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

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Abstract

Glandular odontogenic cyst (GOC) is odontogenic in origin and is an uncommon jaw bone cyst, with a frequency rate ranging from 0.12% to 0.13% of all jaw cysts. It was first described in 1987 by Gardner *et al.*, GOC has a male predominance and occurs primarily in middle-aged individuals. Clinically, the most common site of occurrence is the anterior region of mandible and maxilla. The incidence of recurrence reported is due to incomplete removal of the lining following conservative treatment. This article presents a rare case of glandular odontogenic cyst in a 24-year old male patient in the posterior region of the maxilla.

Keywords: Glandular cyst, maxilla, odontogenic cyst

Introduction

Glandular odontogenic cyst (GOC) is a rare lesion that arises in the tooth bearing areas of the jaws. In 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”. 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 cystic 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].

GOC occurs with a frequency rate of 0.012-1.3% of all the jaw cysts and its prevalence rate is 0.17%. 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. Radiographic appearance is nonspecific, the lesion may appear as unilocular or multilocular radiolucency, usually with well-defined margins and scalloped border [6, 7].

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, with papillary projections of epithelium and absence of inflammation in the subepithelial connective tissue [11]. It has two clinically important attributes: A “high recurrence rate” and an “aggressive growth potential”. The relative rarity of the lesion prompted us to add one more of our case and review the literature.

Case report

A 24-year-old male patient reported with a swelling in the upper left back region of the jaw. The swelling was present since 6 months and increased gradually and attained the present size and was associated with pain on pressure since 2 months. Intra orally a diffuse, non-fluctuant and firm swelling was seen extending from palatal aspect of 25, 26, 27, with associated teeth

being tender on percussion. Aspiration of the lesion was carried out to rule out vascular and cystic lesions. The straw-colored aspirate was obtained. A provisional diagnosis of the odontogenic keratocyst was given. A clinical differential diagnosis of a calcifying epithelial odontogenic cyst, ameloblastoma was made.



Fig 1: Diffuse swelling in the palatal aspect with respect to 25, 26, 27

Radiographic examination



Fig 2: Orthopantomogram shows well- defined unilocular radiolucency extending from 25, 26 and 27 with root resorption and displaced roots with 26, 27 and 28

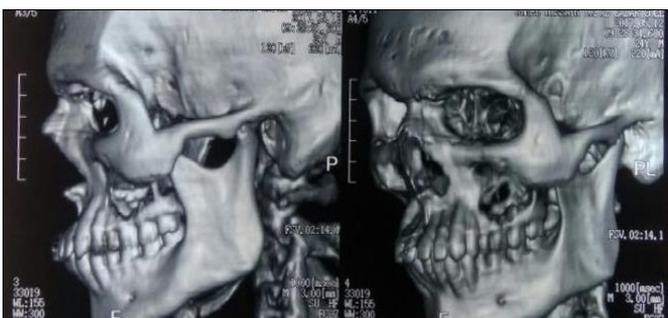


Fig 3: 3D CT Scan revealed unilocular radiolucency in the upper left back region of jaw. Measuring 2x3cm superio inferiorly and extending superiorly from the zygomatic arch and inferiorly at the periapex of 26, 27, 28. Antero posteriorly it extends from inferior nasal concha to the mesial aspect of 28.

Histopathology

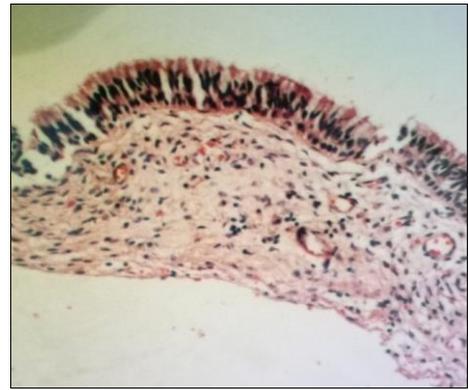


Fig 4: The epithelial layer showed eosinophilic cuboidal and ciliated columnar cells (H and E, magnification 40X)

