



## Glandular Odontogenic Cyst withan Odontome: An Unusual Association

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### ABSTRACT:

*Glandular odontogenic cyst (GOC) is a relatively rare jaw cyst with frequency of only 0.2%. It is common in 5th -6th decade of life with mean age of occurrence of 45.7 years. It has slight male predominance (M: F; 1.3:1) and 70% of the cases occur in mandible. It is locally aggressive and tends to cause bone expansion, cortical perforation, root resorption and has high rate of recurrence. Previously it was thought to have glandular origin due to its histological similarities with low grade mucoepidermoid carcinoma and presence of intraepithelial duct like structures. With advances in modern immunohistochemical markers sialogenic origin is now not supported and odontogenic origin is thought of, but these studies do not confirm the definite odontogenic origin of GOC due to overlapping of markers used. The additional support for their odontogenic origin is provided by their association with other odontogenic tumours such as Ameloblastoma and keratocystic odontogenic tumour (KCOT). Reported here is a case of GOC in association with an odontome which will strengthen the odontogenic origin of this relatively rare jaw cyst.*

**Key words:** Glandular odontogenic cyst, Odontome.

### INTRODUCTION

Glandular odontogenic cyst was first reported in 1987 by Padayachee and van Wyk as sialo-odontogenic cyst, because of its

histopathologic resemblance to salivary gland tissue, such as presence of mucous secreting cells and intraepithelial duct like structures with mucous material.<sup>1</sup>Gardner et al (1988) reported a case of

hybrid GOC with ameloblastoma, and suggested the term glandular odontogenic cyst, as its association with ameloblastoma was a strong evidence of the odontogenic origin of this cyst.<sup>2</sup>

World Health Organization (WHO) Histological Typing of Odontogenic Tumour by Kramer et al (1992) also termed this lesion as GOC and classified it under the heading of developmental odontogenic cysts. WHO 1992 defined GOC as “a cyst arising in the tooth-bearing areas of the jaws and characterized by an epithelial lining with cuboidal or columnar cells, both at the surface and lining the cyst-like spaces within the thickness of the epithelium”.<sup>3</sup>

Although it is a well recognized entity the controversy is still unresolved about its origin, nevertheless it is an important lesion to recognize and diagnose, because of its aggressive behavior and a tendency to recur after conservative treatment.<sup>4</sup> Its association is reported with ameloblastoma and kerato-cystic odontogenic tumor.<sup>2,5,6</sup>

It appears that GOC may have a wide clinicopathologic spectrum, ranging from a small simple cyst to a somewhat neoplastic one resembling low-grade mucoepidermoid carcinoma (MEC), with local aggressive potential and recurrence.<sup>13</sup> The cyst also has histopathological similarities with lateral periodontal cyst (LPC) and botryoid odontogenic cyst (BOC), hence in spite of detailed histological features described by Gardner et al<sup>2</sup> and WHO<sup>3</sup>, a precise diagnosis of GOC is some time difficult.

Kaplan et al (2005) proposed the criteria for histological diagnosis of GOC. These were divided into major and minor criteria, wherein they suggest at least focal presence of each of the major ones mandatory for diagnosis, while the minor criteria supported the diagnosis but were not mandatory.<sup>14</sup>

**The major criteria include:<sup>14</sup>**

- I. Squamous epithelial lining, with a flat interface with the connective tissue wall, lacking basal palisading.
- II. Epithelium exhibiting variations in thickness along the cystic lining with or without epithelial spheres or whorls or focal luminal proliferation.
- III. Cuboidal eosinophilic cells or hob-nail cells.
- IV. Mucous (goblet) cells with intraepithelial mucous pools, with or without crypts lined by mucous producing cells.
- V. Intraepithelial glandular, microcystic or duct-like structures.

**The minor criteria include:**

- I. Papillary proliferation of the lining epithelium.
- II. Ciliated cells.
- III. Multicystic or multiluminal architecture.
- IV. Clear or vacuolated cells in the basal or spinous layers.

These criteria can be used for narrowing down the differential diagnosis or for definite diagnosis of GOC. In the present case diagnosis of GOC was made based on these criteria.

