

Glandular Odontogenic Cyst – A Case Report

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Abstract: Glandular odontogenic cyst is a rare cyst of odontogenic origin .It may present as a unilocular or multilocular radiolucency having unpredictable and aggressive behavior. It has a chance of recurrence after surgical excision. It tends to occur in middle aged males. Here we report a case of glandular odontogenic cyst in a 25 year old man.

Keywords: glandular odontogenic cyst, maxilla, sialo odontogenic cyst

I. Introduction

Glandular odontogenic cyst is a rare developmental cyst.Padayachee and Van Wykin presented the first two cases of this cyst as a sialo-odontogenic cyst in 1987[1].After that Gardner et al in 1988 categorized this cyst as a separate entity under the term ‘glandular odontogenic cyst’[2].The glandular odontogenic cyst is included in the World Health Organization histology under the terms “glandular odontogenic cyst or sialo-odontogenic cyst”[3].High et al introduced the term ‘polymorphous odontogenic cyst’ in 1996 because of its varied histological appearances[4].

Only 111 glandular odontogenic cysts have been reported in the English literature[5]. Glandular odontogenic cyst most commonly occurs in anterior mandible with male predilection in middle age group. In some cases it is asymptomatic, while in some cases it causes pain and may present as a slow growing swelling causing tooth displacement[6,7].It may present as an unilocular radiolucency or multilocular radiolucency with scalloped borders⁸. Histologically, GOC shows a non-keratinized stratified squamous epithelial lining, focal plaque like thickenings within the lining, microcysts or intraepithelial crypts containing mucin, mucous cells and hyaline bodies, eosinophilic cuboidal or columnar cells that may be ciliated, papillary projections of epithelium and absence of inflammation in the subepithelial connective tissue.[1,2,8,9]Treatment includes curettage and enucleation, although some authors believe that marginal resection is the better modality since the cyst has a chance to recur[10].Here we discuss a case of glandular odontogenic cyst with its clinical and radiological features.

II. Case Report

A 25 year old man reported to the Department of Oral Medicine and Radiology with chief complaints of swelling in the left side of the face and pain in the same region for the past two months. The swelling gradually increased to reach the present size .No similar type of swelling present elsewhere in the body. His past medical history revealed nothing significant. Past surgical history revealed that the patient underwent surgery in his left upper jaw at his fifteenth age but unable to produce the old records.

On extra oral examination,(figure 1) obvious facial asymmetry was present on left side of the face due to swelling. The swelling was diffuse, extending superiorly 5mm from the infra orbital rim, inferiorly up to vermilion border of lip, anteriorly from the ala of the nose, posteriorly to 5mm in front of the ear lobe ,margins were ill defined and surface was smooth. No secondary changes were noted in the swelling. Intraorally(figure 2& 3)swelling was present in the alveolar mucosa in relation to 22,23,24,25 with obliteration of buccal vestibule. On palpation, inspectory findings were confirmed and the swelling was firm, tender and fluctuant. Aspiration of the swelling yielded a straw colored fluid(figure 4).

Digital panoramic radiograph revealed an unilocular radiolucency with well defined borders in relation to 23, 24, 25 causing apical root resorption in 24, 25 and mesial displacement of 25(figure 5). Pulp vitality test showed positive response in 23, 26 and negative response in 24,25.

Cone beam Computed Tomogram was taken which revealed an expansile osteolytic lesion with corticated borders causing destruction of nasal bone medially and palatal bone inferiorly. The lesion also invaded the left maxillary sinus (figure 6). In 3D reconstruction view, an osteolytic hypodense lesion with corticated borders measuring 31.3mm& 31.4mm causing apical root resorption in 24,25 and 26.(figure 7). A provisional diagnosis of periapical cyst and differential diagnosis of dentigerous cyst was made. Incisional biopsy was done in the periapical region of 24, 25 and the specimen was sent for histopathological evaluation. Histopathological finding was consistent with glandular odontogenic cyst. The section showed non keratinized

stratified squamous epithelial lining of variable thickness. The lining epithelium showed the presence of mucous cells, intra epithelial crypts, and pseudoglandular structures filled with eosinophilic material. The connective tissue fibrous wall appeared dense with chronic inflammatory infiltrate and cholesterol clefts. Periapical bone was also evident. The cyst was enucleated and the specimen was sent for histopathological evaluation (figure 8). The final diagnosis was consistent with preoperative diagnosis of glandular odontogenic cyst.

III. Discussion

Glandular odontogenic cyst is a very rare cyst and only few cases have been recorded in the literature. Glandular odontogenic cyst usually occurs in anterior mandible, our case differs from the literature since the cyst was located in anterior maxilla. It usually presents as a slow growing swelling, similarly our patient also reported that the swelling gradually grew for the past six months.

