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CODEN: IJBABN **Case Report**

Glandular odontogenic cyst: A rare clinically perplexing entity

Surya Narayan Das, Swarnav Patnaik, Punyasloka Pati, Biranchi Narayan Biswal and Rachna Rath

Department of Oral Pathology and Microbiology, S.C.B Dental College and Hospital, Cuttack, Odisha-753007India

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***Correspondence Info:**
Dr. Punyasloka Pati
Post Graduate Student,
Department of Oral Pathology and Microbiology,
S.C.B Dental College and Hospital, Cuttack, Odisha-753007 India

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Abstract

Glandular odontogenic cyst (GOC) is a rare and infrequent jaw bone cyst of odontogenic origin described in 1987 by Gardener *et al* as a distinct entity. It is a cyst having an unpredictable, potentially aggressive behaviour and has the tendency to grow very large with relatively high recurrence rate. It poses a diagnostic challenge as it can be clinically and histopathologically confused with lateral periodontal cyst, botryoid odontogenic cyst, radicular and residual cysts with mucous metaplasia. The present case report describes GOC in a female patient. Careful histopathological examination is needed to diagnose GOC, and a long-term follow-up is advocated.

Keywords: Glandular odontogenic cyst, mucous cell, botryoid odontogenic cyst.

1. Introduction

Glandular odontogenic cyst (GOC) was first documented as 'sialo-odontogenic cyst' by Padayachee and Van Wyk in 1987. They published two cases that resembled both the botryoid odontogenic cyst and the central mucoepidermoid tumor of jaws. After careful analysis, they concluded both lesions as separate entities. Gardner *et al*., in 1988 described its histopathological features, biological behaviour and established it as a distinct entity and proposed the term 'GOC'. [1,2]

The pathogenesis of GOC represents one of the three possibilities. (1) A true cyst of glandular origin from either entrapped salivary gland primodia or undifferentiated primitive epithelial rests that differentiates into glandular epithelium. (2) An odontogenic primodial origin cyst in which the epithelial lining undergoes prosoplasia (metaplasia from a less specific differentiation to a more specific differentiation) into glandular epithelium. (3) Low-grade mucoepidermoid carcinoma that forms an initial single cystic space instead of the usual multicystic spaces.[3]

The most common mode of presentation is a slow growing intraosseous lesion in the anterior mandible (nearly 87.2%) and most of these are seen crossing the midline. This cyst most is commonly seen in middle-aged adults with a mean age of 49.5 years. Small cysts may be asymptomatic while large cysts often produce clinical expansion. Radiographically, the lesion may appear as a unilocular or, more commonly, multilocular radiolucency. [4,5]

Characteristic histopathological features are variable thickness of nonkeratinized stratified epithelium, with superficial layer of epithelium consisting of columnar or cuboidal cells, occasionally seen with cilia and numerous goblet cells. The epithelium has glandular or pseudoglandular structure, with intra-epithelial crypt or microcyst formation. [6]

GOC being very rare, we present a unique case report citing clinico-histopathological features of this uncommon cyst.

2. Case report

A 75year old female patient reported with chief complaint of swelling in lower front tooth region for last 6 months. Intraorally there was well circumscribed labial

swelling measuring about 5 x 3 cm present in the anterior mandibular region extending from 42 to 34 obliterating the labial vestibule. The swelling was firm, nontender, nonpulsatile and smooth on palpation. The overlying mucosa was normal in colour & texture. There was no evidence of caries or non vitality of the related teeth. Radiographic examination revealed a large, unilocular radiolucency extending from 42 to 34 region [Figure 1a,b]. A provisional diagnosis of an odontogenic cyst was made although possibility of ameloblastoma and giant cell granuloma was considered in the differential diagnosis.

Incision biopsy was performed & microscopic examination revealed that the cyst was lined by a nonkeratinized stratified squamous epithelium with flat epithelial connective tissue interface [Figure 1c]. The superficial layer of the epithelial lining consisted of cuboidal and in some areas columnar cells with hair like projections/ cilia. Mucous/goblet cells with glandular microcystic or duct-like (pseudoglandular) structures were also present in the superficial part of the epithelium. [Figure 1d]. On the basis of precise histopathology, a final diagnosis of GOC was made. The lesion was surgically excised considering the aggressive nature of it and post-operative follow up has been uneventful.

