Case report

CLINICOPATHOLOGICAL SPECTRUM OF GLANDULAR ODONTOGENIC CYST - REPORT OF THREE CASES

ABSTRACT:

Glandular odontogenic cyst (GOC) is rare locally aggressive lesion of jaw constituting 0.012% to 1.3% % of all odontogenic cysts. It commonly occurs in mandible (70%) than maxilla (30%). It arises from rest of dental lamina. Cysts radiologically appear as unilocular or multilocular radiolucency with a well-defined corticated border which mimics dentigerous cyst and lateral periodontal cyst. Histopathologically intraepithelial microcysts, presence of hobnail cells, epithelial lining is of variable thickness with sometimes apocrine snouting are observed. Clinical and radiographic features are nonspecific, and to its similarities with other lesions, it can be misdiagnosed. Treatment modalities of GOC are curettage, enucleation or marsupialization depending on aggressiveness of lesion.

In this case series three cases of GOC have been reported of which two are in mandible and one is in maxilla. Diagnosis was confirmed by histopathology and immunohistochemistry (IHC) markers which were strongly positive for CK19 and negative for CK 18.

KEY WORDS:

Glandular odontogenic cyst, IHC, central mucoepidermoid carcinoma odontogenic cyst.

INTRODUCTION:

GOC is rare odontogenic cyst first reported by Padayachee and van Wykassialo in 1987. Due to presence of glandular tissue, they termed it as "Sialo odontogenic cyst."[1] In 1988 Gardner et al termed it as glandular odontogenic cyst. In 1992 WHO categorized this entity under developmental odontogenic cyst.[2] High et al (1996) termed it as 'polymorphous odontogenic cyst.[3] Incidence rate of GOC ranges from 0.012% to 1.3% of all the jaw cysts and its prevalence is 0.17%.[4]

Radiological finding revealed unilocular or multilocular radiolucencies with sclerotic borders which is non-specific and may mimic other cysts. Final diagnosis was confirmed on the histopathology.[5]

Though GOC is rare, correct diagnosis has clinical importance due to its aggressiveness and high recurrence. Curettage and enucleation are the treatments for GOC but due to high recurrence marginal resection is preferred.[1] Aim of this report is to emphasize on clinical, radiologic features and Histopathological criteria for GOC with IHC characteristics.

CASE PRESENTATIONS:

TABLE -1 – CASES:

Sr. no.	Entity	Case- 1	Case-2	Case-3
1	Clinical features (fig-a and b)	51 yrs /Male Site - Posterior	14 yrs / Female Site - Posterior	21yrs Female E/O - diffuse swelling

		mandible E/O – bony hard swelling over left mandibular ramus I/O - firm swelling extending from distal of 37 to ramus of mandible	mandible E/O - swelling over left body of mandible I/O - swelling extending from 34 to mesial of 36 with obliteration of buccal vestibule	on right zygomatic region. I/O - labial swelling extending from distal of 11 to mesial of 13 and swelling in right anterior palatal region.
2	Radiological findings(fig-c)	multilocular radiolucencies with well corticated borders extended from distal of 37 to mid ramus area.	unilocular radiolucency with well corticated borders in inter radicular area with 35 and 36	well defined radiolucency extending from 16 to 23 involving maxillary sinus with displacement of teeth
3	Histopathological findings(fig-d)	Fulfilled all the major and two minor Kaplan	Fulfilled all the major and two	Fulfilled all the major and two

		criteria	minor Kaplan criteria	minor Kaplan criteria
4	IHC markers CK19 CK18	 Positive Negative	 Positive Negative	 Positive Negative

DISCUSSION:

GOC is considered to be a rare cyst, till date less than 300 cases have been discribed.[3] It mainly affects fifth and sixth decades of life.[6] In the present series affected age group is second to sixth decade. In the literature most of the GOCs have slight male predilection (57 : 43) [7]. In this series we found two females and one male was affected. It has predominance for anterior mandible than maxilla. If seen in maxilla it is evident in anterior maxilla.[8] Reported cases were two in posterior mandible and one in anterior maxilla.

