CASE REPORT

Glandular odontogenic cyst mimicking a radicular cyst: A case series

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Abstract

Glandular odontogenic cyst (GOC) is considered an odontogenic cyst of developmental origin, which because of its histological features had been termed sialo-odontogenic cyst. Although clinically and radiologically they bear similarity to relatively innocuous periapical lesions, their biological behavior is comparable to aggressive tumor-like conditions-like keratocystic odontogenic tumors. Their aggressive nature, ability to attain large dimensions, and relatively high recurrence potential entail that they should be viewed as different propositions from the treatment perspective. Due to the rarity of these lesions, there are no well-established surgical treatment protocols so far. Nevertheless, cases histologically confirmed as GOCs warrant critical review and extended follow-up. This paper presents two reports of glandular odontogenic cysts which involved the posterior maxilla and their treatment outcomes.

Keywords: Cyst, glandular odontogenic cyst, odontogenic cyst, periapical cyst, sialo-odontogenic cyst

Introduction

Glandular odontogenic cyst (GOC) is a rare odontogenic cyst first introduced by Gardner et al.[1] and later recognized by the WHO, in 1992, as a developmental cyst of odontogenic origin. This cystic lesion unique to the jaws intriguingly bears semblance to glandular structures and hence has also been described as sialo-odontogenic cyst. It occurs over a nonspecific age range with an average of 50 years and equal sex predilection.[2,3] Experience over the years has resolved the issue of the origin of these cysts which is now considered odontogenic rather than sialogenic. Collateral appearance of these cysts with odontogenic tumors when they are termed hybrid lesions further reinforces their odontogenic origin.

Although classified as a cyst, the clinical point of interest has been its biological behavior. They have shown a high propensity for recurrence after conservative management. Hence, it is important to diagnose these relatively rare lesions which closely mimic radicular cysts in the clinical presentation but need a more guarded approach in management. Review of literature from 1987 to 2003 reports 60 cases of GOC.[4] According to Kasaboglu et al., only 0.012% of the cysts seen in the oral cavity have fulfilled the stringent criteria for GOC microscopically.[5]

These lesions, therefore, pose a dual challenge, diagnostic, and therapeutic to the clinician. This paper presents 2 cases of GOC with their treatment outcomes.

Case Reports

Case 1

A 22-year-old male presented with history of swelling in the left infraorbital region which was gradually increasing in size and associated with dull intermittent pain for 3 months. There was no history of trauma. On examination, there was a well-circumscribed swelling in the left infraorbital region extending from left infraorbital rim to the corner of the mouth and from the lateral margin of the nose to the malar prominence. The skin over the swelling was normal, pinchable and showed no neurosensory deficits. Intraorally, there was a swelling in the left maxilla extending from the left lateral incisor to the distal of left second molar. There was considerable palatal expansion and minimal buccal expansion.

Swelling was bony hard with areas of egg shell cracking and perforation. 23, 24, 25, 26, and 27 were nonvital, and hence root canal treated. The pre-operative radiograph showed a well-circumscribed unicocular radiolucent lesion with corticated border extending from 22 to 27 [Figure 1]. 28 was missing. It was provisionally diagnosed as a radicular cyst. Aspiration was positive and yielded a serosanguinous fluid. Under general anesthesia, a buccal mucoperiosteal flap was raised. The expanded buccal cortical plate showed areas of perforation [Figure 2]. The cyst was deroofed and enucleated.
Histopathological examination revealed a nonkeratinized stratified squamous epithelium of varying thickness with flat epithelial, connective tissue interface. At focal areas, the lining showed cuboidal cells with eosinophilic cytoplasm along with mucous goblet cells. In certain areas, microcysts were seen opening onto the surface by crypts giving a papillary appearance. It was concluded as an infected GOC. Special staining with Alcian blue and periodic acid-Schiff stain showed the presence of mucous cells in the epithelial lining of the odontogenic cyst [Figure 3].

Follow-up for over 2 years has shown good clinical and radiological signs of healing with no evidence of recurrence.

Case 2

A 28-year-old male patient reported with a history of intermittent swelling involving right side of face over the last 2 years. The patient had undergone extraction of several infected teeth in this quadrant during this course attributing them to be the cause of the swelling. Over the last 3 months, however, the swelling persisted, was painful and gradually increasing in size. On examination, 11-16 were missing. There was a bony hard swelling with areas of fluctuance extending from the 11 to 16 region. Orthopantomogram showed an ovoid well-corticated circumscribed radiolucent lesion extending from 11 to 16 region [Figure 4].

Computed tomographic scans showed extensive buccal cortical perforation and sinus obliteration [Figure 5]. It was provisionally diagnosed as a residual cyst and enucleated.

Histopathological examination showed the complete surface layer of the lining showing cuboidal cells with eosinophilic cytoplasm along with numerous goblet cells suggestive of an infected GOC [Figure 6]. Follow-up radiographs after 2 years showed no evidence of recurrence.

