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ATYPICAL PRESENTATION OF GLANDULAR ODONTOGENIC CYST IN THE ANTERIOR MANDIBLE: A CASE REPORT

[1]Dr. Rashmi Saikhedkar, *[2]Dr. Reshna Roy and [3]Dr. Sukriti Paul

[1]Professor, Department of Oral & Maxillofacial Surgery, College of Dental Science & Hospital, Rau, Indore, Madhya Pradesh-453331.

[2]Associate Professor, Department of Oral Pathology & Microbiology, College of Dental Science & Hospital, Rau, Indore, Madhya Pradesh-453331.

[3]Senior Lecturer, Department of Oral Medicine and Radiology, College of Dental Science & Hospital, Rau, Indore, Madhya Pradesh-453331.

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***Corresponding Author**
Dr. Reshna Roy
Associate Professor, Department of Oral Pathology & Microbiology, College of Dental Science & Hospital, Rau, Indore, Madhya Pradesh-453331.

ABSTRACT

Background: Glandular odontogenic cyst (GOC) is an uncommon developmental odontogenic cyst of jaws with a relative frequency between 0.012 and 1.3%. Generally, this cyst is encountered in the anterior region of the mandible and is more common in females as per as literature. **Result:** GOC is often misdiagnosed with that of other odontogenic cysts such as lateral periodontal cyst (LPC) and others, Periapical cyst or if it has malignant transformation then it might turn to mucoepidermoid carcinoma. This case report presents a rare case of infected glandular odontogenic cyst in a 30 year old patient. **Conclusion:** This cyst is well known entity in the World Health Organization(WHO). It can be confused with Periapical or Lateral Periodontal Cyst so long term follow-up is often needed. Malignant transformation of GOC to mucoepidermoid carcinoma has also been reported. Moreover Immunohistochemistry can be applicable for confirmation of diagnosis.

KEYWORDS: glandular odontogenic cyst, mucous cell, sialo-odontogenic cyst, epidermoid cell.

INTRODUCTION

The glandular odontogenic cyst (GOC), also known as sialo-odontogenic cyst derived from the rests of dental lamina consisting of a stratified squamous epithelium containing numerous mucus-secreting cells. It is a entirely different entity that was described by Padayachee and van Wyk in 1987 which was intrabony in nature multilocular radiolucency.[1]

Gardner et al. in 1988 characterized histopathological features and biological behaviour of glandular odontogenic cyst and established it as a distinct entity and proposed term 'GOC'. He established.[2] GOC as a distinct identity and proposed the term GOC. Sialo-Odontogenic Cyst also known as Infected glandular odontogenic cyst is termed as Polymorphous Odontogenic Cyst. According to High et al, GOC has the capacity to regrow and is aggressive in nature.[3] According to Magnusson et al these cysts comprised 0.012% of total 5800 jaw cysts which were studied and reported in a 19-year period. Among these, four of the cases were reported in males and three in females. Age range for occurrence for GOC is reported to be 30-75 years, the peak being around glandular odontogenic cyst is 55 years.[4] to Morais et al, the most preferred site for occurence of glandular odontogenic cyst is the anterior mandibular region in approximately 85% of the cases. GOC are known to cross the midline.[5]

Few other studies report that mandibular anterior region is preferred site seen for occurrence of GOC almost 80%, while in maxilla only 20% of the case of GOC are reported. The approximate location of the lesion is evident in mandible about 80% and in maxilla about 20%. The ratio is found to be 1:2(maxilla: mandible).[6]

This particular cyst as glandular odontogenic cyst is previously combined with Botryoid cyst because of its glandular element to it. Recently in WHO, this cyst was added due to uncertain histogenesis.

Glandular Odontogenic cyst is generally also considered as large intraosseous cyst which often consists of two elements in it like mucus secreting cells and stratified squamous epithelium like layer. Shear et al termed Glandular odontogenic cyst as "Polymorphous Odontogenic cyst". Immunohistochemistry is required to confirm diagnosis of this GOC cyst.

After various years of discussion and reviews on glandular odontogenic cyst, this cyst draws the attention towards pooling of material on various grounds which lead to increase in

number of cases. Among which it is clearly seen that authors found this cyst to be quite aggressive in nature.

Case Description: A 30 year old female patient had reported to the Department of Oral Medicine & Radiology with a chief complaint of swelling on lower front teeth region w.r.f 31,32,33 to 41,42,43 which is also associated with vestibular swelling for the same since few weeks. The lesion was slow growing with intermittent pain. There was obvious swelling in the chin region from one side angle of the mouth to other. Swelling is hard on palpation and executed bluish hue. On intraoral swelling examination vestibular obliteration due to swelling was seen from one side mandibular canine to other side mandibular canine region i.e 31,32,33 to 41,42,43.Intraoral swelling was fluctuant. The teeth were firm and non-vital. The gingiva and mucosa appeared normal .On Medical examination, patient was suffering from asthma since 2 years and did not had any functional problems.

Radiographic examination: An (OPG) was taken which showed a large well defined, unilocular, radiolucent lesion involving the right to left anterior mandibular region extending 31,32,33,41,42,43 region. The size of the lesion as per OPG findings was approximately 5-6cm.No teeth displacement is seen as such. Defined sclerotic margins are also observed. Excisional biopsy was performed under local anaesthesia and this incision extended from tooth 31,32,33 to 41,42,43. The fluid on aspiration with a 18 gauge needle showed a brown coloured fluid with blood traces in between. A well informed written consent was obtained. The site was opened up, the fluid was drained out. Some cystic lining was obtained which was sent for biopsy. The result of biopsy revealed extracted cyst lining in bits and pieces . Area was debrided and sutures placed. The patient was followed to the clinic and suture removal was done. But the patient did not had any pain or signs of recurrence noted around that area. (As quoted with fig:1).