**CASE REPORT**

A 14 year old female patient presented with a complaint of pain and swelling in lower left jaw since 15-20 days. The pain was continuous and throbbing in nature mimicking acute pulpitis. There was no relevant dental as well as medical history. Extra oral examination revealed small, round to oval swelling approximately 2 X 2 cm in size, over left mandibular body causing slight facial asymmetry. (fig: 1)

Intraoral examination showed round to oval swelling of approximately 2 X 1.5 cm size, over buccal cortical plate extending from 34 to mesial of 36, obliterating the buccal vestibule. The swelling was of bluish color with diffuse borders. On palpation the swelling was soft to firm in consistency. (fig. 2)

Orthopantomogram (OPG) showed radiolucent lesion approximately of size 2X 1.5 cm. present between and apical to roots of 35 and 36. The radiolucency was unilocular, with well-defined sclerotic (radiopaque) borders. A radiopaque mass resembling partially formed crown of a tooth was present in the alveolar crest region between 35 and 36. There was displacement and resorption of roots of 35 and 36. Secondly there was presence of supernumerary tooth bilaterally in lower first premolar region. (fig 4. a)

On aspiration of lesion clear, low-viscosity, straw coloured fluid, admixed with blood was obtained with the collapse of buccal wall of swelling. (fig 3)

This suggested perforation of cortical plate of jaw bone. Surgical enucleation of the lesion was performed. The specimen comprised of two soft

tissue and one hard tissue bits. The hard tissue grossly reassembled the cuspal portion of a developing crown (fig 5. 3).

On microscopic examination soft tissue bit from central part of the lesion (fig:5.2 ) showed cystic cavity lined by non-keratinised stratified squamous epithelium of variable thickness, the connective tissue epithelial junction was flat and in some areas epithelium was separated from underlying connective tissue. The superficial cells of cystic lining were tall columnar with cilia resembling the hobnail cells interspersed with mucous producing goblet cells which were PAS positive. The deeper part of the epithelium showed round to oval cells without basal cell palisading. In some areas intraepithelial, microcyst or duct like structures were seen (fig 6: C & D). The deeper part of connective showed proliferation of odontogenic epithelial island (fig 6: F).

The soft tissue bit obtained from peripheral part of the lesion (fig: 5.1) showed thin elongated strip hard tissue formation along the fibrous capsule. This hard tissue consisted of irregularly arranged areas of dentinal tubules interspersed with soft tissue and resembled with an odontome (fig 6. E).

The cystic lining on histopathological evaluation satisfied all five major and two of four minor criteria for diagnosis of GOC as given by Kaplan et al (2005). The final diagnosis was made as "Glandular Odontogenic Cyst associated with odontome", based on clinical, radiographic and histopathological features.



Fig:1Extraoral picture showing slight swelling in lower left mandibularregion.



Fig 2: Intraoral picture showing round to oval swelling in lower left vestibule associated with 34,35 and 36.





Fig 3: Aspiration revealed 1 ml of clear, fluid which was admixed with blood giving straw color.



Fig4(a) : Unilocular radiolucent lesion with sclerotic borders approximately of size 2X 1.5 cm. between 35 and 36. displacement and resorption of roots of 35 and 36 can be seen. Also there was presence of supernumerary teeth bilaterally in 34 and 44.



Fig 4(b) : The radiopaque mass resembling partially formed crown of a tooth present in the alveolar crest region

