Glandular odontogenic cyst of the mandible: A case report and literature review

Candice Jacinta Antao[1], Ajit Dattatray Dinkar[2], Manisha Maruti Khorate[3], Karla Carvalho[4]

From [1]Ex-Post Graduate Student, [2]Ex-Professor and Head, [3]Professor, Department of Oral Medicine and Radiology, [4]Lecturer, Department of Oral and Maxillofacial Pathology, Goa Dental College and Hospital, Bambolim, Goa, India

Correspondence to: Dr. Candice Jacinta Antao, Department of Oral Medicine and Radiology, Goa Dental College and Hospital, Bambolim, Goa, India. E-mail: candice.antao@gmail.com

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ABSTRACT

Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst that has gained special attention primarily due to its aggressive potential and the microscopic features, it shares with central mucoepidermoid carcinoma of the jaws. This relatively new entity has a prevalence rate of 0.17% and lacks pathognomonic clinicoradiographic features, making the correct diagnosis a major challenge. This report describes a case of GOC presenting as a solitary osteodestructive lesion in the anterior mandible of a 54-year-old male. Cone-beam computed tomography (CBCT) images revealed a multilocular radiolucent lesion with bicortical expansion, cortical thinning, and perforation, indicating an aggressive potential. Histopathology revealed pseudoglandular epithelium with daughter cysts within the stroma, suggesting an invasive nature. The patient was diagnosed and treated in our institution. In view of the high probability of recurrence of GOC, CBCT evaluation and histopathological examination are essential. An aggressive treatment approach is recommended followed by long-term follow-up.

Key words: *Glandular odontogenic, Mucous cells, Sialo-odontogenic*

Glandular odontogenic cyst (GOC) is a relatively rare cystic lesion that accounts for 0.012–1.3% of all the jaw cysts [1,2] and has a prevalence rate of 0.17% [3]. This relatively new entity has attracted the attention of clinicians and pathologists primarily due to its significant predilection toward localized aggressiveness and the microscopic features, it shares with lateral periodontal cyst, botryoid odontogenic cyst, and central mucoepidermoid carcinoma of the jaws [3].

Cases of GOC with an aggressive potential, presenting with cortical perforation and a relatively high rate of recurrence, have been reported [3,4]. Moreover, the clinicoradiographic features of this cyst are non-pathognomonic making its recognition practically impossible [2]. Correct diagnosis is a major challenge and is of extreme clinical importance.

This report describes a case of GOC in the anterior mandible of a 54-year-old male, with CBCT evidence of cortical perforation, indicating an aggressive potential.

CASE REPORT

A 54-year-old male patient reported to our institution with the chief complaint of a swelling in the lower jaw for 5 months. The swelling was initially small in size and gradually increased to its present size. The swelling was associated with pain in the mandibular anterior teeth. The pain was mild in intensity, intermittent in duration, throbbing in nature, and non-radiating and did not have any aggravating or relieving factors. The patient underwent root canal treatment for the same 1 month ago at a local dental clinic, but there was a limited resolution. The patient denied any history of trauma to the area and there was no relevant medical history.

On examination, there was no evidence of an extraoral swelling. However, intraorally, there was a mild swelling in the anterior mandible causing partial obliteration of the labial vestibule. The swelling had diffuse margins, extending from the left canine up to the right premolar region, measuring approximately 2.5 × 1 cm in dimensions (Fig. 1a). On palpation, the swelling was tender, firm in consistency, and non-pulsatile. The associated teeth ($^{\#}$33,$^{\#}$32,$^{\#}$31,$^{\#}$41,$^{\#}$42,$^{\#}$43,$^{\#}$44, and $^{\#}$45) were non-tender on percussion and non-mobile. The overlying skin was intact and afebrile, with no evidence of ulceration or discharge.

