CASE REPORT

Volume-7 | Issue-5 | May-2018 | PRINT ISSN No 2277 - 8179

INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

GOC OF THE ANTERIOR MAXILLA- A RARE CASE

Dental Science

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ABSTRACT

Background: Glandular odontogenic cyst (GOC) is an uncommon developmental Odontogenic cyst of jaws often misdiagnosed because of its overlapping histopathological features with that of other Odontogenic cysts. Although none of the clinical or radiographic features of GOC are unique or pathognomonic, the lesion has a potentially aggressive behavior.

Case report: Here in we report a case of Glandular Odontogenic cyst of Maxilla in a 26 year old male patient highlighting the clinical and histopathological findings to enhance our knowledge in the diagnosis of this rare entity.

Conclusion: This case illustrates that careful clinical radiological and histopathological evaluation is required for the diagnosis and aggressive treatment of this highly recurrent cyst.

KEYWORDS

Glandular Odontogenic Cyst, Lateral Periodontal Cyst, Sialo Odontogenic Cyst

Introduction:

Glandular odontogenic cyst is a rare odontogenic jaw cyst with a frequency rate being 0.012% to 1.3% of all jaw cysts. It was first documented by Padayache and Van wyk in 1987 as "SIALO-ODONTOGENIC CYST".[1] The name was changed to "GLANDULAR ODONTOGENIC CYST" by Gardner et al in 1988 because of the lack of evidence of salivary gland origin,[2]and the term was later adopted by the World Health Organization (WHO) in 1992.[3] GOC mainly occurs in the anterior region of the mandible, followed by the anterior region of the maxilla. It occurs primarily in the middle-aged people, female : male ratio being 19:28.[4,5,6,7]

Case Report:

A 26 year old male patient reported to the department of Oral Medicine and Radiology, for the evaluation and management of an asymptomatic swelling in the anterior part of the maxilla since two weeks. The swelling was asymptomatic and present from past three months with a history of trauma. The upper right central incisor was extracted as the patient complained of pain. However, there was no apparent resolution of the swelling and gradual enlargement was experienced.

Extra oral photograph showed swelling on right side of the face causing moderate asymmetry. Intra oral photograph showed well-defined swelling on right upper alveolar region of the maxilla causing gross asymmetry. On palpation, an oval shaped mass on the labial aspect of the right anterior maxilla, extending from the right maxillary lateral incisor to the right maxillary first premolar region was noticed. The gingival and alveolar mucosa were of normal appearance (Fig. 1)
Orthopantamogram determined oval shaped unilocular radiolucent lesion in the maxillary anterior region extending from the left central incisor to the right canine and superiorly approaching the hard palate. The clinical and radiographic differential diagnosis included Botroid Odontogenic cyst and Adenomatoid Odontogenic tumour. Incisional biopsy of the specimen was made, that consisted of irregular fragments of soft tissue measuring from 0.3 to 1 cm in dimension.

Microscopic examination showed cystic cavity lined by Non-keratinized stratified squamous epithelium of variable thickness, the connective tissue epithelial junction was flat (Fig.2). Papillary proliferation of the lining epithelium was seen in some areas (Fig.3) that showed plaque-like thickening (Fig.4). The superficial cells of cystic lining were cuboidal cells interspersed with mucous producing goblet cells and also in some areas intraepithelial microcyst or duct like structures were seen (Fig.5).

Alcian blue staining showed positivity for mucous cells, mucous pooling and mucous secreting goblet cells within the papillary projections of epithelial lining (Fig.6).

The final diagnosis was made as Glandular Odontogenic Cyst based on the clinical, radiographic and histopathological features.

