Case report

Glandular odontogenic cyst: case report and review of diagnostic criteria

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ABSTRACT

The glandular odontogenic cyst (GOC) is an uncommon jaw bone cyst of odontogenic origin with unpredictable and potentially aggressive behaviour. It also has the propensity to grow to a large size and tendency towards recurrence. GOC can be easily misdiagnosed microscopically as a central mucoepidermoid carcinoma. This paper reports a case of GOC in a 56-year-old male and reviews the main criteria for accurate diagnosis. The diagnosis of GOC can be extremely difficult due to the rarity of the cyst and lack of clear diagnostic criteria.

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1. Introduction

The glandular odontogenic cyst (GOC) is a rare developmental cyst of the jaws first described in 1988 by Gardner et al. (1988). In 1987, Padayachee and Van Wyk reported two cases that were similar to the botryoid odontogenic cyst, but with a gland element and suggested the name “sialo odontogenic cyst”. This cyst has an uncertain histogenesis and was recently listed by the World Health Organization as a developmental odontogenic epithelial cyst. It is characterized by an epithelial lining with cuboidal or columnar cells, both at the surface and lining, with crypts or cyst-like spaces within the thickness of the epithelium. The “glandular odontogenic cyst” (GOC) has two clinically important attributes: it has “a high recurrence rate” and it displays “an aggressive growth potential” (Jankowski, 2010).

The most common location of GOC is the anterior mandibular region in approximately 85% of the cases, especially beyond the midline. GOC occurs mostly in middle aged men. Clinically, this lesion is generally painless, slow growing and its size can vary from less than 1 cm in diameter to large dimensions (Krishnamurthy et al., 2009). Small cysts may be asymptomatic, while large ones can cause bone expansion accompanied by pain and paraesthesia. An association with impacted teeth, resorption and tooth displacement is common. The rate of recurrence of GOC described in literature ranges from 21% to 55% depending on the different treatment options (Boffano et al., 2010).

Radiologically, GOC may be unilocular with a well-defined border, but occurs more often as a multilocular cyst with well-defined radiopaque margins (Krishnamurthy et al., 2009). The histological features of GOC strongly suggest an origin from the remains of dental lamina. The microscopic features are a cystic cavity lined with non-keratinized, stratified, squamous epithelium, localized plaque-like thickenings of the epithelium, variable numbers of mucous-secreting cells in the surface layer of the epithelium, a tendency to subepithelial fibrous tissue formation, multiple cysts and the absence of inflammation. The superficial layer of the epithelium consists of eosinophilic cuboidal cells, which make the surface irregular (Oliveira et al., 2009; Oliveira Neto et al., 2010). Treatment recommendations for GOC in the literature are inconsistent and not evidence based due to the rarity of the condition, ranging from minor procedures, such as enucleation and curettage, to major surgery, such as marginal resection, peripheral ostectomy, and partial jaw resection (Oliveira Neto et al., 2010).

The authors describe a new case of GOC in a male patient, along with the immunohistochemical profile. A literature review was performed through a search of the Medline database on GOC in order to establish guidelines for correct diagnosis based on clinical, histological and immunohistochemical features, as this cyst has features that overlap with botryoid odontogenic cyst and mucoepidermoid carcinoma.

