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Case Report

Dentistry Section

Glandular Odontogenic Cyst of Maxillary Anterior Region: Report of a Rare Case

AMITA ADITYA[1], ANTARA KORE[2], SHRUTI PATIL[3], DAISY HAPPY[4], VAISHNAWI SAWANT[5]

ABSTRACT

Glandular Odontogenic Cyst (GOC) is a rare odontogenic cyst that indicates the pluripotency of the oral epithelium due to the exhibition of glandular characteristics. It is more prevalent in the mandibular anterior region and shows a predilection in males. The cyst was first described by Padayachee and van Wyk in 1987. It has non distinct clinical and radiographic features but very characteristic histopathological features. Here, the authors report a rare case of GOC occurring in a 36-year-old male patient in the maxillary anterior region. An intraoral periapical radiograph revealed a large unilocular radiolucency from the apex of 11 to the mesial aspect of 15. The cyst was enucleated under local anaesthesia, and endodontic treatment was performed for the associated teeth. No recurrence was seen at the two-month follow-up.

Keywords: Anterior maxilla, Pluripotent, Radiograph, Sialo-odontogenic cyst

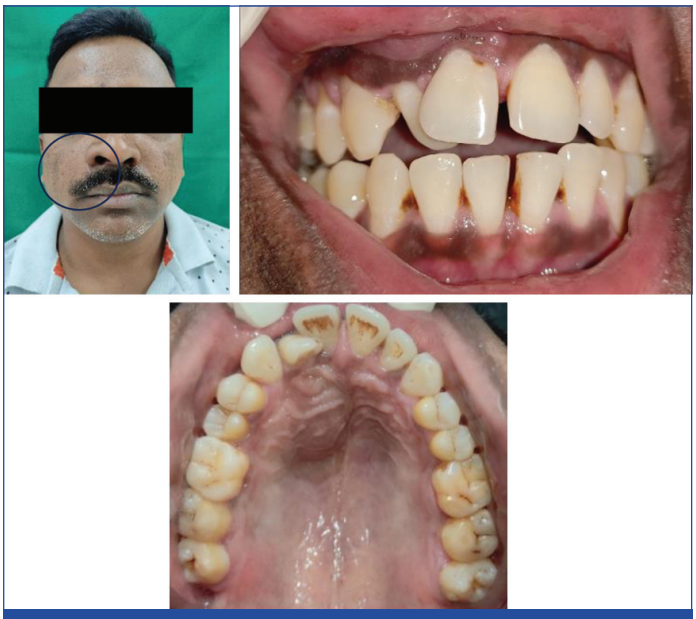
CASE REPORT

A 36-year-old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in the upper front region of the jaw since one and a half. The patient had been apparently alright two months prior, then started noticing a small swelling on the right-side of the upper jaw, which gradually increased to its present size. The swelling was painless, and the patient noticed tooth mobility. There was no history of trauma. The patient has been under medical care for hypertension since last four years and also mentioned a history of tobacco chewing 1-2 times a day since 10 years.

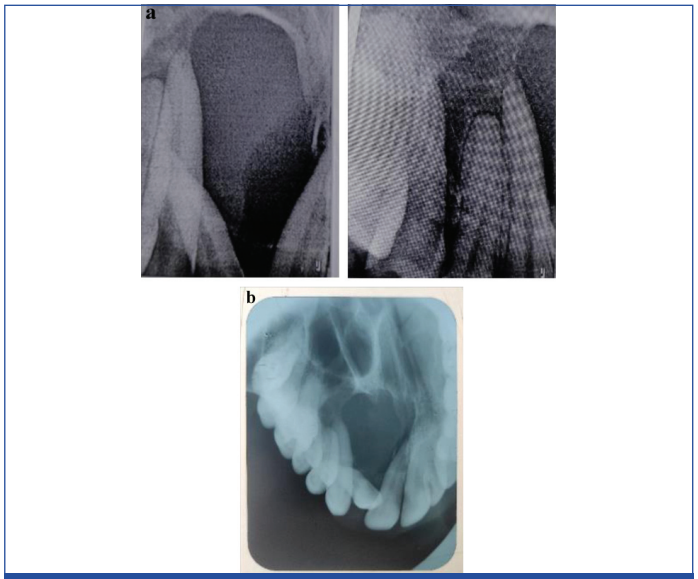
Clinical examination revealed an extraoral diffuse swelling on the right-side of the face, extending mediolaterally from the nasal septum to 2 cm distal to the right ala of the nose and supero-inferiorly from the right infraorbital margin to the right upper corner of the angle of the mouth. The colour was similar to the adjacent skin, and the surface structure was smooth. It was afebrile, soft to firm in consistency, and non tender on palpation. Lymph nodes were non palpable and non tender on extraoral palpation [Table/Fig-1].

