Case Report

Maxillary Glandular Odontogenic Cyst: An Uncommon Entity at an Unusual Site

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Abstract

The glandular odontogenic cyst (GOC) is a rare developmental cyst that was described in 1988 by Gardner, et al. This lesion demonstrates non-specific clinical course and radiological findings. Hence, often confused with other lesions, but specific histopathlogical features help in establishing its correct diagnosis. This article presents a rare case of a maxillary glandular odontogenic cyst in a 70-year-old female patient.

Keywords: Cyst, developmental, maxilla, mucous cell, odontogenic cyst, sialo-odontogenic cyst

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Introduction

Glandular Odontogenic Cyst (GOC) is an extremely rare lesion occurring in the jawbones.[1] It was first discussed at the meeting of the International Association of Oral Pathologists in 1984, but was first documented as 'sialo-odontogenic cyst' by Padayachee and Van Wyk in 1987.[2] Gardner, et al. in 1988, histologically found that the epithelial lining of this cyst was odontogenic in nature and gave the term "glandular odontogenic cyst" which was later adopted by WHO.[3,4]

It most commonly affects anterior mandible of middle aged males, but other regions of the jaw can also be affected.[5] Its clinical and radiographic findings are non-specific. It usually presents as a slow growing asymptomatic swelling, with a mean age of 46.7 years and 50 years, respectively.[6] Radiographically, it can be presented an unilocular or multilocular lesion, with a well-defined borders, which may or may not be scalloped.[3]

The cystic lining is made up of non-keratinized epithelium with papillary projections, nodular thickenings, mucous-filled clefts, and "mucous lakes." The cystic lining shows the presence of cuboidal epithelial cells, or "hobnail" cells within the epithelium. Inflammation within the subepithelial connective tissue is usually absent.[1,4] The case is presented due to its rarity, as well as its occurrence in an unusual location, which prompt us to add this rare case in the existing literature.

Case Report

A 70-year-old female reported to the department with a chief complaint of a swelling in the left maxillary anterior region of the jaw, over the past six months. The patient gave the history of gradual onset of the swelling which has slowly increased to the present size. The swelling was also associated with mild and intermittent pain, occasionally. She had also consulted a local dentist 2 months back, and the yellowish fluid was aspirated. Certain medications were also prescribed (patient unaware of it) and the swelling subsided then after. Since last 15 days, similar symptomatic swelling developed in the same region. The patient also gave a history of extraction by the same local dentist in the same region 9 months back, due to the pain and mobile teeth.

Extraoral examination revealed slight facial asymmetry with fullness of left middle third of the face. No other abnormality was detected extra-orally. Intra-oral examination revealed a single diffuse swelling in relation to 21 to 23 regions, measuring about 3 × 2 cm in its greatest diameter. The overlying mucosa was normal with areas of slight bluish tinge (Figure 1A). On palpation it was soft and tender at the center, but firm and non-tender at the periphery. Many missing teeth were observed with edentulous left maxillary arch.

The radiographic investigations advised were intraoral periapical radiograph, panoramic radiograph (Figure 1B) and maxillary occlusal (Figure 1C). The radiographs revealed a large oval multilocular radiolucent with an ill-defined borders of approximate 3 × 1.5 cm in greatest diameter in relation to 22 and 23. Few areas at the periphery of the lesion also showed ill-defined borders also involving the left maxillary sinus. Based on the history and findings, a differential diagnosis of lateral periodontal cyst, botroid odontogenic cyst was made and was provisionally diagnosed as residual cyst.

Aspiration revealed a brownish blood tinged coloured fluid, and was further surgically excised under local anesthesia. Histopathological examination revealed a non-keratinized stratified squamous cystic epithelium with areas of pseudostratified ciliated columnar epithelium of varying thickness (Figure 2A). Plaque like epithelial thickenings with mucous-filled clefts were also appreciated in a few areas. The connective tissue capsule was moderately collagenous with chronic inflammatory cells and cholesterol clefts (Figure 2B). Intraepithelial crypts formation with PAS positive mucous cells were also observed (Figure 2C). A diagnosis of a glandular odontogenic cyst (secondary infected) was made.

