Glandular Odontogenic Cyst: a Case Report and Literature Review

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ABSTRACT

Background: The glandular odontogenic cyst is now a well-known entity comprising < 0.5% of all odontogenic cysts with a recent review tabulating about 200 cases in the English literature. Glandular odontogenic cyst shows epithelial features that simulate salivary gland or glandular differentiation. The importance of glandular odontogenic cyst relates to the fact that it has a high recurrence rate and shares overlapping histologic features with central mucoepidermoid carcinoma. The purpose of this paper is to describe the clinical, radiological, and histopathological features of a case of glandular odontogenic cyst with the course of treatment and 9-years follow-up, followed by a review of the literature.
Methods: A 63-year-old male was referred for further investigation of a mandibular radiolucency observed by his general dental practitioner. The main complaint was a murmuring sensation in the lower jaw right side. Radiological examination revealed a well-defined, unilocular, radiolucent lesion, involving the right mandible with 17 and 68 mm in mediolaterally and anteroposterior dimension, respectively.
Results: A total enucleation of the cystic lesion and surgical extraction of tooth #46, #47 and #48, was performed under local anaesthesia. Histopathologic examination revealed a glandular odontogenic cyst.
Conclusions: Glandular odontogenic cyst shows no pathognomonic clinico-radiographic characteristics, and therefore in many cases it resembles a wide spectrum of lesions. Diagnosis can be extremely difficult due to histopathological similarities with dentigerous cyst, lateral periodontal cyst and central mucoepidermoid carcinoma. Therefore a careful histopathological examination and a long-term follow-up (preferably seven years) are required to rule out recurrences.

Keywords: jaw cyst; oral surgical procedures; review.

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INTRODUCTION

The glandular odontogenic cyst (GOC) is a rare, locally aggressive type of developmental odontogenic cyst. Over the last three decades, several case reports and case series have been reported, and recent publications tabulated about 200 cases in the English literature [1,2]. Thus, GOCs, although rare, is now a well-known entity comprising < 0.5% of all odontogenic cysts [3-5].

The cyst was originally reported by Padayachee et al. [6] who, in 1987, described two cases of unusual odontogenic cysts with features of botryoid odontogenic cyst (lateral periodontal cyst) and central mucoepidermoid carcinoma (CMEC) but with a glandular element, and proposed the term "sialo-odontogenic cyst" [6]. In 1988, eight additional cases were described by Gardner et al. [7] preferring the term "glandular odontogenic cyst" because the cyst epithelium wall was odontogenic and contained mucin elements with absence of salivary tissue [8]. In 1992, the World Health Organization (WHO) included GOCs in the classification as a developmental odontogenic cyst defined as a developmental odontogenic cyst with epithelial features that simulate salivary gland or glandular differentiation [5]. The odontogenic origin has been confirmed immunohistochemically by numerous investigators [9-12]. It is not uncommon to encounter jaw cysts that exhibit some of the features described in GOC. Some microscopic features of GOC are similar to metaplastic changes in dentigerous cysts or lateral periodontal cyst, but also CMEC, which is why caution should be exercised in histopathological diagnosing [5, 13].

We hereby report the course of treatment and long-term outcome of a rare case of glandular odontogenic cyst in a 63-year-old male followed by a review of the literature.

Demographic

GOCs occurs most commonly in middle-aged adults, with highest prevalence at fifth and sixth decades of life [13-16], however, there are also reports in paediatric patients [15]. The cyst shows no gender predilection [13-16]. It has been reported that in South African population GOCs has a strong male predominance which may reflect the difference in gender distribution in different population groups [14,17].

Anatomic location

In 73.2 to 80% of the lesions, the cyst is located in the mandible and 20 to 26.8% in the maxilla, and approximately 60% in the anterior region of the jaws [13,16,18,19]. When the maxilla is affected, GOCs tend to occur in the globulomaxillary relationship [13,20].

Signs and symptoms

Lesions are commonly associated with swelling/expansion in 43.5 to 87% which is the most common presenting complaint [13,16,19,21], although about 75% are asymptomatic [16,21].