2. Case report

A 56-year-old white male was referred to the Department of Oral-Maxillofacial Surgery, School of Dentistry, Federal University of Rio Grande do Norte, Brazil, with a four month history of
a swelling in the anterior region of the mandible and pain on compression (Fig. 1). There had been a significant increase in the symptoms in the previous month. There was no significant past medical history. He reported no history of extraoral trauma to the region and no tobacco or alcohol use. The intraoral examination revealed swelling in the anterior mandible region. The lesion exhibited tenderness upon palpation with crepitis in some regions and signs of fluctuation. The mass was covered by intact mucosa and there were no signs of infection or sensory loss (Fig. 2). The panoramic radiograph revealed a well-defined, unilocular, radiolucent lesion extending from sagittal midline (region of the symphysis) to the right mental foramen laterally, over the mandibular canal (Fig. 3). An occlusal radiograph clearly showed the expansion and erosion of the lingual and buccal cortex, resulting in a thinning of these bone plates. Provisional diagnoses of unicystic ameloblastoma, central giant cell lesion and keratocystic odontogenic tumour were given. Aspiration yielded a serous brownish-red fluid. Incisional biopsy was performed under local anaesthesia and the specimen was sent for histopathological examination, which revealed fragments of cyst wall lined with stratified squamous epithelium of varying thickness, along with epithelial whorls in some areas. Some of the underlying cells displayed clear cytoplasmic processes, and the luminal surface was covered by pseudo-cilia in other areas (Figs. 4–6). The cyst capsule was dense connective tissue with sparse mononuclear infiltrate. PAS staining revealed numerous PAS-positive mucous cells and a small number of pseudo-glandular structures were seen throughout the epithelial lining. Immunostaining revealed that the epithelium of the lesion stained for CKs-7, 8, 10, 13 and 19 (Figs. 7 and 8). The connective tissue wall consisted of numerous cholesterol clefts, mild chronic inflammatory cells and hemorrhaging. The histopathological diagnosis was GOC. Under local anaesthesia, the
lesion was enucleated in one piece and the buccal cortical bone was removed through an intraoral incision, followed by careful curettage to avoid damage to the inferior alveolar nerve. Peripheral ostectomy was also performed to eliminate extensions of the cyst (Figs. 9 and 10). On macroscopic examination, there was one fragment of tissue 2.8 x 1.5 cm in dimension, brownish in colour and with a firm consistency. The histological examination of the surgical specimen confirmed the diagnosis of GOC. Eight months following the surgical procedure, a postoperative panoramic radiograph was taken, which revealed evidence of bone healing and complete resolution.

3. Discussion

Glandular odontogenic cyst (GOC) is an uncommon jaw bone cyst of odontogenic origin with approximately 114 cases reported in
In analysis of 12 cases and a Medline search, Shen et al. (2006) found a slight predominance among males and the third decade of life. In agreement with the literature, the anterior region of the mandible was affected in the patient described here. However, while the literature reports that most GOCs have slow growth (Oliveira et al., 2009), the patient in the present case reported fast growth of the cyst.

There is no pathognomonic radiological feature for GOC. Thus, radiographically, this entity may appear as a unilocular or multilocular radiolucent lesion in either jaw, with well-defined and sclerotic borders in some cases and ill-defined borders in others, often reaching large dimensions. Expansion and thinning of cortical plates are sometimes observed on occlusal radiographs (Oliveira Neto et al., 2010). These features were observed in the present case, in which the panoramic radiograph revealed a unilocular radiolucent lesion in the anterior region of the mandible. According to Tran et al. (2004), lesions that appear as unilocular radiolucencies located in the mandible include lateral periodontal cysts, simple bone cyst, keratocystic odontogenic tumour, ameloblastoma and central giant cell lesion. A lateral periodontal cyst does not usually exceed 1 cm in diameter. A simple bone cyst is a pseudocyst, as it does not have an epithelial lining and occurs as a result of impaired bone repair due to trauma to the anterior region of the mandible. The patient described here reported no incidents of trauma. A keratocystic odontogenic tumour also presents as a radiolucent area with well-defined cortical margins and predominance in the mandible with a tendency towards growing in the medullary spaces of the bone without causing obvious bone expansion. A central giant cell lesion is a benign localized proliferation that is osteolytic, sometimes aggressive and more frequent in the mandible. Unicystic ameloblastoma is a variant of ameloblastoma presenting as a cyst, with more than 90% of cases involving the mandible (Oliveira Neto et al., 2010). Tran et al. (2004) report that aneurysmal bone cysts have clinical and radiographic features similar to GOC, but are rarely found in the jaws and, when present in the oral cavity, are most often found most the region of the mandibular molars in patients under 20 years of age. It should be stressed that clinical and radiographic findings of GOC vary and are often not pathognomonic, as reported by Shen et al. (2006).