Radiographic evaluation with a panoramic radiography and cone-beam computed tomography (CBCT) revealed a single, well-defined multilocular radiolucent lesion with corticated margins extending across the midline (Fig. 1b). Three-dimensional reconstructed view of the lesion is shown in Fig. 1c. The lesion appeared to involve the periapical region of endodontically treated mandibular teeth. Superiorly, the lesion involved the crestal bone in the inter-radicular region of the anterior teeth and inferiorly, the lesion was in close approximation to the right mental foramen. Expansion of the labial and lingual cortical plates throughout the extent of the radiolucency, with cortical thinning and perforation,

was prominent. Internal structure displayed discrete radiopaque structures at the base, suggestive of extruded root canal obturating material. Root resorption of the left lateral incisor and right canine and premolars was noted (Fig. 2a and b).

Based on the history, clinical, and radiographic findings, a provisional diagnosis of a radicular cyst was given, with a differential diagnosis of keratocystic odontogenic tumor and central giant cell granuloma. Incisional biopsy was conducted under local anesthesia and aspiration yielded 0.5 cc of a serous yellow-colored fluid. Protein estimation of the aspirate was 5.6 g/dl. The specimen was sent for histopathological evaluation. Microscopic examination revealed a pseudoglandular stratified squamous non-keratinized cystic epithelium (3–4 cell layers thick) with a mild arcading pattern lining crypt-like areas of mucous pools and showing luminal proliferations. Intraepithelial areas of eosinophilic coagulum and clear cells were seen. Within the stroma, another cystic area lined by pseudostratified columnar epithelium with a scattering of clear cells and few goblet cells were seen. This multicystic feature indicates the presence of daughter cysts. Connective tissue stroma also showed multiple daughter cysts and deeper stroma showed multinucleated giant cells and chronic inflammatory cell infiltrate (Fig. 2c). Based on the overall features, a final diagnosis of GOC was given.

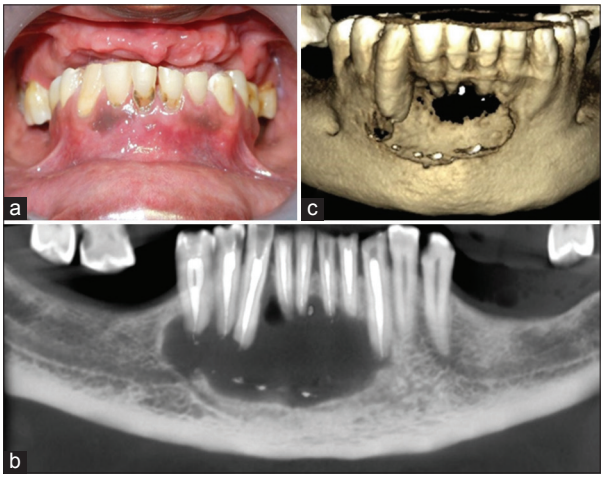

Figure 1: (a) Intraoral photograph showing a mild swelling in the anterior mandible. (b) Cone-beam computed tomography panoramic view showing a solitary multilocular radiolucent lesion with corticated margins, measuring 34.8 × 12.3 × 17.7 mm in dimension, extending across the midline from the mandibular left canine up to the mandibular right second premolar. (c) Three-dimensional reconstructed view of the lesion

Considering the aggressive behavior of the cyst, a less conservative treatment approach was planned. Under general anesthesia, a crevicular incision was given around the necks of the mandibular anterior teeth and subperiosteal dissection was done to expose the cystic lesion. The cystic lesion was excised and curetted, and total extraction of the mandibular teeth was carried out. Peripheral ostectomy was done, the bone around the cystic margins was removed and bony prominences were rounded up. The excised specimen was analyzed histopathologically, and the report was consistent with the incisional biopsy diagnosis of GOC. Post-operative recovery was uneventful (Fig. 3). The patient has been under regular follow-up for the past 3 years, with no evidence of recurrence (Fig. 4).