Radiographic features

Cortical bone perforation appears in up to 50% of the lesions [13,16,18,19]. The cyst presents as a well-defined unilocular radiolucency in 53.6 to 61.5% of cases, and in 30.4 to 46.4% as a multilocular radiolucency. The margins of the radiolucency are usually well defined with a corticated rim in 94.5% of the lesions [13,16,18,19,21]. Despite the fact that there is a tendency for GOCs to be unilocular, it has been stated that the number of unilocular and multilocular lesions is almost equal and that the radiographic appearance of GOCs varies and is not pathognomonic [19]. There have been reports of GOCs mimicking other cysts; 10.7% of the lesions mimicked dentigerous relationship, lateral periodontal relationship, and cysts in globulomaxillary relationship [13]. Root resorption has been reported in 13.9 to 30% of lesions and tooth displacement in 24.4 to 50% of lesions [16,17,19].

CASE DESCRIPTION AND RESULTS

Clinical features

A 63-year-old male was referred to a private hospital "Kaebekirurgisk Klinik" in Copenhagen, Denmark on November 2013 from his general dental practitioner for further investigation of a mandibular radiolucency observed in a routine intraoral periapical radiograph. The chief complaint was a murmuring sensation in the lower jaw right side. Medical and family history was inconspicuous. On extraoral examination, there was no swelling and no paraesthesia of the lower lip, and the patient did not have any functional problems. Intraoral examination revealed good oral hygiene with no swelling or asymmetries. The gingiva and the mucosa appeared normal. No teeth were tender on

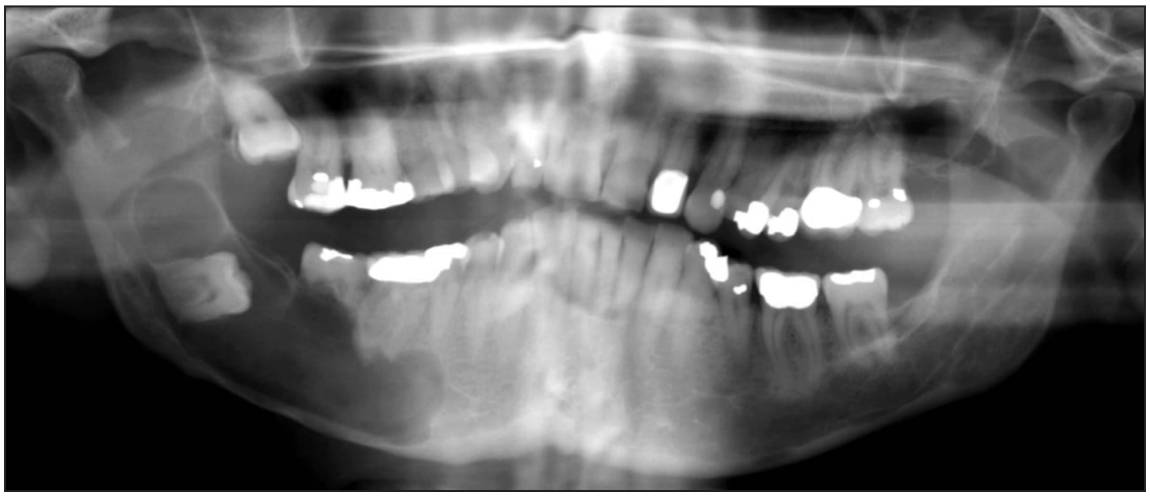

Figure 1. Orthopantomogram showing lesion in the mandible right side.

percussion and the only periodontal pocket > 5 mm was found on tooth #47. No discharge of pus or any inflammatory fluid was present.

Radiologic features

Radiological investigations included panoramic radiograph (OPG) and cone-beam computed tomography (CBCT) scan. OPG revealed a well-defined, unilocular, radiolucent lesion, involving the right mandible extending from the ramus to tooth #45. Displacement of tooth #48 towards the mandibular angle was noted. The root-complex of tooth #47 was resorbed and only a part of the crown remained. The mandibular canal was not identifiable on the OPG (Figure 1).

The CBCT scan showed a unilocular, homogeneous hypodense lesion, extending mediolaterally from the ramus to the inferior border of the mandible and further to the periapical region of tooth #45. The lesion measured 17 mm in mediolaterally dimension inferior for tooth #47 (Figure 2), and 68 mm in anteroposterior dimension (Figure 3). Minor expansion of the medial portion of the mandible was found and thus cortical resorption/thinning of the lingual border inferior to tooth #47 was noted. Inferior displacement of the mandibular canal was also noted with some part only having very thin bone separating the canal from the lesion (Figure 2).

