

CASE REPORT

Glandular odontogenic cyst in maxilla: A case report and literature review

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Received: 18-07-2013

Accepted: 07-08-2014

ABSTRACT

Glandular odontogenic cyst (GOC) is an uncommon jaw bone cyst of odontogenic origin described in 1987 by Gardner *et al.* It is a cyst having an unpredictable and potentially aggressive behavior. The increased recurrence rate can be due to its multilocularity and incomplete removal of the lining following conservative treatment. Clinically, the most common site of occurrence is the anterior region of mandible. GOC has a slight male predilection and occurs primarily in middle-aged patients. This article presents a case of glandular odontogenic cyst in a 30-year-old female patient in the posterior region of the maxilla, which is quite rare.

Key words: Glandular cyst, maxilla, odontogenic cyst

INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare lesion that arises in the tooth bearing areas of the jaws. Padayachee and Van Wyk initially reported it as a sialodontogenic cyst in 1987 based on the possibility of salivary gland origin but its odontogenic origin was first described in 1988 by Gardner *et al.*, who proposed the name GOC because the cyst wall epithelium was odontogenic and contained mucin elements with no evidence of salivary tissue involvement.^[1-3] The term mucoepidermoid cyst or mucous producing cyst was used by Sadeghib in 1991 due to the microscopic findings of mucus producing cells and squamous cells.^[4,5] In the 1992 World Health Organization (WHO) typing of odontogenic tumors, GOC was defined as “a cyst arising in the tooth-bearing areas of the jaws characterized by an epithelial lining with cuboidal or columnar cells both at the surface and lining crypts or cyst-like spaces within the thickness of the epithelium”.^[6]

GOC is relatively rare lesion with a frequency rate of 0.012-1.3% of all the jaw cysts and its prevalence rate is 0.17%.^[7] It has two clinically important attributes: A “high recurrence rate” and an “aggressive growth potential”.^[8]

GOC primarily occurs in middle-aged patients with slight male predilection and the most common site of occurrence is mandibular anterior region where it usually presents as a painless, slow-growing swelling. Radiographically, these cysts are described as well-defined, unilocular or multilocular without specific diagnostic characteristics.^[9] Histologically, GOC shows a non-keratinized stratified squamous epithelial lining, focal plaque like thickenings within the lining, microcysts or intraepithelial crypts containing mucin, mucous cells and hyaline bodies, eosinophilic cuboidal or columnar cells that may be ciliated, papillary projections of epithelium and absence of inflammation in the subepithelial connective tissue.^[1,5,10,11] The relative rarity of the lesion prompted us to add one more of our case and review the literature.

CASE REPORT

A 30-year-old female patient reported with a swelling in the upper right back region of the jaw. The swelling was present since 8 months which was painless and increased gradually in size. Extraoral examination showed slight asymmetry with fullness of the right side of the face. Intraorally a diffuse, non fluctuant and firm swelling was seen with normal overlying mucosa extending from the buccal aspect of 11-17 obliterating the vestibule [Figure 1]. The teeth were tender on percussion. Panoramic radiographic examination revealed a well-defined unilocular radiolucency extending from 15-17. 16 was carious with root resorption and roots of 15 and 17 were displaced [Figure 2]. A provisional diagnosis of radicular cyst was made. Gross examination showed a smooth to rough mass measuring 2.5 × 2 cm and the cut section showed a cystic

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10.4103/0973-029X.140923



Figure 1: Clinical photograph shows swelling in the upper right back region of the jaw



Figure 2: Orthopantomogram shows radiolucent area in 15, 16, 17 region



Figure 3: Gross specimen showing a smooth lobulated mass

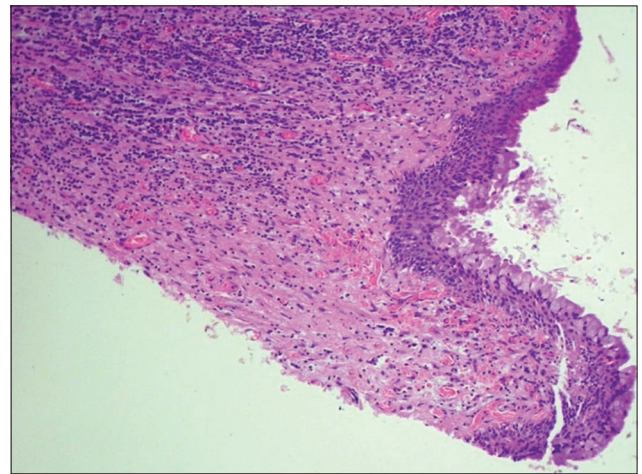


Figure 4: Photomicrograph shows lining epithelium (H&E stain, $\times 100$)