Discussion :

GOC has gained special attention by clinicians and pathologists due to two reasons. First, is the histomorphologic resemblance of GOC with the Lateral Periodontal Cyst (LPC), Botroid Odontogenic Cyst (BOC) and Central Mucoepidermoid Carcinoma (CMEC) of the jaws, so it is also known as SIALO-ODONTOGENIC CYST or MUCOEPIDERMOID CYST.[8]Second is the potential aggressive behaviour of the lesion. The histogenesis of this lesion is unclear. Initially it was suggested to originate from intra osseous salivary gland tissue but recently an odontogenic origin is suggested because of histopathological features and cytokeratin profile that arises from cell rest of serre of dental lamina[9]

The main clinical finding in this disease will be asymptomatic slow growing swelling, the clinical picture however is non-specific and Radiographically, GOC does not display specific or pathognomonic features. It may present as a multilocular with scalloping margin or unilocular radiolucencies with well defined margin.[10,11]

Histopathologically GOC demonstrates[12,13]

- A nonkeratinized stratified squamous epithelial lining of variable thickness.
- Presents a flat interface between epithelium and the underlying connective tissue.
- In more or less extensive areas; the superficial layer of epithelium consists of eosinophilic cuboidal cells, columnar cells, or ciliated cells that form papillary projections and folds.
- Within the thickness of epithelium, there are intraepithelial gland like structures consisting of mucous cells and mucin filled crypts or microcysts lined by cuboidal cells that are presumed to be

formed from the folding of the lining epithelium.
- Focal thickenings or plaques of epithelium where cells form whorls or spheres are also seen.
- The sub epithelial connective tissue is usually free of inflammation and may demonstrate irregular shaped calcifications or islands of odontogenic epithelium rarely.
- Pools of mucicarminophilic material, mucous cells, goblet cells, and vacuolated cells in variable numbers are seen within the epithelium. Mucin pooling can be confirmed by PAS and Alcian blue stain.
- Special stains used are PAS, Alcian blue and mucicarmine

Further, immunohistochemical findings in GOC suggest that its biologic behaviour may be associated with dysregulation of cell death in the lining epithelium indicated by increased expression of anti apoptotic protein bcl-2. Inspite of its aggressive behaviour and recurrent nature, the numbers of Ki-67 positive cells are lower, which suggest that biologic behaviour of GOC is not associated with cell proliferation. Incomplete removal due to its multicystic configuration, tendency of epithelium to separate from connective tissue or growth through cancellous spaces of bone may account for its high recurrence rate which demands more aggressive treatment procedures and follow up of 5-7years is needed.[9,10]

To conclude, GOC is a rare and aggressive lesion with a high recurrence rate. According to a recent review only less than 170 cases of GOC were reported in English literature so that causes lack of knowledge about this cyst which makes its diagnosis difficult. Careful clinical radiological and histopathological evaluation must be carried out. CT scans are recommended because they provide accurate information about locularity as well as expansion of the lesion, cortical integrity and involvement of the contiguous soft tissue. The purpose of reporting this case was to add to the existing knowledge about this rare cyst which still has an uncertain nature.

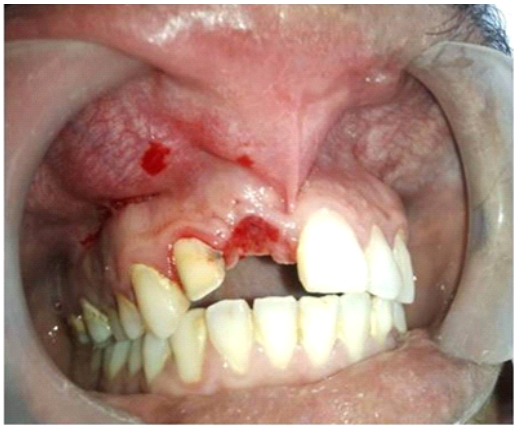
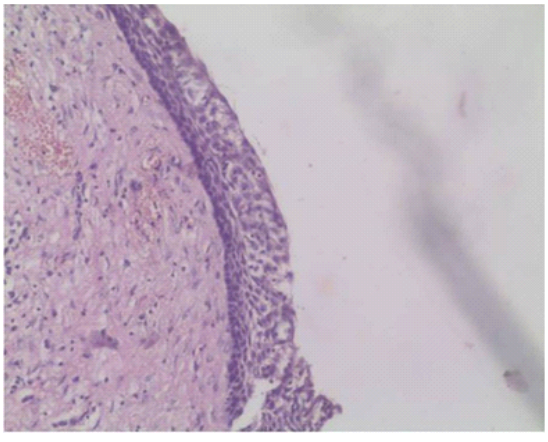


FIGURE 1- INTRA ORAL PHOTOGRAPH SHOWING WELL-DEFINED SWELLING ON RIGHT SIDE OF THE MAXILLA CAUSING GROSS ASYMMETRY.