Discussion

After its first description in 1987, Gardner, et al. in 1988 coined

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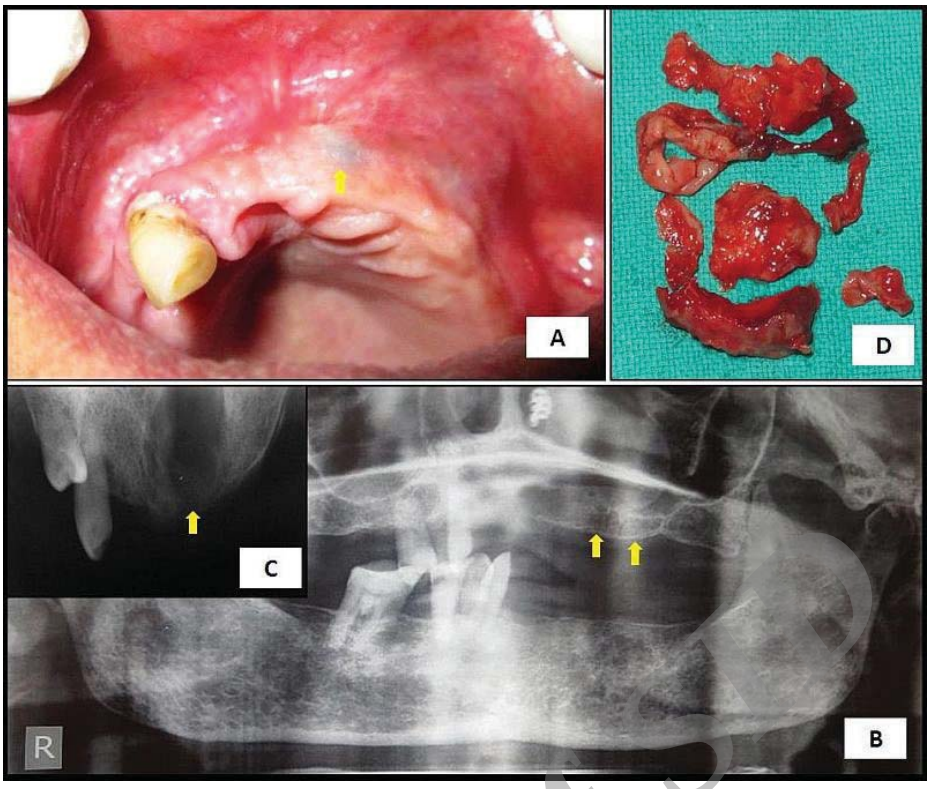

Figure 1. A) Intraoral clinical photograph shows swelling in relation to 21 to 23, with bluish tinge; **B)** Panoramic radiograph reveals a large multilocular radiolucency in the same region, involving left maxillary sinus; **C)** Maxillary occlusal radiograph showing radiolucency of approximate 3 × 1.5 cm in its greatest diameter with ill-defined borders; **D)** Gross specimen showing multiple bits soft tissue.

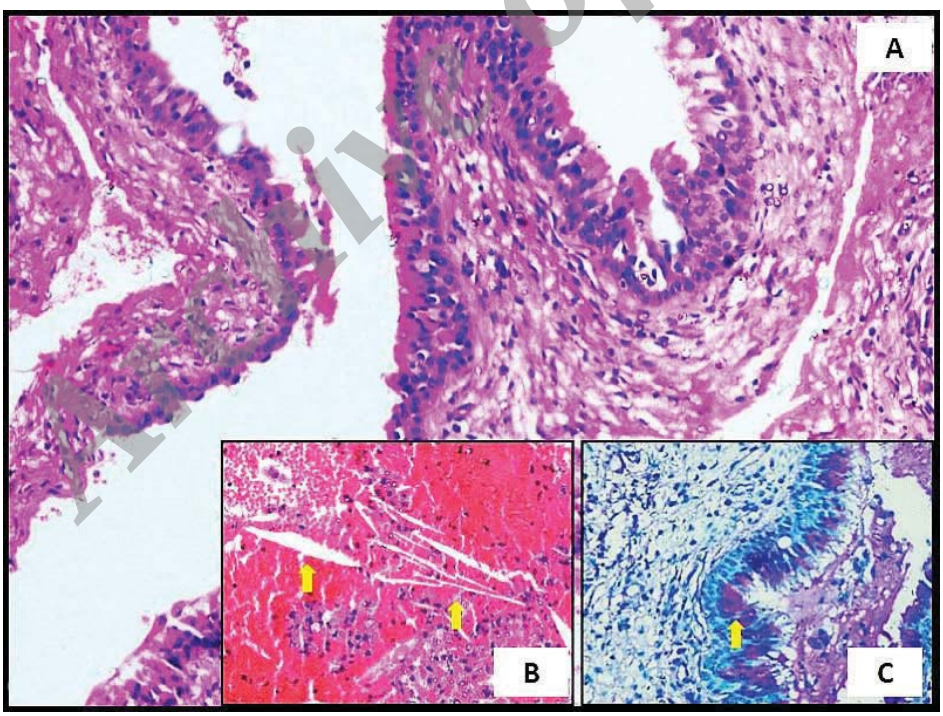

Figure 2. A) Photomicrograph showing cystic epithelium lining of variable thickness with mucous-filled clefts and inflamed capsule (H&E stain, ×200); **B)** Connective tissue capsule showing cholesterol clefts (H&E stain, ×200); **C)** PAS positive mucous cells (PAS stain, ×200)

the term "glandular odontogenic cyst" after reporting 8 cases of this lesion which affected either jaws in patients who had a wide age distribution.[3] In 1992, the World Health Organization accepted GOC as a distinct pathological entity and classified it as a developmental odontogenic cyst.[3] In 1996, a new terminology was proposed by High, et al. as "Polymorphous odontogenic cyst" based on its varied histologic appearance.[8,9]

