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Glandular Odontogenic Cysts in Maxilla: Literature Review and Report on Cases

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Study Area: Mashhad, Iran
Coordinates: 36°18'N; 59°36'E

Key words: Apparent diffusion coefficient map, MR imaging.

Abstract

Glandular odontogenic cysts are rare developmental lesions affecting the mandible, especially its anterior region, more frequently seen in males. This cyst has a high recurrence rate and its radiographic image is associated with its rate of recurrence. There is some overlap between features of radiographic and histopathological images of glandular odontogenic cysts and those of some lesions such as mucoepidermoid carcinoma. Moreover, diagnosis of this cyst is very important due to its invasive potential. This article is based on two patients reported with glandular odontogenic cysts in the maxilla, which is an uncommon location for this lesion. The first patient was a male, 63-yrs. with a glandular odontogenic cyst in the maxillary anterior region. The second reported patient was a female, 21 yrs. where lesion observed in the maxillary posterior region.

Introduction:

Glandular odontogenic cyst (GOC) is a rare developmental cyst (Krishnamurthy *et al.*, 2009; Anchlia *et al.*, 2015; Salehinejad *et al.*, 2011). First of all it was reported by Padayachee & Van Wyk (1987), who referred to it as sialo-odontogenic cyst. However, the World Health Organization (WHO) changed its name to "Glandular Odontogenic Cyst" in 1992 (Kramer *et al.*, 1992). GOC is more common in the mandible, especially in the mandibular anterior region; unfortunately, if GOC occurs in the maxilla, it will affect the globulomaxillary region (White & Pharoah, 2014).

In patients, the lesion could occur in a wide age range, with an average age for developed GOC is 50 yrs. (Tran *et al.*, 2004). Such cysts are more often seen in men (Kaplan *et al.*, 2008), and its radiographic image shows a unilocular and multilocular appearance with well-defined cortical borders that may have smooth or scalloped edges (White & Pharoah, 2014). Histopathological examination of GOC reveals a thick epithelium having cylindrical surface cells with cilia, a large number of microcytes containing mucin, and duct-like spaces in the epithelium (Neville *et al.*, 2008). Although GOC is a rare lesion, its correct clinical diagnosis is of high importance since it has invasive potential, a high incidence of cortical perforation, and a high recurrence probability (Kaplan *et al.*, 2008). Since this cyst is rarely observed in the maxilla (Lyrio, *et al.*, 2009), this article aims to report two cases of GOC in the maxillary anterior region of a male patient and in the

maxillary posterior region of a female patient.

Observed cases:

Case-1:

Patient is of 63 yrs. man who became aware of a swelling under his denture in the maxillary anterior region adjacent to midline on the right side a year ago. He maintained the swelling was drained when the denture experienced pressure, oozing a blood-like liquid. However, since last month, a new swelling has been formed once again in the same region under the denture. According to the patient, he was not having any notable medical history. The mucosa on the swelling was normal and of the same colour as the adjacent mucosa (Fig.-1). In the complete image taken, a single well-demarcated lucent lesion with a cortical border was observed in the maxillary anterior region near the midline area on the right side (Fig.-2).

Figure-1: Cyst-like swelling in the maxillary anterior region on the right side adjacent to the midline

A "Cone Beam Computed Tomography" (CBCT) examination was performed to study the effects of the cyst

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Figure-2: The lucent region with cortical borders in the maxillary anterior region

Figure-3: CBCT image. Destruction of the cortical bone on the palatal side and erosion of the cortical bone on the buccal side on the nasopalatine canal (Fig.-3). The prepared CBCT image showed erosion of the palatal wall and destruction of the buccal wall. Since the wall of the nasopalatine canal was observed on CBCT cross-sectional images, the probability of the cyst being a nasopalatine canal cyst was ruled out. The differential diagnoses that were considered for this lesion included residual cyst, odontogenic keratocyst, "Calcifying Odontogenic Cyst" (COC), and central giant cell granuloma. The patient was referred to the surgical ward for a biopsy procedure.

Histopathological examination of the prepared sample showed a developmental odontogenic cyst. The inner surface of the cyst was made of stratified squamous epithelium, but parts of it consisted of ciliated cylindrical epithelium, mucous cells, and glandular areas could be seen in-between the epithelial cells. The juncture of the epithelium with the connective tissue was smooth and the connective chorion of the cyst contained collagen fibers. Inflammatory infiltration of lymphoplasmacytic cells was observed at the depth of the lesion in the cross-sections of the striated muscles and nerves. The lesion was diagnosed as a developmental odontogenic cyst of the glandular odontogenic type.

