Case Report & Review

Bilateral glandular odontogenic cyst of the maxilla: A rare case report and review of literature

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ABSTRACT

The glandular odontogenic cyst (GOC) is a recently recognized rare developmental odontogenic cyst having an aggressive behavior and accounts for 0.012 to 1.3% of all jaw cysts. In 1992, the World Health Organization recognized this cyst as glandular odontogenic cyst (GOC). Clinically, GOC usually presents as a painless, slow-growing swelling that tends to affect the anterior part of the jaws. Many authors have suggested that the cyst mainly occurs in 4th and 5th decades and presents as an expansion of jaws with or without pain or Paraesthesia. The dental professionals must be aware of the clinical significance of this lesion as the aggressive nature of the lesion has been reported and at least 25% to 55% recur following curettage. Hence the treatment of choice is excision rather than enucleation in order to obviate recurrence. Here, we report a case of bilateral GOC in the anterior region of the maxilla, in a 29-year-old male patient, which is very unusual as it is the first bilateral case to be reported in the literature.

Keywords: *Glandular odontogenic cyst, maxilla, bilateral*

Introduction

Padayachee, and Van Wyk described in 1987 two cystic lesions of the jaws with histologic features that did not fit into any known classification of cysts. [1] The histology was suggestive of glandular differentiation and also presented features of the botryoid variant of lateral periodontal cyst. The authors coined the term “sialo-odontogenic cyst,” denoting an association with salivary glands. In 1988, Gardner et al reported a series of these cysts and called the lesion glandular odontogenic cyst (GOC) because the cyst wall epithelium contained only mucin elements with no evidence of salivary tissue involvement. [2] The GOC is included in the WHO histologic typing of odontogenic tumors under the terms GOC or Sialo odontogenic cyst. [3] The second edition of the World Health Organization's (WHO) histological classification of odontogenic tumours in 1992 recognized it as "a cyst arising in the tooth-bearing areas of the jaws characterised 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". [3, 4, 5, 6]

The glandular-odontogenic cyst (GOC) is considered a developmental, uncommon jawbone lesion of odontogenic origin,derived from the rests of dental lamina. It is more common in males, and no race or ethnic predilection has been found. [7] These cysts tend to occur in a wide age range; the mean age being 49.5 years. [8] Generally GOC’s are intaosseous and

clinically produce painless slow growing swellings. Most of the lesions were located in the anterior jaw with mandibular involvement in 80% of the cases. [9] Radiographic findings of these cysts reveal unilocular or multilocular radiolucency with a well-defined border and often scalloped margins. [10, 11, 12]

Histologically, GOC is a polycystic structure and is lined in parts by non-keratinized stratified squamous epithelium of varying thickness. The epithelium has a glandular or pseudoglandular structure, with goblet or mucous producing cells as well as intra epithelial crypts or microcysts containing mucous. Sometimes these crypts form duct-like structures filled with PAS positive material. Ghost cell keratinization also has been described. The interface between the epithelium and the connective tissue is flat. [13]

Several treatment modalities have been used. These include curettage, enucleation with careful dissection of the margins and local block excision. The prognosis of this cyst still remains unclear. However, the aggressive nature of the lesion has been reported, and at least 25% to 55% recur following curettage. It is imperative to follow up the patient carefully for recurrent lesion several years after curettage or enucleation, since cases have been reported as long as seven years after original treatment. [14] We report a case of bilateral GOC in the anterior region of the maxilla, in a 29-year-old male patient, which is very unusual as it is the first bilateral case to be reported in the literature.

Case Report

A 29-year-old male patient was referred to our hospital for the complaint of pain in the right anterior maxillary region for the past 4-5 days. Patient was asymptomatic earlier, and the pain aggravated on chewing hard food. Extra orally, the swelling was not obvious and did not produce any facial asymmetry. Intraorally mucosa over the swelling appeared normal. Tenderness on percussion was elicited in relation to 14 and 15. Radiography revealed periapical radiolucency in relation to 14, 15, 16 and 24, 25, 26 measuring 5x6 cm in size. An excisional biopsy of the lesion was performed from both right and left maxilla and the specimen was submitted for histopathological examination. Macroscopically the submitted tissue was slimy in consistency and whitish grey in color.