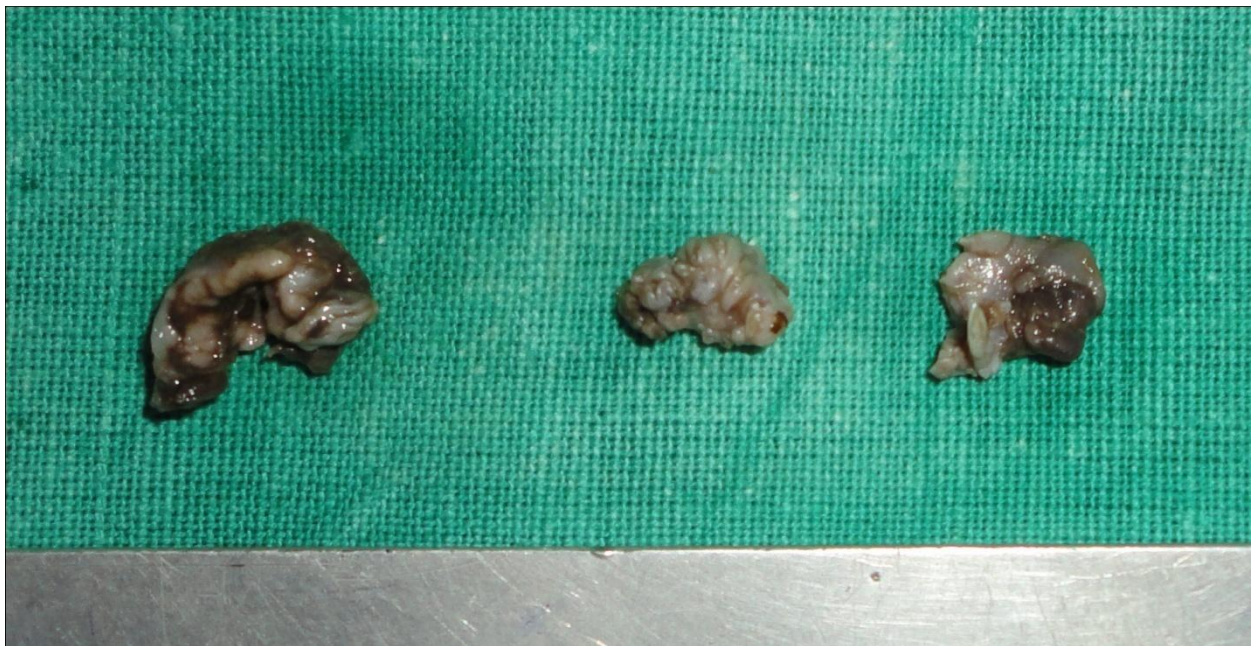


Fig 5 (1, 2, 3): Three bits of tissue 1: lower portion of lesion, 2: central portion and 3: crown like hard tissue embedded in lesional soft tissue



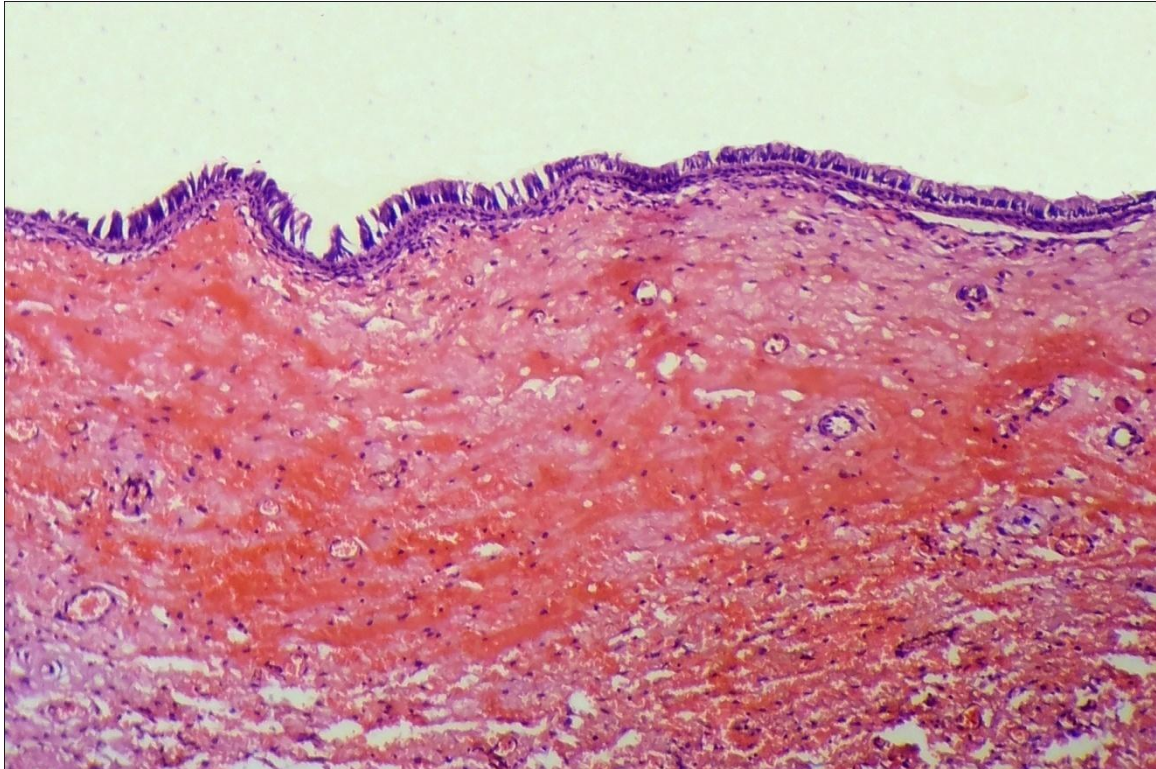


Fig 6.a (4X)