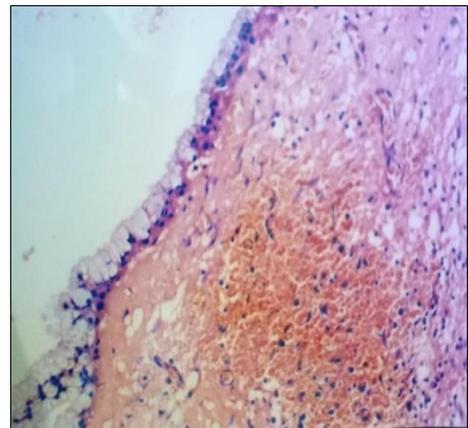


Fig 5: epithelial lining exhibiting superficial cuboidal cells with numerous goblet cells and underlying connective tissue tissue showing chronic inflammatory cells predominantly lymphocytes (H and E, magnification 10X)

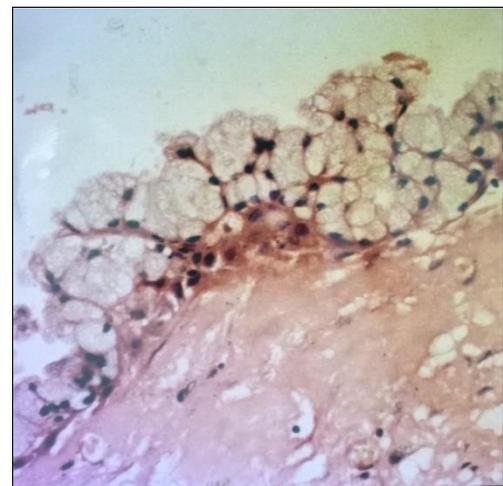


Fig 6: Shows squamous epithelial lining with numerous goblet cells (H and E magnification 40X)

Histopathological examination

Biopsy examination revealed pseudostratified ciliated columnar epithelial lining overlying the connective tissue capsule. The epithelial lining consists of columnar to cuboidal cells referred to as hobnail with cilia or filiform extension of cytoplasm. Epithelium has a glandular structure with microcyst. Microcyst contains a PAS positive material.

Numerous goblet cells are seen in the superficial part of epithelium. The epithelium shows plaque-like thickenings and whirling pattern of squamous epithelial cells. Other areas of epithelium showed pseudo-glandular pattern with few ciliated and mucus cells. Histopathology was suggestive of glandular odontogenic cyst.

Table 1: Study of gender and age, radiographic characteristics treatment

Sl. No	Study Title	No of cases	Gender & age	Location	Radiographic characteristics	Cortical plate integrity Expansion-E Perforation-P	Treatment	Reccurrence	Follow Up
1	Purohit <i>et al.</i> ^[11]	1	30/F	Ant Maxilla	Unilocular	NR	Enucleation + curettage	NR	NR
2	C.Fowler <i>et al.</i> ^[12]	46	Mean age 51 years. M:F =1:1	36-mand Ant mand-19 Max-9 7 Ant maxilla	Unilocular-27 Multilocular-14 5 cases-nr	E+20 cases P+3 cases	Enucleation/curettage	19.6%	8.75yrs
3	Guruprasad <i>et al.</i> ^[15]	1	17/F	Maxilla	Unilocular	E+	Enucleation+curettage	NR	1year
4	Shahnaz S.Tambawala <i>et al.</i> ^[16]	1	17/F	Post Mandible	Multilocular	E+	curettage	NR	3months
5	Faisal M <i>et al.</i> ^[18]	1	11/M	Ant+Post mandible	Multilocular	E+ P+	Enucleation with curettage	NR	1 year
6	Araujo de Morais <i>et al.</i> ^[9]	1	56/M	Post mandible	Unilocular	E+	Enucleation+curettage	NR	8 months
7	John J Frazier ^[10]	1	13/M	Maxilla	Unilocular	NR	Enucleation	NR	3 months
8	Robert D Foss ^[11]	1	46/F	Ant Mandible	Multilocular	E+ P+	Segmental Mandibular resection	Reported	NR
9	Vanishree M <i>et al.</i> ^[12]	1	17/F	Ant Maxilla	Unilocular	NR	Enucleation with curettage	NR	1 year
10	Boffano <i>et al.</i> ^[13]	2	68/M 82/F	Post mandible Post mandible	Multilocular Unilocular	E+ E+	Curettage Enbloc resection	NR NR	2 yrs 6 yrs
11	Anuthama Krishnamurthy <i>et al.</i> ^[14]	2	42/F 21/M	Ant Mandible A+P mandible	Multilocular multilocular	E+ E+	Enbloc resection Enbloc resection	NR NR	2 years NR
12	Kaplan <i>et al.</i> ^[15]	111	14-75 Mean-45yrs	Mand-70% Max-30% Ant- 24% Post-25%	Unilocular-53% Multilocular-46%	E-88% P-61%	Curettage/enucleation -83% Resection - 17%	35.9% 0%	Mean 2.7yrs
13	Present case	1	24/M	Post maxilla	unilocular	E+	Enucleation with curettage	NR	3 months

