Case Report

Glandular Odontogenic Cyst of the Posterior Maxilla

Jahanshah Salehinejad DDS MS[1], Shadi Saghafi DDS MS[1], Reza Zare-Mahmoodabadi DDS MS[1], Narges Ghazi DDS MS•[1], Hamed Kermani DDS[2]

Abstract

The glandular odontogenic cyst is an uncommon developmental odontogenic cyst described as a distinct entity by Gardner et al. in 1988. The Glandular odontogenic cyst occurs more commonly in middle-aged people and has a predilection for the mandible. Only histopathological examinations allow for certain diagnosis of the cyst. The increased recurrence rate can be due to its multilocularity and incomplete removal of the lining following conservative treatment. This article presents a case of glandular odontogenic cyst in a 28-year-old male patient in the posterior region of the maxilla, which is quite rare.

Keywords: cyst, glandular, odontogenic cyst

Introduction

Glandular odontogenic cyst (GOC) is an uncommon developmental cyst.[1] In 1987, Padayachee and Van Wyk[2] reported the first two cases of this cyst as a "sialo-odontogenic cyst". This cyst has been established as a distinct entity by Gardner et al.[3] in 1988, under the term "glandular-odontogenic cyst". In 1992, GOC was included in the list of histologic typing of odontogenic tumors by the World Health Organization (WHO) with the terms GOC or sialo-odontogenic cyst.[1]

Clinically, the most common site of occurrence is the anterior mandible. GOC has a slight male predilection and occurs primarily in middle-aged patients.[1,4] Radiographic examination reveals a well-defined unilocular or multilocular radiolucency, often with scalloped margins.[5] Histologically, GOC shows certain characteristic features that include nonkeratinized stratified squamous lining epithelium, focal thickenings (plaques) within the lining, eosinophilic cuboidal or columnar cells that may ciliated, papillary projections of epithelium, mucous cells, interepithelial gland-like structures, and absence of inflammation in the subepithelial connective tissue.[2,3,5] Treatment of GOC includes curettage and enucleation, although some authors prefer marginal resection due to a tendency of the cyst to recur after curettage or enucleation.[4]

The incidence of this lesion in the maxilla, particularly the posterior region, is quite rare. Earlier reports indicate that GOCs have been found predominantly in the anterior mandible.[1] Sittitavornwong et al., based on 64 analyzed cases of GOC in the literature, have stated an overall prevalence for mandibular involvement and well-documented predilection toward anterior occurrence when the maxilla is involved.[6] The aim of this article is to present a rare case of GOC, which is uncommon in the posterior maxilla.

Case Report

In November 2008, a 28-year-old man was admitted to the School of Dentistry, University of Mashhad, Mashhad, Iran with a five-year history of painless mild swelling of the right maxilla. The patient's medical history was insignificant. Intraoral examination showed a very mild swelling in the buccopalatal region of the maxilla that extended from the right second premolar to the right third molar. The overlying mucosa was of normal color and appearance. A panoramic radiograph demonstrated a well-defined, unilocular radiolucency present in an intraradicular position between the roots of the second premolar, and first and second molar of the right maxilla, causing root displacement of the second premolar and slight root resorption of the first and second molar (Figure 1).

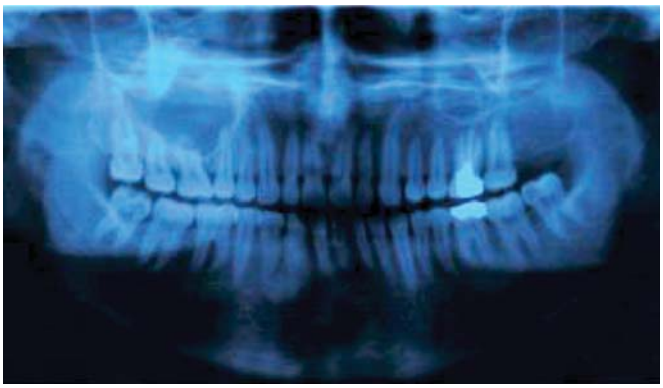

Figure 1. Panoramic radiograph shows a well-defined unilocular radiolucency extending from the right second premolar to the right third molar of the maxilla.

A well-defined unilocular radiolucent lesion was also observed in a periapical film. The lesion was enucleated via a Caldwell-luc approach under local anesthesia. Peripheral ostectomy was performed to a depth of approximately 2 mm. The gross specimen was composed of an elastic, brownish-cream cystic mass that measured 35×25×10 mm. Histopathologic examination of the biopsy tissue showed a cystic lesion with luminal epithelium and surrounding connective tissue. The cyst was lined by nonkeratinized stratified squamous epithelium. Epithelial thickness varied between 4 to 6 cells, along with papillary projections into the cyst's lumen (Figure 2A). The epithelial superficial layer showed eosinophilic cuboidal and ciliated columnar cells (Figure 2B). Focal epithelial thickenings (plaques) were also observed (Figure 3A). Glandular-like structures and mucous cells were seen throughout the lining of the epithelium (Figures 2A, 3A, and 3B). The underlying connective tissue consisted of densely fibrous tissue without inflammation. The patient was diagnosed with GOC. Two years after surgery, the