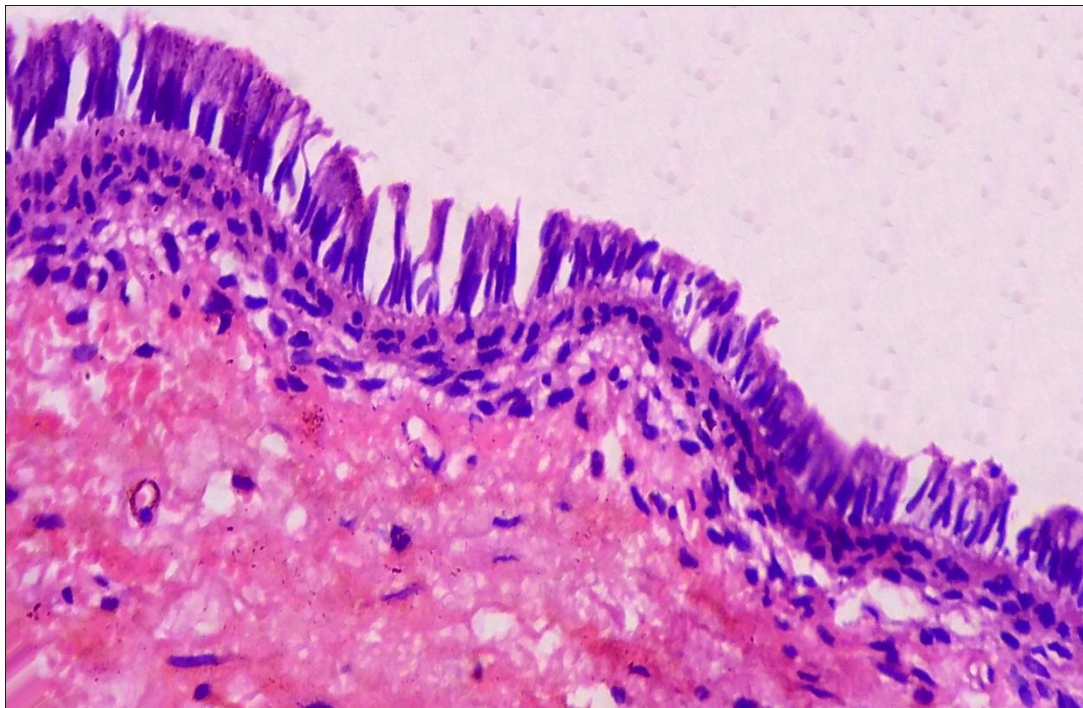


Fig 6.b (10X)