Incision biopsy was performed under local anaesthesia by use of an envelope incision. The marginal incision extended from tooth #46 to #48. A mucoperiosteal flap was elevated and bone was removed to access the lesion. Aspiration of the lesion revealed brown liquid and incisional biopsy was performed. The wound was closed using a resorbable suture (4-0 Vicryl - Ethicon Inc; New Jersey, USA). The result of the biopsy revealed only cyst lining with mild chronic inflammation. Two weeks later enucleation of the cystic lesion and extraction of tooth 46#, #47 and #48 was performed.

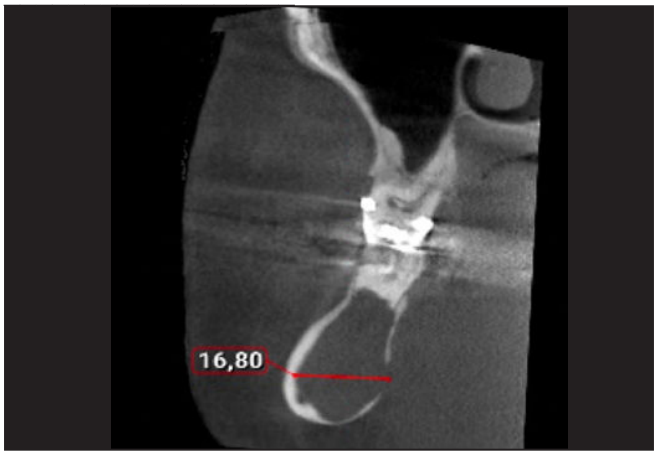

Figure 2. Preoperative cone-beam computed tomography showing measurement of ≈ 17 mm in mediolaterally dimension inferior for tooth #47 and minimal bone separating the mandibular canal from the lesion.

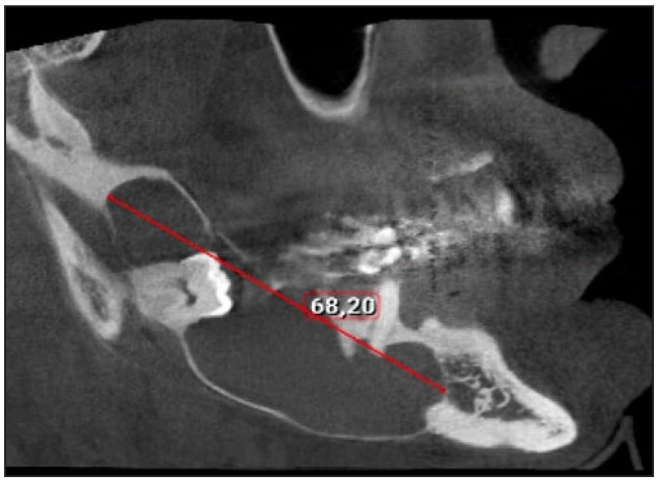

Figure 3. Preoperative cone-beam computed tomography showing measurement of ≈ 68 mm in anteriorposterior dimension of the lesion.

Under local anaesthesia a marginal incision was made with a releasing incision at the mesial aspect of tooth #45. Extraction of tooth #46 and #47 was performed. Tooth #48 was surgically removed following osteotomy of the buccal bone and sectioning of the tooth together with enucleation of the cystic lining. The enucleated specimen was sent for histopathological evaluation. No macroscopic damage to the inferior alveolar nerve was noted. Closure was completed using resorbable suture (4-0 Vicryl - Ethicon Inc; New Jersey, USA). No intra- or postoperative adverse events were noted.

The patient was followed regularly at the interval of one week, two weeks, four weeks, five months and one year. Minor paraesthesia of the right side of lower lip was noted during the first week to five months follow-up, but the patient reported no paraesthesia at one year follow-up; no recurrence has been noted 1 year after surgery (Figure 4). OPG was taken at one-year follow-up before the patient was referred to his general dental practitioner for further follow-up for at least five years. The patient was recently seen for a clinical evaluation and an OPG nine years after initial management was taken (Figure 5).

Histologic features

Histopathologic examination revealed specimens lined with non-keratinised squamous epithelium exhibiting variable structure with a few focal thickenings and a sharp and flat epithelium-connective tissue interface. Intraepithelial glandular/duct-like or microcystic structures lined by cuboidal cells were frequent findings (Figure 6) as were superficial eosinophilic cuboidal cells, in some areas appearing as “hob-nail” cells (Figure 7). Clear vacuolated cells were seen in suprabasal areas of parts of the epithelium (Figure 7) and in some areas mucous goblet cells were seen within the epithelial lining. In some areas slight to moderate chronic inflammation was seen in the underlying connective tissue.