The clinical and radiographic aspects of the present case suggested provisional diagnoses of unicystic ameloblastoma, central giant cell lesion or keratocystic odontogenic tumour. However, aspiration revealed a serous brownish-red fluid and the clinical diagnosis was changed to a cystic lesion. Krishnamurthy et al. (2009) report that clear, low-viscosity aspiration fluid may be a helpful clinical indication of GOC, and pre-operative aspiration and fluid inspection may be advisable. In contrast, brownish-red aspirate was obtained in the case these authors report, which was attributed to blood, as observed in the present case as well.

Shen et al. (2006) report that, while the histogenesis of GOCs remains uncertain, most authors believe that these cysts originate from odontogenic epithelium. Important evidence in support of such a concept lies in the morphology of the epithelium. GOC arising in tooth-bearing areas has led to suggestion of an odontogenic epithelial origin, while, histopathologically, salivary components have been noted. The thin, cuboidal or columnar epithelium is reminiscent of reduced enamel epithelium. The plaque-like or spherical structures are also commonly seen in odontogenic lesions, such as lateral periodontal cyst and gingival cyst in adults, and rarely in dentigerous cyst and adenomatoid odontogenic tumour (AOT). Intraepithelial duct and mucous-producing cells with or without cilia have been ubiquitously reported in odontogenic lesions.

Kaplan et al. (2008) stated that “due to similarities in microscopic characteristics between GOC and lesions such as botryoid cyst, radicular and dentigerous cysts with mucous metaplasia and more importantly low-grade mucoepidermoid carcinoma, a definitive diagnosis can be difficult to make”. Nevertheless, they added that a diagnosis of GOC had to be based on the histopathological diagnosis of the five major features. These are squamous epithelium, varying thickness, cuboidal eosinophilic (“hobnail”) cells, mucous (goblet) cells and intraepithelial glandular or duct-like structures. The histological diagnosis of the present case fulfilled the histopathological criteria for GOC proposed by these authors.

According to Jankowski (2010), although it is unlikely that any reports used such strict criteria, this statement clearly indicates that the histopathology alone may be considered to be insufficiently specific in each and every case of GOC. Nevertheless, it does raise the possibility that GOCs were underreported in many if not all of the SR included reports. This could have occurred because the histopathologists assigned a diagnosis of GOC only to those lesions that they were absolutely satisfied and fulfilled the requirements of this diagnosis. The immunohistochemistry studies carried out by Shen et al. (2006) revealed CK expression of GOCs with a high degree of tissue specificity. The epithelium of the GOCs stained for CKs AE1, 7, 8/18, 10/13, 14 and 19, with slight changes in patterns, but.

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**Table 1**

<table>
<thead>
<tr>
<th>Clinical features (Thor et al., 2006)</th>
<th>Radiographic aspects (Oliveira et al., 2009)</th>
<th>Histological criteria (Thor et al., 2006)</th>
<th>Immunohistochemical aspects (Oliveira et al., 2009)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age: median 49</td>
<td>Unilocular or multilocular radiolucent lesion (Shen et al., 2006)</td>
<td>Stratified squamous epithelium of varying thickness, along with epithelial whorls in some areas</td>
<td>CK7: surface layer epithelial</td>
</tr>
<tr>
<td>Sex: male</td>
<td>Well-defined borders</td>
<td>Mucous cells and cuboidal eosinophili/hobnail cells and ciliated cells</td>
<td>CK8: surface, suprabasal layer</td>
</tr>
<tr>
<td>Location: mandible (anterior region)</td>
<td>Others aspects: peripheral osteosclerosis</td>
<td>Intraepithelial microcysts/duct-like</td>
<td>CK10: suprabasal and plaque</td>
</tr>
<tr>
<td>Usually asymptomatic</td>
<td>Occasionally: root resorption</td>
<td>Lining width variations, focal proliferation</td>
<td>CK14: suprabasal, basal and plaque</td>
</tr>
<tr>
<td>Grow: slow</td>
<td>and displacement of tooth</td>
<td>Minor criteria: mucous-lined crypts, papillary projections and clear-vacuolated cells</td>
<td>CK19: all layers</td>
</tr>
</tbody>
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The literature by 2010. According to Shen et al. (2006), GOC has a frequency rate of only 0.012–1.3% of all cysts. This paper reports a case of GOC in a 56-year-old male and discusses the main aspects for the proper correct diagnosis through a literature review.
no staining for CKs 16 and 20 was visualized, which shares a similar distribution to CKs with odontogenic epithelium. The authors also found that the two CKs were strongly expressed in GOC epithelium in spite of small differences in different specimens and different cell layers, histochemically indicating that the epithelium of a GOC may be odontogenic origin in nature. The present case had similar results, with positivity for CK 7, 8, 10, 13, 14 and 19.

