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CASE REPORT

Glandular odontogenic cyst involving the posterior part of maxillary sinus, a rare entity

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Abstract The Glandular Odontogenic Cyst (GOC) was first coined by Gardner et al. [2], in 1988 as an odontogenic origin, is a rare developmental lesion considered a distinct entity because of its uncommon clinical and histopathological characteristics. This lesion can involve either jaws, but the anterior region of the mandible is the most affected area. It strikes distinct age groups, with an average patient age of 50 years. Radiographically, GOC does not display specific or pathognomonic features. It may present as a multilocular or unilocular radiolucencies. The cyst has an aggressive nature and high tendency of recurrence, so long-term follow-up should be carried out. The treatment is controversial, varying from conservative methods to block excision. It is believed that the low prevalence of GOC in the literature is because of not only its rarity, but principally to the fact that its main characteristics are also found in other pathological entities, thereby generating controversial diagnoses. The aim of this paper is to present a rare case of Glandular Odontogenic Cyst (GOC), which is uncommon in the posterior maxilla, that mimicks the lateral odontogenic cyst/botroid odontogenic cyst/Central Muco-epidermoid carcinoma. Owing to its tendency to recur, the lesion needs careful and meticulous planning for its surgical removal.

Keywords The glandular odontogenic cyst · Posterior part of maxillary sinus · Surgical procedure

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Introduction

Glandular Odontogenic Cyst (GOC) is relatively a rare entity, that was first described by Padayachee and Van Wyk [1] in 1987 under the term 'sialo-odontogenic cyst' attributing its possible etiology to salivary glands. The term 'Glandular odontogenic cyst' (GOC) was first coined by Gardner et al. [2], in 1988 as an odontogenic origin. Currently recognized by the World Health Organization [3] in 1992. Its frequency rate is only 0.012% to 1.3% of all jaw cysts. GOC has gained special attention by clinicians and pathologists due primarily to two reasons. First, is the histomorphologic features between GOC, Lateral Periodontal Cyst (LPC), Botroid Odontogenic Cyst (BOC), and Central Mucoepidermoid Carcinoma (CMEC) of the jaws [1,4,5,6] second, is the potential aggressive behaviour of the lesion. Radiographically, GOC does not display specific or pathognomonic features. It may present as a multilocular or unilocular radiolucencies [7]. The cyst has an aggressive nature and high tendency of recurrence, so long-term follow-up should be carried out. The treatment is controversial, varying from conservative methods to block excision. The recurrence rates appear to be correlated with the conservative approach [8,9].

Because of the lack of sufficient clinicopathologic information to elucidate the natural history of this enigmatic cyst, reporting of new cases should be encouraged. The following report describes a case of glandular odontogenic cyst in posterior part of the maxillary sinus which is an extremely rare site and this location is never mentioned in the scientific literature.

Case report

A 60-year-old female patient reported to our department of Oral and Maxillofacial Surgery, for the evaluation and management of an asymptomatic, soft, intra-oral left upper swelling of posterior part of the maxilla. She reported that it had been present for the past 6 months, but asymptomatic, and only became painful 3 months earlier following an infection in the upper left second and third molar tooth which were mobile. The teeth maxillary left second and third molars were extracted by a general dental practitioner. However, there was no apparent resolution of the swelling and gradual enlargement was experienced.

Extra-orally there was no significant facial asymmetry (Fig. 1). Intra-oral examination showed a moderate left upper alveolar soft swelling (Fig. 2). There was

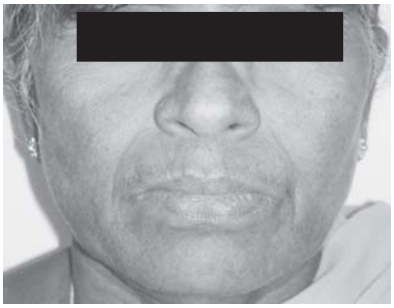

Fig. 1 Patient at initial examination. No significant facial asymmetry

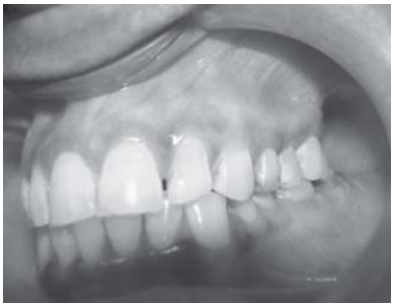

Fig. 2 Intra-oral left buccal region showing vestibular obliteration

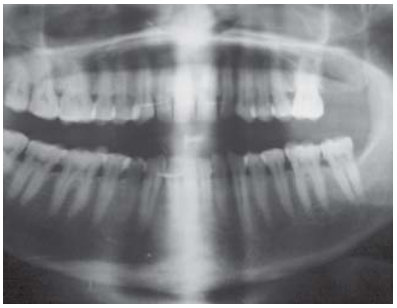

Fig. 3 Orthopantomogram shows haziness in left posterior part of maxillary sinus

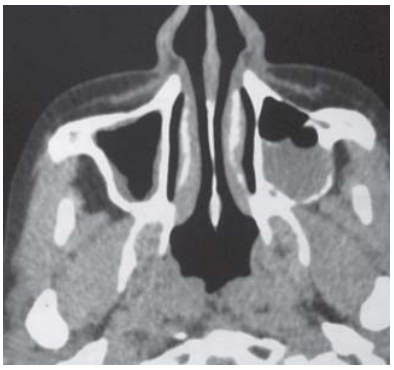

Fig. 4 Axial CT scan showing lesion involving posterior part of the maxillary sinus