DISCUSSION

In 1987, Padayachee and van Wyk reported two cases that appeared similar to botryoid odontogenic cyst. They coined the

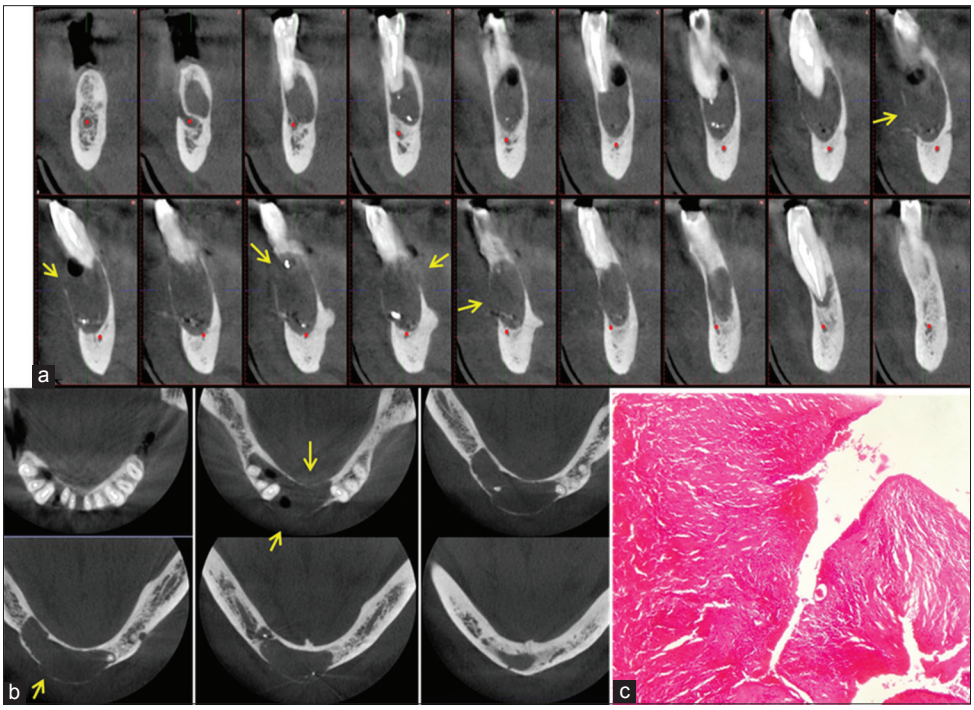

Figure 2: (a) Cone-beam computed tomography sagittal sections showing cortical thinning, perforation (arrows), and the close approximation of the lesion to the right mental foramen. (b) Cone-beam computed tomography axial sections showing bicortical expansion and cortical perforation (arrows). (c) Photomicrograph (H and E at 40x magnification) showing pseudoglandular stratified squamous non-keratinized cystic epithelium with crypt-like areas with mucous pools

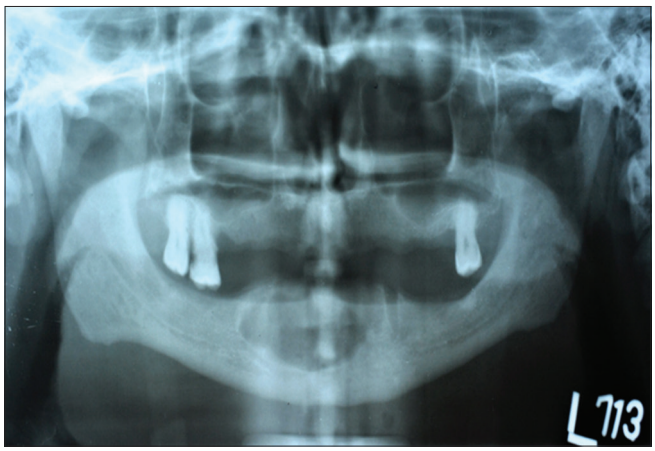

Figure 3: Post-operative panoramic radiograph showing the surgical bony defect following cyst excision and peripheral ostectomy