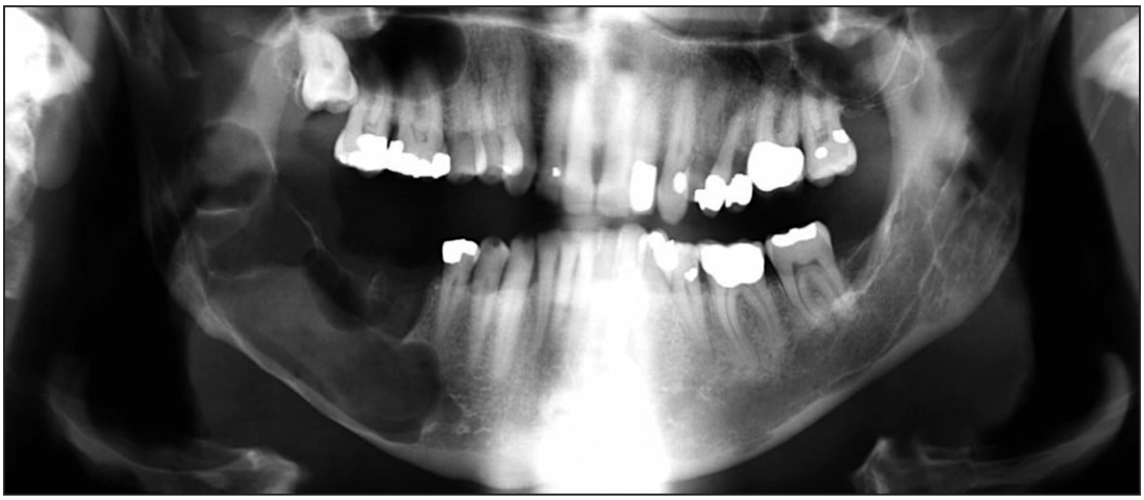

Figure 4. One-year postoperative orthopantomogram showing lesion with healing bone at the base of the mandible.

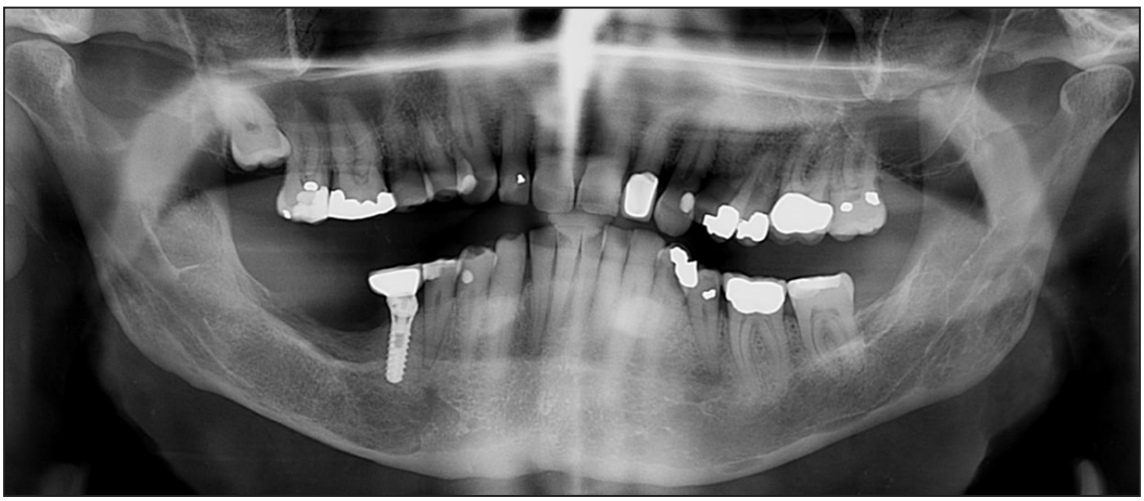

Figure 5. Nine-years postoperative orthopantomogram showing fully healed lesion.

