

Glandular Odontogenic Cyst: A Case Report

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ABSTRACT

Background: Glandular Odontogenic Cyst (GOC) is a rare developmental cyst of the jaws. The GOC's importance relates to the fact that it exhibits a propensity for recurrence similar to the odontogenic keratocyst, and that it may be confused microscopically with central mucoepidermoid carcinoma (CMEC). Numerous histopathologic features for the GOC have been described, but the exact microscopic criteria necessary for diagnosis have not been universally accepted. Furthermore, some of the microscopic features of GOC may also be found in dentigerous, botryoid, radicular, and surgical ciliated cysts. The histological features of GOC strongly suggest an origin from the remains of dental lamina. Radiographically, GOC presents as well-defined radiolucencies with uni- or multilocular appearance. This report describes a case of GOC occurrence in the posterior mandibular ramus region in a 50-year-old female which is a rare combination of site and gender for occurrence.

Keywords: Glandular odontogenic cyst, oral surgery, dentistry.

INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare developmental cyst of odontogenic origin. Padayachee and Van Wyk initially reported it as a sialo-odontogenic cyst in 1987 but its odontogenic origin was first described by Gardner et al. in 1988 who also proposed the name 'GOC'.^{1, 2} The term 'polymorphous odontogenic cyst' for this cyst was proposed by High et. al in (1996), because of its aggressive growth pattern.³ Sadeghib in 1991 used the term mucoepidermoid cyst or mucous producing cyst due to the microscopic findings of mucus producing cells and squamous cells.² Moreover, WHO histological typing of odontogenic tumors also includes glandular odontogenic cyst under the terms, GOC or Sialoodontogenic cyst⁴. Magnusson et al. observed that only 0.012% of the cysts seen on the oral cavity have fulfilled the criteria of GOC microscopically².

Although rare, this cyst is a well-known clinical entity and is important to recognize and diagnose due to its aggressive behaviour and tendency to recur. This cyst reports a slight male predilection with a male: female ratio of 1.3: 1, 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 maxilla². Radiographic examination reveals a well-defined cyst like unilocular or multilocular radiolucency, often with scalloped margins and sclerotic borders. In addition, there may be root resorption and tooth displacement with cortical perforation, leading to extension of the cyst into the adjacent soft tissues.

Histologically, GOC is characterized by a cyst wall lining of non-keratinized epithelium with papillary projections, nodular thickenings, mucous-filled clefts, and "mucous lakes." In addition, it

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includes cuboidal basal cells, which are sometimes vacuolated³. GOC treatment includes curettage and enucleation, although some authors believe marginal resection to be a more reliable treatment due to the tendency of the cyst to recur after curettage and enucleation^{3 1}. The relative rarity of this lesion is the reason behind presentation of this case. In this case, we are presenting a unique case of GOC in a 50 year old female patient in the posterior mandibular region.

CASE REPORT

A 50 year old female patient referred to the Outpatient Department of Oral and Maxillofacial Surgery at Karnavati School of Dentistry, Gandhinagar, by a local dentist after discovering a well-defined radiolucent pathology in the left ramal region of the mandible on a routine panoramic view. The patient reported mild dull pain over the left posterior mandibular region for a period of 1 month with no aggravating or relieving factors. An extraoral examination revealed no significant clinical findings. Intraorally, there was mild obliteration of the buccal vestibule in the left lower second molar region with no other relevant clinical finding. Past medical, and family history were non-contributory and there was no previous history of trauma. Panoramic radiograph revealed a large unilocular radiolucent extending from the distal border and apex of the lower left second molar into the ramus to approximately 6 mm away from the sigmoid notch. There was no expansion of the posterior or inferior borders of the ramus, as seen on the panoramic radiograph. (Figure 1)



Fig 1: Preoperative panoramic radiograph showing unilocular radiolucency with well defined border in lower left ramus region.

Based on the clinical and radiographic findings, a provisional diagnosis of keratocystic odontogenic tumor due to minimal expansion with considerable bony involvement was proposed. The lesion was enucleated under general anaesthesia and the specimen was sent for histopathological examination, which showed stratified squamous epithelium with variable thickness, cubic and ciliated cells in the superficial lining with intraepithelial cysts and mucous cells and cyst capsule of a dense connective tissue with sparse mononuclear infiltrate. The final diagnosis was glandular odontogenic cyst.