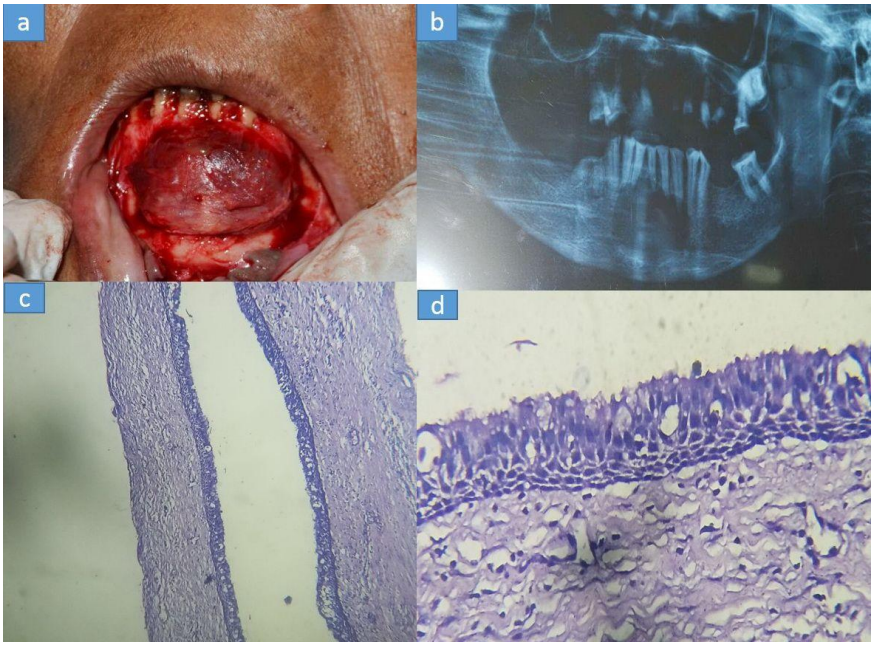

Figure 1

a) Solitary well circumscribed swelling involving anterior mandible with respect to 42 -34 tooth region.
b) Orthopantogram showing unilocular radiolucent lesion in anterior mandible
c) Histopathology showing non-keratinized stratified squamous epithelial lining with underlying non-inflamed fibrous capsule (H & E stain, 100X).
d) Microscopic evidence of presence of cuboidal/columnar cells with cilia, goblet cells & microcystic areas in the stain, 400X).

3. Discussion

GOC is a rare lesion with a frequency rate of only 0.012%to 1.3% of all jaw lesions. Hence this lesion is seldom suspected on clinical and radiological examination. Some researchers believe that GOC is often misdiagnosed because of the overlap of its histological features with other odontogenic cysts, such as botryoid or lateral periodontal cysts or central low-grade mucoepidermoid carcinoma. Furthermore, the radiographic appearance of GOC varies and is not pathognomonic.[7-9]

There is a slight male predilection and a lesion occurs mostly in middle-aged patients. The most common clinical finding is that of a painless swelling. The anterior mandible is the preferred site of occurrence for this cyst. Radiographically the lesion presents as a unilocular or multilocular radiolucency, usually with well-defined borders. [4] The differential diagnosis includes botryoid cysts, keratocysts, residual cysts, central mucoepidermoid carcinoma (CMEC) and ameloblastoma. Our case agreed with the above findings except that it occurred in a female patient at a later age group.

The histopathological characteristic features of GOC given by Gardner *et al*. and 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. The sub-epithelial connective tissue is usually free of inflammation. Our case depicts all the classical characteristic features of GOC.[2,10]

On the basis of histopathology GOC should be differentiated from lateral periodontal cyst (LPC) and CMEC as they exhibit overlapping of histological features. The lining epithelium in LPC exhibits focal thickenings and glycogen-rich epithelial cells, similar to those observed in GOC. However, the identification of ciliated epithelium, presence of superficial cuboidal cells and intraepithelial microcysts or duct-like structures with mucous cells specifically differentiate GOC from LPC and botryoid odontogenic cyst (BOC) and favours the diagnosis of GOC.[11]