Authors' affiliations: [1]Department of Oral and Maxillofacial Pathology, Dental Research Center, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran, [2]Department of Oral and Maxillofacial Surgery, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran.
•**Corresponding author and reprints:** Narges Ghazi DDS MS, Department of Oral and Maxillofacial Pathology, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran. Tel: +98-511-882-9501,
E-mail: nargess_ghazi@yahoo.com.
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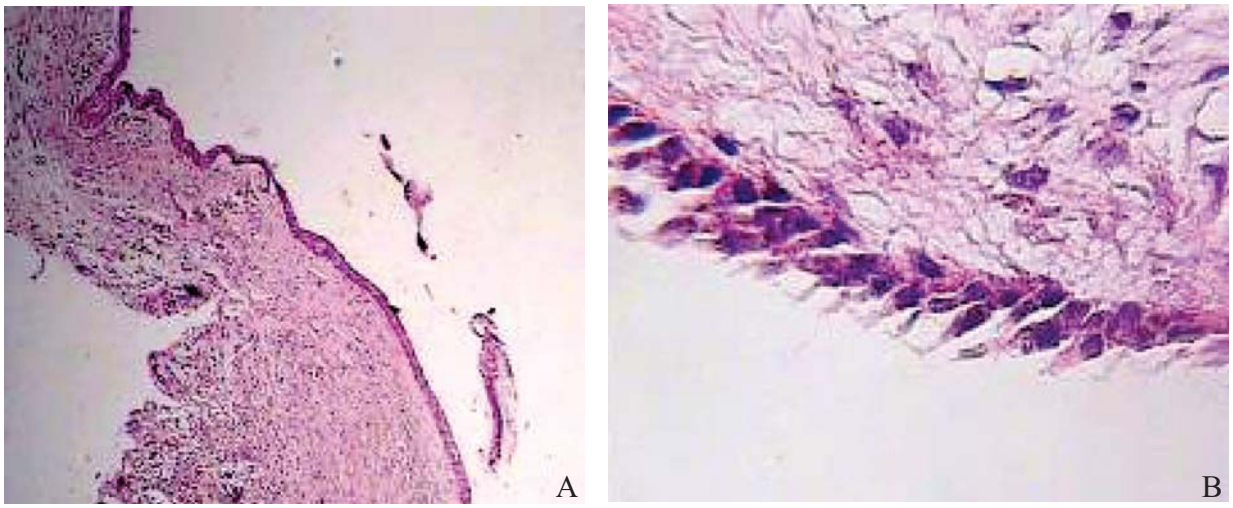

Figure 2. A) Photomicrograph showing crypt formation, papillary projections and mucous cells (H&E, original magnification 40×). **B)** The epithelial superficial layer showed eosinophilic cuboidal and ciliated columnar cells (H&E, original magnification 400×).

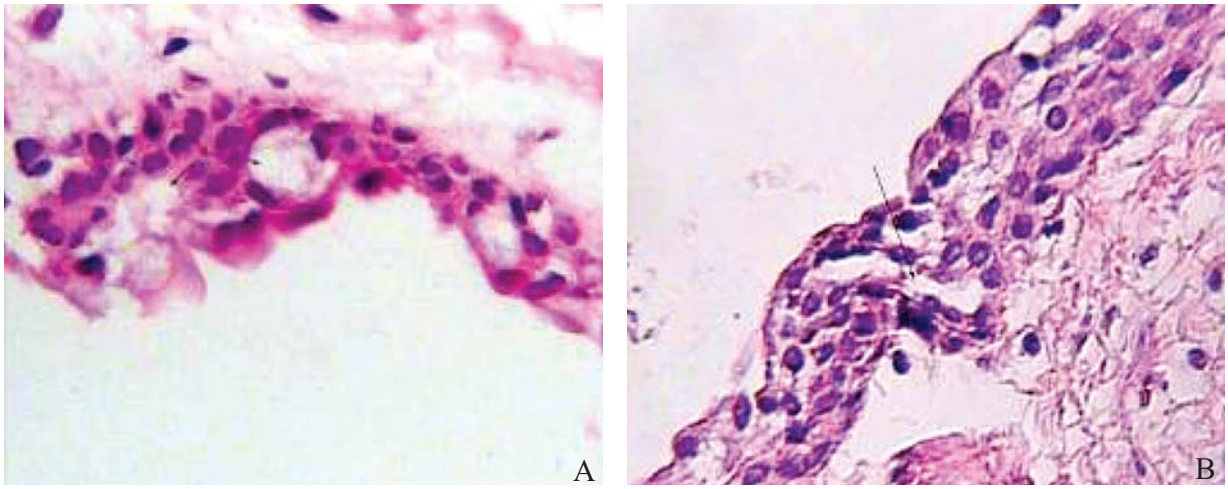

Figure 3. A) Mucous cells (arrows) and epithelial thickening (plaques) are seen in this section (H&E, original magnification 400×). **B)** Cyst lining composed of gland-like structures (arrow, H&E, original magnification 400×).