The common clinical feature of GOC is an asymptomatic swelling, [9] In the series, extra oral and intraoral asymptomatic swelling were observed with displacement of teeth in one case.

Radiographically GOC may appear as a unilocular or multilocular radiolucent lesion with well-defined borders, root resorption and displacement of the teeth.[1] In this series similar findings were observed with radio opacity in one case.

Clinical and histopathological finding of GOC overlap with other cysts or tumors, like dentigerous cysts and radicular cysts, botryoid cysts, lateral periodontal cysts, and mucoepidermoid carcinoma. This may have negative impact on treatment and patient prognosis.[6] To overcome this, Kaplan et al (2005) has suggested histopathologically five major and four minor criteria and concluded that all the major criteria should be fulfilled for diagnosis and the minor ones may aid in the same.[3] They are as follows:

TABLE - 2 - Kaplan et al criteria (2005)

Major criteria	**Minor criteria**
• squamous epithelium lining lacking basal cell palisading	• papillary proliferation of the lining epithelium
• focal luminal proliferation	• ciliated cells
• hob nail cells	• multi luminal/multicystic architecture
• goblet cells with or without crypts lined by mucous producing cells	• clear / vacuolated cells in basal or spinous layers
• intraepithelial microcysts	

But Fowler et al. did not segregate the major and minor criteria for GOC but proposed ten distinguishing features, out of which seven features should be fulfilled for the diagnosis of GOC[3] they are as follows:

Surface eosinophilic cuboidal cells, also called “hobnail cells”

Intraepithelial microcysts or duct-like spaces

Apocrine snouting of hobnail cells

Clear or vacuolated cells

Variable thickness of the cyst lining

Papillary projections or “tufting” into the cyst lumen

Mucous goblet cells

Epithelial spheres or plaque-like thickenings

Cilia on the surface of eosinophilic cuboidal cells

Multiple compartments

In this case series all five major and two minor Histopathological Kaplan criteria and eight distinguishing features proposed by Fowler et al were fulfilled. Special stains like mucicarmine and periodic acid-schif (PAS) can be used for presence of mucin and mucous cells[9,] we used PAS stain and mucicarmine for confirmation.

Histopathologically GOC should be differentiated from dentigerous cyst, lateral periodontal cyst (LPC) and l mucoepidermoid carcinoma.[1]

Presence of intraepithelial duct like spaces lined with mucous cells, ciliated cells, glandular or pseudo glandular structures, microcysts or mucous pools in the epithelium differentiate GOC from LPC.[10]

Presence of microcysts, focal luminal epithelial proliferation and clear cells differentiate GOC from dentigerous cyst.[11]

Cuboidal cells, epithelial whorls, and intraepithelial and microcysts are not typical for central MEC which differentiate GOC from MEC. Immunohistochemical marker CK18 studies differentiate GOC from MEC.[1] The presence of MAML2 (mastermind-like 2) gene in low-grade central MEC may helpful to differentiate GOC from MEC.[6]

By Immunohistochemical studies,Odontogenic nature of GOC can be confirmed by cytokeratin - 7, 13, 14 and 19.[12] In the series, all the cases were strongly positive for CK 19. GOC can be differentiated from Low grade Mucoepidermoid Carcinoma by Negative expression of CK 8 & 18.[13] In the case series all the cases were negative for CK 18.

To assess the proliferate nature of the GOC IHC staining can be done using Ki-67, p53.[14]

Treatment of GOC includes curettage with and without peripheral osteotomy, enucleation or marsupialization.[5] GOC recurrence is particularly associated with conservative surgical procedures, so adjunctive therapies like peripheral osteotomy, cryotherapy, or the application of Carnoy's solution are involved.[6] Treatment of choice for one case was en bloc resection due to ramus involvement and two cases were treated with enucleation.

GOC has high recurrence rate (21 % to 55%,) this attribute to the aggressive nature of cyst.[3] Three to seven years follow up is advisable .[2] In our cases 2 years follow up was done, recurrence was not evident.

