AfricanJournalofBiological Sciences

Glandular Odontogenic Cyst (GOC) – A review on well known rare entity.

Author details :

1) Author name: **Akilan R**
Affiliation: Senior Lecturer, Department of OMFS, Sree Balaji Dental College & Hospital, Chennai

2) Author name: **Rohini R**
Affiliation: Senior Resident, Department of Dentistry, GIMS, Greater Noida

Article History
Volume 6, Issue 5, 2024
Received: 22 May 2024
Accepted: 29 May 2024
doi:*10.33472/AFJBS.6.5.2024. 8413-8417*

Abstract:

The occurrence of a Cyst in jaws and in Maxillofacial region have been studied through variety of literatures amongst which Glandular Odontogenic cyst is considered to have rare incidence comprising of < 0.5% of all odontogenic cysts which has been showcased in a recent review tabulating around 200 cases in English literatures. The nomenclature probably is due to the fact that Glandular odontogenic cysts has its epithelial features resembling salivary glands or glandular differentiation. The notable features of this cyst includes its higher recurrence rate and its histopathological features sharing resemblance with Mucoepidermoid Carcinoma and other odontogenic lesions.

Keywords:

Glandular odontogenic cyst, mucoepidermoid carcinoma, hobnail cells, high recurrence

Introduction:

The definition of a 'Cyst' can be clearly explained as "A pathological (uni- or multilocular) sac that may or may not be lined by an epithelium and filled with a fluid, semifluid, or gaseous contents and not created by the accumulation of pus". Their occurrences in facial bony structures and their soft tissue counterparts has been well documented throughout literatures[1].

The Glandular Odontogenic Cyst (GOC) represents a rare and intriguing entity within the spectrum of odontogenic cysts, captivating the interest of oral and maxillofacial surgeons, pathologists, and researchers. Over the years, numerous studies have sought to elucidate the clinical, radiographic, histopathological, and molecular characteristics of GOC, shedding light on its diagnostic challenges, management considerations, and potential prognostic factors.

Historically, the glandular odontogenic cyst was initially reported by Padayachee et al., in 1987 who described odontogenic cysts having features resembling lateral periodontal cyst and central mucoepidermoid carcinoma and gave the term 'sialo odontogenic cyst'[2] and in
1988 Gardener et al gave the term 'Glandular Odontogenic cyst'. In 1992, the WHO has included GOC under developmental odontogenic cysts [3].

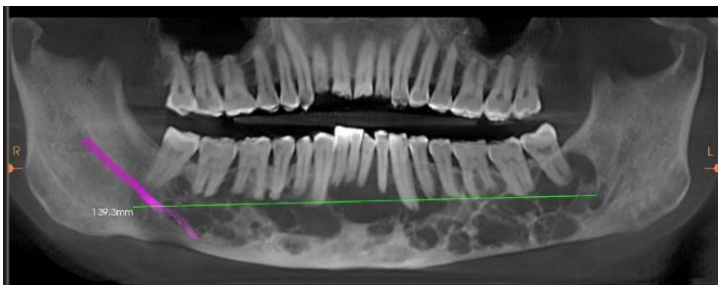
Clinical Characteristics:

Initial reports of GOC highlighted its predilection for the anterior mandible, although cases involving the maxilla have also been documented. Clinically, GOC often presents as a slow-growing, painless swelling, with radiographic findings typically demonstrating a welldefined radiolucency with smooth borders. However, its nonspecific presentation poses challenges in distinguishing it from other cystic lesions, necessitating a comprehensive diagnostic approach.The prevalence of GOCs is observed higher in middle aged adults with no gender predilection mostly in fifth and sixth decades of life[5][10]. Lesions are commonly associated with swelling/expansion in 43.5 to 87% which is the most common presenting complaint although about 75% are asymptomatic[11].

Radiographical features:

The radiographical appearanes includes rounded or oval lesion which can be unilocular or multilocular lesions usually with clear demarcated borders. The inconsistent clinical nature and intraosseous predilictions of these lesions demonstrate the importance of radiographic examinations like CT, CBCT for 3-dimesional analysis. Though GOC has the tendency to be unilocular, the ratio of unilocular and multilocular presentation has found to be equal with inconsistent radiographic features. Root resorption has been reported in 13.9 to 30% of lesions and tooth displacement in 24.4 to 50% of lesions[4].

(a)Axial CBCT section



(b) Reconstructed OPG

Histopathological Features:

Histologically, GOC exhibits a spectrum of epithelial patterns, including cuboidal, columnar, stratified squamous and pseudostratified epithelium, often interspersed with mucous cells. This diverse histological appearance underscores the complexity of its diagnosis and underscores the importance of careful microscopic examination. Additionally, immunohistochemical analysis has been employed to delineate its cellular origin and differentiate it from other odontogenic cysts and tumours. Kaplan et al and Fowler et al proposed numerous microscopic major and minor criteria.