Case-2:

Patient was a 21 yrs. female with a swelling in the palatal area of the maxillary posterior region who referred to the Mashhad School of Dentistry (Fig.-4). The swelling developed one year back. Vitality tests of the teeth in the region were positive. In the periapical image, on the right side of the first molar region, the periodontal ligament was slightly loose (Fig.-5). In the complete image, a well-demarcated radiolucency was observed in the maxillary posterior region from the apex of the first premolar to the distal of the first molar (Fig.-6). Possible diagnoses for this patient included odontogenic Keratocyst and Calcifying Odontogenic Cyst (COC).

Figure-4: Clinical view- swelling in the palatal area of the maxillary posterior region on the right side.

Figure 5: Periapical view: well-demarcated radiolucency of the apices of teeth 4-6 showing increased PDL width.

Figure 6: 2nd patient: noncortical well-demarcated radiolucency from the distal tooth 4 to the distal tooth 6

The histopathological examination included an incomplete excision of the cystic wall that consisted of 2-6 layers of stratified squamous epithelium together with a number of mucous cells and mucin-containing pools. The stroma of the cyst consisted of fibrovascular connective tissue, and inflammatory infiltration of lymphoplasmacytic cells was observed. Sections of adipose tissue, vessels, nerves and of the minor salivary glands were obtained from a part of the sample. Based on pathological and radiographic findings, the final diagnosis was that the cyst was of the glandular odontogenic type.

Discussion:

GOC is evidenced as a rare lesion that accounts for 0.02 to 1.2 percent for all jaw lesions (Manor *et al.*, 2003). There were no reports of GOC until Padayachee & Van Wyk (1987) who reported the first GOC lesion (Macdonald-Jankowski, 2010). In a study conducted by Sharifian & Khalili (2007) on odontogenic cysts among 1,277 Iranian patients from 1987 to 2007, GOC was found in about 0.3 percent of the total cases of jaw cysts. How far the gender is concerned, the GOC is more prevalent among men in all age decades but, in women, it is more common in the fifth age decade, which is the general peak decade for GOC. This anomaly may reflect hormonal changes in women during the perimenopausal period (Macdonald-Jankowski, 2010). However, higher prevalence has been reported in women in some references (Tran *et al.*, 2004).

The second patient mentioned in this article was a 21 yrs. female. In a study by Khosravi *et al.* (2013) conducted in Isfahan on demographic distribution of odontogenic cysts, 85 percent of the GOC cases were observed in women and only 14.29 percent in men and the average age of the patients was 50. However, report of Sharifian & Khalili

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(2007) evidenced for equal numbers of male and female patients. A large percentage of patients reported in the study by Macdonald-Jankowski (2010) were in the second half of their fifth age decade, but the target groups were of lower age decades in East Asia and the Sahara Desert. According to a study by Kaplan *et al.* (2008) GOC was observed in the age range of 14-75 year. However, Faisal *et al.*, (2015) reported a GOC case in the mandible of an 11-year-old youngster. Our first and second patients were, respectively, of higher and lower age than the peak age for glandular odontogenic cyst development.

As per White & Pharoah (2014) Glandular odontogenic cysts are more prevalent in the anterior region. In the West (North America and Europe), GOC is reported to be more inclined to affect the anterior regions of both jaws (Macdonald-Jankowski, 2010). In a study by Khosravi *et al.*, (2013) 28.57 percent of GOC lesions were seen in the maxillary anterior region and 14.20 percent in the maxillary posterior region. Although GOC is formed in the mandible four times more compared to the maxilla (Padayachee & Van Wyk, 1987), yet it was observed in the maxilla of both of our patients (in the first patient in the maxillary anterior edentulous region, and in the second patient in the maxillary posterior region).

As for shape and border, 53 percent of the lesions were also seen to be unilocular (Tran *et al.*, 2004). The rate of multilocular lesions in GOC was seen higher compared to other odontogenic cysts (Manor *et al.*, 2003). In 95 percent of the patients the border was reported to be well-defined (Kaplan *et al.*, 2008), with reported sclerotic margins (Manor *et al.*, 2003) and scalloped edges (Kaplan *et al.*, 2008) observed in merely 7.7 and 13 percent of the cases, respectively. In both of our patients, the lesions were unilocular with well-defined cortical borders.

The case report reviews revealed that most lesions in male patients developed exclusively in the maxillary anterior region, were unilocular, where the patients were younger than 40 years of age (Manor *et al.*, 2003; Noffke & Raubenheimer, 2002; Mascitti *et al.*, 2014; Kim *et al.*, 2012; Bhat *et al.*, 2014). In our representative first patient, the lesion was unilocular but he was 63 yrs. Koppang *et al.*, (1998) reported a lesion in the maxillary anterior region of a 69-yrs. man. Further, Patron *et al.* (1991) reviewed the case of a 52 yrs. patient who had been treated for unilocular ameloblastoma of the maxillary anterior region, and reintroduced it as a glandular odontogenic cyst.