Microscopy of the biopsied specimen showed a non-keratinized stratified squamous epithelial cystic lining with a connective tissue capsule. The cystic lining is of variable thickness exhibiting focal plaques of epithelium (Fig. 1) with a flat connective tissue interface. The superficial layer of cystic lining consists of columnar cells, goblet cells, mucous cells & few ciliated cells that form papillary projections. (Fig. 2) Within the thickness of the epithelium there are intra epithelial gland like structures which in some areas showed mucin filled crypts (mucicarmine positive) (Fig. 3) and in other areas showed PAS positive material. (Fig. 4) Connective tissue revealed the formation of a daughter cyst and moderately collagenous with few endothelium lined vascular channels. (Fig. 1) Few areas of irregular calcifications along with mild chronic inflammatory cell infiltration are also present. The diagnosis of glandular odontogenic cyst was given correlating with the clinical, radiographic, and histopathologic features.

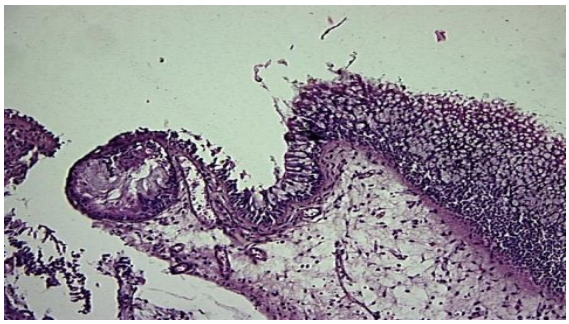

Fig. 1 Photograph showing nonkeratinised stratified squamous epithelial lining along with daughter cyst within the moderately collagenous connective tissue stroma [H&E,40x]

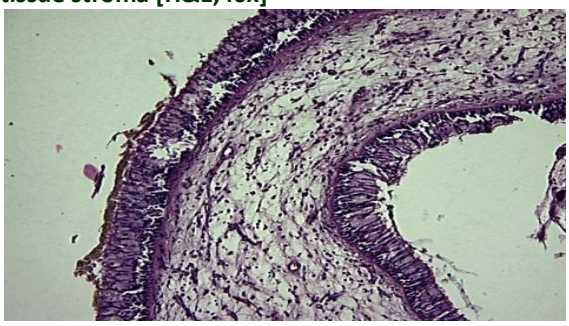

Fig. 2 Photograph showing nonkeratinised stratified squamous epithelial lining with goblet cells, mucous cells and intraepithelial clefts [H&E,40x]

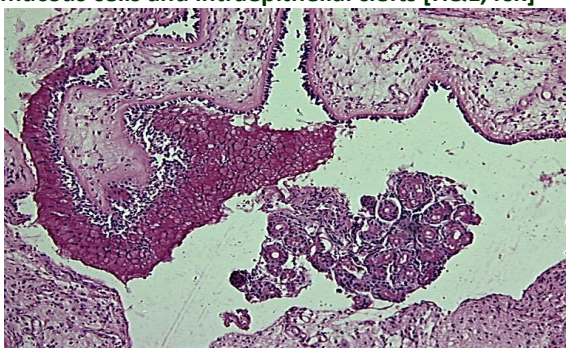

Fig. 3 Photograph showing special stain[Mucicarmine stain, 10x]

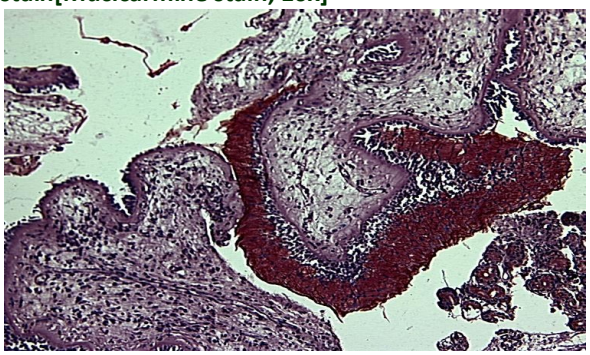

Fig. 4 Photograph showing special stain [Per-iodic Acid Schiff Stain, 10x]