Fig 2: Intraoperative image showing cystic enucleation.



Fig 3: Enucleated specimen from the bone cavity

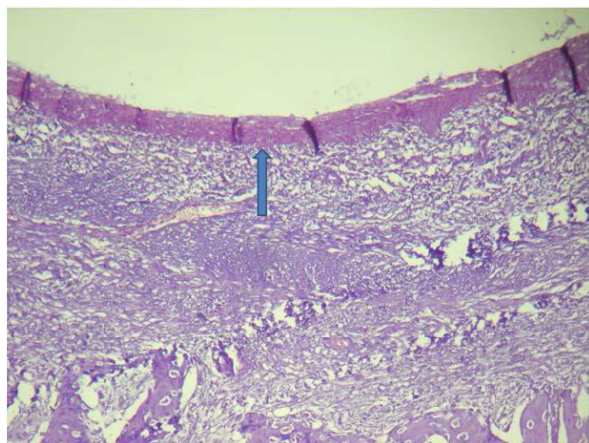


Fig 4: Cyst cavity lined by stratified squamous epithelium (arrow) (H&E 100X).

Postoperative recovery was uneventful. Patient is currently subjected under a long-term follow-up. One year follow-up showed improved bone healing with no signs of relapse.

DISCUSSION

The GOC is a rare entity with relatively low frequency of 0.012–0.03% and prevalence rate 0.17%. Magnusson et al. in their study evaluated 5900 cases of jaw bone cysts and found only seven cases of GOC, i.e., about 0.12%^{2,5}. In another similar study by Van Heerden et al., only 1.3% cases were reported². As there are very few cases of GOC reported in English literature, its incidence, clinical manifestations, radiological findings, histological features, treatment and prognosis has no unified opinion.⁶ Glandular odontogenic cyst usually occurs in anterior mandible, our case differs from the literature since the cyst was located in posterior mandible. Glandular Odontogenic cyst more commonly affects males than females, which also differs with the literature.⁶

GOC recognition based on physical and radiological examinations alone is practically impossible, a fact that the authors of all previous studies harmoniously stress upon. Only histopathological examinations allow for a certain diagnosis of the cyst. Since it occurs as an intraosseous cyst in periapical region, the following lesions such as dentigerous cyst, periapical cyst, odontogenic keratocyst, radicular cyst, myxoma, ameloblastoma, central giant cell lesion, fibrous dysplasia, and central mucoepidermoid carcinoma (MEC) have to be considered in the

differential diagnosis.⁶ Histologically the presented case consisted of certain characteristic features of GOC like stratified squamous epithelium varying in thickness and cuboidal or ciliated epithelium with mucus-producing cells on the surface. Owing to the fact that lateral periodontal cyst (LPC) and CMEC exhibit substantial overlap between histological features, their histopathological differentiation becomes difficult and must be performed with considerable care. In particular the differentiation of low-grade CMEC from GOC is more important and difficult. However, the identification of intra epithelial microcysts or duct-like structures, epithelial whorls, ciliated cells, and superficial cuboidal cells in GOC differentiates it from low-grade CMEC. Likewise, the absence of duct like spaces with mucous cells and ciliated epithelium in the histological sections of LPC, favors the diagnosis of GOC.^{2,5,6}

Radiologically, GOC may appear as intraosseously localized, multilocular or unilocular radiolucent lesion with well-defined borders. In some instances it may also present scalloped, and peripherally osteosclerotic borders, together with root resorption and displacement of the teeth.² In the presented case, radiographic examination shows unilocular with well-defined borders without any root resorption and displacement of teeth. Several studies indicate that GOC is relatively an aggressive lesion with a high tendency for erosion or perforation of cortical plate and chance of recurrence. Hence treatment varies from curettage, enucleation, en bloc and partial osteotomy⁶. For our patient the cyst was enucleated and she is under follow up to monitor recurrence.

CONCLUSION

In conclusion, glandular odontogenic cyst (GOC) being such a rare entity, this paper may enhance the existing knowledge about GOC and may guide readers and clinicians to pay special attention to similar cases when encountered in clinical work.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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