NR- Not Reported

Discussion

The GOC is a rare lesion, with a frequency rate ranging from 0.12% to 0.13% of all jaw cysts and has a high recurrence potential.^(7,8) H.H. Araujo de Morais *et al.*, reported the frequency rate of 0.012- 1.3% of all jaw cyst ^[9,10].

Glandular odontogenic cyst (GOC) is an uncommon jaw bone cyst of odontogenic origin with approximately 114 cases reported in the literature by 2010. Mascitti *et al.*, (2013) reported 111 cases of GOC with majority of cases in 4th and 5th decades of life which was coincident with that of MacDonald-Jankowski (2010). However their study reported the age at presentation of GOCs in the East Asian and sub-Saharan African population was nearly a decade younger. According to Fowler *et al.* the mean age at diagnosis was 51 years with a range of 20–82 years (N = 45). According to Kaplan *et al.*, GOC occurs within a wide age range of 14-75 years, but has never been reported in children less than 10 years of age. Most GOCs in literature are reported in patients over 30 years of age, with a mean age of 45.7yrs ^[11].

This paper reports a case of GOC in a 24-year-old male and highlights the main aspects with respect to diagnosis through a literature review. There is no significant gender predilection. This cyst is most often located in the mandible (70%), whereas only 30% of cases are located in the maxilla (Boffano *et al.*, 2010). 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. However, while the literature reports that most GOCs have slow growth (Oliveira *et al.*, 2009), the patient in the present case reported fast growth ^[13]. The most common site of occurrence is the anterior region of mandible followed by anterior region of maxilla and posterior region of mandible ^[12].

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 sclerotic borders or ill-defined borders, often reaching large dimensions. Sometimes, the occlusal radiographs shows expansion and thinning of cortical (Oliveira Neto *et al.*, 2010) ^[12].

The origin of GOC is very distinctive. However, Mark and Stern *et al.*, have proposed one of three possibilities:

1. A true cyst of glandular origin can arise either from entrapped salivary gland primordia or undifferentiated primitive epithelial rests that develop into the glandular epithelium.
2. An odontogenic primordial origin cyst in which the epithelial lining undergoes prosoplasia (metaplasia from a smaller specific differentiation to an added specific differentiation) into the glandular epithelium.
3. Low-grade mucoepidermoid carcinoma that forms an initial single cystic space as an alternative to the usual multicystic spaces.¹³

In the present case the patient is a middle-aged male. The most common site of occurrence is the anterior region of mandible followed by anterior region of maxilla and posterior region of mandible. Occurrence of GOC in posterior region of maxilla is rare. About four cases have been reported in literature till date and ours is probably the fifth case.

A diagnosis based only on clinical and radiological aspects, is very difficult to evaluate because of similarities with various other intrabony pathologies, hence a histopathological evaluation becomes mandatory ^[12]. On clinical and radiological examination, a differential diagnosis of other lesions such as dentigerous cyst, odontogenic keratocyst and ameloblastoma can be made.