According to Toida et al. (1994) it is important to differentiate GOC from central mucoepidermoid carcinoma (MEC), particularly the low-grade and predominantly cystic type. Vered et al. (2010) report that, in cases for which the differential diagnosis of GOC or central MEC cannot be made based on morphological features alone, especially when the biopsy sample is small, extensive maspin immunolabeling in the epithelial-mucous cells (in both the cytoplasm and nuclei) may be used as a tool to favour the diagnosis of central MEC over GOC. These lesions share some histopathological features, but have a wide range of biological behaviour that may differ among lesions of the same type, and among different types of lesions. This may reflect the pluripotential character of the odontogenic epithelium, which has the ability to develop diverse types of lesions. In light of this, no definite separation can be made between central LGMEC and GOC, but a more frequent immunopositivity of the epithelial-mucous cells favours a diagnosis of central MEC. Table 1 displays clinical, radiographic, histological and immunohistochemical criteria for the correct diagnosis of GOC, but it should be stressed that diagnosis of this cyst should be based on histological and immunohistochemical aspects.

Several methods of treatment of GOC, both conservative and aggressive, have been suggested. Conservative options include enucleation, marsupialization, curettage with and without peripheral ostectomy, curettage with adjuvant Carnoy solution and cryotherapy. However, several authors have recently recommended a more aggressive approach (marginal resection to partial jaw resection) as the treatment of choice for GOCs. According to Boffano et al. (2010) in addition to the treatment modality used, the rate of recurrence is directly related to the size and locular nature of the lesion. Thor et al. (2006) reported that most cases of recurrence were of large multilocular lesions with cortical perforations. In the present case, there was no cortical perforation and conservative treatment associated to peripheral ostectomy was performed.

Due to the propensity of a GOC to recur and become large, it is important for patients diagnosed with this cyst to be followed carefully (Jankowski, 2010, Shen et al. 2006). Shen et al. (2006) also reported that it should be emphasized that the relative rarity of this cyst make evaluation of the behaviour and prognosis is difficult. Over half of the published cases had been followed up for periods of less than two years or were very recent cases, whereas recurrence often develops after the third year of the postoperative period.

4. Conclusion

The clinical and radiographic findings of GOC are varied and often not pathognomonic, but, following the literature review, the present study suggests criteria for the correct histological diagnosis of GOC. Additionally, the authors refer to the difficulty of definitive histopathological diagnosis in cystic lesions with the presence of gland-like structures. We agree with Kaplan et al. (2008) criteria when reported, that microscopically GOC is characterized by an epithelial lining with cuboidal or columnar cells at both the surface and lining, and crypts or cyst-like spaces within the thickness of the epithelium. On radiographic study, it is typically radiolucent, unilocular or multilocular, with well-defined borders. Its occurrence may be associated with impacted or displaced teeth, as well as root resorption. Expansion is observed in the majority of cases, with thinning, erosion or perforation of the cortical plates in 67%.

Competing interests

None.

References