The differentiation of low-grade CMEC from GOC is more important and difficult. However, superficial cuboidal cells, epithelial whorls, ciliated cells and intraepithelial microcyst or duct-like structures are not found in CMEC. The central MEC and the GOC are distinct entities with different cytokeratin (CK) profiles and the differential expression of CKs 18 (CMEC) and 19 (GOC) could be useful adjunctive tools in differentiating these two entities. [12]

The aggressive biologic behaviour of GOC and its propensity for recurrence might be associated with cell kinetics in the lining epithelium. Tosios *et al*. (2000) investigated the expression of bcl-2 protein, Ki-67 antigen and p53 protein in GOCs & concluded that the increased expression of the anti-apoptotic bcl-2 may be associated with deregulation of cell death in the lining epithelium of the GOCs; however Ki-67 and p53 status did not seem to play a significant role in cell proliferation. [13]

Treatment by enucleation or curettage alone is associated with a high recurrence rate. Small unilocular lesions can be treated by enucleation. Surgical treatment of large lesions should include enucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw resection in multilocular cases. Follow up should be continued for at least 3 years & up to 7 years in cases with features associated with increased risk of recurrence rate due to its intrinsic biological behavior, multilocularity of the cyst and incomplete removal of the lining following conservative treatment.[14]

4. Conclusion

Although the GOC is a rare entity, the general dentist should be aware of its presence, and GOC should be included in the differential diagnosis of anterior mandibular swellings. As clinical radiographic correlation is not sufficient; this case draws the importance of histological evaluation of tissue removed from the lesion. Due to its aggressive behaviour and the tendency for recurrence, adequate treatment and long-term follow-up is advocated.

References

[1] Padayachee A, Van Wyk CW. Two cystic lesions with features of both the botryoid odontogenic cyst and the central mucoepidermoid tumour: Sialo-odontogenic cyst? *J Oral Pathol* 1987; 16: 499-504.

[2] Gardner DG, Kessler HP, Morency R, Schaner DL. The glandular odontogenic cyst: An apparent entity. *J Oral Pathol* 1988; 17: 359-66.

[3] Mark RE, Stern D. Oral and maxillofacial pathology. A rationale for diagnosis and treatment. 2nd ed, Vol. New Delhi: Quintessence Publishing co, Inc; 2012.

[4] Shafer WG, Hine, Levy. Shafer's textbook of Oral Pathology.5th ed. India: Elsevier; 2005.

[5] Manor R, Anavi Y, Kaplan I, Caldren. Radiological features of glandular odontogenic cyst. *Dentomaxillo fac Radiol* 2003; 32:73-9.

[6] Shear M. Cysts of the Oral Region. 3rd ed. Mumbai: Varghese Publishing House; 1992.

[7] Van Heerden WF, Raubenheimer EJ, Turner ML. Glandular odontogenic cyst. *Head & neck*. 1992 Jul 1; 14(4):316-20.

[8] Waldron CA, Koh ML. Central mucoepidermoid carcinoma of the jaws: report of four cases with analysis of the literature and discussion of the relationship to mucoepidermoid, sialodontogenic, and glandular odontogenic cysts. *J Oral Maxillofac Surg* 1990; 48:871– 877.

[9] Altini M, Shear M. The lateral periodontal cyst: an update. *J Oral Pathol Med* 1992; 21: 245–250.

[10] Fowler CB, Brannon RB, Kessler HP, Castle JT, Kahn MA. Glandular odontogenic cyst: Analysis of 46 cases with special emphasis on microscopic criteria for diagnosis *Head and Neck Pathol* 2011; 5:364-75.

[11] Kaplan I, Anavi Y, Hirshberg A. Glandular odontogenic cyst: A challenge in diagnosis and treatment. *Oral Dis* 2008; 14:575-81.

[12] Prabhu S, Rekha K, Kumar GS. Glandular odontogenic cyst mimicking central mucoepidermoid carcinoma. *J. Oral Maxillofac. Pathol.* 2010 Jan 1; 14(1):12.

[13] Tosios KI, Kakarantza Angelopaulou E, Kapranos N. Immunohistochemical study of bcl-2 protein, ki67 antigen and p53 protein in epithelium of glandular odontogenic cysts and dentigenous cysts. *J Oral Pathol Med* 2000; 29:139-44.

[14] Kaplan I, Gal G, Anavi Y, Manor R, Caldron. Glandular odontogenic cyst: Treatment and recurrence. *J Oral Maxillo fac Surg* 2005; 63:435-41.