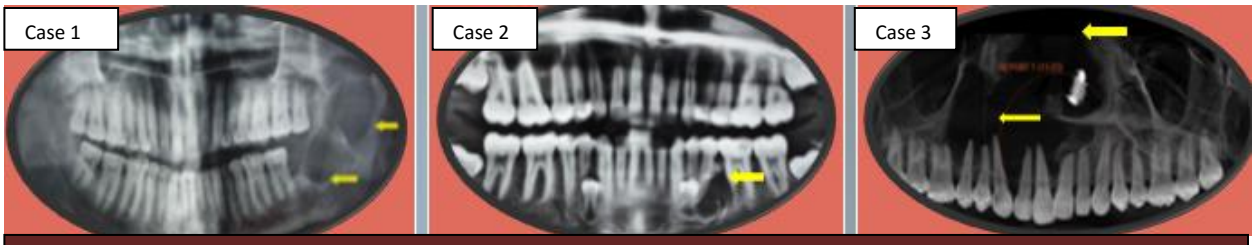
CONCLUSION:

GOC is a diagnostic challenge to oral pathologist due to clinical and histopathological overlapping,

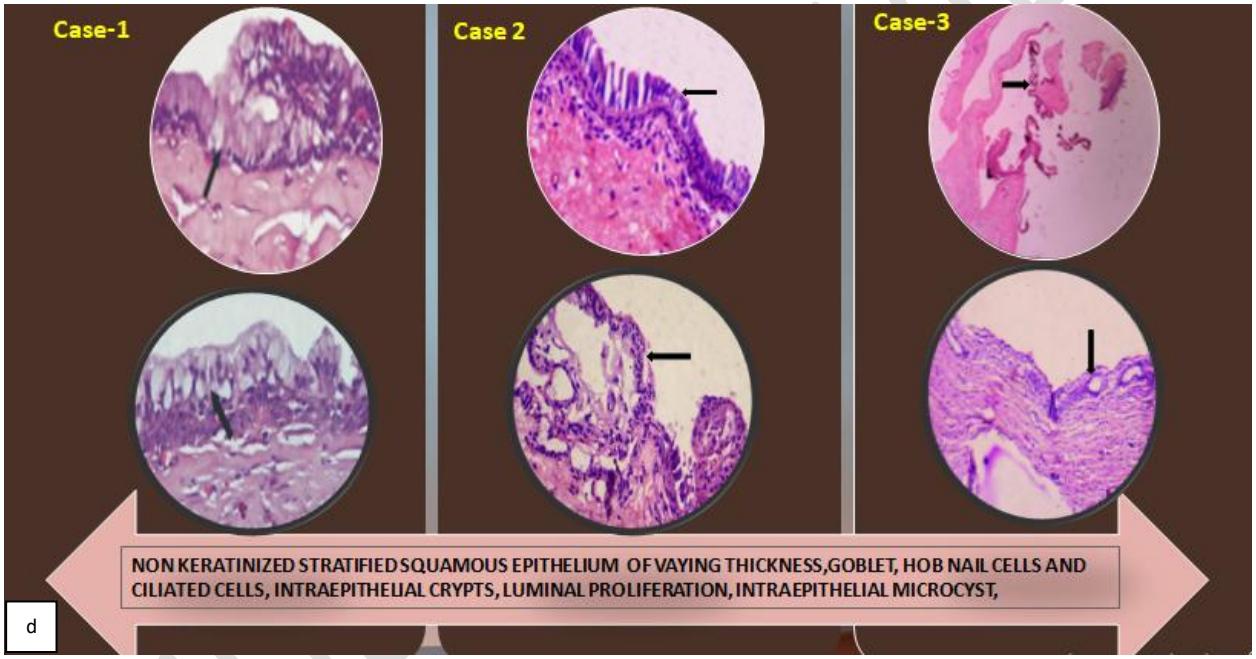
So a careful clinical, radiological and histopathological evaluation must be carried out with IHC to ensure correct diagnosis and treatment.

a- EXTRA ORAL FEATURES

b- INTRAORAL FEATUTES



C –RADIGRAPHIC FEATURES



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