Discussion

GOCs have been described as slow growing lesions with a predilection to involve the anterior mandible and a reported involvement of 85%. Both the cases reported in this series, however, involved the posterior maxillary dentoalveolar segment and extended into the maxillary sinus. The extensiveness of these lesions involving the maxilla can be attributed to the ease of expansion into the maxillary airspace before causing perceivable clinical expansion.

Radiographically, they may be uni or multilocular radiolucent lesions often crossing the midline and may be quite extensive. It is impossible to offer a radiological diagnosis as they closely mimic other unilocular lesions such as periapical granuloma,
Involvement of multiple teeth, poor response to vitality of these teeth, bicortical expansion, and presence of cortical perforation are clues towards a more vigorous lesion clinically though, and hence warrant meticulous histopathological study of features.

About 39.3% of cases show cortical perforation and 14.3% show thinning and erosion; both of which are much higher than in odontogenic keratocysts. A higher percentage of cortical compromise (71.3%) has been noted in recurrent lesions. Cortical integrity can hence be an important criteria in treatment planning.[6,7] Both the cases reported above were large and involving a span of more than two teeth, unilocular but showed extensive buccal cortical thinning and perforation.

Histopathology remains the mainstay in the diagnosis of this intrabossaeous cyst.[8] The histopathologic features of GOC may be a blend of cystic and tumor-like features just like its clinical behavior. Cystic features include a cavity lined by a nonkeratinized, stratified squamous epithelium varying in thickness with subepithelial fibrous connective tissue layer and microcysts in the adjacent bone marrow spaces. On the other hand, localized plaque like thickenings of the epithelium, the presence of variable numbers of mucus-secreting cells in the surface layer of the epithelium, sometimes forming crypt-like invaginations or gland-like areas within the layer of epithelium lead to appearances that mirror odontogenic and salivary tumors.[9] It is obvious that some of its microscopic features overlap with well-differentiated mucoepidermoid carcinomas of the jaws, gingival cysts of adults, and lateral periodontal cysts (particularly the botryoid form).

The disease is proliferative and aggressive and has a high rate of recurrence. The high rate of recurrence can be attributed to the thin lining, presence of microcysts, and tubular extensions of the cystic lining into the surrounding marrow which may escape clearance and high mitotic capacity of the cells very similar to OKC.[9,10] Although microcysts were seen in certain sections in case1, both cases showed an uneventful post-operative course with no evidence of recurrence. The involvement of almost all the teeth in the quadrant, extensive buccal cortical perforation, and palatal expansion, and close to complete obliteration of the maxillary sinus, however, highlights their aggressive behavior.

Many authors follow the major and minor criteria suggested by Kaplan et al. as a guideline to diagnosis GOC, particularly to differentiate it from low-grade mucoepidermoid carcinomas. The major criteria include variably thick lining with or without spherical whorled epithelium, cuboidal eosinophilic cells called “Hob nail” cells, mucous “goblet” cells with intraepithelial mucous pools with or without crypts leading to glandular duct-like structures. The minor criteria include papillary projections, ciliated cells, and clear or vacuolated cells in the basal or spinous layer.[8] As indicated in the presented cases, special stains with Alcian blue and periodic acid-Schiff stain are mandatory to demonstrate the positive reaction of the mucous cells and mucous pools which are pathognomonic of these lesions.

GOC will have to be conceived as a cyst with tumor like behavior. Several management procedures that have been tried for OKC have been extrapolated to GOC. Studies have shown that enucleation and curettage can lead to high recurrence rates (25%) similar to that for OKC. To get optimum results, it is imperative to speculate the possibility of GOC in large unilocular or multilocular periapical cyst like lesions, ensure complete removal of the thin cystic lining with special attention to the interadicular cysts when teeth are retained, sacrifice of

Figure 5: Case 2 - Pre-operative coronal section of computed tomographic scan showing unilocular cystic lesion obliterating right maxillary sinus

Figure 6: Case 2 - H and E section showing complete surface layer of the lining having cuboidal cells with eosinophilic cytoplasm along with numerous goblet cells suggestive of an infected glandular odontogenic cyst
at least 2 mm of encompassing bone to eradicate microcysts and tubular extensions, and minimum follow-up of 2-5 years. As both the cases, in this series were unilocular large lesions involving the maxilla with the oroantral partition still intact, they were enucleated with bony clearance. Follow-up of over 2 years has shown no evidence of recurrence and good bone fill.

**Conclusion**

GOC will have to be considered in the differential diagnosis of seemingly benign periapical lesions. The greater incidence reporting in the last decade clearly can be attributed to the higher histopathological recognition of these lesions enabled by use of special stains to demonstrate the characteristic cellular features. These lesions will have to figure in the differential diagnosis of clinicians treating large uni and multilocular periapical lesions, and viewed from a different treatment perspective for better long-term outcomes.

**References**