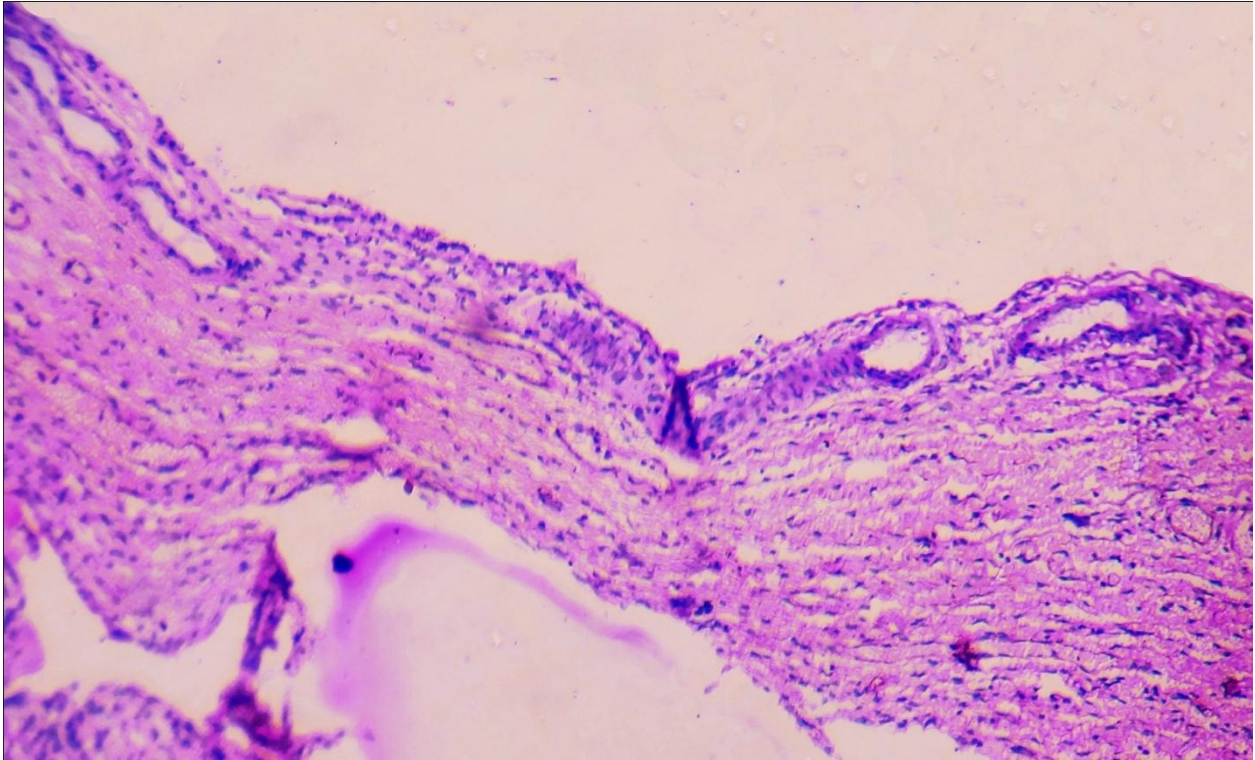


Fig 6.c (4X)

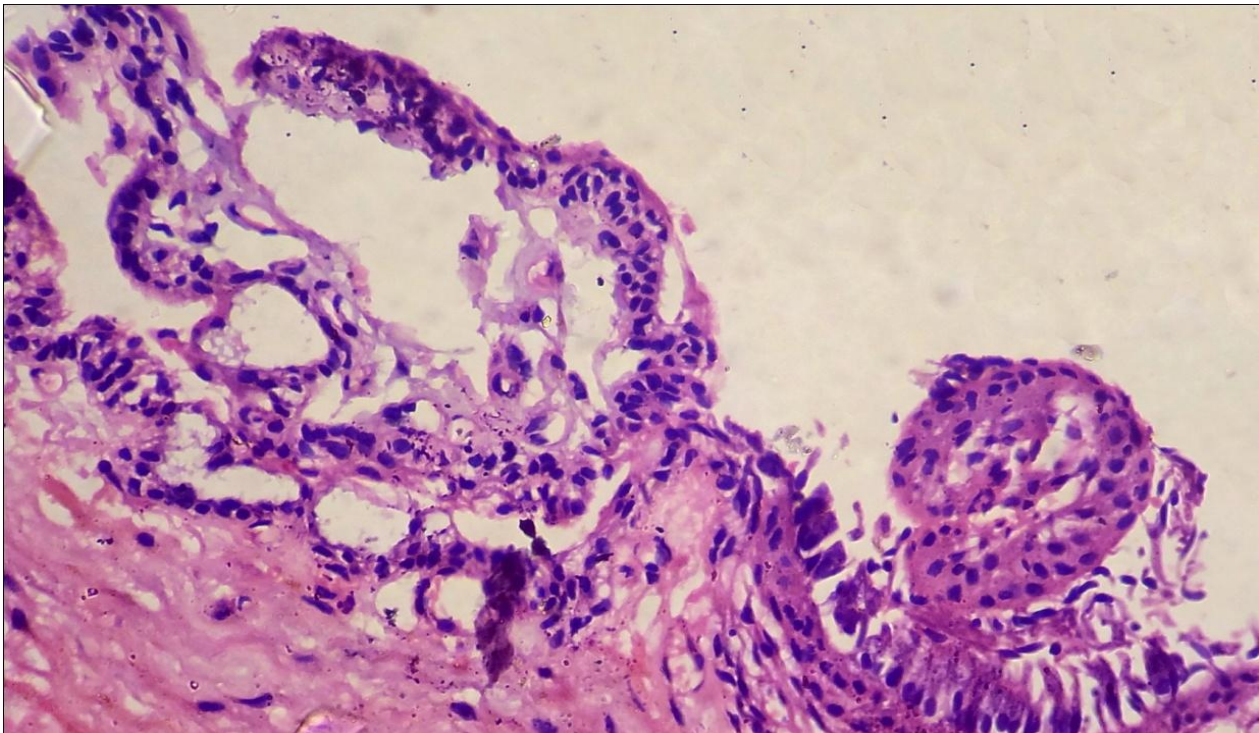


Fig 6.d (10X)