Discussion

GOC is a rare lesion with a frequency rate of only 0.012%[6] to 1.3% of all jaw lesions. [15, 16] It is therefore seldom suspected on clinical and radiological examination. Some researchers believe that GOC is often misdiagnosed because of the overlaping of its histological features with other odontogenic cysts, such as botryoid or lateral periodontal cysts or central low-grade mucoepidermoid carcinoma. [17-20]

GOCs shows predilection for males in most global groups, except for the sub-Saharan African global group. Most of the cases occur in the fifth decade. Men predominate in the second to the fourth decades, whereas women predominate in the fifth. This anomaly may reflect the hormonal changes occurring in females during the perimenopause or menopause. [21, 22, 23] Swelling, pain and discovery as an incidental finding were the most common clinical presentations encountered. Histopathologically GOC shows the following features: [2, 3, 20, 24, 25, 26] Nonkeratinized stratified squamous epithelial lining of highly variable thickness presenting a flat interface with the underlying connective tissue. The superficial layer of epithelium consists of eosinophilic cuboidal cells or “hob-nail” cells, columnar cells, or ciliated cells that form papillary projections and fronds. Pools of mucicarminophilic material, mucous cells, larger granular cells, and vacuolated cells in variable numbers are seen within the epithelium. Within the thickness of epithelium, there are intraepithelial gland like structures consisting of mucous cells and mucin filled crypts or microcysts lined by cuboidal cells that are presumed to result from folding of lining epithelium. Focal thickenings or plaques of epithelium where cells form whorls or spheres are also

seen. The sub epithelial connective tissue is usually free of inflammation and may present irregular shaped calcifications or islands of odontogenic epithelium.

GOCs should be distinguished from Lateral Periodontal Cyst (LPC), Botryoid Odontogenic Cyst (BOC), and Central Mucoepidermoid Carcinoma as they exhibit considerable overlapping of histological features. LPC is a developmental odontogenic cyst lined by thin non-keratinized epithelium and also exhibits focal epithelial thickenings and glycogen rich epithelial cells, similar to those observed in GOC's. [27, 28] BOC is a locally aggressive polycystic variant of LPC, shows similar histomorphologic feature with those of GOC, like epithelial plaques and areas of glycogen rich clear cells.[29] However, the identification of ciliated epithelium and duct like spaces with mucous cells specifically differentiate it from LPC and BOC and favors the diagnosis of GOC. [3, 30]

To differentiate low grade Central Mucoepidermoid Carcinoma (CMEC) from GOC (multicystic variant) is more important and difficult. Significant histological overlapping exists between GOC and CMEC. However, superficial cuboidal cells, epithelial whorls, ciliated cells, and intraepithelial microcyst or duct like structures are not typical for CMEC and their presence or absence can help in establishing a definitive diagnosis. [31] Immunostaining with CK-18 and 19 and their positivity in GOC may help in differentiating GOC from CMEC. [32] Certain studies demonstrated that the use of IHC for p-53 and Ki-67 can help in differentiating GOC from CMEC. GOC exhibited decreased p-53 positivity and increased Ki-67 index when compared to CMEC. [31]

Rare findings include an association with Ameloblastoma, [2] squamous odontogenic tumor like hyperplasia, solid epithelial down growths into the cyst wall, [24] satellite microcysts, [33] hyaline bodies [34] and epithelial ghost cell calcification. [35]

It is of interest to note that in this case the cyst was bilateral and in the anterior maxillary region which is seldom encountered, and is necessary to distinguish from surgical ciliated cyst (SCC). SCC is lined by pseudo stratified ciliated columnar epithelium of respiratory type but can be totally or focally replaced by squamous, cuboidal or columnar cells. Also there may be sub-epithelial hyalinization and foci of squamous metaplasia of luminal epithelium in areas of inflammation. The connective tissue wall is fibrous with or without inflammatory infiltrate.

The aggressive biologic behavior 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. [2o, 25, 36, 37] Further, immune-histochemical findings in GOC suggest that its biologic behavior may be associated with dysregulation of cell death in the lining epithelium indicated by increased expression of anti apoptotic protein bcl-2. But in spite of its aggressive nature, the cyst frequently recurs; the numbers of Ki-67 positive cells are lower, which suggest that biologic behavior of GOC is not associated with cell proliferation. Incomplete removal due to its multicystic configuration, tendency of epithelium to separate from connective tissue or growth through cancellous spaces of bone may account for its high recurrence rate. [25]

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