During intraoral examination, distal tipping of 11 and mesio-palatal tipping of 12 were observed. Midline spacing and Grade I mobility were noted with the maxillary right anterior teeth, and tenderness on percussion was found in the maxillary right lateral incisor, canine, and first premolar. A single partially well-defined intraoral swelling was seen extending from the labial aspect of 11, 12, 13, obliterating the labial vestibule and extending palatally to the midpalatal region up to the middle third of the palate, and mediolaterally from the midpalatal region to 14. The swelling appeared ovoid in shape, approximately 3×2 cm in size, with a smooth surface [Table/Fig-1]. On palpation, it appeared soft, fluctuant, tender, non-mobile, and non compressible, with a slight rise in local temperature. Considering the history and the clinical presentation, a provisional diagnosis of an infected odontogenic cyst was made, with a differential diagnosis of a benign salivary gland tumour in the right anterior maxillary region (Pleomorphic adenoma of minor salivary gland).

Intraoral periapical radiographs and a topographic occlusal radiograph of the region revealed tipping of teeth and a large unilocular radiolucency from the apex of 11 to the mesial aspect of 15, partially surrounded by a scalloped, hyperostotic border [Table/Fig-2a,b]. For further radiographic evaluation, cone beam computed tomography was performed, revealing a well-defined, unilocular, expansile radiolucency measuring 18.40 mm supero-inferiorly and

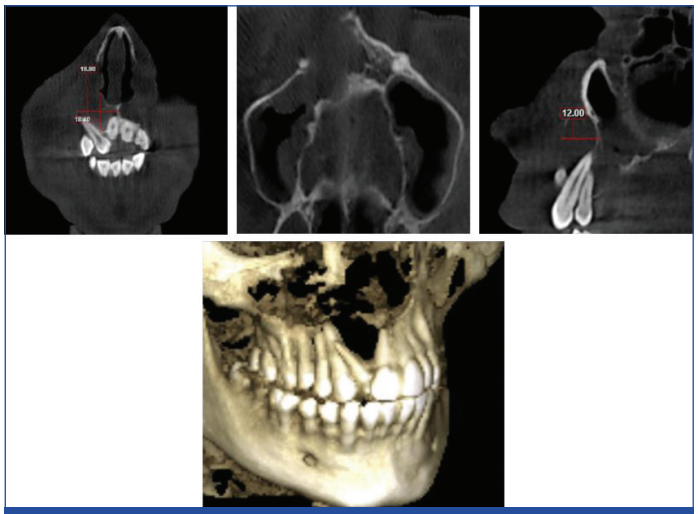


[Table/Fig-1]: Clinical presentation of the lesion.



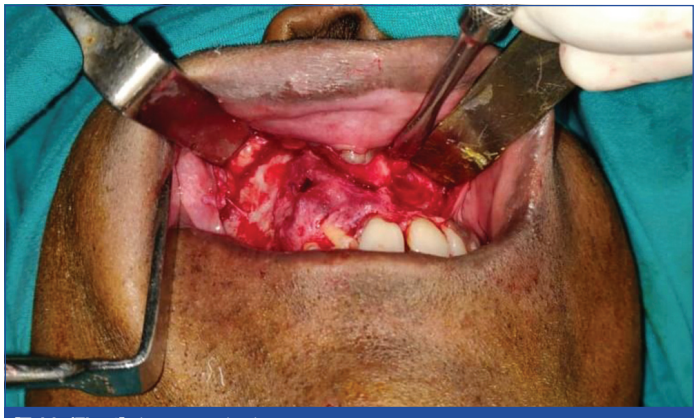
[Table/Fig-2]: Intraoral periapical radiograph: (a) and topographic occlusal radiograph; (b) revealed tipping of teeth and large unilocular radiolucency from apex of 11 to mesial aspect of 15.