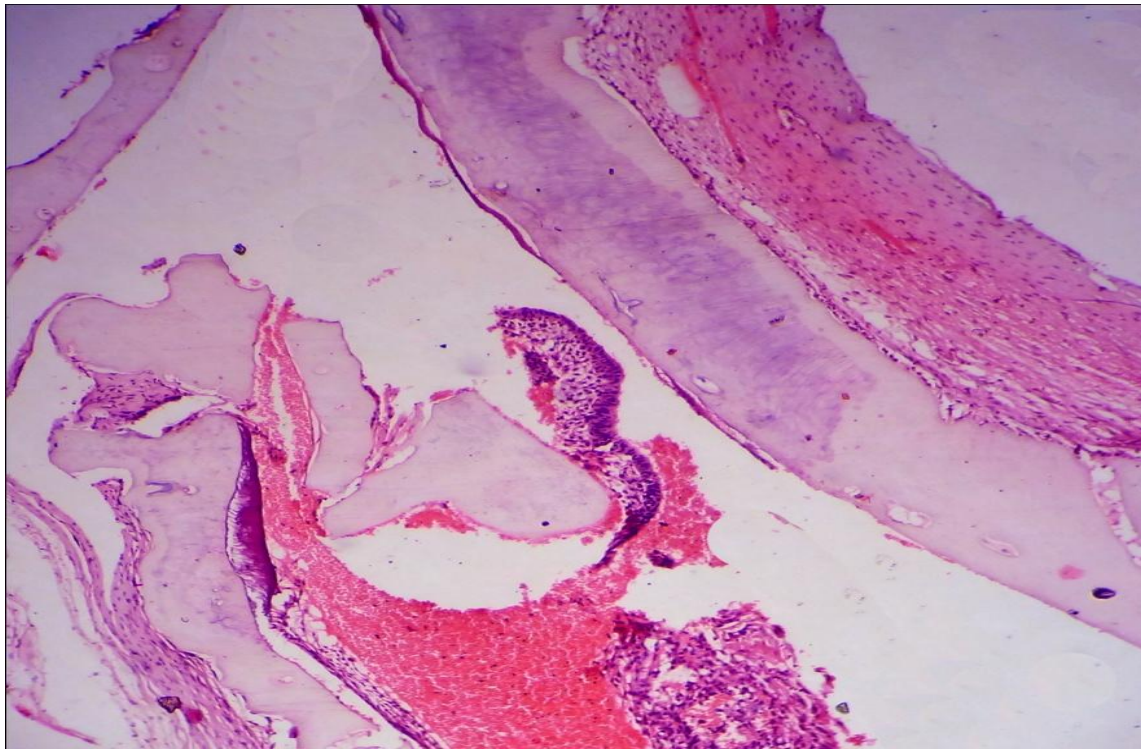


Fig 6.e (4X)

