Case Report

Gardner's Cyst Enswathing the Maxillary Antrum: Report of A Rare Case and Review of Literature

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ABSTRACT

Glandular odontogenic cyst (GOC) was named so by Gardner and the credit of discovery can be attributed to the work of Padayachee and Van Wyk (1987). The incidence of GOC is said to be between 0.012% and 1.3%. Even so, a little over 100 cases are reported in English literature. Mandible is more commonly affected than maxilla (20%) with almost 80% cases reported, with an anterior predilection. Even though GOC affecting maxilla is discussed in the literature, to the best of our ability, we could find that, in India, less than five cases affecting the maxillary sinus is ever reported, with none explaining about such a huge cyst that has encompassed the whole of the ipsilateral maxillary sinus. The aim to publish this case report was to understand the rarity in pathology, which GOC encompasses. Such rare cases if reported need to be published for the knowledge, prompt diagnosis, and appropriate treatment planning. Any pathology in the head and neck region should be seen with an eagle's eye for appropriate management to increase patients' quality of life.

KEYWORDS: *Glandular odontogenic cyst, maxillary antrum cyst, rare cyst*

INTRODUCTION

Gardner's cyst or Glandular odontogenic cyst (GOC) is a very rare (0.012%–1.3%) developmental cyst with aggressive growth potential. The mandible is found to be affected four times more often than the maxilla. A case of a sizeable cyst with unabridged extension into the maxillary sinus is discussed. Kaplan *et al.*[1] showed that the cyst habitually affects middle-aged men, whereas our patient is a young female.

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Only 20% of cases have been reported so far in literature about GOC in maxillary antrum. In Indian scenario, hardly less than five cases have been reported so far.[2] The important thing that needs to be noticed is the aggression

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of pathology, as we all know that maxillary antrum is a hollow cavity with high vascularity, which gives it the pathway of least resistance to increase in size. Such cases if unnoticed or untreated can lead to various fatal complications as the pathway to cranial base through sinuses and orbital region is very near to approach.

The aim to publish this case report was to understand the rarity in pathology, which GOC encompasses. Such rare cases if reported need to be published for the knowledge, prompt diagnosis, and appropriate treatment planning.

Case Report

A female patient aged 24 years reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of swelling in the midpalatal region since a year. On elaborating, the swelling was gradual in onset, slowly enlarging, and was not associated with pain, fever, or discharge from nose or locally. There was no evidence of infraorbital paresthesia. The patient gave a history of fractured 21,One and half years ago as a result of trauma. She gave a history of betel nut chewing occasionally. No significant medical history was present.

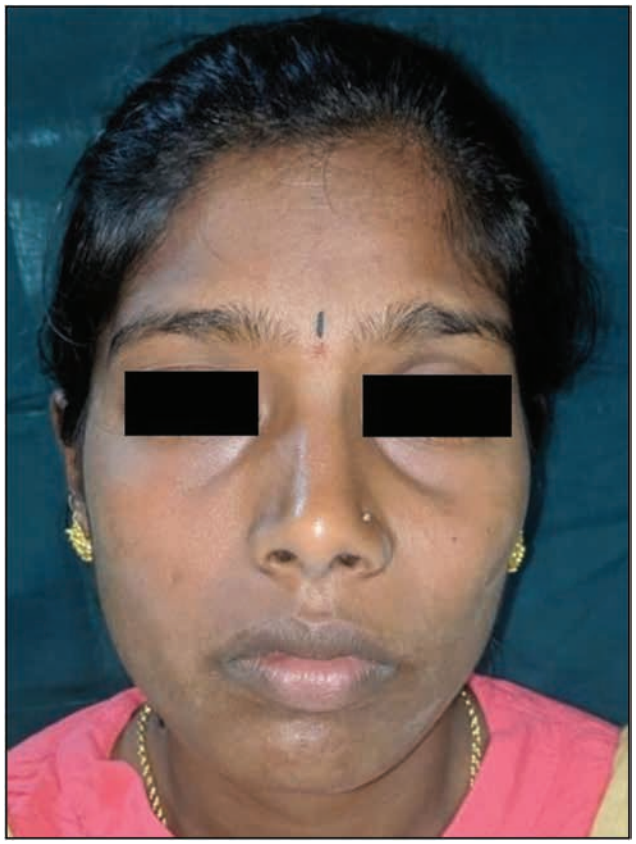

Figure 1: Frontal view showing a diffuse swelling in relation to the left malar region

On extraoral examination, a diffuse swelling was present in the left malar region extending mediolaterally 1 cm from left ala of nose to 4 cm medial to tragus and superoinferiorly 2 cm from left infraorbital region to 1.5 cm above the left corner of the mouth [Figure 1]. Skin over the swelling was normal, and the swelling was firm, non-fluctuant and non-tender. It was noted that the patient has an adequate mouth opening.