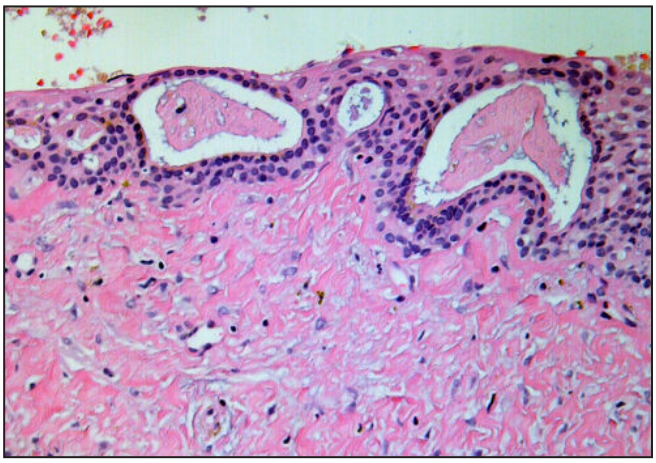

Figure 6. Variable thickness of cyst lining with microcysts lined by eosinophilic cuboidal cells (hematoxylin and eosin stain, original magnification x400).

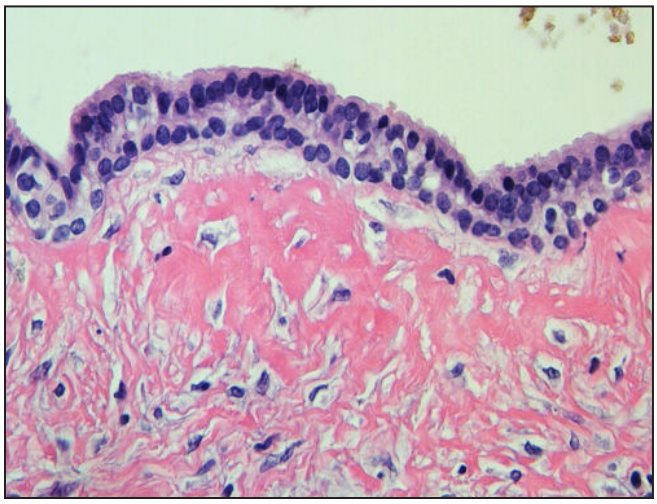

Figure 7. Cyst lining with surface eosinophilic cuboidal cells (hobnail cells) and clear (vacuolated) cells in basal and parabasal layers. Cilia are also noted (hematoxylin and eosin stain, original magnification x400).

DISCUSSION

GOCs is a rare lesion comprising approximately < 0.5% of all odontogenic cyst [3,4]. Recent publications tabulated about 200 cases in the English literature [1,2]. The cyst is rarely suspected on clinical and radiological examination and the radiographic appearance varies and is therefore not pathognomonic [19]. The lesion typically presents radiographically as a unilocular or multilocular radiolucency with a well-defined corticated rim which may have a scalloped border [5,13,16,18,19,21]. Despite the fact that there is a tendency for GOCs to be unilocular, it has been stated that the number of uniocular and multilocular is almost equal [19]. GOC is typically associated with the roots of multiple teeth, and tooth displacement or tooth resorption is common [5,8]. Tooth displacement is more commonly seen than root resorption [17]. The aggressive potential of GOC is often seen in either cortical thinning or perforation [12]. GOCs can mimic other cysts; dentigerous cyst and lateral periodontal cyst [13]. Therefore, the recognition of this cyst based on clinical and radiological examination is impossible since the radiograph appearance of GOCs varies and is not pathognomonic [19,21]. Association with an impacted tooth is extremely rare, and extreme caution should be exercised in diagnosing GOC when in a dentigerous relationship [5].

The microscopic features of GOC have been well documented, and WHO now includes a definition of this lesion and lists numerous characteristic microscopic features of GOC [5]. The histogenesis of GOC remains uncertain. It was initially proposed to develop from intraosseous salivary gland tissue [6]. GOC is now believed to be a developmental odontogenic cyst that arises from remnants of the dental lamina [5].

Kaplan et al. [12,18] were the first to describe the number of microscopic features that are necessary for diagnosis of GOC [12,18]. The group listed major and minor microscopic criteria for GOC based on the frequency of each feature in reported cases from the literature [12,18]. Based on their analysis, it was suggested that the presence of each of the major criteria must be present for diagnosis and the presence of minor criteria supports the diagnosis but are not mandatory (Table 1). Practical applicability of major and minor microscopic criteria may encounter some difficulties [13]. Fowler et al. [13] also investigated microscopic features that were necessary for diagnosis in problematic cases of GOC.

