

Odontogenic glandular cyst: a case report

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Abstract: Glandular Odontogenic Cyst (GOC) is a rare developmental cyst of the jaws. The histological features of GOC strongly suggest an origin from the remains of dental lamina. Radiographically, GOC presents as well-defined radiolucencies with uni- or multilocular appearance. A case of GOC in a 54-year-old black female is presented here. Clinical, histological and imaging features were evaluated. Due to the high tendency of recurrence and the aggressive potential of GOC, careful clinical and radiological evaluation must be carried out. CT scans are recommended because they provide accurate information about locularity of the lesion, cortical integrity, expansion of the lesion and involvement of the contiguous soft tissue. (*J Oral Sci* 51, 467-470, 2009)

Keywords: Glandular odontogenic cyst; odontogenic cysts; jaw cysts; computed tomography.

Introduction

In 1987, Padayachee and Van Wyk (1) described two multilocular mandibular cystic lesions that were similar to botryoid odontogenic cysts but with a glandular element. A year later, Gardner et al. (1988) reported eight other cases with histopathological features mimicking mucoepidermoid carcinoma. (2) They called the lesion Glandular Odontogenic Cyst (GOC).

The GOC is a rare developmental cyst of the jaws. The World Health Organization (WHO) includes GOC as a developmental odontogenic epithelial cyst under the terms glandular odontogenic cyst or sialo-odontogenic cyst. It occurs mostly in middle-aged men, especially in the anterior mandible. GOC can be asymptomatic or may cause pain, slow-growing swelling and tooth displacement. (3-12)

Radiographically, GOC presents as well-defined radiolucencies with uni- or multilocular appearance (3,6,9-11,13). Loss of cortical integrity and root resorption may occur (3,7,11,12). Computed tomography (CT) and magnetic resonance imaging (MRI) are recommended for diagnosis, surgical planning and follow-up (3,12,14).

The histological features of GOC strongly suggest an origin from the remains of dental lamina, the microscopic features being a cystic cavity lined by nonkeratinized, stratified, squamous epithelium, localized plaque-like thickenings of the epithelium, variable numbers of mucous-secreting cells in the surface layer of the epithelium, tendency to subepithelial fibrous tissue formation, multiple cysts and absence of inflammation. The superficial layer of the epithelium consists of eosinophilic cuboidal cells, which make the surface irregular (5,7,9,12).

The cyst has an aggressive nature and high tendency of recurrence, so long-term follow-up should be carried out. The treatment is controversial, varying from conservative methods to block excision. The recurrence rates appear to be correlated with the conservative approach (3,6-9,12,13).

Case Report

A 54-year-old black female was referred to the Stomatology Department of Heliópolis Hospital, São Paulo, Brazil, with a swelling in the anterior region of the

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mandible and pain on compression. The patient reported 8 months of evolution, with significant increase in the last 6 months. No medical reports were mentioned. Oral examination showed edentulous jaws and normal color and the appearance of covering mucosa, despite remarkable mass.

Occlusal radiography revealed a unilocular radiolucency with well-defined borders involving the symphysis region and mandibular body (Fig. 1A). Axial and coronal CT (bone window) showed a large, hypodense and multilocular lesion with cystic pattern and well defined borders, extending from the sagittal midline to the mental foramen laterally, with inferior limits ranging from the alveolar process to the mandibular base (Figs. 1B, 1C). Severe buccal expansion and thin septa within the lesion were noted. In some areas, the buccal cortical bone was perforated. No involvement of soft tissues was depicted (Fig. 1D).

The lesion was enucleated and the material sent for

histopathological examination, which showed stratified squamous epithelium with variable thickness, cubic and ciliated cells in the superficial lining with intraepithelial cysts and mucous cells and cyst capsule of a dense connective tissue with sparse mononuclear infiltrate. The final diagnosis was again glandular odontogenic cyst (Fig. 2).

After 9 months, recurrence was observed during follow-up. The patient was subjected to curettage, a conservative approach, under general anesthesia.

The microscopic analysis showed a cystic capsule with stratified squamous epithelium of variable thickness, mucous and ciliated cells. There were intraepithelial cysts and the connective tissue was dense and well vascularized, confirming the diagnosis of glandular odontogenic cyst (Fig. 2).

The patient recovered well and had no complaints one month later. Unfortunately, the patient did not return for further follow-up evaluation.