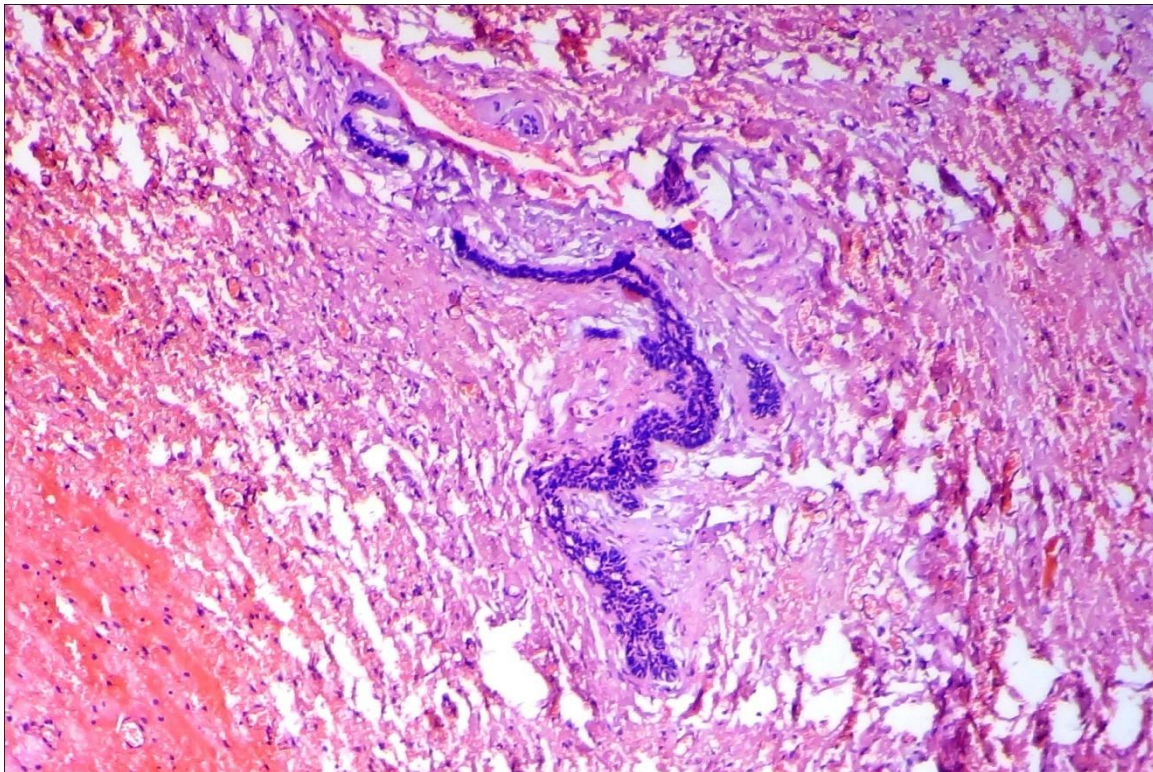


Fig 6.f (4X)

Fig 6 : a & b: Mucous (goblet) cells, Cuboidal eosinophilic or hob-nail cells and ciliated cells c: Epithelial lining with intraepithelial ductal structures and flat interface with the connective tissue wall, lacking basal palisading. d: Intraepithelial glandular, micro cystic or duct-like structures with luminal proliferation, e: decalcified section of odontome, f: odontogenic epithelial island in the connective tissue.

## DISCUSSION

In 1987 when Padayachee and Van Wyk first time reported a case of sialo-odontogenic cyst, it was thought to originate from salivary tissue entrapment within the jaw bones.<sup>1</sup> Gardner et al (1988)<sup>2</sup> and WHO (1992)<sup>3</sup> histological typing of odontogenic tumours described GOC as independent histopathological entity with probable odontogenic origin. Today, more than 115 cases of glandular odontogenic cysts have been published in the literature but only few cases of glandular odontogenic cyst are found to be associated with odontogenic tumour.<sup>16</sup>

The first such case of GOC associated with ameloblastoma was published by Gardner et al (1988). It showed histopathological features of both GOC and ameloblastoma in mixed plexiform and follicular pattern and was concluded to be an evidence to support an odontogenic rather than a sialogenic origin.<sup>2</sup> M Hisatomi et al (2000) diagnosed similar a case of GOC in association with ameloblastoma.<sup>5</sup> Kumaraswamy Naik L R et al (2008) claimed to diagnose a similar case of GOC in association with unicystic ameloblastoma in a 14-year-old boy. But, it did not fulfill all the histopathological diagnostic criteria given by Kaplan et al.<sup>17</sup>

Jung Hoon Yoon et al reported a case of GOC partly lined by epithelium with features of KCOT. In this particular case the cystic epithelium was predominantly thin and non-keratinized squamous, with cuboidal or ciliated epithelium with other features of GOC such as goblet cells, epithelial plaque of swirl-like arrangement and

intraepithelial micro cysts or duct-like structures. Also the histological features of KCOT like uniform layer of stratified squamous epithelium, with a hyperchromatic, palisaded basal cell layer and a corrugated parakeratotic surface of the lining epithelium.<sup>18</sup> Present case of GOC in association with odontome will help in strengthening the odontogenic origin of these cysts.

It comprises of 0.2 % of all odontogenic cysts.<sup>15</sup> It is most commonly seen in mandible in approximately 70% of the cases with more prevalence to anterior as compared to posterior region. The present case reports its unusual association with an odontome.

GOC is more common in males as compared to females with M: F ratio of 1.3:1. The majority of cases were reported in patients older than 30 years, with a mean of 45.7 years. GOC can occur within a wide age range of 14–75 years, but has never been reported in children <10 years of age.<sup>7</sup> Clinically, this lesion is generally painless, slow growing and its size can vary from less than 1 cm in diameter to large dimensions.<sup>8</sup> Small cysts may be asymptomatic, while larger ones can cause bone expansion accompanied by pain and paraesthesia.<sup>9</sup>

Radiographically it typically presents as a radiolucent lesion. The lesions can be unilocular (53.8%) or multilocular (46.2%), with well-defined borders in 95% of cases. Scalloped or sclerotic borders were described in 13% of cases.<sup>8,11</sup>



