\textsuperscript{5,210,12,13,15,16,19} Gardner and Morency\textsuperscript{11} suggested that curettage or enucleation might be the treatment of choice for GOC, provided that the clinician closely monitors the patient for several years and the lesion does not occur in the posterior maxilla because of the difficulty of controlling recurrences in this region. A long-term follow-up should be carried out.\textsuperscript{7,12,15,18} A period of 3 years is recommended by the literature, but some authors suggest that the patients should be followed for at least 5 years after treatment.\textsuperscript{3} Prognosis is good.\textsuperscript{16}

In the present case, a total enucleation of the lesion was conducted followed by sacuereation of the osseous walls, with the aim of minimizing the possibility of recurrence. Eighteen months after surgery, the area was clinically well-healed, and the patient was rehabilitated (Fig 8). Panoramic radiograph did not show any sign of recurrence (Fig 9). However, it was advised that follow-up should continue.

REFERENCES