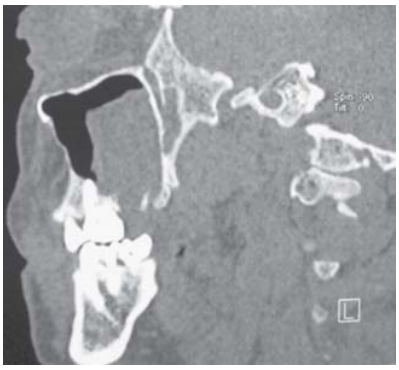

Fig. 5 Sagittal view CT scan showing lesion involving posterior part of the maxillary sinus

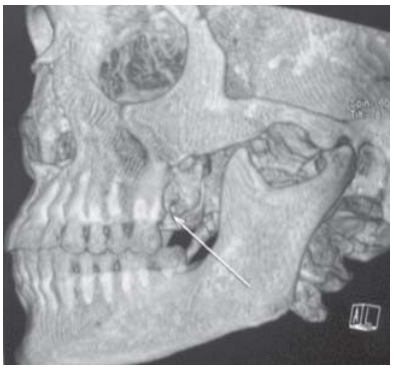

Fig. 6 3D image CT scan shows bony involvement of the buccal cortex in second molar region

no sensory loss, obvious signs of infection, and limitation of mouth opening, her nasal mucosa was intact. On palpation, an egg-shell mass on the labiobuccal aspect of the left posterior maxilla, extending from the left maxillary canine to the left maxillary second molar region. The gingival and alveolar mucosa were of normal appearance, but palpation of the apical region of first and second molar revealed focal dehiscence of bone. Orthopantomogram shows unilocular radiolucency in the left upper first, second and third molar region involving the posterior part of the maxillary sinus (Fig. 3). Computed tomography of the middle third of the face showed a cystic lesion with dense content, measuring about 2cm x 2cm size, located deep in the left maxillary sinus next to the posterior lateral wall of the left maxillary sinus (Fig. 4). Sagittal view shows the involvement of posterior part of maxillary sinus without any perforations in posterior border of maxilla (Fig. 5). Three dimensional view shows perforations of the cortical bone on lateral aspect of the maxilla in the second molar region (Fig. 6). The radiographic differential diagnosis included an odontogenic keratocyst, ameloblastoma, odontogenic myxoma, central giant cell granuloma, and Botroid odontogenic cyst; and last on our differential diagnosis list was GOC.

An incisional biopsy was carried out under local anesthesia. Aspiration yielded 0.5 cc of brownish - red colored fluid. A reflection of a full-thickness mucoperiosteal flap demonstrated perforated cortical bone covering the lesion. Grossly, the specimen consisted of irregular fragments of soft and hard tissue measuring from 0.3 to 1 cm in dimension. Histologic examination showed a non-keratinized cyst lined by thin cuboidal epithelium that exhibited cilia in parts and focal epithelial thickenings (plaques). The lining projected into the cyst's lumen in areas. Glandular epithelium was also identified within selective areas, including a few of the thickened areas. Mucicarmine positive mucous cells and clear cells were well identified within the glandular areas and also replaced part of the uniform lining epithelium (Fig. 7). Biopsy report revealed 'glandular odontogenic cyst of the posterior maxillary sinus'. Owing to the clinical, radiological and pathological findings a final diagnosis of 'Glandular odontogenic cyst of the posterior maxillary sinus' was made. Considering the age of the patient, clinically slow growing nature and being present mostly within the confines of the antrum it was decided to treat the case by enucleation followed by chemical application of carnoy's solution of those areas which were exposed to soft tissues.

The surgery was performed under general anesthesia. Mucogingival incision (caldwel luc's procedure) placed from left upper lateral incisor to the left first molar (Fig. 8). Mucoperiosteal flap raised, lesion was exposed (Fig. 9) and complete enucleation of the lesion including the sinus lining was done (Fig. 10). Carnoy's solution applied in all those areas where soft tissue involvement was present. A nasal antral window is created by trans nasal antrostomy into the inferior meatus. Iodoform gauze impregnated with topical antibiotic is passed through the antral window and pulled to the nares. The antrum was packed lightly for infection and hemostatic control with the remaining gauze. Then mucogingival incision was closed with 4-0 vicryl (Fig. 11). Postoperative recovery was uneventful. Excisional biopsy was sent to histology and pathology for analysis. The pathological diagnosis was 'glandular odontogenic cyst'.

The patient returned for 1-month follow-up, where she had no gross extra-oral swelling, and intra-orally the wound had healed well with no evidence of infection. Regular follow-up was done 3rd month, 6th month.