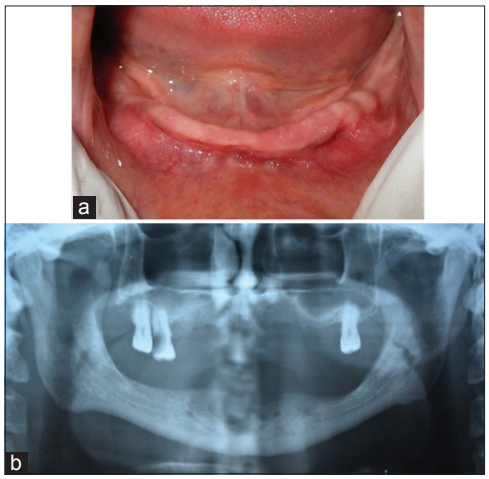

Figure 4: Images at 1-year follow-up (a) Intraoral view and (b) panoramic radiograph showing bone remodeling and no evidence of recurrence

term "Sialo-odontogenic cyst" due to the presence of a glandular element with mucous cells and pools of mucin in the epithelial lining. These mucous pools were lined by eosinophilic cuboidal cells which resembled salivary gland ducts [2,3]. In 1988, Gardner first proposed the term "GOC." He proposed the cyst to be a rare entity of odontogenic origin, as the cystic lining was composed of odontogenic epithelium and mucin elements, without evidence of salivary tissue involvement [2]. Several names have been proposed for this lesion such as a sialo-odontogenic cyst, GOC, and mucoepidermoid odontogenic cyst [5] due to the histomorphologic resemblance of GOC with the lateral periodontal cyst, botryoid odontogenic cyst, and central mucoepidermoid carcinoma of the jaws [6]. In 1992, GOC was accepted as a distinct pathological entity by the World Health Organization reflecting the theory that these cysts were most likely of odontogenic origin and the term "GOC" was included in "Histological Typing of Odontogenic Tumors" and is still used until this day [1,7,8]. Although the issue of the origin of GOC has not been completely resolved, most of the existing evidence supports an odontogenic origin rather than origin from intraosseous salivary gland tissue (sialogenic origin) [5,6]. Immunohistochemical studies have also attempted to clarify the lesion's origin [4,8]. Besides, several cases of hybrid lesions of GOC with other odontogenic tumors are also an indicator of odontogenic origin, whereas the lack or minimal of expression of markers do not support a sialogenic origin [4].

Krishnamurthy *et al*. detected only 0.012% of the cases of GOC in the oral cavity [3]. While Luczak *et al.* [1] reported that 1.3% all the cysts of the oral cavity were determined by Van Heerden as cases fulfilling the criteria of GOC microscopically. A review of literature shows that there are <200 reported cases of GOC till date [2,6,9].

GOC occurs commonly after the 4th decade of life as a slow-growing swelling with a prevalence rate of 0.17% [2,3,7]. In the survey conducted by Chrcanovic and Gomez, which is stated to be the largest survey on GOC in the literature, 58 publications reporting 169 GOCs were studied and they observed a slight male predilection with a male-to-female ratio of 1.15:1 (males: 90% and females: 78%). The mean age ± SD of the patients was found to be 48.1 ± 13.1 years (min-max: 12–90 years). Literature suggests the anterior region of the mandible to be the most commonly affected area, followed by the anterior region of the maxilla. However, reports of bilateral occurrence and posterior location are also reported [2,3,7]. Occasionally, the lesion may present with symptoms of pain due to compression of a neurovascular bundle or secondary infection [10]. Figueiredo *et al*. reported that in the initial stages, GOC may grow asymptomatically within the jaws and cause swelling with cortical expansion only at a later date [2]. The present case displays most of the features described in the literature.

Radiographic appearance of GOC is quite variable and is non-pathognomonic, making its recognition quite difficult. GOC may appear as a unilocular or multilocular radiolucent lesion with well-defined peripheral osteosclerotic borders. Occasionally, scalloped border, root resorption, and displacement of the teeth may be evident [4,7]. As the radiographic features of GOC are non-specific, GOC can be confused with lesions such as a radicular cyst, keratocystic odontogenic tumor, ameloblastoma, and central giant cell granuloma. Careful radiographic evaluation is essential and CT or CBCT is recommended, as it provides accurate information about locularity of the lesion, cortical integrity, degree of expansion, and involvement of the adjacent soft tissue [7]. CBCT images of the present case revealed cortical perforation indicating an aggressive potential. In the review conducted by Kaplan *et al*., 85.4% of the GOCs encroached on the cortical plates [4].