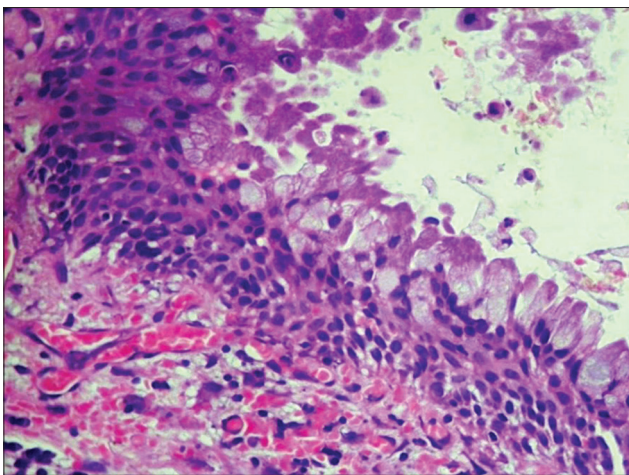


Figure 5: Photomicrograph shows surface ciliated columnar epithelium (H&E stain, $\times 400$)

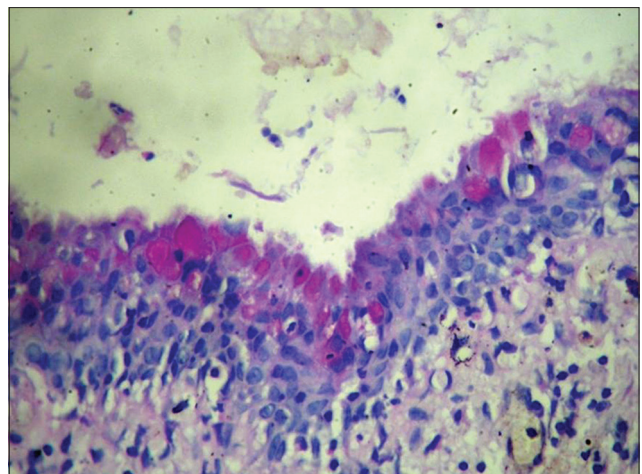


Figure 6: Photomicrograph shows special stained mucous cells (PAS stain, $\times 400$)

wall filled with necrotic material [Figure 3]. Histopathologic examination, revealed a cystic cavity lined by a non keratinized stratified squamous epithelium with surface layer composed of ciliated columnar cells [Figures 4 and 5]. Intraepithelial crypts containing PAS positive mucous cells were also observed [Figure 6]. Plaque like epithelial thickenings were also seen in one area. Subepithelial inflammatory infiltrate was found in the underlying connective tissue.

Immunohistochemistry (IHC) staining was done using CK-18, CK-19, p53, Ki-67. Among them CK-19 and Ki-67 were found positive. [Figures 7 and 8]. A final diagnosis of GOC was made.

DISCUSSION

GOC is a relatively rare entity. Magnusson and co-authors evaluated 5900 cases of jaw bone cysts and found only 7 cases of GOC, about 0.12%; whereas Van Heerden and others reported 1.3% of GOC in their study.^[7,12]

In the present case the patient is a middle-aged female whereas existing literature reports a slight male predilection. The most common site of occurrence is the anterior region of mandible followed by anterior region of maxilla and posterior region of mandible.^[2,11,13,14] Occurrence of GOC in posterior region of maxilla is rare. About three cases have been reported till date and ours is probably the fourth case.

A diagnosis based only on clinical and radiological examination is difficult because of similarities with various other intrabony pathologies, hence a histopathological evaluation becomes mandatory.^[15]

The histopathological characteristics of GOC have been divided into major and minor categories by Kaplan *et al.*^[16] GOC should be distinguished from lateral periodontal cyst, botryoid odontogenic cyst, surgical ciliated cyst, radicular cyst with mucous metaplasia and central MEC as it exhibits considerable overlapping of histopathological features [Figure 9].^[17,18]

Kaplan *et al.*, in their study found that GOC showed positivity for p53 and Ki-67. When compared to MEC, these

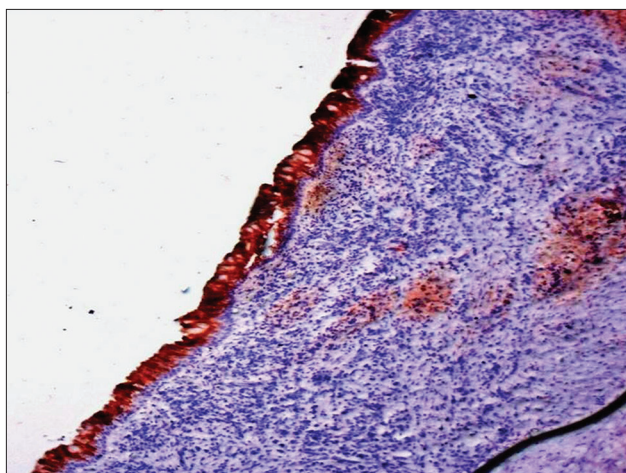


Figure 7: Photomicrograph shows strong positive immunoreactions to the Ki-67 protein (IHC stain, x100)