The histopathological characteristics of GOC have been divided into major and minor categories by Kaplan *et al.*, 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 ^[15]. Histopathological features of the present case are correlated with the major and minor criteria given by Kalpan *et al.*, which includes squamous epithelial lining, flat interface, variation in thickness of the lining presence of epithelial spheres or whorls, no palisading, cuboidal eosinophilic cells or "hob-nail" cells, mucous goblet cells with intraepithelial mucous pools with or without crypts lined by mucous producing cells and interepithelial glandular microcysts or duct-like structures were the major criteria and papillary projections, ciliated cells, and clear or vacuolated cells in basal or spinous layer were the minor criteria ^[8].

Histopathologically, GOC must be differentiated from lateral periodontal cyst(LPC), botryoid odontogenic cyst (BOC), central mucoepidermoid carcinoma (CMEC), and a radicular or dentigerous cyst with mucous metaplasia as they display substantial overlap of histological features ^[14]. LPC is a developmental and odontogenic cyst in origin and the lining shows thin non-keratinized squamous epithelium and also exhibits focal epithelial thickenings and glycogen-rich epithelial cells, comparable to those seen in GOC's ^[14]. BOC is a polycystic variant of LPC and is locally destructive, shows alike histopathological features with those of GOC, such as areas of glycogen-rich clear cells and epithelial plaques. Were as, the identification of duct-like spaces with mucous cells and ciliated epithelium specifically differentiated from LPC and BOC favors the diagnosis of GOC's ^[14]. The differentiation of low-grade central mucoepidermoid carcinoma from GOC particularly its multicystic variant is further important and complicated. Significant histopathological overlap exists among GOC and CMEC. However, superficial cuboidal cells, ciliated cells, epithelial whorls, and duct-like structures or intraepithelial microcyst are not characteristic for CMEC, and their occurrence or non-existence can help in establishing an ultimate diagnosis. In-addition, immuno-staining with cytokeratin-18 and 19 and their positivity in GOC might assist in differentiating GOC from CMEC. Radicular or dentigerous cysts with mucous metaplasia have also to be distinguished with that of GOC. The incidence of clear cells, microcysts, and epithelial spheres appears to be of greater help in the distinctive diagnosis of GOCs associated with an unerupted tooth from dentigerous cysts by means of metaplastic changes. Mucous metaplasia of the epithelial lining of radicular cyst can be differentiated by the findings that radicular cyst is associated with the periapical area, non-vital tooth ^[8].

Two major studies of Kaplan *et al.*, and Fowler *et al.*, have reported a high recurrence rate following conservative surgical management (enucleation, curettage, etc.). According to Kaplan *et al.*, the aggressive biologic behaviour and propensity for recurrence might be associated with cell kinetics in the lining epithelium, i.e. infoldings, microcysts and plaques, which are suggestive of active cell proliferation. These areas of epithelial thickening may be comparable to the proliferative changes seen in dental lamina underlying the odontogenic nature of the GOC. Fowler *et al.*, in their study of 46 cases, reported an average time period from initial treatment to first recurrence of 8 years. Since 2008, the no of cases reporting recurrence have been less with reported recurrence only in two studies of Jefferson *et al.*, and Fowler *et al.*, in cases prior to 2008, Kaplan had reported a recurrence rate of 35.9% ^[15].

Immunostaining with CK-18 and 19 and their positivity in GOC may help in differentiating GOC from CMEC. Certain studies indicate that the use of IHC for p-53 and Ki-67 can help the clinician in differentiating GOC from CMEC. GOC exhibited decreased p-53 positivity and increased Ki-67 index when compared to CMEC ^[15].

Several studies indicate that GOC is relatively an aggressive lesion with a high tendency for erosion or perforation of cortical plate and chance of recurrence. Hence treatment varies from curettage, enucleation, enbloc and partial osteotomy. In addition, marsupialization and curettage in conjunction with Carnoy's solution and cryotherapy have been used. However, some authors prefer marginal or segmental resection because of the high potential for recurrence and the aggressive nature of these lesions ^[14].

Conclusion

A case of GOC in a 24-year-old male patient is being reported here with highlighting the review of literature along with special emphasis on histopathology. 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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