Fig. 7 Histopathological slide shows a non-keratinized cyst lined by thin cuboidal epithelium that exhibited cilia. The lining projected into the cyst's lumen in areas. Glandular epithelium was also identified within selective areas

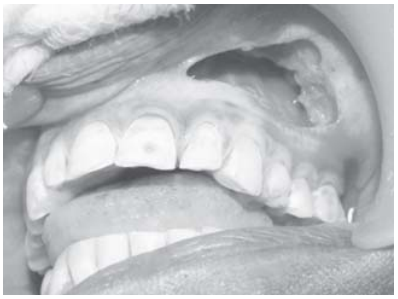

Fig. 8 Shows mucogingival incision from left upper lateral incisor upto the first molar of the same side. Mucoperiosteal flap raised

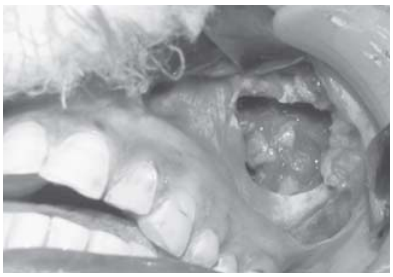

Fig. 9 Shows lesion within the maxillary sinus

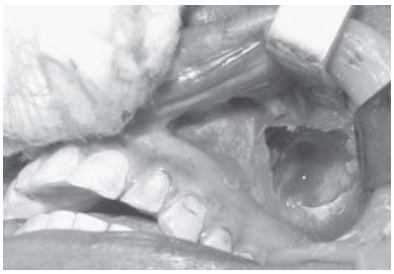

Fig. 10 Shows complete enucleation of the cystic and sinus lining

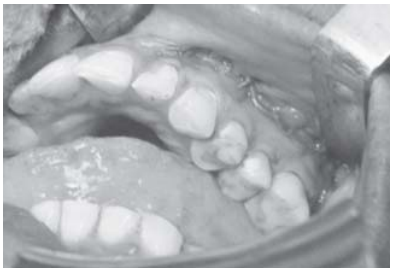

Fig. 11 Shows after enucleation, incision was closed with 4-0 vicryl suture

Discussion

The few cases of GOC reported in the literature has no reliable information regarding predilection for gender, age or most common localisation. Review has also verified equal gender predilection as shown by Hussain K, Edmondson HD, Browne RM [13] when compared with others that favoured slight female predilection [14]. The cyst occurred in a wide age range with the average being in the fourth decade.

The literature, which consider the anterior region of the mandible [10,11,7] the most affected area, followed by the anterior region of the maxilla [12].

The case report shows the involvement of the posterior part of maxilla invading posteriolateral wall of maxillary sinus which is rare. Moreover, there remains a lack of clarification on some issues, such as histogenesis, biological behaviour and the appropriate treatment of these lesions. The main clinical finding in this disease is painless local edema, the clinical picture, however, is non-specific [15]. Lack of consistent clinical manifestations and the intra-antral development of these lesions mean that radiography is essential. Radiographic findings include a rounded or oval lesion, usually with well defined borders [3].

Most authors agree that there are no radiographic features specific of glandular odontogenic cysts. The differential diagnosis is made with botryoid cysts, keratocysts, residual cysts, the central mucoepidermoid carcinoma, and the ameloblastoma.

Histologically it is a polycystic structure with a non-keratinized squamous epithelium and cuboidal or ciliated epithelium with mucus-secreting cells. There is histological similarity with the central mucoepidermoid carcinoma [18]. Certain authors have suggested using immunohistochemical markers to differentiate these tumours [16,17].

Treatment options include curettage, enucleation and fixation with carnoy's solution [19], and en-bloc local excision with primary reconstruction [13] aiming to cure the patient and avoid further surgery, since there is a high recurrence rate with the conservative treatment [9]. Others recommend conservative procedures with postoperative follow-up during 3 to 5 years [15]. Bhatt et al. [20] advised that more conservative methods (excision, enucleation, curettage, thorough extirpation, and cryotherapy) should be applied. In this case performing a conservative procedure of enucleation and thorough curettage followed by the application of carnoy's solution was considered due to the age of the patient, relatively asymptomatic slow growing nature of the lesion and more so the cyst is within the confines of the posterior part of the antrum without involvement of floor of the orbit and lateral wall of the nose. The boundaries of the lesion was accurately studied by computer tomography. The present case was followed for 6 months. There was no science of recurrence and no complication occurred. The presentation of this case can contribute to the previously presented data in the world literature and add further information so that this little understood entity can be better clarified statistically.

Conclusion

GOC is a rare and aggressive lesion with a high recurrence rate. Careful clinical and radiological evaluation must be carried out. CT scans are recommended because they provide accurate information about locularity of the lesion, cortical integrity, expansion of the lesion and involvement of the contiguous soft tissue. According to the extent of the lesion, treatment options include curettage, enucleation and fixation with carnoy's solution [19], and en-bloc local excision with primary reconstruction.

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