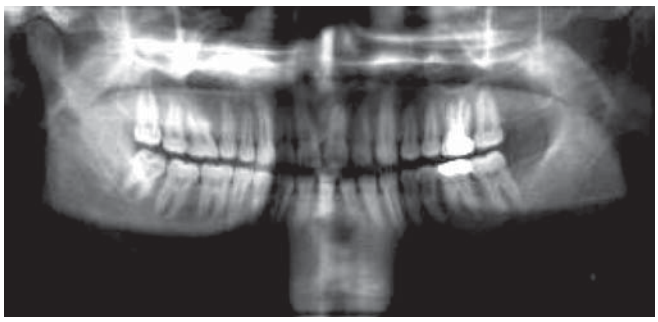

Figure 4. Postoperative panoramic radiograph after two years. Note improved healing of the bone in the area of the pre-existing lesion.

patient was seen for a follow-up examination. Panoramic radiograph showed bone formation in the area of the preexisting lesion (Figure 4).

Discussion

As it has been remarked in the introduction the GOC is an uncommon developmental cyst. Frequency rate of GOC is 0.012% to 1.3% of all the jaw cysts and its prevalence is 0.17%.[1]

Clinically, the most common site of occurrence is the mandible (85%), especially in the anterior region,[4] followed by the anterior region of maxilla.[7] The posterior region of maxilla is a rare area of occurrence for GOC.[4] The clinicopathological features of 64 cases of GOC in the English-Language-Literature from 1966 to 2006 were reviewed by Sittitavornwong et al. Thirteen cases occurred in the maxilla and only two of them were identified in the posterior portion.[6] Nair[5] and Prabhu[8] later reported another two cases of this lesion in the posterior site of maxilla. Recently a case has been described in the posterior part of maxilla invading posteriolateral wall of maxillary sinus,[7] so the present case is probably the sixth case of GOC in the posterior portion of maxilla in the English-Language-Literature.

GOC does not display specific or pathognomonic radiographical features. It may present as a multilocular or unilocular radiolucency with well-defined borders. The recognition of this cyst on the base of physical and radiological examination is practically impossible. Only the histopathological examination allow for certain diagnosis of the cyst.[6,9]

GOC shows certain histological characteristic, which have been divided into major and minor categories by Kaplan and et al.[10]

Major criteria including:

1- Squamous epithelial lining, flat interface

2- Variations in thickness of the lining with or without epithelial "spheres" or "whorl", no palisades
3- Cuboidal eosinophilic cells or "hob-nail' cells
4- Mucous "goblet" cells with interepithelial mucous pools with or without crypts lined by mucous-producing cells
5- Interepithelial glandular microcystic or duct like structures

Minor criteria are as follows:
1- Papillary projections
2- Ciliated cells
3- Multicystic or multiluminal architecture
4- Clear or vaculated cells in basal or spinous layer

The present case showed certain of the abovementioned characteristic criteria such as non-keratinized stratified squamous epithelium of varying thickness, gland-like structures, eosinophilic cuboidal or occasionally ciliated columnar cells that form papillary projections, presence of mucosal cell areas and focal thickenings (plaques). These histological features have also been described by other authors.[2,3,5]

In GOC, mucous cells are remarkably abundant and the papillary fronds or projections are most exceptional.[11] GOC has many histological features similar to low-grade central mucoepidermoid carcinoma (MEC). In fact, it has been suggested that many cases formerly diagnosed as central MEC can be examples of GOC, as well as some low-grade MECs would have originated from GOCs.[12,13]

Pires et al. have demonstrated cytokeratin (CK) expression in GOC and central MEC. They found differences in CKs 18 (30% vs. 100%) and 19 (100% vs. 50%). The authors have suggested that GOC and central MEC are distinct entities in which expression of CKs 18 and 19 could be useful adjunct tools in differentiating between these two lesions.[13] Immunohistochemical studies using cytokeratin 7, 13, 14, and 19 and their positivity strongly support the odontogenic nature.[14] The detection of osteodentin and negative reaction for epithelial membrane antigen (EMA) in the glandular structure show that these features are not of glandular origin and support the concept of odontogenic differentiation in GOC.[1]

The reported treatment of GOC ranges from a conservative approach (enucleation, marsupialization, curettage with or without peripheral ostectomy, curettage with adjuvant Carnoy's solution, or cryotherapy) to marginal resection and segmental resection. Some authors prefer marginal and segmental resection due to a tendency of the cyst to recur after conservative treatment.[4,15]

It has been demonstrated that the rate of recurrence increases with the radiographic complexity of the cyst.[10] It was reported that the majority of patients who were found with recurrent disease were noted to have large multilocular lesions with cortical perforations, whereas those patients with smaller unilocular lesions appeared to be more amenable to curettage and peripheral ostectomy.[15]

The present case showed no sign of recurrence after two years.

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