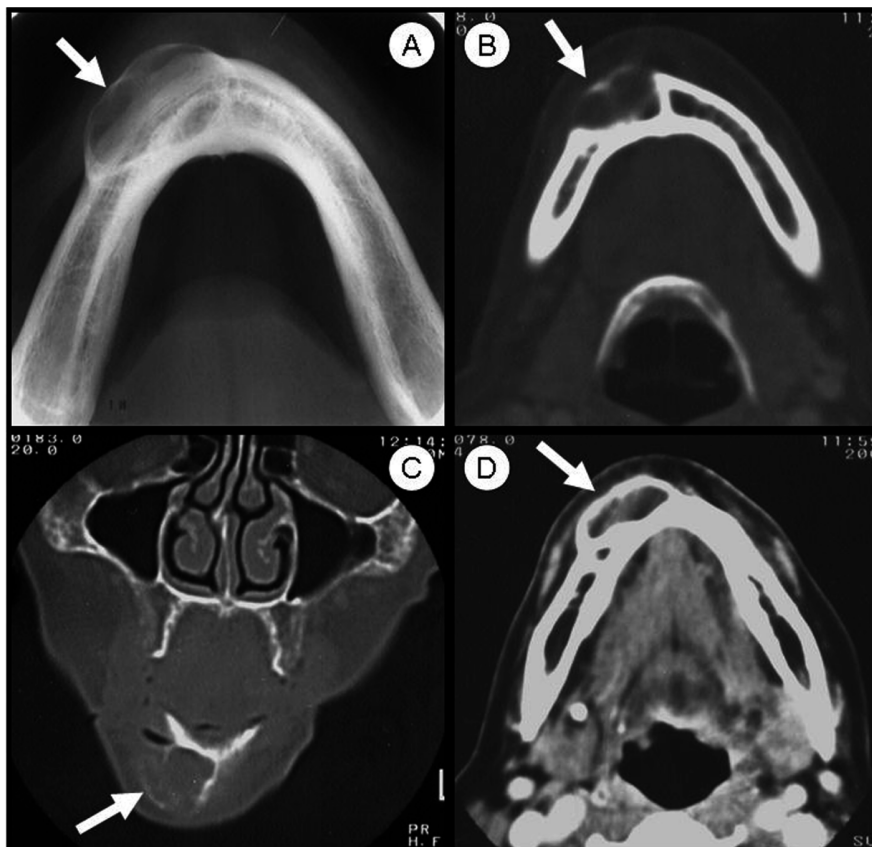


Fig. 1 Occlusal radiography and computed tomography images. (A) occlusal radiography demonstrating a unilocular radiolucency with well-defined borders; (B and C) Axial (B) and coronal (C) CT (bone window) showing a large, hypodense and multilocular lesion with cystic pattern and well-defined borders (arrows); (D) Axial CT (soft tissue window) showing no involvement of soft tissues (arrow).

Discussion

A case of GOC, a rare developmental cyst of the jaws, is hereby presented. (3-12)

Similar to previous studies, our case had mandibular involvement, with swelling and pain as complaints. The radiological and histological features were also in accordance with previous reports, showing a well-defined radiolucency with well-defined borders (3,6,9-11,13) and remains of dental lamina (5,7,12) respectively.

The disagreement was related to gender predilection and the growing time: the literature shows predilection for men and a slow-growing process (3-12), while the present case was reported in a woman who reported rapid growth.

The difference in radiographic features of the case reported was due to the discrepancy between conventional radiographic exam diagnosis and CT exam diagnosis: the conventional images showed a unilocular radiolucency, as mentioned in the literature. (3,6,9-11,13) However, the CT scan revealed a multilocular lesion and cortical bone

perforation, validating the need for multiple plane images in cases of GOC. The CT clarified the limits of the lesion and the involvement of the contiguous soft tissue, and proved helpful in treatment planning. (3,12,14)

The aggressive nature of the lesion was evident, especially because of the recurrence and the significant increase in size since the first diagnosis. This behavior supports the belief that conservative treatment may lead to recurrence, and invasive techniques should be considered, such as marginal resection and segmental resection. As mentioned before, the cyst has an aggressive nature and high tendency for recurrence and the treatment remains controversial (3,6-9,12,13).

In conclusion, GOC is a rare and aggressive lesion with a high recurrence rate. Careful clinical and radiological evaluation must be carried out. CT scans are recommended because they provide accurate information about locularity of the lesion, cortical integrity, expansion of the lesion and involvement of the contiguous soft tissue.