Glandular odontogenic cyst (GOC) is a rare developmental lesion which has morphological similarities to other odontogenic cysts, which make its diagnosis challenging for oral diagnosticians and pathologists.[6] It has an occurrence rate of 0.012% to 1.3% and a prevalence rate of 0.17%.[5] It usually affects middle

Table 1. Major and minor histopathological diagnostic criteria for GOC

Minor criteria
a. Squamous epithelial lining with a flat interface with the connective tissue wall but no basal palisading; b. Variable thickness of cystic epithelial lining with or without epithelial "spheres," "whorls," or focal luminal proliferation; c. Presence of cuboidal eosinophilic cells or "hobnail" cells; d. Presence of mucous (goblet) cells with intraepithelial mucous pools, with or without crypts lined by mucous-producing cells within the epithelium. e. Presence of Intraepithelial glandular, microcystic, or duct- like structures.
Minor criteria
a. The lining epithelium shows Papillary proliferations b. Presence of ciliated cells c. Presence of multicystic or multiluminal appearance. d. Presence of clear or vacuolated cells in the basal or spinous layers of epithelium.

aged individuals, with the peak frequency in the sixth decade of life. Yet, there are cases which have also been reported in younger age group.[5–8] Male to female ratio is 1.3:1 to 2:1, and the most common site of involvement is anterior mandibular region.[3–5] The patient usually presents with an asymptomatic swelling having a slow growth rate.[5–8] Sometimes, the lesion may be symptomatic due to compression of a neurovascular bundle or secondary infection (however, is uncommon).[5,6] In the present case, GOC was reported in a 70-year-old female who presented with a symptomatic swelling in the anterior maxillary region. This symptomatic nature in the present case can be due to aspiration done by a local dentist, which gradually got secondarily infected over a period of time. This finding was also confirmed during the histopathological examination, where chronic inflammatory cells with cholesterol clefts were appreciated. Clinically, it is usually soft to fluctuant but may also be bony hard in consistency.[4,7] The overlying mucosa is generally normal in appearance except in a few cases, where a bluish or purple tinge was also observed.[5] In a review done in 111 cases by Krishnamurthy, et al. in 2009, aspiration yields a clear watery fluid indicative of GOC but, a brown coloured viscous fluid can also be aspirated,[5–8] also observed in the present case.

Similar to the clinical findings, the radiographic findings of GOC are also non-specific.[2,3] It is usually round to oval shaped well defined intraosseous radiolucent lesion, which may appear as a multilocular or unilocular.[5] Sometimes, also shows irregular to scalloped borders, and may also cause root resorption, displacement of tooth and cortical expansion and perforation.[4,8] It is located below the apex of the tooth and may reach the interproximal are and shows no relationship with the periodontium.[5] Radiographically, it bears resemblance with lateral periodontal cyst, botryoid cysts, keratocysts, residual cysts, central mucoepidermoid carcinoma and ameloblastoma.[9] In the present case, the radiographs revealed an oval shaped multilocular radiolucency with irregular borders and peripheral sclerosis (few areas) involving maxillary sinus.

The cystic epithelium is non-keratinized stratified squamous type and shows the presence of glandular or pseudoglandular structures, formation of intraepithelial crypt or microcyst.[8] The cells of the epithelial lining are cuboidal or columnar, which are occasionally ciliated. The epithelium may also show the presence of epithelial thickening or plaques with Goblet cells.[5] The sub-epithelial connective tissue is usually free of inflammation. Histopathological diagnosis of GOC can be established by certain major and minor criteria, proposed by Kalpan, et al. (2005) and Brannon, et al. (2011). Atleast focal presence of major criteria is mandatory, and the presence of minor criteria can only aid in establishing the correct diagnosis (Table 1).[8] Histological differential diagnosis includes lateral periodontal cyst, botryoid odontogenic cyst, dentigerous cyst or radicular cyst with mucus prosoplasia and most importantly mucoepidermoid carcinoma.[5–8] The present case had all the characteristic features of GOC, but shows the presence of chronic inflammatory cell infiltrate and cholesterol clefts, thus was diagnosed as GOC (secondary infected). Immunohistochemical analysis can also assist in its diagnosis by differentiating it with other lesions of glandular origin.[9]

Its recurrence rate range from 25% to 55%, which is indicative of its aggressive behaviour.[7] This may be attributed to its thin cystic lining and the presence of microcysts, which make the complete surgical removal difficult.[5] The choice of treatment for GOC depends on the status of the patient, site of involvement and clinician's view. Conservative approaches like enucleation and curettage with or without applying Carnoy's solution is associated with high recurrence rate. However, the recurrence rate of GOC 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. Therefore, large lesions should be treated more aggressively (enucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw resection for multilocular cases) and should be followed for a longer period.[9,10]

In summary, the maxillary glandular odontogenic cyst is a rare odontogenic cyst having an aggressive clinical course, behavior and high recurrence rate. It is often misdiagnosed due to its non-specific clinical, radiological and sometimes histopathological features. It should also be include in the differential diagnosis of unilocular/multilocular radiolucency of the maxillary anterior region. Hence, careful evaluation is essential before confirming its diagnosis, which also requires appropriate treatment planning and its management.

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