The Glandular Odontogenic Cyst of the Anterior Maxilla: Report of Two Cases

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ABSTRACT
Glandular Odontogenic Cyst (GOC) is a rare developmental odontogenic cyst with unique histopathologic features. It is included in the World Health Organization (WHO) histologic typing of odontogenic tumors under the terms glandular odontogenic cyst or sialo-odontogenic cyst. The aim of this article is to report two cases of GOC that occurred in the anterior maxilla in young male patients. The origin, clinical, radiographic and histologic features and treatment of the GOC are also discussed.

Key Words: Glandular odontogenic cyst, Sialo-odontogenic cyst, Anterior maxilla

INTRODUCTION
Padayechee and Van Wyk in 1987 first described a unique cyst of the jaws under the term “sialo-odontogenic cyst,” attributing its possible etiology to salivary glands and emphasizing the presence of mucous cells and cilia as part of the epithelial lining.1 In 1988, Gardner et al. reported the first series of these cysts and coined the term glandular odontogenic cysts (GOC), to emphasize the odontogenic derivation of these cysts.2 GOC has gained special importance by clinicians and pathologists primarily due to 2 reasons. The first is the overlap in the histomorphologic features between GOC, lateral periodontal cyst, Botryoid odontogenic cyst and central mucoepidermoid cyst of the jaws.1 The second reason is the potential aggressive behavior of the lesion.3,4 The GOC most frequently in middle aged people5,6 with slight male predilection.5 Radiographically, it appears as a well defined radiolucency, which may be unilocular or multilocular.5,6 GOC has a predilection for the mandible, affecting both anterior and posterior areas and predilection toward anterior occurrence when the maxilla is involved.5 To improve the clinicopathologic information to elucidate the natural history of this enigmatic cyst, reporting of new cases should be encouraged. We report two rare cases of GOC occurring in the anterior maxilla region.

CASE REPORT
Case 1
A 23-year old male visited Oral and Maxillofacial Surgery clinic with history of a swelling of the anterior maxillary region since 3-5 months. The patient reported an external injury to the anterior teeth 8 years back. The medical history of the patient was non contributory. Intraoral examination revealed a swelling in the anterior maxilla extending from right maxillary lateral incisor to left maxillary canine tooth in the palatal aspect. The right and left maxillary central incisors and lateral incisors, as well as the left canine were sensitive to percussion. Non-vitality of these teeth were confirmed by performing pulp sensibility test. Radiographic examination revealed a well defined unilocular radiolucency present in the root apex of right central and lateral incisor and left central, lateral incisor and canine tooth (Figure 1). Root
canal treatment was carried out as part of the treatment.

Figure 1. Intraoral periapical radiograph showing a unilocular, well defined lesion in the anterior maxilla

Aspiration of the lesion yielded yellowish brown colored fluid without cholesterol crystals or distinctive cellular elements, indicating it to be a benign cystic lesion. The history of trauma and non-vitality of teeth strongly suggested radicular cyst. Therefore, considering the clinical diagnosis of radicular cyst, the surgeon performed an excisional biopsy.

Under general anesthesia, the lesion was enucleated with peripheral ostectomy and chemical cauterisation using Carnoy’s solution (Composition: mixture of absolute alcohol (6ml), chloroform (3ml), glacial acetic acid (1ml), ferric chloride (1mg).7 Apicoectomy of (which tooth) was also performed. Intraoperatively, the cystic lining was thin and the bone margin was thinned out which indicated an aggressive cystic lesion. Therefore, peripheral ostectomy and chemical cauterisation using Carnoy’s solution was performed as for any other aggressive cystic lesion.

The surgical specimen consisted of multiple fragments of pale brown soft tissue and the largest bit was measuring around 3x1x0.3 cm in dimension. Routine sections were prepared and stained with hematoxylin-eosin stain, periodic acid-Schiff (PAS) and mucicarmine stain. Histological examination showed a cystic cavity lined by epithelium of varying thickness with a flat interface between the epithelium and underlying connective tissue (Figure 2).