Table 1. The major and minor criteria listed by Kaplan et al. [12]

Major criteria	Minor criteria
1. Squamous epithelial lining, with a flat interface with the connective tissue wall, lacking basal palisading 2. Epithelium exhibiting variations in thickness along the cystic lining with or without epithelial ‘‘spheres’’ or ‘‘whorls’’ or focal luminal proliferation 3. Cuboidal eosinophilic cells or ‘‘hobnail’’ cells 4. Mucous (goblet) pools, with or without crypts lined by mucous-producing cells 5. Intraepithelial glandular, microcystic, or duct-like structures	1. Papillary proliferation of the lining epithelium 2. Ciliated cells 3. Multicystic or multiluminal architecture 4. Clear or vacuolated cells in the basal or spinous layers

The authors concluded that not all of Kaplan et al. [12,18] major criteria need to be present for diagnosis, but more likely a combination of specific microscopic features [13]. Therefore, diagnosis is not necessarily corresponding with their major and minor criteria [13]. Fowler et al. [13] listed ten histologic parameters to distinguish GOCs from other lesions with a similar histopathological appearance (GOC mimickers). The presence or absence of the ten histologic parameters was based and adapted from previously reported features of GOC (Table 2) [13]. It was suggested, following statistical analysis that a reliable diagnosis of GOC can be made when at least 7 of 10 following criteria are present [5,13]. Fowler et al. [13] concluded that eosinophilic cuboidal cells (hobnail cells) are necessary for diagnosis but are not pathognomonic of GOC in the absence of other microscopic parameters. Moreover, the presence of intraepithelial microcysts, clear (vacuolated) cells, epithelial spheres, variable thickness, and multiple compartments are superior in distinguishing GOCs from GOC mimickers [13].

GOCs also shares overlapping histologic features with CMEC, a rare malignant intraosseous neoplasm. The relationship of GOC and CMEC has been previously discussed by several investigators [13,22-26]. Some authors speculate that GOC and CMEC represent a biological spectrum of the same disease [13,27]. This speculation is supported by the aggressive radiologic presentation and high recurrence rate often seen in GOCs [13,27,28].

Fowler et al. [13] reported three cases in which islands resembling CMEC were noted within the cyst wall. In two of these cases, the CMEC-like islands invaded bone which otherwise were classic GOCs microscopically. It has been suggested that these CMEC-like islands within the cyst wall most likely have no clinical significance [13]. Nevertheless, it may propose the possibility that GOC and CMEC are related or that CMEC could develop from a pre-existing GOC [5,13]. It has also been proposed that many cases previously diagnosed as CMEC could have been GOC because of similar histological overlap [21].

This issue raises a diagnostic dilemma because the distinction between these lesions is critical for treatment planning and patient prognosis. Recently it has been discovered that most mucoepidermoid carcinoma (MEC) has a t(11:19)(q21:p13) translocation which results in fusion of MECT1:MAML2 gene [29-31]. This translocation has also been reported in CMEC [32-34]. Bishop et al. [34] partially resolved this controversy by establishing that GOCs lack the MAML2 gene rearrangements that are often seen in CMECs, though the number of cases tested was small. However, later investigations found that these rearrangements can be negative in approximately 32% of CMECs [27,35].

Table 2. Histological parameters and description listed by Fowler and colleagues [13]

Histological parameters	Histological description
Surface eosinophilic cuboidal cells	Also called ‘‘hobnail cells’’. These cells are present on the surface of the cyst lining and resemble cuboidal cells of the reduced enamel epithelium that lines dental follicles and dentigerous cysts.
Intraepithelial microcysts or duct-like spaces lined by a single layer of cuboidal to columnar cells similar to surface cells	Sometimes the microcysts are lined by mucous goblet cells. These microcysts may contain mucous pools, eosinophilic material, or may appear to be empty. In areas, the microcysts may open onto the surface of the lining epithelium.
Apocrine snouting of hobnail cells	Sometimes the hobnail cells demonstrate ‘‘pinching off’’ of the surface similar to decapitation secretion seen in cells that line apocrine gland ducts.
Clear or vacuolated cells	These cells contain clear cytoplasm and may be present in the basal and/or parabasal layers. The clear cytoplasm is due to glycogen in some cases. In areas of attenuated cyst lining, clear basal cells may be directly subjacent to the surface eosinophilic cuboidal cells.
Variable thickness of the cyst lining	This was recorded as positive only if marked variability in the thickness of the cyst lining was present
Papillary projections or ‘‘tufting’’ into the cyst lumen	These papillary projections sometimes are formed by several microcysts opening onto the surface of the cyst lining, but may also be formed independent of microcysts.
Mucous goblet cells	These cells may be present singly or in small clusters on the surface or within the cyst lining. They may also line microcysts.
Epithelial spheres or plaque-like thickenings	These are identical to those seen in lateral periodontal cysts or botryoid odontogenic cysts. Sometimes the epithelium in these plaques exhibits swirling or spherule formation.
Multiple compartments	Multiple cystic spaces similar to those seen in botryoid odontogenic cysts.
Cilia	These are true cilia on the surface of eosinophilic cuboidal cells, and are distinct from apocrine snouting.

