Glandular Odontogenic Cyst of the Maxilla: A Case Report

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INTRODUCTION

The glandular odontogenic cyst (GOC) is a developmental cyst of the jaws first described in 1988 by Gardner et al. 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.” High et al., (1996) proposed the term ‘polymorphous odontogenic cyst’ for this cyst because of its aggressive growth pattern. GOC is a rare lesion with an incidence of 0.2% of odontogenic cysts. It affects males twice as frequently as females and the mandible almost three times as frequently as maxilla. The age range is 14-75 years, and the mean age is 45.7 years. Clinically, this lesion is generally painless, slow growing and its size can vary from less than 1 cm in diameter to large dimensions. Small cysts may be asymptomatic, while large ones can cause bone expansion accompanied by pain and paresthesia. Radiographically GOC presents as well defined radiolucencies with uni or multilocular appearances. Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) are recommended for diagnosis, surgical planning and follow up. GOC is characterized histologically by “an epithelial lining with cuboidal or columnar cells both at the surface and lining or cyst-like spaces within the thickness of the epithelium” Many histological features of GOCs are similar to those of low-grade central mucoepidermoid carcinomas. Since GOC is a very rare lesion and its occurrence in the maxilla is unusual, here we present a unique case of a GOC in 45 year old female patient in the anterior maxilla and reviews the main criteria for diagnosis.

Key words: Odontogenic cyst, Maxilla, Goblet cells

CASE HISTORY

A 45 year old female patient reported with a chief complaint of pain in relation to the upper left front tooth region since 1 month. The patient was asymptomatic earlier and there was no associated swelling intra orally or extraorally. Tenderness on percussion was elicited on 21, 22 and 23. Medical and dental histories were unremarkable. Intraoral radiography revealed well defined round radiolucency in relation to 21, 22, 23 measuring about 1.5x1.5 cm (Figure 1). Enucleation of the lesion was performed, and the specimen was submitted for histopathological examination.

Macroscopically the submitted tissue was brownish in color, soft in consistency and the cut section revealed a well formed cystic cavity surrounded by a fibrous capsule. Microscopically epithelial lining with variety of patterns around cystic spaces were noticed. Cuboidal or columnar epithelium with 2-4 cells layer with frequent inclusions of PAS positive goblet mucous producing cells as single or in cluster. (Figures 2 and 3).

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Stratified squamous epithelium of 5-6 cell thickness, epithelium of 1-2 cell thickness with plaque formation and ciliated columnar epithelium were also seen. (Figure 4) In some areas epithelium showed cells with surface eosinophilic material (hobnail cells), clear vacuolated cells and intraepithelial microcyst formation. Supporting connective tissue wall was highly vascularized with loosely arranged collagen fibers consisting of focal collection of chronic inflammatory cells. Based on the histopathological features a diagnosis of Glandular Odontogenic Cyst was given. The patient was followed up for 6 months without any signs of recurrence.

**DISCUSSION**

GOCs are relatively uncommon cysts first reported in 1987. Ever since then, they have remained as interesting controversy for the researchers all over the world. The lesion was initially referred to as a “sialo-odontogenic cyst” and believed to have salivary gland origin, but due to lack of evidence the term “glandular odontogenic cyst” was later adopted by the World Health Organization in 1992. Frequency rate of GOC is 0.012% to 1.3% of all the jaw cysts and its prevalence is 0.17%. There is a slight male predilection and lesions occur mostly in middle aged patients. In our case the patient was a middle aged female. GOCs displayed a predilection for the mandible, and for the anterior sextants of both jaws. Therefore, their minimal association with unerupted teeth can be readily understood, as the majority are distant from the third molars, which account for the great majority of unerupted teeth.

The main clinical finding is painless local edema. The clinical picture, however, is non-specific. Rarely the lesion may cause pain due to compression of a neurovascular bundle or secondary infection. Depending on the site of the lesion there may also be paresthesia, or a feeling of pressure on the dental arcade.