Histopathological features: Histopathologic examination revealed specimens lined with non-keratinised squamous epithelium exhibiting variable structure and a flat epithelial connective tissue interface. The connective tissue stroma consists of mucous and epidermoid cells scattered in and around the stroma. Also the stroma is loaded with chronic inflammatory cells infiltration of which mostly are predominantly lymphocytes. Due to the presence of chronic inflammation, the epithelium is atrophied. The epithelium under 10x magnification appears slightly corrugated.

FIGURES

Fig 1: OPG showing scalloped borders w.r.t 31,32,33,41,42,43

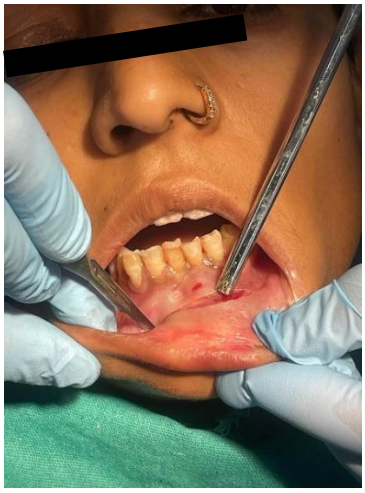

Fig 2: Pre-operative View

Fig 3: During procedure

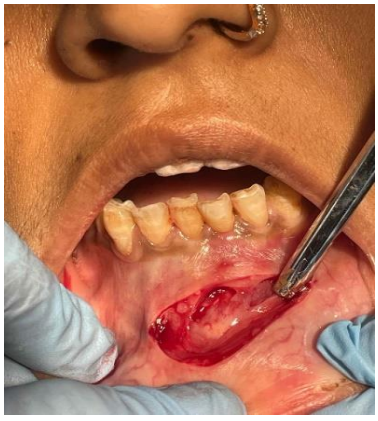

Fig 4: Post-Operative View

Fig 5: Suture placed

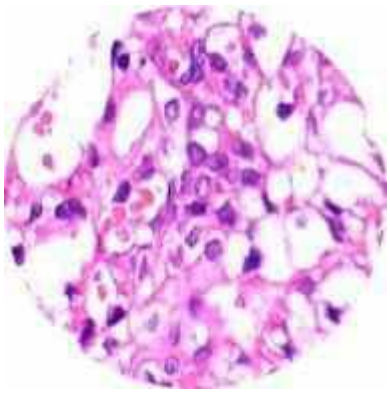

Fig 6: Under 40X magnification presence of mucous and epidermoid

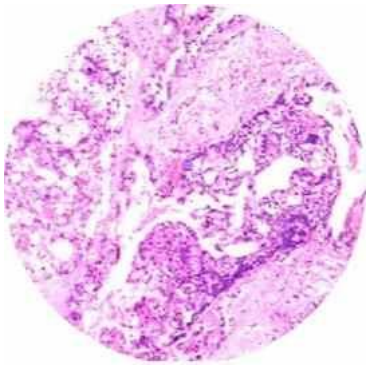

Fig 7: Under 10X magnification presence of chronic inflammatory cells

DISCUSSION

It is a rare type of odontogenic jaw cyst which has histological features similar to salivary gland tumor. According to Campos et al, this jaw cyst occurs in anterior mandible region[7] and this is similar to our reported case where the lesion is located in the anterior mandibular region w.r.f 31,32,41,42. Glandular odontogenic cyst seemed to occur in unerupted tooth or displaced tooth as found by Gomez et al.[8] And this is very similar to clinical finding that glandular odontogenic cyst occurs in unerupted tooth.

Moreover, in glandular odontogenic cyst its been reported that swelling is associated with cortical expansion and this is similar in our reported case that swelling was found with respect to anterior mandibular region.[9]

According to Magnusson et al, radiographic examination showed a well-defined radiolucency with a scalloped border and it is reported in a review of 7 cases of GOC.[10] This is exactly similarly observed in our reported radiological findings where glandular odontogenic cyst has properly defined scalloped borders.

As per in article mentioned in Shaik et al suggested that aspiration fluid of GOC are typically of serous brownish red coloured fluid from glandular odontogenic cyst in mandible[11] .This is in accordance to the fluid aspirate picture in this case report.

Majority of GOC are found in female patients and this was reported in Kadam et al, Kaur et al, Tambawala et al[12,13,14] and this is alike to our reported case where glandular odontogenic cyst is evident in female patient. Glandular odontogenic cyst is a relatively aggressive lesion and shows a greater signs of recurrence about 30-50%.[15,16]

Although the histogenesis is not so clear. Glandular odontogenic cyst (GOC) earlier it was reported to rise from intraosseous salivary gland tissue but now it is evident that this cyst develops from remnants of dental lamina.[17], In our reported case also, the histogenesis is bit unclear.

Most authors in their studies have reported to agree to the finding of GOC, doesnot have any specific radiological investigation.[18]

The line of treatment of glandular odontogenic cyst can be curettage, enucleation or marsupialisation. Our reported case was treated by a complete surgical excision. The patient was called for follow-up for any further investigations if needed.

CONCLUSION

Glandular odontogenic cyst is one of the most aggressive lesion which can cause bone resorption abruptly. The purpose of this case presentation is to enhance knowledge and know in details about this cyst as it is often misdiagnosed both on histological as well as clinical findings and this fact will challenge the work of a oral pathologist. This cyst is well known

entity in the World Health Organization(WHO). It can be confused with Periapical or Lateral Periodontal Cyst so long term follow-up is often needed.

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Conflict of Interest: N/A.

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