18.80 mm mesio laterally, with ill-defined, irregular margins and a partially corticated border. In the axial view, expansion of the palatal cortical plate and destruction of the labial cortical plate involving the nasal floor and floor of the maxillary sinus were observed [Table/Fig-3]. A radiographic diagnosis of an odontogenic keratocyst was made, with a differential diagnosis of unicystic ameloblastoma, globulomaxillary cyst, and adenomatoid odontogenic tumour. No abnormalities were detected in the haematological examination.



[Table/Fig-3]: Cone beam computed tomography images revealing well-defined, unilocular, expansile lesion.

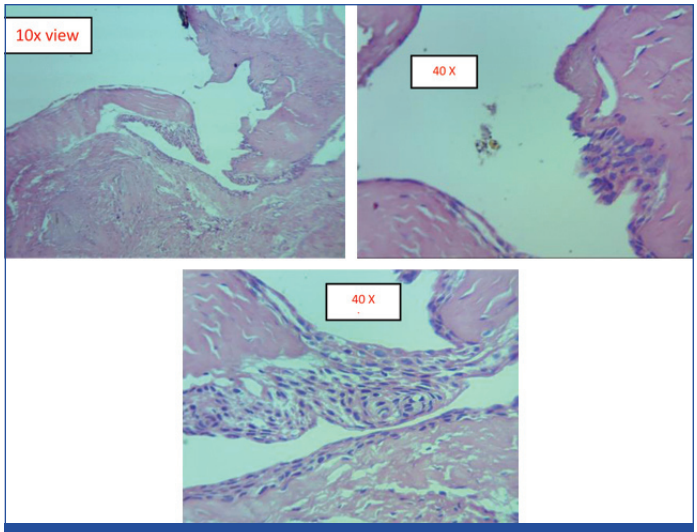
Fine needle aspiration cytology showed chronic inflammatory cells, few squamous cells, and debris, suggestive of an inflammatory cyst. The cyst was then enucleated under local anaesthesia, and endodontic treatment was performed for the associated teeth [Table/Fig-4]. The Haematoxylin and Eosin (H&E) stained section showed a non keratinised thin cystic epithelial lining 2-3 cell layers thick with foci of epithelial thickening and ciliated cells. Hyalinised subepithelial connective tissue, muscle, and bone tissues were also seen. The histopathological features were suggestive of GOC [Table/Fig-5]. Considering the high rate of recurrence of GOC, the patient has been kept under long-term follow-up [Table/Fig-6]. A recent two-month follow-up showed the patient to be completely asymptomatic with no recurrences.



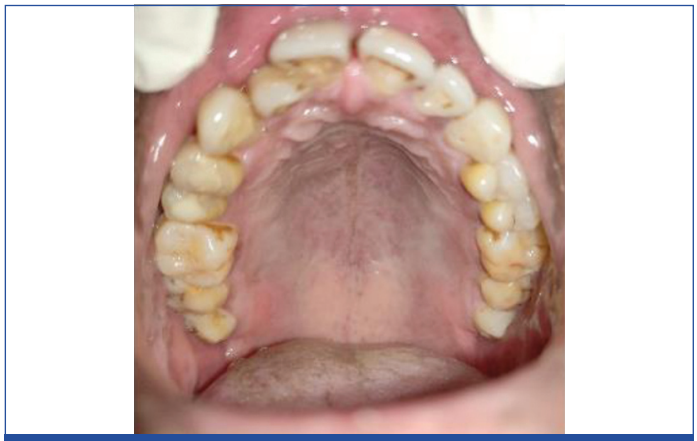
[Table/Fig-4]: Intraoperative Image.