The cystic capsule exhibited minimal inflammation. Eosinophilic cuboidal cells forming the surface layer of the cyst lining, surface ciliated cells and cells with a clear or vacuolated cytoplasm were readily identified. Focal areas of epithelial thickenings or plaques were noticed. Mucous cells were well identified within the glandular areas (Figure 3) and were PAS positive.

A diagnosis of glandular odontogenic cyst was made.- The healing process was uneventful during the follow up period. There is no evidence of recurrence 24 months postoperatively.

Case 2
A 21-year old man visited Oral and Maxillofacial Clinic, for the evaluation & orthodontic correction of malaligned teeth. He also complained of an asymptomatic, mild swelling in the anterior maxilla with mobility and palatal tipping of maxillary central incisors. The patient noticed the mobility of
central incisors and mild swelling of anterior maxilla since 6–8 months which gradually increased to the present size. Intra oral examination revealed a swelling in the anterior palate extending from right maxillary canine, lateral and central incisor to left maxillary central incisor. The maxillary central incisors exhibited extrusion and palatal tipping with grade II mobility. The overlying mucosa was of normal color. There was no relevant medical history.

Radiographically, a unilocular radiolucency was observed in the periapical region of maxillary anterior teeth extending from right maxillary canine, lateral and central incisor to left maxillary central incisor. Displacement of the root was detected in right and left maxillary central incisors (Figure 4).

The maxillary central and lateral incisors were sensitive to percussion and exhibited heat and cold response. Aspiration of the lesion yielded yellowish straw colored fluid without cholesterol crystals or distinctive cellular elements. Subsequently, under general anaesthesia the lesion was enucleated with peripheral ostectomy and chemical cauterisation using Carnoy’s solution and extraction of the involved teeth. The surgical specimen consisting of multiple fragments of brown soft tissue measuring overall 4.2x2.5x2cm was submitted for routine tissue processing and evaluated microscopically. Wax sections of 4µm thickness were prepared and stained with hematoxylin and eosin and Periodic acid-Schiff (PAS). Histologically, an epithelial lining of varying thickness surrounded by a capsule with minimal inflammation was observed. The epithelial lining exhibited plaque like thickening into the capsule. Eosinophilic cuboidal cells formed the surface layer of the cyst lining and also lined the intraepithelial crypts. Surface ciliated cells and mucous producing goblet cells were readily identified (Figure 5).

The mucous cells stained positively with PAS and mucicarmine stain. The histopathological picture of the present case was not fulfilling all the criteria of Kaplan et al. and also histopathology did not suggest any other odontogenic cyst. A diagnosis of glandular odontogenic cyst was made considering the presence of epithelial plaque, mucous cells and mucin filled cystic spaces. The healing process was uneventful. The patient is under regular follow up and there is no evidence of recurrence 26 months postoperatively.

DISCUSSION

The clinical features and the histopathology of the present case are consistent with those reported for GOC in the literature. GOC is a slight predilection for males, and the lesion occurs mostly in middle-aged patients with age range of 14–75 years. The mandible, especially anterior region is the most common site of occurrence for this lesion. Similarly, it has been well documented that in the maxilla, it also has the predilection for anterior region. In this report, both the cases occurred in the anterior region of maxilla in young male patients. Review of the literature has demonstrated that most cases presented as painless swelling, as
in these two cases. Radiographically, it presents as a well defined radiolucency, either unilocular or multilocular.\(^5\) In our cases, both the cysts showed well defined unilocular radiolucency in the apical region of maxillary anterior teeth.

