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Glandular Odontogenic Cyst of the Maxilla- A Rarity: Case Report

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Abstract: Glandular odontogenic cyst (GOC) routinely presents as a painless, slow-growing cystic lesion of the jaw which has an inclination towards anterior part of the mandible. Glandular odontogenic cysts (GOCs) are relatively uncommon intraosseous solitary or multiloculated cysts which originate from the odontogenic epithelium. The interesting fact of GOCs is that they exhibit a propensity for recurrence which is similar to keratocystic odontogenic tumors and that they may be confused microscopically as they exhibit features which are commonly seen in central mucoepidermoid carcinoma. GOC usually occurs in the fourth and fifth decades of life and presents as an expansion of jaws. It shows nonspecific clinical and radiological findings which may resemble other lesions, but histopathological examination helps in its diagnosis. We present a case of asymptomatic glandular odontogenic cyst in a 50-year-old male patient gradually increased in size which was radio-diagnosed as the dentigerous cyst.
Keywords: Glandular Odontogenic Cyst, Dentigerous Cyst, Maxillary swelling, Goblet Cells, Panoramic radiograph, Histopathology

INTRODUCTION

The glandular odontogenic cyst (GOC) is a developmental cyst of the jaws first described in 1988 by Gardner *et al* [1]. In 1987, Padayachee and Van Wyk reported two cases that were similar to the botryoid odontogenic cyst, but with a gland element and suggested the name "sialo odontogenic cyst" [2]. High *et al*., proposed the term 'polymorphous odontogenic cyst' for this cyst because of its aggressive growth pattern.

Glandular odontogenic cyst (GOC) is a rare lesion and has incidence rate between 0.012% and 1.3% of all jaw cysts [3, 4]. The frequency rate of GOC has reported 19 cases in 1995 [5], 54 cases in 2002 [6], and 113 cases until 2009 [7]. GOC also was known as a sialo-odontogenic cyst [8], mucoepidermoid cyst, and polymorphous odontogenic cyst [9-11]. GOC is now considered as an odontogenic cyst and involves middle-aged adults with predilection for anterior region of the mandible. Radiographically, GOC shows unilocular or multilocular radiolucency [7]. Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) is recommended for diagnosis. This cyst is a well-known clinical entity and very important to recognize and diagnose this cyst because due to its aggressive behavior, they recur. This cyst reports a slight male predilection with a male: female ratio of 1.3: 1.6 and the most-commonly affected site is the anterior mandible. It tends to occur over a wide age range of 10-90 years, with a mean age of 49.5 years. The mandible seems to be affected more commonly (87.2%) than the maxilla [12]. GOC is characterized histologically by "an epithelial lining with cuboidal or columnar cells both at the surface and lining or cyst-like spaces within the thickness of the epithelium" Many histological features of GOCs are similar to those of low-grade central mucoepidermoid carcinomas [13] Since GOC is a very rare lesion and its occurrence in the maxilla is unusual, here we present a unique case of a GOC in the maxilla and clinico-histopathological features of this rare cyst.

CASE HISTORY

A 50-year-old Indian male patient reported to our hospital with a complaint of diffuse swelling in the anterior maxilla for the past 4 years (Figure1). The patient was asymptomatic and had no history of discharge. The swelling had gradually increased in size. Medical history was not significant. The swelling was bony hard and non-tender on palpation. Overlying skin was normal. Intraoral examination revealed an expansile swelling crossing midline. The swelling was firm in consistency; non-tender on palpation with obliteration of labial vestibule. The overlying mucosa was normal color and appearance.

The panoramic radiograph revealed well–defined, unilocular radiolucency with sclerotic borders extending from 13 to 25 regions along with impacted supernumerary tooth within the radiolucent lesion (Figure 2). A well-defined unilocular radiolucent lesion was appreciated on CT scan extending from the right canine to the left second premolar of the maxilla (Figure 3). The provisional diagnosis of the dentigerous cyst was given. Aspiration yielded a thick straw color fluid. Enucleation of the cyst was done under general anesthesia with an intraoral approach using crevicular incision extending from maxillary right premolar to left premolar. The cyst was dissected and enucleated completely and sent for histopathological examination. On macroscopic examination, cystic lumen and well-defined borders with the supernumerary tooth was seen.

Histopathological examination (HPE) revealed the presence of cystic lumen with epithelial lining and supporting connective tissue capsule (Figure 4). The cyst wall is lined by the stratified squamous epithelium of varying thickness along with mucous cells, epithelial whorls in some areas and few goblet cells (Figure 5). The superficial layer of the epithelium showed eosinophilic cuboidal and columnar ciliated cells (Figure 6). The connective tissue wall consisted of numerous cholesterol clefts, moderate chronic inflammatory cell infiltrate, and hemorrhage. Based on these findings, histopathology was suggestive of GOC.