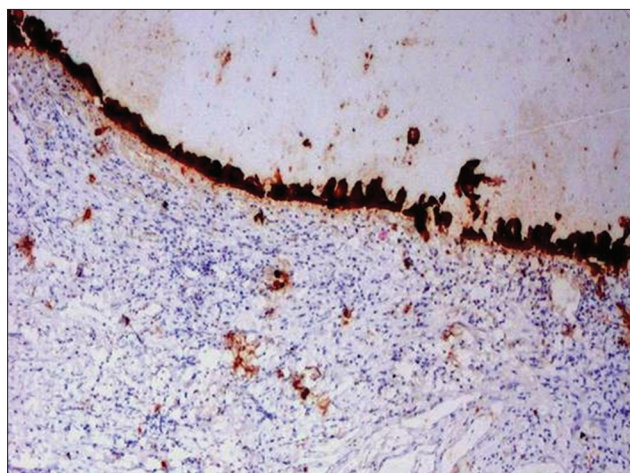


Figure 8: Photomicrograph shows strong positive immunoreactions to the CK-19 protein (IHC stain, x100)

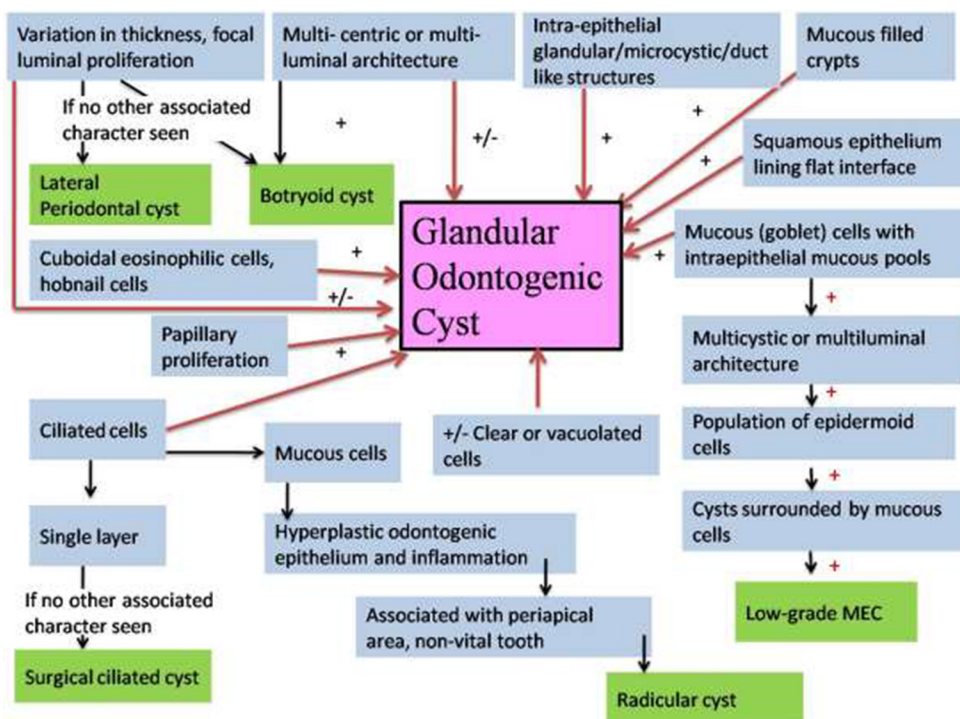


Figure 9: Histopathological differential diagnosis of glandular odontogenic cyst¹⁷

markers could be auxiliary aids in the differential diagnosis of these lesions. According to various authors, positive immunostaining with CK-18 and CK-19 in GOC may further help in differentiating GOC from central mucoepidermoid carcinoma (MEC). A recent study found significantly higher expression of both cytoplasmic and nuclear MASPIN in the mucous cells in low grade MEC (16.5% cytoplasmic, 1.7% nuclear) as compared with GOC (1.5% and 0.3%) or odontogenic cysts with mucous metaplasia (1% and 0.4%).^[17,18]

Several treatment modalities have been used which include curettage, enucleation with careful dissection of the margins and local block excision. The prognosis of this cyst still remains unclear. However, the aggressive nature of the lesion has been reported and the recurrence rate is directly related to the size of the lesion. 14.4% of the small lesions recur in contrast to 85.6% of the large lesions.^[16] Therefore, large lesions should be treated more aggressively and followed for a long period.^[17,19]

CONCLUSION

GOC is a rare cyst in maxillary region. It is important to consider histopathological features for its diagnosis since it bears resemblance to lesions like MEC. IHC provides an additional tool for its differential diagnosis.

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How to cite this article: Purohit S, Shah V, Bhakhar V, Harsh A. Glandular odontogenic cyst in maxilla: A case report and literature review. *J Oral Maxillofac Pathol* 2014;18:320-3.

Source of Support: Nil. **Conflict of Interest:** None declared.