Furthermore, MAML2 rearrangements those were not present in primary GOCs where subsequently detected in recurrent GOCs or apparent CMECs arising from GOCs [36,37]. Reddy et al. [27] argued that MAML2 rearrangement inconsistencies have made molecular analysis unreliable in differentiating between these two entities.

Pires et al. [9] investigated expression of cytokeratin 18 and 19 (CKs 18 and 19) in GOC and CMEC. It has been suggested that CKs 18 and 19 could be useful in differentiating between the two entities. The group concluded that all CMEC expressed CKs 18 whereas GOCs expressed CKs 19 consisting with previous studies [10,11,21]. Ultimately, histologic features must be correlated with clinical and radiologic information to render an accurate diagnosis. Reddy et al. [27] emphasised that location and clinical signs are important distinguishing parameters between GOCs and CMECs. In contrast to the typical presentation of GOCs, CMEC usually present as painful swellings in the mandibular posterior body-ramus complex, often in association with impacted teeth [32].

Enucleation, curettage and marsupialization prior to enucleation are the most common treatment for GOC but is associated with a recurrence rate of 21.6 to 50% [13,16,18]. Fowler et al. [13] reported a 50% recurrence rate for the lesions with an average length of follow-up of 8.75 years. Kaplan et al. [28] reported a lower recurrence rate of 29.2%, within 0.5 to 7 years, with a mean follow-up of 2.9 years. Chrcanovic et al. [16] reported a recurrence rate of 21.6%, within 0.1 to 20 years, with a mean follow-up of 4.5 years.

Most cases of GOCs have been treated by conservative procedures such as enucleation or curettage; however, GOC shows a high recurrence rate, and the risk of a recurrence increases with size, multilocular appearance and comprised cortical integrity [16,17]. Marsupialization and decompression may be performed for larger lesions to promote shrinkage prior to enucleation or curettage [28]. Lesions have been reported to recur after three years [11], eight years [13] and ten years [38]. Long-term follow-up is advocated and some authors suggest at least 3-year follow-up, and preferably 7 years for GOCs [28]. Because of its local aggressive behaviour and tendency for recurrence, some authors have advocated block resection, particularly for larger or multilocular lesions [28,38]. Thor et al. [38] did a follow-up of a GOC for 13 years. The authors treated recurrence of the same cyst 11 times with conservative surgery during the first ten years of follow-up [38]. Lastly, a block resection of the GOC was performed, resulted in no subsequent recurrences [38]. The former supports the findings of Kaplan et al. [28] which showed that recurrence was associated with conservative surgery such as enucleation or curettage and none of the patients treated by peripheral ostectomy or marginal resection had a recurrence.

The case presented illustrates successful conservative approach and enucleation of a large mandibular lesion with 9 year follow-up. Long-term post-treatment follow-up of large lesions is recommended because of the slow nature of bone healing.

CONCLUSIONS

Glandular odontogenic cyst is a rare odontogenic cyst, with less than 200 cases reported world-wide till date. Though rare, the cyst is now relatively well known among oral and head and neck pathologists, and World Health Organization now includes a definition and numerous characteristics of glandular odontogenic cyst.

Glandular odontogenic cyst shows no pathognomonic clinico-radiographic characteristics, and therefore in many cases it resembles a wide spectrum of lesions. Diagnosis can be extremely difficult due to histopathological similarities with dentigerous cyst, lateral periodontal cyst and central mucoepidermoid carcinoma, and therefore a careful histopathological examination and a long-term follow-up - preferably seven years - are required to rule out recurrences.

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