According to World Health Organisation's classification of odontogenic cysts – 5th edition 2022, it emphasizes that even if characteristic, not all features are present in all cases but more features provide more confidence in the diagnosis rather it is considered to have seven of ten characteristic histopathological features are necessary to make GOC a diagnosis[3][5].

Amongst these presence of 'hobnail cells' in the lining epithelium seemed to be most characteristic features [6].

Molecular Pathogenesis:

While the exact pathogenesis of GOC remains elusive, molecular studies have provided insights into its genetic alterations and potential oncogenic pathways. Mutations in the PTCH and SMO genes, commonly associated with the Hedgehog signaling pathway, have been implicated in the development of GOC, suggesting a possible role in its pathogenesis. Furthermore, aberrant expression of various molecular markers, including Ki-67, p53, and Bcl2, has been reported in GOC, offering potential prognostic indicators for disease behavior and recurrence [7]. Demonstrating the *MAML2* rearrangement for intraosseous MEC is important in the differential diagnosis of these two lesions, though further studies are required to confirm this phenomenon [6].

Management Strategies:

Surgical enucleation with thorough curettage of the surrounding bone remains the mainstay of treatment for GOC. However, due to its propensity for recurrence, long-term follow-up is essential to monitor for disease persistence or progression. Recent advances in imaging modalities, such as cone-beam computed tomography (CBCT), have facilitated preoperative assessment and surgical planning, optimizing outcomes and reducing the risk of recurrence.

Enucleation, curettage and marsupialization prior to enucleation are the most common treatment for GOC but is associated with a recurrence rate of 21.6 to 50% .Thor et al[8] with their experiences highlights that GOC lesions managed conservatively have higher recurrence rate than those managed by peripheral ostectomy and block resection surgical methods[9].

Conclusion:

The literature on Glandular Odontogenic Cyst encompasses a diverse array of studies spanning clinical, radiographic, histopathological, and molecular aspects of this intriguing lesion. While significant strides have been made in elucidating its diagnostic features and management principles, many questions regarding its pathogenesis and behaviour remain unanswered. Continued collaborative research efforts are essential to unravelling the complexities surrounding GOC, ultimately enhancing our understanding and management of this rare odontogenic cyst.

References:

1. Shear M, Speight PM, editors. Cysts of the oral and maxillofacial regions. 4th ed. Oxford: Blackwell Munksgaard; 2007.
2. Padayachee A, Van Wyk CW. Two cystic lesions with features of both the botryoid odontogenic cyst and the central mucoepidermoid tumour: Sialo-odontogenic cyst? J Oral Pathol. 1987;16:499–504.
3. El-Naggar AK, Chan JKC, Takata T, Grandis JR, Slootweg PJ. The fourth edition of the head and neck World Health Organization blue book: editors' perspectives. Hum Pathol. 2017 Aug;66:10-12

4. Gorgis R, Christian Krarup SA, Reibel J, Nørholt SE. Glandular Odontogenic Cyst: a Case Report and Literature Review. J Oral Maxillofac Res. 2023 Jun 30;14(2):e4. doi: 10.5037/jomr.2023.14204. PMID: 37521326; PMCID: PMC10382194.
5. Fowler Craig B., Brannon Robert B., Kessler Harvey P., Castle James T., Kahn Michael A. Glandular odontogenic cyst: analysis of 46 cases with special emphasis on microscopic criteria for diagnosis. Dec;2011 Head Neck Pathol. 5(4):364. doi: 10.1007/s12105-011-0298-3
6. Soluk-Tekkesin M, Wright JM. The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2022 (5th) Edition. Turk PatolojiDerg. 2022;38(2):168-184. doi: 10.5146/tjpath.2022.01573. PMID: 35578902; PMCID: PMC9999699.
7. Tosios KI, KakarantzaAngelopaulou E, Kapranos N. Immunohistochemical study of bcl-2 protein, ki67 antigen and p53 protein in epithelium of glandular odontogenic cysts and dentigenous cysts. J Oral Pathol Med. 2000;29:139–44.
8. Thor A, Warfvinge G, Fernandes R. The course of a long-standing glandular odontogenic cyst: marginal resection and reconstruction with particulated bone graft, platelet-rich plasma, and additional vertical alveolar distraction. J Oral Maxillofac Surg. 2006 Jul;64(7):1121-8.
9. Kaplan I, Gal G, Anavi Y, Manor R, Calderon S. Glandular odontogenic cyst: treatment and recurrence. J Oral Maxillofac Surg. 2005 Apr;63(4):435-41
10. Chrcanovic BR, Gomez RS. Glandular odontogenic cyst: An updated analysis of 169 cases reported in the literature. Oral Dis. 2018 Jul;24(5):717-724
11. Mascitti M, Santarelli A, Sabatucci A, Procaccini M, Lo Muzio L, Zizzi A, Rubini C. Glandular odontogenic cyst: review of literature and report of a new case with cytokeratin-19 expression. Open Dent J. 2014 Feb 7;8:1-12.