Lesions vary in size from 0.5 to 2 cm (Kaplan *et al.*, 2008). One type of size classification for GOC is based on the number of affected teeth: if more than two teeth are affected, the GOC is considered large, but it is considered small if fewer than two teeth are affected (Kaplan *et al.*, 2005). Based on this classification system, the ratio of large to small lesions is four to one (76 percent of the lesions are large) (Kaplan *et al.*, 2008). In the study carried out by Marco, 7.3 percent of the lesions had affected the

anterior and posterior regions simultaneously (Mascitti *et al.*, 2014). In our first patient, the lesion was confined to the anterior region.

Based on previous studies, most patients suffer from painless swellings (Sittitavornwong *et al.*, 2006). Our both the patients had painless swellings. Multilocular lesions have a greater inclination for cortical expansion. In 61 percent of the cases, GOC is accompanied by perforation of the cortical bone and in 24 percent of the cases by cortical bone thinning (Kaplan *et al.*, 2008).

It has been reported that GOC is accompanied by odontogenic keratocyst and ameloblastoma (Yoon *et al.*, 2006; Hisatomi *et al.*, 2000; Kasaboglu *et al.*, 2006; Geist *et al.*, 2004) as well as by impacted teeth.

Lesion size is related to the rate of recurrence where about 86 percent of cases of recurrence are related to large lesions. The GOC may recur more than once, and lesions that cause perforation are more likely to recur and are often multilocular (Kaplan *et al.*, 2008). Recurrence may occur even after seven years, and a minimum follow-up period is of three years which usually extends for longer than seven years, i.e., a high risk of recurrence (Kaplan *et al.*, 2005). Thro *et al.* (2006) followed up the cases of 13 patients with GOC and noticed recurrence in 11 of them during the first ten years. Manor *et al.* (2003) studied complete radiography, the first conventional radiography for GOC, and prepared CT specimen to study perforation in five patients. They suggested CT be performed for large lesions, especially multilocular lesions and/or lesions that affect structures outside of the jaws such as the sinuses and the nasal floor or orbit. Some studies have shown discrepancies between diagnoses based on conventional radiography and those based on CT, but Cano *et al.* (2012) reported there were no differences between these two. Oliveira *et al.* (2009) stated in their article that, in conventional radiography, the image of the patient's lesion was observed as unilocular radiolucency, but CT scan of the patient showed a multilocular lesion with cortex perforation. Hisatomi *et al.* (2000) stated a wide range of imaging modalities such as MRI were helpful in GOC accompanied by ameloblastoma.

In our first patient, complete radiography was first employed. CBCT was then used to study perforation of the nasal floor and cortex. The lesion was unilocular both in the complete and the CBCT images, but perforation was observed in the latter.

Clinically and pathologically, GOC varies from a small benign pathologic lesion to a destructive malignant neoplasm such as a central mucoepidermoid carcinoma (Kasaboglu *et al.*, 2006). Some researchers believe this cyst cannot be diagnosed because its histological image overlaps those of other odontogenic cysts such as botryoid and/or lateral odontogenic cysts and low-grade mucoepidermoid carcinoma (Manor *et al.*, 2003; Sittitavornwong *et al.*, 2006). In both of our patients,

odontogenic keratocyst was considered one of the possible diagnoses. Moreover, giant cell granuloma was similar to the glandular odontogenic cyst (Tran *et al.*, 2004). Hisatomi *et al.* (2003) stated in their study that MRI could not distinguish GOC from dentigerous, radicular, and nasopalatine cysts.

GOC is an odontogenic cyst and must be distinguished from nasopalatine cysts, which are fissural cysts (Kim *et al.*, 2012). For our first patient, who had a lesion in the maxillary anterior region, we employed the CBCT technique to distinguish the GOC from a possible nasopalatine cyst. It was revealed that the cyst was not a nasopalatine one because the canal wall had not been destroyed.

The GOC is treated by curettage or enucleation, although some researchers prefer marginal resection to prevent recurrence inclination after curettage or enucleation (Kasaboglu *et al.*, 2006). Based on the radiography images, the differential diagnoses considered for the first patient included residual cyst (related to an infected tooth that had been extracted earlier), odontogenic keratocyst, calcifying odontogenic cyst (COC), and central giant cell granuloma (based on the lesion's location and shape). The differential diagnoses considered for the second patient, who had a lesion in the maxillary posterior region, were odontogenic keratocyst and calcifying odontogenic cyst. Due to the similarities in histopathological images and invasive potential, and because of the high recurrence rate of glandular odontogenic cysts, especially in the first patient in whom perforation had also happened, it is necessary to diagnose the cyst correctly, treat it appropriately and employ follow-up procedures for the patient in a regular manner.

Conclusions:

Due to the invasive potential and high recurrence rate of the glandular odontogenic cyst, and because of its great similarities to other jaw cysts, it must be correctly diagnosed, adequately treated, and regularly followed up for several years..

Acknowledgment:

The authors are thankful to the subjects (two patients) who had allowed us to use their medical data as a part of this report.

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