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Glandular odontogenic cyst: A case report

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Abstract

Glandular odontogenic cyst (GOC) is a rare and uncommon jaw bone cyst of odontogenic origin. It was described as distinct entity in 1987 by Gardener *et al.* It is a cyst having an unpredictable, potentially aggressive characteristic, and has the tendency to grow in large size with relatively high recurrence rate. There are many similar diagnostic symptoms to glandular odontogenic cyst as it can be clinically and histopathologically confused with lateral periodontal cyst, botryoid odontogenic cyst, radicular and residual cysts with mucous metaplasia, and low-grade mucoepidermoid carcinoma and it is difficult to differentiate between them. The present case reports a 35-year-Female patient came to the hospital with a complaint of painless swelling on her chin and lower anterior jaw bone. The swelling was noted by the patient 3-month earlier and had slowly increased in size. Careful histopathological examination is needed to diagnose GOC, and a careful long-term follow-up is advocated.

Keywords: Glandular odontogenic cyst, histopathology and surgical treatment

Introduction

Glandular Odontogenic Cyst (GOC) is an uncommon jaw bone cyst of odontogenic origin, first described as a distinct clinicopathologic entity in 1988 by Gardner *et al.* [1] In 1987, Padayachee and Van Wyk [2] reported two cases that were similar to Botryoid Odontogenic Cyst (BOC) but with a glandular element and proposed the term 'Sialo odontogenic cyst'. The GOC is included in the WHO histologic typing of odontogenic tumors under the terms GOC or Sialo odontogenic cyst [3] Magnusson *et al.* observed that only 0.012% of the cysts seen in the oral cavity have fulfilled the criteria of GOC microscopically [4].

The prevalence of GOC varies from 0.012% to 1.3% of all jaw cyst with a mean of 0.17%. Its clinical importance arises from two properties, "high recurrence rate and "aggressive growth pattern." [5] Clinically, the most common preferred site of occurrence is the mandible, especially in the anterior region. The cyst presents commonly as a slow growing intraosseous lesion in the anterior mandible and frequently crossing the midline. Small cysts may be asymptomatic; however, large cysts often produce clinical expansion, which sometimes can be associated with pain and rarely paresthesia may be seen. Clinical and histological diagnoses are significant as GOC have shown a tendency towards recurrence similar to the odontogenic keratocyst, and its resemblance microscopically with the central mucoepidermoid carcinoma (CMEC) [4] In this article, we summarized clinical, radiological, and histopathological features of case diagnosed as GOC.

Case Report

A 35-year-Female patient presented to the hospital with a complaint of painless swelling on her chin and lower anterior jaw bone. The swelling had a 3-month history of presentation and had slowly increased in size. The patient denied any significant medical history or family history. Extra oral examination revealed a slight facial asymmetry with a slight expansion of both buccal and lingual cortices in the anterior mandible. Neither cervical lymphadenopathy nor sign of inflammation were noted. The expansion was more pronounced on facial surface than on lingual surface. The overlying oral mucosa was intact. The right central incisors, lateral incisors, and lower anterior teeth had grade 3 mobility. Panoramic radiograph revealed 4× 3 cm, multilocular radiolucency with corticated, scalloped border. The main lesion extended from roots of premolar on the right side to distal root of molar on left side. (Fig 1).

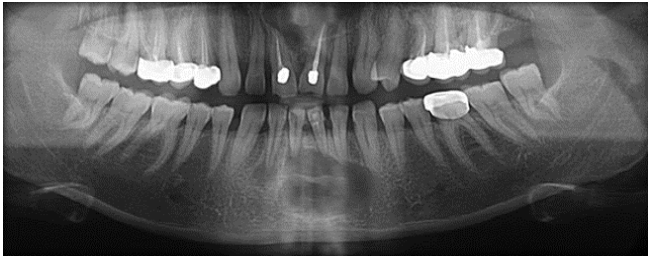


Fig 1: Panoramic radiograph

An incisional biopsy was taken for histopathological examination, revealed nonkeratinized stratified squamous epithelium lining with a flat interface with subsequent stroma. The epithelium showed plaque-like thickenings and whirling pattern of squamous epithelial cells. Other areas of epithelium showed pseudo-glandular pattern with few ciliated and mucous cells. Histopathology was suggestive of glandular odontogenic cyst. (Figure 2).