Radiographically, attempt was made to divide these cysts into small- and large-lesions. A lesion was classified as small if it involved not more than 2 adjacent teeth and was limited to the alveolar bone. A lesion was classified as large if it involved more than 2 teeth and extended beyond the alveolar bone or into adjacent anatomic structures, such as the mandibular ramus, maxillary sinus, or nasal cavity. An association with impacted teeth, root resorption and tooth displacement is common. Present case was small (<2 teeth) unilocular lesion associated with root displacement and resorption. Cortical bone perforation was also present in this case. Small lesions were uncommon (large: small - 4:1). Root resorption or tooth displacement was present in 22–24%, of cases. Cortical plate perforation was reported in 61% of the cases. This feature is an indication for the aggressive potential of GOC and is associated with high rate of recurrence.<sup>7</sup>

The microscopic features of GOC are a cystic cavity lined with non-keratinized, stratified, squamous epithelium of variable thickness, superficial layer of the epithelium consists of eosinophilic columnar to cuboidal cells sometime called as “hobnail” cells. The surface epithelium also has mucous producing goblet cells. The glandular or pseudoglandular structures, with intraepithelial crypts or microcysts are present which are lined by similar cells as on the surface. It is important to differentiate GOC from lateral periodontal cyst, botryoid cyst, radicular and dentigerous cysts with mucous metaplasia, surgical ciliated cyst and low-grade

mucoepidermoid carcinoma.<sup>19</sup> Some histopathological features of GOC like, localized plaque-like thickenings of the epithelium, subepithelial fibrous tissue formation with formation of multiple cysts, islands of odontogenic epithelium and the absence of inflammation suggest some similarities with lateral periodontal cysts or botryoid odontogenic cyst. Present case had more similarity with lateral periodontal cyst but presence of histological features such as hobnail cells, goblet cells, glandular duct like structure and absence of plaque like thickening of lining epithelium and absence of inflammatory cell infiltrate supported diagnosis of GOC. Also the biological behavior such as rapid growth, cortical plate perforation, root resorption and displacement support our diagnosis.

Although histogenesis was thought to be from salivary gland, the present histological features of GOC strongly suggest the odontogenic origin.<sup>7,11,12</sup>

To differentiate this cyst from low grade MEC Fabio Ramosa Pires et al (2004)<sup>21</sup> and Motais et al (2012)<sup>16</sup> studied the cytokeratin expression in GOC and MEC. They found GOC to be positive for CK 19 were as low grade MEC. Thus they concluded that CK 18 and 19 could be useful markers in differential diagnosis of GOC and also confirmed the odontogenic origin of GOC. Present case was negative for CK 18 but showed weak positivity for CK 19.

L Zhang et al (2010) studied immunohistochemical expression of proteins involved in sonic hedgehog



pathway which previously were studied as the pathogenic mechanism in KCOT development. They found positive expression of proteins SHH, PTCH, SMO and GLI1 in GOC epithelium thus pointing towards a neoplastic pathogenesis.<sup>22</sup>

Treatment recommendations for GOC in the literature are inconsistent and not evidence based due to the rarity of the condition. They are ranging from minor procedures, such as enucleation and curettage for small and unilocular lesions, to major surgery, such as marginal resection, peripheral ostectomy, and partial jaw resection for large and multilocular lesions.<sup>11</sup> The rate of recurrence of GOC as described in literature ranges from 21% to 55%. Recurrence is common and directly related with the size of the lesion only 14.4% of the small lesions recur in contrast to 85.6% of the large lesions.<sup>14</sup> Also cortical bone perforation, root resorption and multilocular lesions are found to have higher rate of recurrence.<sup>20</sup> So, it is advised to maintain the follow up for at least 3 years, preferably 7 years.<sup>14</sup> Present case was enucleated and under follow-up since 18 months without clinical and radiographic evidence of recurrence.

#### CONCLUSION:

Glandular odontogenic cyst was found to be associated with odontogenic tumour and that was the reason Gardner et al first time proposed the odontogenic origin of GOC. Although, few cases have been reported in the literature to show the association of GOC with ameloblastoma and KCOT, but there is no case report of its

association with Odontome. So, this case report is an addition to the association of GOC with odontogenic lesions, which further strengthens the odontogenic origin of the GOC. The association of GOC with other odontogenic neoplasms, aggressive biological behavior and positivity for proteins such as SHH, PTCH, SMO and GLI1 associated with odontogenic neoplasms may suggest its consideration as a neoplasm.

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