Microscopically, GOC has characteristic features listed by Gardner.\(^2\) Kaplan et al. proposed a list of microscopic criteria for GOC such as non-keratinized stratified squamous epithelium, epithelial whorls or spheres within the lining, eosinophilic cuboidal or columnar cells which are occasionally ciliated and presence of mucous cells with microcystic areas.\(^3\) The morphology of epithelial lining in GOC strongly suggests an odontogenic epithelial origin, especially from remnants of dental lamina.\(^2,6\) The thin, cuboidal epithelium in some areas which was also appreciated in our cases resembles the reduced enamel epithelium. Also, mural or luminal plaques are observed in other cysts of odontogenic origin such as lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC) and gingival cyst of adults.\(^4\) Immunohistochemical studies also provide strong support for the odontogenic origin of GOC.\(^9,10\)

Odontogenic cysts such as radicular and dentigerous cyst may resemble GOC microscopically when they exhibit mucous metaplasia in the epithelial lining.\(^5\) In the present report, the second case did not exhibit all the criteria suggested by Kaplan et al.\(^5\) But, GOC should be considered in the differential diagnosis whenever there is presence of mucous cells, epithelial plaque and mucin filled duct like spaces.

Some of the histopathologic features exhibited by GOC are similar to LPC such as focal plaque like epithelial thickenings and glycogen-rich epithelial cells. However, the LPC is a less aggressive lesion with limited growth potential and low postsurgical recurrence.\(^11\) The GOC shares some similar clinical features with BOC such as age, anatomic distribution and multilocularity.\(^6\) Also, histologically it exhibits overlap with those of BOC, like thin epithelial lining and epithelial thickenings.\(^12\) The presence of duct-like spaces with mucous cells and ciliated epithelium specifically differentiate GOC from LPC and BOC.\(^13\)

The most significant histopathologic differential diagnosis for GOC is central mucoepidermoid carcinoma (MEC), especially the low-grade variant\(^14\) and it is very important to distinguish between the two. Radiographically, it can present as either a unilocular or multilocular radiolucency with well defined borders.\(^8\) Significant histological overlap is observed between GOC and central MEC. It has been speculated that GOC may represent the most benign end of the spectrum of central MEC, necessitating close and prolonged follow-up of all lesions.\(^4\) One of the distinguishing features in GOC is the typical thin epithelial lining without any solid epithelial proliferation as seen in MEC.\(^15\) In addition, MEC do not show the swirling spherical aggregates (epithelial plaque) that are often seen in GOC.\(^16\) Difference in cytokeratin expression of GOC and central mucoepidermoid carcinoma may play some role in differentiating between the two lesions. The negative reactivity with cytokeratin 8 and 18 in GOC may be significant in its differentiation from central mucoepidermoid carcinoma.\(^10,17\) Also, difference in percentage of cells that stain positively with anti-CK 18 and 19 in GOC and central mucoepidermoid carcinoma has been demonstrated.\(^18\)

GOC is considered as potentially aggressive in nature and shows frequent perforation and thinning of cortical plates\(^5\) and high recurrence rate following conservative surgical treatment.\(^3,4\) Recurrence rate varying from 21%\(^11\) to 55%\(^3\) has been reported. Recurrence may be partly related to the thinness of the cyst wall and to the presence of microcysts making complete removal difficult.\(^4\) Long term follow up is advisable in these cases as recurrence has been reported even 7 years after the initial treatment.\(^1\)

The treatment of choice varies form a curettage to local block excision.\(^3,4,6,9\) Kaplan et al. in his review of 111 cases of GOC has reported that treatment by enucleation or curettage carries the highest risk for recurrence, especially in large and multilocular lesions. Peripheral ostectomy or marginal resection can decrease the risk.\(^5\)

Even though this entity often present diagnostic challenges for pathologists due to the histologic overlap with other established lesions, it definitely exhibits microscopic features and biologic behavior distinct enough to be regarded as a separate entity. Because of the scarcity of cases with long term follow up, the prognosis of this cyst remains unclear. Therefore, it is imperative that
patients be followed carefully. Analyses of additional cases of this rare odontogenic lesion may add clarity to our current understanding of the glandular odontogenic cyst.

REFERENCES


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