Clinicoradiological examination does not guarantee a reliable diagnosis due to lack of characteristic features of GOC. Histopathological examination, however, can aid in achieving the correct diagnosis [1]. According to Kaplan *et al*., GOC exhibits major and minor histopathological criteria. They suggested that the presence of the major histopathological criteria is mandatory for diagnosing GOC, while the minor criteria can further support the diagnosis [4]. The major criteria include a lining of non-keratinized squamous epithelium with a flat interface, which may show the presence of focal luminal proliferation

or "spheres" or "whorls" with the absence of basal palisading, cuboidal eosinophilic ("hobnail") cells, and mucous (goblet) cells with intraepithelial mucous pools, with the presence of glandular or pseudoglandular structures. Minor criteria include papillary proliferation, presence of ciliated or clear (vacuolated) cells in the epithelium, and multicystic architecture [4]. Fowler *et al*. recorded 10 microscopic parameters for each of the 67 cases they analyzed. The presence of seven or more parameters was considered as highly predictive of a diagnosis of GOC, while the presence of five or less parameters was highly predictive of a non-GOC diagnosis or "GOC mimickers" [8]. The present case showed most of the characteristic histopathological features of GOC described above, thus fulfilling the criteria presented by Kaplan *et al*. and Fowler *et al*.

The low prevalence of GOC in the literature is believed to be not only due to its rarity but also due to overlapping of histological features, leading to misinterpretation [8]. GOC tends to mimic a wide clinicopathologic spectrum ranging from odontogenic cysts to destructive malignant neoplasms such as central low-grade mucoepidermoid carcinoma [3]. A detailed microscopic examination is essential in arriving at the final diagnosis.

There are articles, wherein GOC has been mismanaged by root canal treatment on a pre-assumption that it was a periapical cyst [11]. Dental history of our patient suggests that our patient too suffered a similar misdiagnosis and underwent root canal treatment for all the anterior teeth. Conducting an incisional biopsy followed by thorough microscopic evaluation can aid in preventing mismanagement.

The high recurrence rate of GOC is attributed to several factors. The aggressive nature associated with the multilocularity of the cyst, presence of daughter cysts, and the tendency of separation of the thin epithelium from the underlying capsule during surgical enucleation are directly responsible for a higher tendency for recurrence [7]. The size of the lesion is another feature correlated with recurrence probability. 85.6% of the large lesions recur in contrast to 14.4% of the small lesions. Hence, an aggressive management of large lesions and long-term follow-up has been recommended [4,10]. Another reason for relapse is the conservative treatment approach [7]. The presence of microcysts/daughter cysts and their invasive potential makes the complete surgical removal difficult [3]. Multicystic lesions treated conservatively by curettage or enucleation, demonstrated, an increased recurrence rate of 55% with an average of 4.9 years [7]. The present case presented with features such as large size, multilocular shape, cortical perforation, and multiple daughter cysts, all indicating a strong potential for recurrence.

The treatment of GOC is highly controversial and ranges from curettage and enucleation to *en bloc* resection and partial ostectomy [2,7]. Depending on the size and nature of the lesion, authors have suggested enucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw resection for multilocular cases [10]. The present case underwent peripheral ostectomy due to the radiopathological signs of aggressive behavior. The patient has been under regular follow-up visits till date, with no evidence of recurrence. The purpose of reporting this case was to contribute to the previously presented data in the world literature and add to the existing knowledge about this uncommon cyst which still has an uncertain nature.

CONCLUSION

GOC is a rare cyst of odontogenic origin, displaying non-specific clinicoradiographic characteristics. It is necessary to raise the level of awareness of this uncommon lesion among dental practitioners. It resembles a wide spectrum of lesions creating a major diagnostic challenge. CBCT evaluation is essential and can aid in treatment planning, as this lesion demonstrates cortical changes due to its aggressive behavior. The invasive potential and high probability of the recurrence of GOC necessitate a careful histopathologic examination and an aggressive treatment approach followed by a long-term follow-up.

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