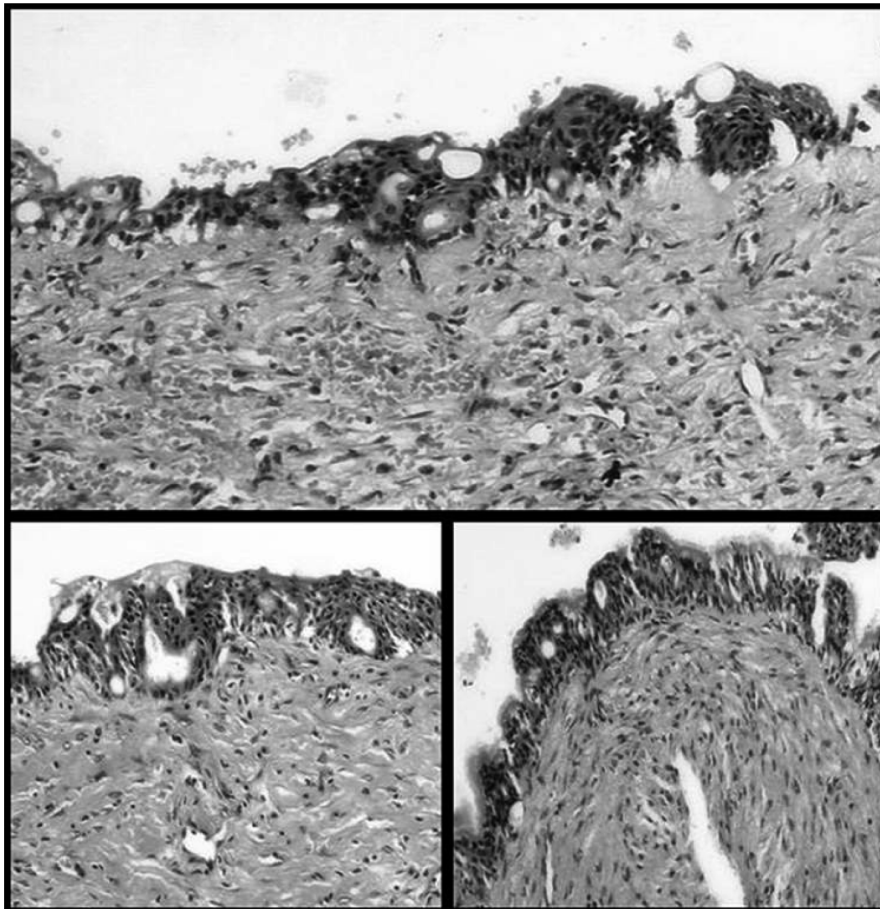


Fig. 2 Microscopic analysis showing a cystic capsule with stratified squamous epithelium of variable thickness, mucous and ciliated cells. Intraepithelial cysts and dense and well vascularized connective tissue can be observed.

References

1. Padayachee A, Van Wyk CW (1987) Two cystic lesions with features of both the botryoid odontogenic cyst and the central mucoepidermoid tumour: sialo-odontogenic cyst? *J Oral Pathol* 16, 499-504.
2. Gardner DG, Kessler HP, Morency R, Schaffner DL (1988) The glandular odontogenic cyst: an apparent entity. *J Oral Pathol* 17, 359-366.
3. Manor R, Anavi Y, Kaplan I, Calderon S (2003) Radiological features of glandular odontogenic cyst. *Dentomaxillofac Radiol* 32, 73-79.
4. Ertas U, Buyukkurt MC, Gungormus M, Kaya O (2003) A large glandular odontogenic cyst of the mandible: report of case. *J Contemp Dent Pract* 15, 53-58.
5. Tran PT, Cunningham CJ, Baughman RA (2004) Glandular odontogenic cyst. *J Endod* 30, 182-184.
6. Sittitavornwong S, Koehler JR, Said-Al-Naief N (2006) Glandular odontogenic cyst of the anterior maxilla: case report and review of the literature. *J Oral Maxillofac Surg* 64, 740-745.
7. Kaplan I, Gal G, Anavi Y, Manor R, Calderon S (2005) Glandular odontogenic cyst: treatment and recurrence. *J Oral Maxillofac Surg* 63, 435-441.
8. Velez I (2006) Glandular odontogenic cyst. Report of two cases and review of the literature. *N Y State Dent J* 72, 44-45.
9. Osny FJ, Azevedo LR, Sant'Ana E, Lara VS (2004) Glandular odontogenic cyst: case report and review of the literature. *Quintessence Int* 35, 385-389.
10. Kasaboglu O, Basal Z, Usubutun A (2006) Glandular odontogenic cyst presenting as a dentigerous cyst: a case report. *J Oral Maxillofac Surg* 64, 731-733.
11. Qin XN, Li JR, Chen XM, Long X (2005) The glandular odontogenic cyst: clinicopathologic features and treatment of 14 cases. *J Oral Maxillofac Surg* 63, 694-699.
12. Thor A, Warfvinge G, Fernandes R (2006) The course of a long-standing glandular odontogenic cyst: marginal resection and reconstruction with particulated bone graft, platelet-rich plasma, and additional vertical alveolar distraction. *J Oral Maxillofac Surg* 64, 1121-1128.
13. Noffke C, Raubenheimer EJ (2002) The glandular odontogenic cyst: clinical and radiological features; review of the literature and report of nine cases. *Dentomaxillofac Radiol* 31, 333-338.
14. Farman AG, Kushner GM, Gould AR (2002) A sequential approach to radiological interpretation. *Dentomaxillofac Radiol* 31, 291-298.