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Case Report

GOC in Maxillary Anterior Jaw: An Infrequent Case Report

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Abstract

Padayachee and Van Wyk in 1987 first documented the case of Glandular odontogenic cyst (GOC) and regarded this as 'sialo-odontogenic cyst' they published two cases that mimicked botryoid odontogenic cyst and the central mucoepidermoid tumor of jaws. Glandular odontogenic cyst (GOC) is counted as infrequent developmental odontogenic cyst of jaw that has the incidence of 0.012 to 1.3% only GOC is very rare with only 111 cases have been documented in the English literate till date. This case report in unique as this patient presented with the painless swelling in maxillary anterior region.

Keywords: Glandular odontogenic cyst; maxillary anterior region; pathology; radiolucency; decompression.

INTRODUCTION

Padayachee and Van Wyk in 1987 first documented the case of Glandular odontogenic cyst (GOC) and regarded this as 'sialo-odontogenic cyst' they published two cases that mimicked botryoid odontogenic cyst and the central mucoepidermoid tumor of jaws [1]. According to the second edition of the World Health Organization's (WHO) histological classification of odontogenic tumours in 1992 it is "a cyst arising in the tooth-bearing areas of the jaws and characterised by an epithelial lining with cuboidal or columnar cells both at the surface and lining crypts or cyst-like spaces within the thickness of the epithelium" [2].

Glandular odontogenic cyst (GOC) is counted as infrequent developmental odontogenic cyst of jaw that has the incidence of 0.012 to 1.3% only GOC is very rare with only 111 cases have been documented in the English literate till date. GOC is dangerous owing to its a high recurrence rate and an aggressive growth potential [3].

Clinically, it is an asymptomatic slow growing swelling with predilection of occurrence in mandibular anterior region. It usually occurs in middle ages and is slightly more frequent in males [3].

On radiographic analysis these cysts present as unilocular or multilocular radiolucency with a well-defined border. Treatment of GOC includes curettage and enucleation, although some authors believe marginal resection to be more reliable treatment, due to tendency of the cyst to recur after curettage and enucleation [4-6].

According to Slootweg GOC's histopathologically resembles to a well-differentiated (low-grade) mucoepidermoid carcinoma but it requires entirely different management [7].

Although only a recently recognized clinical entity, the GOC is nevertheless an important lesion to recognize and diagnose, because of its potential for aggressive behaviour and a tendency to recur. Since this lesion was first recognized, an adequate number of case series have been published to permit a systematic review (SR) to be performed to determine not only the clinical and radiological features and outcomes of this lesion globally, but also any variation in its presentations within specific communities [1]. Therefore, clinical and radiological features, if they exist with any degree of specificity, should be identified and be recruited to refine further the diagnosis of those cases for which histopathological diagnosis is ambiguous. For example, such a radiological criterion, marginal definition, has been applied to distinguish between the two fibro-

osseous lesions, the fibrous dysplasia, a hamartoma; and the ossifying fibroma, a neoplasm [8].

This case report in unique as this patient presented with the painless swelling in maxillary anterior region.

CASE REPORT

A 58 years old male patient reported to the department of oral and maxillofacial surgery with the chief complaint of pain and swelling in upper front region of face since 3 months of reporting (Figure 1 and 2). The swelling kept on increasing in size and the upper anterior teeth were TOP positive. After initial clinical examination, CBCT evaluation was done along with biopsy and FNAC was performed.

Figure 1 and 2: Images of the patient

On CBCT evaluation, there was a well corticated multicystic osteolytic lesion, suggestive of benign Odontogenic tumour, which could be differentially diagnosed as central giant cell granuloma, odontogenic keratocyst, ameloblastoma. There is evidence of a well defined osteolytic radiolucency, with soft tissue density in the anterior maxilla basal bone extending from periapex of 13-23 mesio-distally, crossing the midline anterio-posteriorly. The lesion extends from the periapical region till floor of nasal fossa, superio-inferiorly. There is expansion of both the palatal and labial cortical plates. There is evident thinning and focal effacement of the adjoining labial cortical plate at the level of apical thirds of alveolus and basal maxilla. There is partial to complete effacement of palatal cortical plate. The lesion superiorly is noted to be communicating with the nasal cavity with thinning, effacement/perforation of the nasal floor bilaterally/ Oro-nasal communication. Anterior extent of the lesion could not be defined due to labial plate effacement measuring around 30.8 mm x 37.4 mm x 21.2 mm in its greatest antero-posterior, transverse and supero-inferior dimensions (Figure 3).