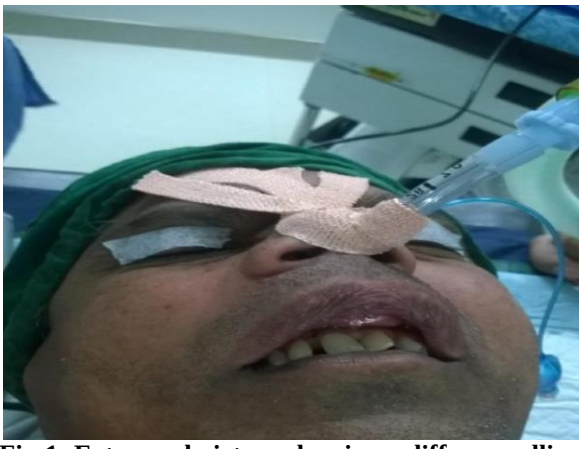

Fig-1: Extra oral picture showing a diffuse swelling on the anterior maxilla

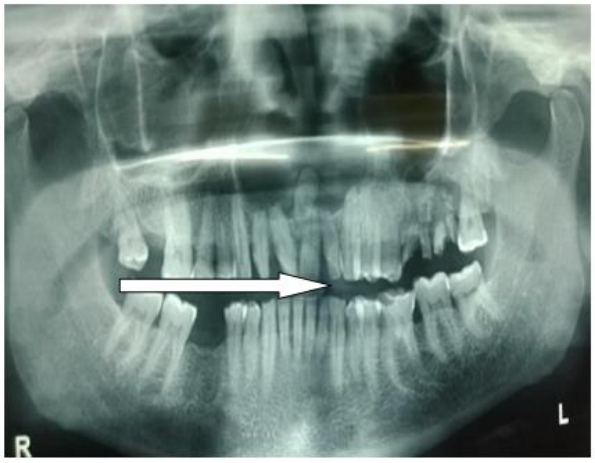

Fig-2: Panoramic radiograph showing well–defined, unilocular radiolucency with sclerotic borders extending from 13 to 25 regions along with impacted supernumerary tooth within the radiolucent lesion.

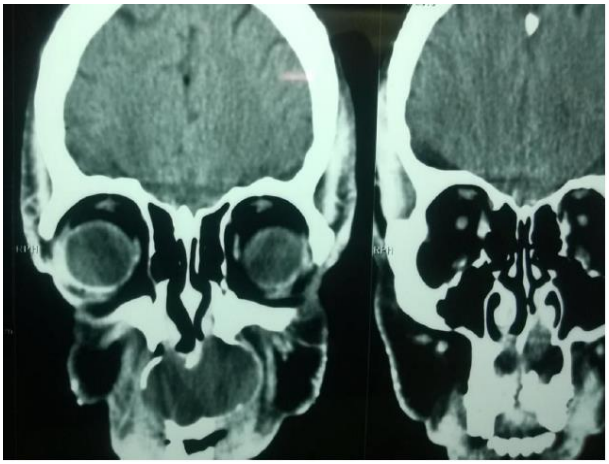

Fig-3: Coronal section of CT scan showing a well-defined unilocular radiolucent lesion extending from the right canine to the left second premolar of the maxilla

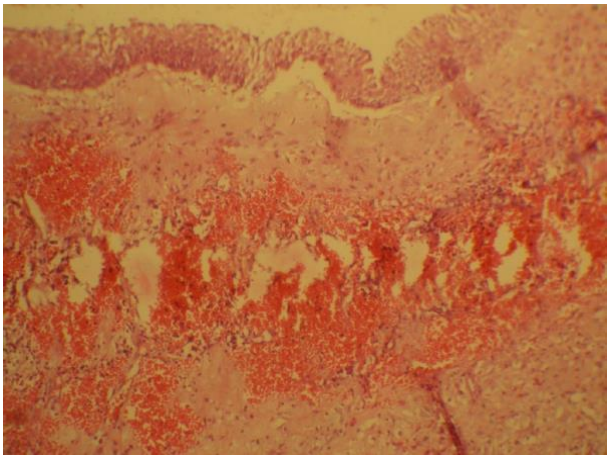

Fig-4: Cyst cavity lined by stratified squamous epithelium with flat epithelial and underlying connective tissue along with areas of hemorrhage (10X)

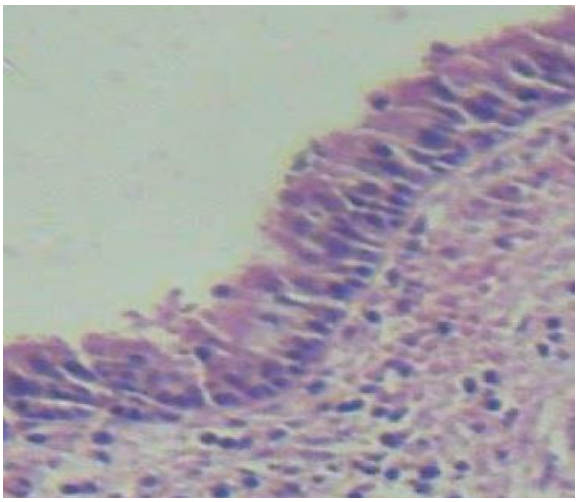

Fig-5: Epithelial lining with goblet cells (40X)