Glandular Odontogenic cyst more commonly affects males than females, which is in accordance with the literature. It usually presents as an unilocular or multilocular radiolucency with well defined borders. In our case also, there was an unilocular radiolucency with well defined borders.

Glandular Odontogenic cyst usually does not have distinct clinical and radiological features. So the diagnosis of the cyst based on clinical and radiographical features is not possible. Histopathological diagnosis is mandatory. Since it occurs as an intraosseous cyst in periapical region, the following lesions such as dentigerous cyst, periapical cyst, odontogenic keratocyst, radicular cyst, myxoma, ameloblastoma, central giant cell lesion, fibrous dysplasia, and central mucoepidermoid carcinoma (MEC) have to be considered in the differential diagnosis.

Aspirated fluid in the present case was straw colored with low viscosity. Literature reveals that colorless fluid with low viscosity clinically suggests Glandular Odontogenic cyst. The microscopic features of glandular odontogenic cyst is similar to lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC), and the central Mucoepidermoid carcinoma. Lateral Periodontal cyst is a developmental odontogenic cyst lined by thin non keratinized epithelium and also shows focal epithelial thickenings and glycogen rich epithelial cells similar to glandular odontogenic cyst[12,13]. BOC is a locally aggressive polycystic variant of LPC[12] shows similar histological features similar to GOC like epithelial plaques and areas of glycogen rich clear cells[14]. However the presence of ciliated epithelium and duct like spaces with mucous cells specifically differentiated from LPC and BOC and helps in the diagnosis of GOC[3,15,16].

The differentiation of low grade Central Mucoepidermoid Carcinoma from GOC especially its multicystic variant is more significant and difficult. There is a considerable histological overlap exists between GOC and CMEC. However, superficial cuboidal cells, epithelial whorls, ciliated cells, and intraepithelial microcyst or duct like structures are not pathognomic for CMEC and their presence or absence can help in establishing a definitive diagnosis[17]. Immunostaining with CK-18 and 19 and their positivity in GOC may help in differentiating GOC from CMEC[18]. Certain studies indicate that the use of IHC for p-53 and Ki-67 can help the clinician in differentiating GOC from CMEC. GOC exhibited decreased p-53 positivity and increased Ki-67 index when compared to CMEC[17]

Several studies indicate that GOC is relatively an aggressive lesion with a high tendency for erosion or perforation of cortical plate and chance of recurrence. Hence treatment varies from curettage, enucleation, en bloc [19] and partial osteotomy[20,21] For our patient the cyst was enucleated and he is under follow up to monitor recurrence.

IV. Conclusion

We discussed the clinical, radiological and histopathological features of glandular odontogenic cyst. This case report will add to information about glandular odontogenic cyst which is rare type of odontogenic cyst.

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Figure 1



Extra oral examination revealing swelling in the left side of the face

Figure 2



Intra oral examination showing swelling in the periapical region of 23,24,25 with obliteration of buccal vestibule.

Figure 3



Mirror image of the periapical region

Figure 4



Aspiration of the cyst showing straw coloured fluid

Figure 5



Shows unilocular radiolucency in peri apical region of 23,24,25 with well defined borders causing apical one third root resorption of 24 ,25 and mesial displacement of 25

Figure 6



Coronal section of Cone Beam Computed Tomography Showing expansile osteolytic lesion with corticated borders causing destruction of nasal bone medially and palatal bone inferiorly, the lesion also invaded the left maxillary sinus.

Figure 7



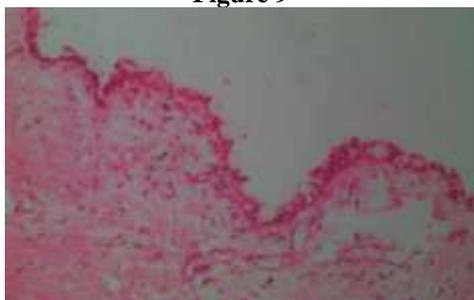
.In 3D reconstruction view, an osteolytic hypodense lesion with corticated borders measuring 31.3mm& 31.4mm causing apical root resorption in 24 ,25 and 26.

Figure 8

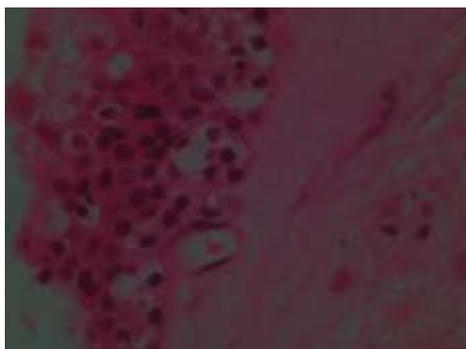


Macroscopic appearance of the excised cyst .

Figure 9



Histopathological photograph



The section showed non keratinized stratified squamous epithelial lining of variable thickness. The lining epithelium showed the presence of mucous cells, intra epithelial crypts, and pseudoglandular structures filled with eosinophilic material.