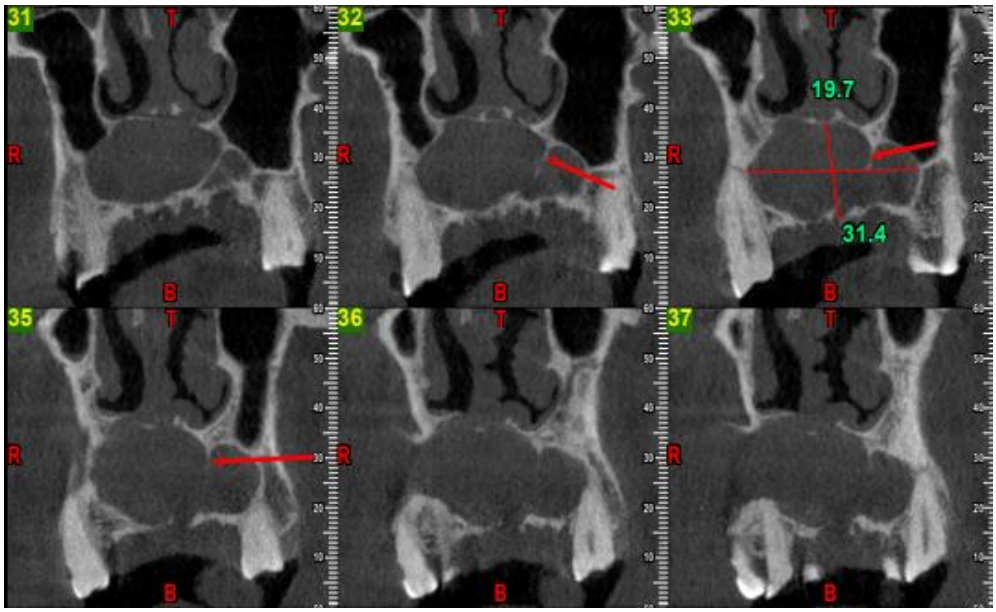

Figure 3: CBCT image

The cytology report stated the presence of extravasated RBCs with few lymphocytes in background and the histopathological report came out to be Odontogenic Cyst (Suggestive of Glandular Odontogenic Cyst) (Figure 4).

Histopathological Diagnosis:	:	Submitted H & E stained section shows pseudostratified ciliated columnar epithelium overlying fibrocellular connective tissue stroma. The overlying epithelium is of variable thickness with presence of goblet cells in the superficial layer with flat juxtra epithelial junction. The underlying fibro cellular connective tissue stroma shows parallel arrangement of collagen fibers with spindle shaped fibroblasts interspersed. Chronic inflammatory infiltrates chiefly composed of plasma cells and lymphocytes are seen. Blood vessels of various shapes and sizes engorged with RBCs are observed.
Impression:	:	Odontogenic Cyst (Suggestive of Glandular Odontogenic Cyst) Follow up is required.

Figure 4: Biopsy Report

The patient was earlier treated for MI around 3 years back and was currently suffering from hemiplegia and on informing about the risk associated with the surgery of such patient under general anesthesia, the patient and his family denied the surgical procedure under GA.

Considering his medical condition, decompression initially was considered as a treatment option for the patient and it was planned that when the size of the lesion will reduce, enucleation under LA will be considered.

Treatment and follow up

The decompression was started immediately after the biopsy report and then iodoform packing (Figure 5) was done after every 15 days, followed by regular radiographic evaluation.

Figure 5: Iodoform dressing

OPG was performed after every 2 months (Figure 6) and when the size of lesion reasonably decreased along with diminished extraoral and intraoal swelling (Figure 7) an enucleation was performed by raising a full thickness mucoperiosteal flap (Figure 8) from 25 to 15 and creating a lateral bony window. The sample was again send for final Histopathological examination after completely resecting the cyst margins. At last, primary closure of the cystic cavity was done.

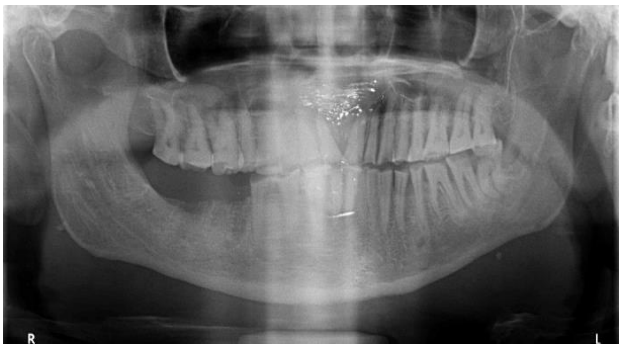

Figure 6: OPG done after 6months of decompression

Figure 7: Decrease in extraoral swelling

Figure 8: full-thickness flap raised

DISCUSSION

GOC is an infrequent cyst and only 111 cases of GOC have been reported in the literature so far with Magnusson *et al*., observing that only 0.012% of the cysts seen on the oral cavity have fulfilled the criteria of GOC microscopically [3, 8].

The aggressive biologic behavior of GOC and its propensity for recurrence might be associated with cell kinetics in the lining epithelium. Tosios *et al*., (2000) investigated the expression of bcl-2 protein, Ki-67 antigen and p53 protein in GOCs. The authors concluded that the increased expression of the anti-apoptotic bcl-2 may be associated with deregulation of cell death in the lining epithelium of the GOCs, whereas Ki-67 and p53 status did not seem to play a significant part in cell proliferation [10].

Radiographically, the GOC is localized intraosseously and may appear as a multilocular or unilocular radiolucent lesion with well-defined borders. Sometimes it may present with peripheral osteosclerotic border and scalloping, root resorption and displacement of the teeth. The clinical and radiographic findings of GOC are varied and often not pathognomonic and usually, it presents as asymptomatic slow growing lesion but occasionally may be accompanied by pain [11, 12].

Treatment by enucleation or curettage alone is associated with a high recurrence rate. Small unilocular lesions can be treated by enucleation. Surgical treatment of large lesions should include enucleation with peripheral ostectomy for unilocular cases and marginal resection or partial jaw resection in multilocular cases. Marsupialization followed by second phase surgery is an option for lesions approaching vital structures. Follow up should be continued for at least 3 years (up to 7 years in cases with features associated with increased risk of recurrence rate due to its intrinsic biological behavior, multilocularity of the cyst and incomplete removal of the lining following conservative treatment) [13].

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