DISCUSSION

The GOC is a relatively rare odontogenic cyst of the maxillofacial region [1]. It was initially named "Sialo-odontogenic Cyst," as it showed mucous cells and pools of mucin in the epithelial lining, which was often lined by eosinophilic cuboidal cells resembling salivary gland ducts [2]. The name "GOC" was suggested by Gardner DG et al., in 1988, owing to the fact that the cyst wall epithelium was odontogenic in nature and contained mucin elements, with the absence of salivary tissue [2]. The rarity of its occurrence, with an incidence of 0.012%-1.3% and the absence of any characteristic clinical and radiographic features, makes it a



[Table/Fig-5]: H&E stained section showed non keratinised thin cystic epithelial lining 2-3 cell layer tick with foci of epithelial thickening and ciliated cells.



[Table/Fig-6]: Postoperative, one month recall clinical presentation.

difficult entity to diagnose, and to date, only a few cases have been reported [3-5].

The GOC is an uncommon lesion, comprising only 0.012% to 1.3% of the cystic lesions of the jaw [6]. It is aggressive in nature and most commonly occurs in the mandibular symphyseal region, with only a 30% incidence seen in the maxilla [7]. Large lesions are associated with clinical symptoms of pain and numbness, whereas smaller lesions are largely asymptomatic. It is important for the lesion to be distinguished from dentigerous cyst, ameloblastoma, and odontogenic keratocyst owing to its higher recurrence in comparison with other lesions. This aggressive nature of the cyst is speculated to be due to penetration of the bone cortex, which leads to a high rate of recurrence after conservative therapy [8].

Radiographically, it may appear as unilocular or multilocular radiolucency with a well-defined margin and may mimic dentigerous cyst and ameloblastoma. In the present case, the clinical and radiographic presentation mimicked any regular odontogenic cyst. It was only after the histopathological examination that the actual character of the lesion was revealed. Immunohistochemical confirmation confirms the odontogenic nature of GOC. Histopathologically, it shows very distinct features, for which major and minor microscopic criteria have been suggested by Kaplan for the diagnosis [7]. It needs to be differentiated from low-grade central mucoepidermoid carcinoma because of similar histopathological features [9].

For treatment, marginal resection or peripheral ostectomy is recommended as they result in a low recurrence rate [10,11]. The risk of GOC recurrence varies between 21% and 55% [12]. In the present case, since the authors had done enucleation, they intended to keep the patient under long-term follow-up in order to monitor any recurrences. Similar cases from the last 20 years have been tabulated in [Table/Fig-7] [12-28].