EG:FIGURE 2- NON KERATINIZED CYSTIC LINING WITH BASAL CUBIODAL CELLS(H&E,10x)

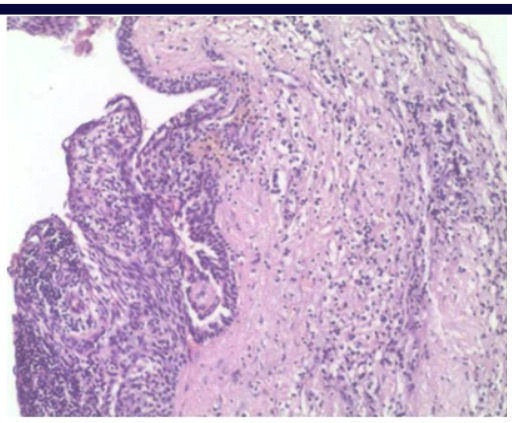


FIGURE 3- PAPILLARY PROJECTIONS OF EPITHELIAL LINING.(H&E,10X)

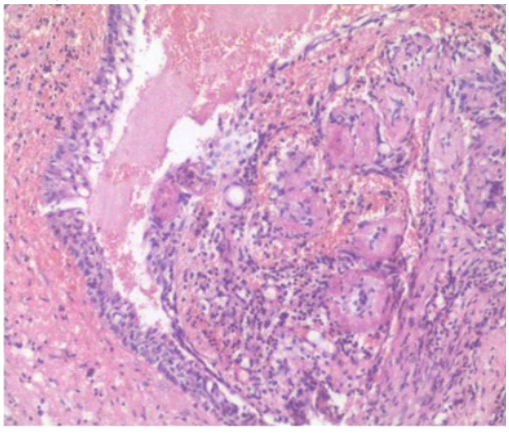


FIGURE 4 - EPITHELIAL PLAQUE-LIKE THICKENING. (H&E,40X)

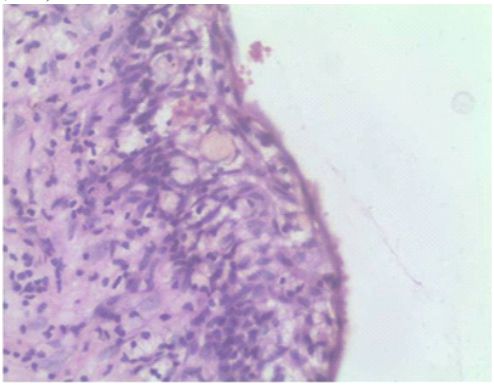


FIGURE 5 - NUMEROUS MUCOUS CELLS IN THE CYST LINING.(H&E,40X)

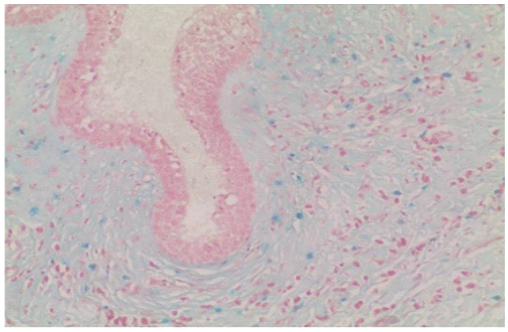


FIGURE 6- ALCIAN BLUE STAIN SHOWING POSITIVITY FOR MUCOUS CELLS AND MUCOUS POOLING AND MUCOUS SECRECTING GOBLET CELLS WITHIN THE PAPILLARY PROJECTIONS OF EPITHELIAL LINING

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