Radiographically, the GOC is a localized lesion and may appear as a multilocular or unilocular radiolucent lesion with well-defined borders. Sometimes it may present with peripheral osteosclerotic border and scalloping, root resorption and displacement of the teeth. Expansion and

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**Figure 1:** Orthopantomogram showing well-defined unilocular radiolucency in the anterior maxilla

**Figure 2:** Photomicrograph showing Periodic acid–Schiff (PAS) positive material within the mucous cells (PAS stain, ×400)

**Figure 3:** Photomicrograph showing mucous producing goblet cells in cluster (H and E, ×200)

**Figure 4:** Photomicrograph showing stratified squamous epithelium of varying thickness with highly vascular connective tissue (H and E, ×200)
thinning of cortical plates are sometimes observed on occlusal radiographs.1,10

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.11

The microscopic features of GOC are variable. Numerous histopathological features for the GOC have been described, but the exact microscopic criteria necessary for the diagnosis have not been accepted universally. Kaplan et al added that a diagnosis of GOC had to be based on the mandatory presence of the following five major features: Squamous epithelium, varying thickness, cuboidal eosinophilic (hobnail) cells, mucous (goblet) cells, and intraepithelial glandular or duct like structures.3 The diagnosis is made when the superficial layer of the epithelial lining consists of columnar or cuboidal cells sometimes referred to as ‘hobnail’, occasionally with cilia or filiform extensions of the cytoplasm. Furthermore, the epithelium has a glandular or pseudoglandular structure, with intra-epithelial crypts or microcysts or pools lined by cells similar to those on the surface. These microcysts may open onto the surface of the epithelium giving a papillary or corrugated appearance. Numerous goblet cells may be present, mainly in the superficial part of the epithelium. Occasionally, the epithelium is thinner, similar to reduced enamel epithelium. Epithelial thickenings or plaques may be present either in this thin epithelium or in the stratified squamous epithelium.11,12

The microscopic differential diagnosis of GOC includes lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC), and the central Mucoepidermoid Carcinoma (MEC). The plaque-like epithelial thickening in LPC made authors to believe that the GOC could be the clinical microscopic variant of LPC. But the presence of ciliated epithelium and duct like spaces with mucous cells differentiate GOC from LPC and BOC.3,13 It has been suggested that central MEC (CMEC), especially the low-grade variant is regarded as the most important histopathological differential diagnosis from GOC. It has been speculated that GOC may represent the most benign end of the spectrum of central MEC. However, CMECs do not show superficial cuboidal cells, epithelial whorls, ciliated cells and intraepithelial microcysts or duct-like structures.3,14 Certain authors have suggested using immunohistochemical markers to differentiate these lesions. The distinction between GOC and MEC were established based on immunohistochemical studies using cytokeratin (CK). CK7, 13, 14, and 19 positivity and negative reaction for epithelial membrane antigen (EMA) supported odontogenic origin. Some authors suggested expression of CK 8, 18, and 19 might be useful tool in differentiating these lesions.13

It has been demonstrated that the rate of recurrence increases with the radiographic complexity of the cyst.7 Because of the nonspecificity of the clinical and radiologic findings of GOC, small, unilocular lesions may be missed. If the lesion is completely enucleated, recurrence is unlikely in these cases. Because large unilocular lesions pose a lower risk of recurrence than multilocular ones and enucleation is recommended for higher risk large, multilocular lesions. Major treatment modalities are indicated (periodical resection, marginal resection, or partial jaw resection), depending on the size of the lesion, integrity of the jaw borders, and proximity of the lesion to vital structures. Follow-up is recommended for all cases for at least 3 years, and up to 7 years in cases with features associated with increased risk.15

CONCLUSION

Purpose of this case presentation is to increase existing knowledge of this rare entity as histopathological diagnosis of GOC is challenging for a pathologist. Our case was unique because of its relative minimally aggressive nature, presenting as a small unilocular radiolucency and its appearance in the maxilla which is an unusual site for glandular odontogenic cyst.

REFERENCES


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