Author	Age/Sex	Presentation	Region affected in maxilla (Teeth)	Radiographic features	Treatment	Follow-up	Recurrence
Noffke C and Raubenheimer EJ [13] (2002)	15/M	Swelling, tooth displ	Max. (22-23)	Uniloc. RL	-	-	No
	17/M	Swelling tooth displ	Max. (12 -17).	Uniloc. RL	-	-	No
Kaplanl I et al., [12] (2005)	15/M	Swelling	Max.	Multiloc. RL, Root resorption	Partial Maxillectomy	5 y	No
	29/M	Swelling, paresthesia Tooth displacement	Max.	Uniloc. RL	Marsupialization	6 y	No
	25/F.	Swelling	Ant. Max.	Uniloc. RL root resorption	Enucleation	4 y	No
	54/M	Swelling, tooth mobility	-	Uniloc. RL tooth displ	Enucleation	3 y	Yes
Qin XN et al., [14] (2005)	37/F	Swelling, pain	(22-24)	Multiloc. RL tooth displ	Curettage	1 y and 10 m	No
	52/F	Swelling	(16-25)	Multiloc. RL	Curettage	5 y	No
	27/F	-	(11-15)	Uniloc. RL	Curettage	No	-
	28/M	Swelling	(21-27)	Uniloc. RL	Curettage	1 y and 10 m	No
	28/M	Swelling, pain	(21-26)	Uniloc. RL	Curettage	3 y and 4 m	No
	40/M	Swelling	(11-16)	Uniloc. RL	Curettage	5 y and 2 m	No
	25/M	Swelling	(13-16)	Uniloc. RL	Curettage	1 y and 2 m	No
	22/M	-	(21-23)	Uniloc. RL	Curettage	No	
	59/F	Swelling	(13-15)	Uniloc. RL	Curettage	4 y	No
Sittitavornwong S et al., [15] (2006)	57/F.	Swelling	(21-24)	Multiloc. RL tooth displ	Excision	3 m	No
Nair RG et al., [16] (2006)	45/F	Swelling, pain	(16-18)	Uniloc. RL	Enucleation	No	
Manzini M et al., [17] (2009)	27/F	Pain, pruritus, Inflammated mucosa	Ant. Max	Uniloc. RL	Enucleation	No	
Prabhu S et al., [18] (2010)	47/F	Swelling	Max.	Uniloc. RL	Enucleation	5 y	No
Amberkar VS et al., [19] (2011)	29/M	Swelling, pain	Max. (13-17, 23- 27)	Uniloc. RL	-	-	-
Guruprasad Y et al., [20] (2011)	17/F	Swelling, pain	Post. Max.	Uniloc. RL	Enucleation and curettage	1 y	No
Chandra S et al., [21] (2016)	70/F	Swelling	Max anterior	Multilocular RL	Surgical excision	-	-
Ogura I et al., [22] (2017)	74/M	Swelling	Max	Unilocular	Cystectomy		
	45/F	Swelling	Max	Unilocular	Cystectomy	-	-
Gurler G et al., [23] (2017)	39/F	Swelling	Post. maxilla	Unilocular	Enucleation	-	-
	33/M	Swelling	Ant. maxilla	Unilocular	Enucleation	-	-
Frazier JJ and Flint DJ [24] (2017)	13/M	-	Ant. maxilla	Unilocular	Enucleation	-	-
Ojha B et al., [25] (2018)	50/M	Swelling	Max	Unilocular	Enucleation		
	50/F	Swelling	Post maxilla	Unilocular	Enucleation		
Changani K et al., [26] (2018)	50/F	Swelling	Ant. Max	Unilocular	Enucleation		
Kothari C et al., [27] (2022)	16/M	Swelling, pain	Post. Max	Uniloc. RL	Enucleation, peripheral osteoctomy	-	-
Arora G et al., [28] (2022)	40/M	Swelling, pain	Ant. maxilla	Unilocular	Enucleation	1 y	No
Present case	36/M	Swelling	Ant. maxilla	Unilocular	Enucleation	2 m	No

[Table/Fig-7]: Summary of glandular odontogenic cyst in maxillary jaw in last 20 years [12-28].
Uniloc: Unilocular; RL: Radiolucency; Ant: Anterior; Post: Posterior

CONCLUSION(S)

Although the glandular odontogenic cyst is an uncommon entity, it must be considered in the differential diagnosis of cystic lesions of the jaw due to its aggressive clinical nature and high recurrence rate. Its non specific clinical and radiographic features pose a diagnostic challenge for oral physicians.

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PARTICULARS OF CONTRIBUTORS:
1. Professor, Department of Oral Medicine and Radiology, Dr. D. Y. Patil Dental College and Hospital, Dr. D. Y. Patil Vidyapeeth, Pimpri, Pune, Maharashtra, India.
2. Postgraduate Student, Department of Oral Medicine and Radiology, Sinhgad Dental College and Hospital, Pune, Maharashtra, India.
3. Medical Data Analyst, Department of Oral Medicine and Radiology, Vasta Bioinformatics Pvt. Ltd., Pune, Maharashtra, India.
4. Professor, Department of Periodontics, Sinhgad Dental College and Hospital, Pune, Maharashtra, India.
5. Postgraduate Student, Department of Oral Medicine and Radiology, Dr. D. Y. Patil Dental College and Hospital, Pimpri, Pune, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Amita Aditya,
Professor, Department of Oral Medicine and Radiology, Dr. D. Y. Patil Dental College and Hospital, Sant Tukaram Nagar, Pimpri, Pune-411018, Maharashtra, India.
E-mail: dr.amita30@rediffmail.com

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