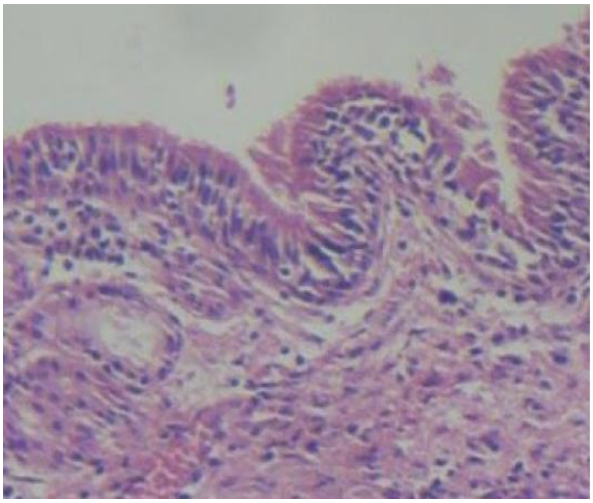

Fig-6: Superficial layer of the epithelium with eosinophilic cuboidal and columnar ciliated cells along with papillary projections (40X)

DISCUSSION

Glandular Odontogenic Cysts are relatively uncommon cysts first reported and documented in 1987. Since the discovery, they have remained as interesting controversy for the researchers throughout the world. The lesion was initially referred to as a "sialo-odontogeniccyst" and believed to have salivary gland origin, but due to lack of evidence the term "glandular odontogenic cyst" by Padayachee but later adopted by the World Health Organization in [2]. Glandular odontogenic cyst (GOC) routinely presents as a painless, slow-growing cystic lesion of the jaw which has an inclination towards anterior part of the mandible. Glandular odontogenic cysts (GOCs) are relatively uncommon intraosseous solitary or multiloculated cysts which originate from the odontogenic epithelium. GOCs usually occur in the fourth and fifth decades of life and presents as an expansion of jaws. It shows non-specific clinical and radiological findings which may resemble other lesions, but histopathological examination helps in its diagnosis. Radiographically, the GOC is a localized lesion and may appear as a multilocular or unilocular radiolucent lesion with well-defined borders. Sometimes it may present with peripheral osteosclerotic border and scalloping, root resorption and displacement of the teeth. Expansion and thinning of cortical plates are sometimes observed on occlusal radiographs [1, 7]. Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) is recommended for diagnosis. This cyst is a well-known clinical entity and is important to recognize and diagnose due to its aggressive behavior and tendency to recur.

Kaplan *et al.* added that a diagnosis of GOC had to be based on the mandatory presence of the following five major features: Squamous epithelium, varying thickness, cuboidal eosinophilic (hobnail) cells, mucous (goblet) cells, and intraepithelial glandular or duct like structures [14].

The diagnosis is made when the superficial layer of the epithelial lining consists of columnar or cuboidal cells sometimes referred to as 'hobnail', occasionally with cilia or filiform extensions of the cytoplasm. Furthermore, the epithelium has a glandular or pseudo-glandular structure, with intra-epithelial crypts or microcysts or pools lined by cells similar to those on the surface. These microcysts may open onto the surface of the epithelium giving a papillary or corrugated appearance with the superficial part of the epithelium showing many goblet cells. Occasionally, the epithelium is thinner, similar to reduced enamel epithelium. Epithelial thickenings or plaques may be present either in this thin epithelium or in the stratified squamous epithelium [15].

The microscopic differential diagnosis of GOC includes lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC), and the central Mucoepidermoid Carcinoma (MEC). The plaque-like epithelial thickening in LPC made authors to believe that the GOC could be the clinical microscopic variant of LPC. But the presence of ciliated epithelium and duct like spaces with mucous cells differentiate GOC from LPC and BOC.

It has been suggested that central MEC (CMEC), especially the low-grade variant is regarded as the most important histopathological differential diagnosis from GOC. It has been speculated that GOC may represent the most benign end of the spectrum of central MEC. However, CMECs do not show superficial cuboidal cells, epithelial whorls, ciliated cells and intraepithelial microcysts or duct-like structures [16].Certain authors have suggested using immunohistochemical markers to differentiate these lesions. The distinction between glandular odontogenic cyst (GOC) and Muco-epidermoid carcinoma (MEC) can be established based on immunohistochemical markers using cytokeratin (CK). CK 7, 13, 14, and 19

postivity and negative reaction for epithelial membrane antigen (EMA) supported odontogenic origin. Some authors suggested expression of CK 8, 18, and 19 might be useful tool in differentiating these lesions.

CONCLUSION

Most of the oral lesions present as swellings which are to be differentiated from one pathological entity from other. In such cases, histopathology forms a definitive diagnostic option. In our case, swelling was seen in the anterior part of the maxilla which is a rare site of occurrence of the glandular odontogenic cyst.

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