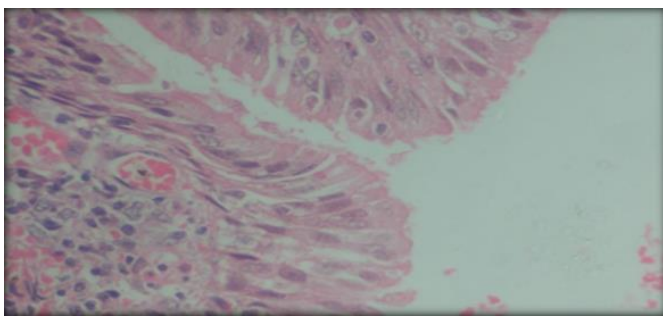


Fig 2: Photomicrograph showing superficial layer of the epithelium with eosinophilic cuboidal and columnar ciliated cells along with papillary projections (40x)

Treatment: One month later, complete enucleation and subsequent curettage were performed under endotracheal general anesthesia. Very thick cystic lining was removed from the cavity. Primary closure was done (Fig. 3, Fig. 4, Fig. 5). Specimen was sent for histopathology investigation, which revealed again glandular odontogenic cyst. One-year follow-up the case has revealed no recurrence on the cyst (Figure 6).



Fig 3: Surgical incision



Fig 4: Surgical extraction

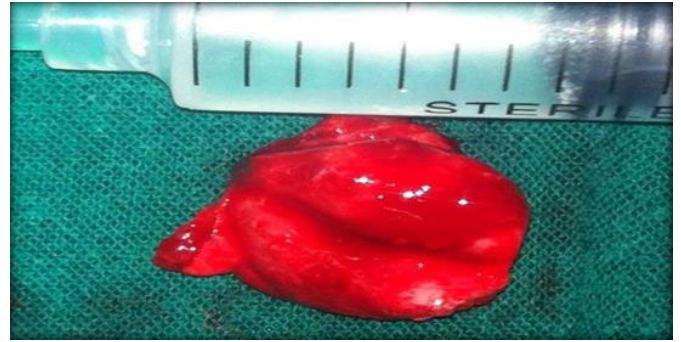


Fig 5: glandular odontogenic cyst



Fig 6: Soft tissue healing after 12 months

Discussion

GOC is a local aggressive cyst from odontogenic origin with a high recurrence rate. The average age at diagnosis of the cyst is 51 with a peak ranged from the 5th to 7th decade. GOC has no gender predilection [6].

GOC has a clear preference for the mandible, with 71.8% of cases in this region and only 28% in the maxilla which is similar to other studies. The anterior mandible was the most common area of involvement. The involvement of anterior jaws both anterior mandible and anterior maxilla was twice that of the posterior jaws [7].

Radio graphically, the lesions typically presented as a radiolucent unilocular or multilocular lesion. Unilocularity of the cyst was the more common presentation with 56% cases. This was similar to other reviews of Kaplan *et al.* and Fowler *et al.* [6]

According to Kaplan *et al* [4], histopathologically it exhibits.

Major criteria

- Non-keratinized squamous epithelial lining with a flat interface
- Presence of “spherules”/knobs or “whorls” or focal luminal proliferations
- Epithelial lining exhibits surface cuboidal eosinophilic cells or “hob-nail” cells
- Mucous/goblet cells with intraepithelial mucous pools with or without crypts lined by mucous producing cells
- Intraepithelial glandular microcystic or duct-like (pseudoglandular) structures.

Minor criteria include

- Papillary proliferation
- Ciliated cells
- Multicystic or multiluminal architecture
- Clear or vacuolated cells in basal or spinous layer.

As a guideline, they suggested that at least the focal presence of each of the major criteria is mandatory, whereas the minor criteria need not be present for the diagnosis but may just support it.

Treatment of GOC includes curettage and enucleation, although some authors believe marginal resection to be a more reliable treatment, due to tendency of the cyst to recur after curettage and enucleation. However, no malignant transformation potential had been seen even though an increased Ki-67 index reported in certain studies^[8,9].

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