On intraoral examination, a well-localized swelling measuring 2 × 1.5 cm approximately, involving the midpalatal region was noted [Figure 2] with slight erythema present over the mucosa. The swelling on palpation was seen to be soft, fluctuant, and non-tender. It was noted that there was a firm swelling present with respect to the buccal cortex of the left maxilla as well, crepitus was elicited on palpation. Electronic pulp testing was carried out, and it was noted that 21–25 teeth did not show any response, 26–28 teeth elicited delayed response. Aspiration of the contents was performed. It yielded a pale yellow-colored fluid. Protein estimation of the fluid was reported to be 8.6 mg/dL [Figure 3].

Panoramic radiography was performed, which showed a unilocular radiolucency with sclerotic borders, extending into the left maxillary sinus, primarily surrounding the roots of 21–26 teeth [Figure 4]. Computed tomography (CT) Paranasal sinus view (PNS) view showed a well-corticated cystic lesion 4 × 4 cm, involving maxilla on left side with extension into the maxillary sinus. Erosion of the anterolateral wall of the maxillary sinus along with palatal roof was noted [Figure 5].

Incisional biopsy was performed via buccal approach, it was noted that the lesion had expanded and eroded the buccal cortex, and was extremely thin as a number 15 blade was used to remove the overlying bone to expose the lesion. The sections were submitted and the report was awaited.

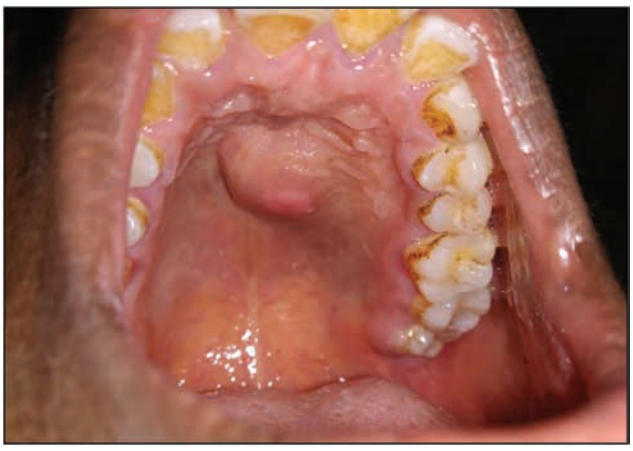

Figure 2: Well-defined palatal swelling crossing the midline

The diagnosis of Gardner's cyst is arrived upon when the superficial layer of the epithelial lining consists of columnar or cuboidal cells, referred to as hobnail, occasionally with cilia or filiform extensions of the cytoplasm. The incisional biopsy histopathologic report in our case came as GOC [Figure 6]. The cystic cavity was seen to be lined by nonkeratinized stratified epithelium with a thickness of 3–4 layers of cuboidal cells, with numerous mucous cells and stellate reticulum-like cells. In few areas, epithelial cells were seen to be proliferating due to inflammation. Separation of the epithelium from underlying connective tissue was also observed in few areas. Supporting connective tissue consisted of loosely arranged collagen fibers moderately infiltrated with chronic inflammatory cells. Deeper to that osteoid tissue was also present. The diagnosis of GOC was made only by histopathologic analysis, which again stresses the role of oral and maxillofacial pathologist in our field. However, immunohistochemical examination was also proposed to patient but it was not performed due to financial concern of the patient. Caldwell-Luc approach [Figure 7] was used to expose the lesion [Figure 8]. The cyst was enucleated [Figure 9] and packing of the antrum was carried out with povidone–iodine impregnated ribbon gauze, followed by which a priorly fabricated acrylic plate [Figure 10] was used to cover the defect and the specimen was send for biopsy and the mucoperiosteal flap elevated was sutured with Vicryl 4-0.

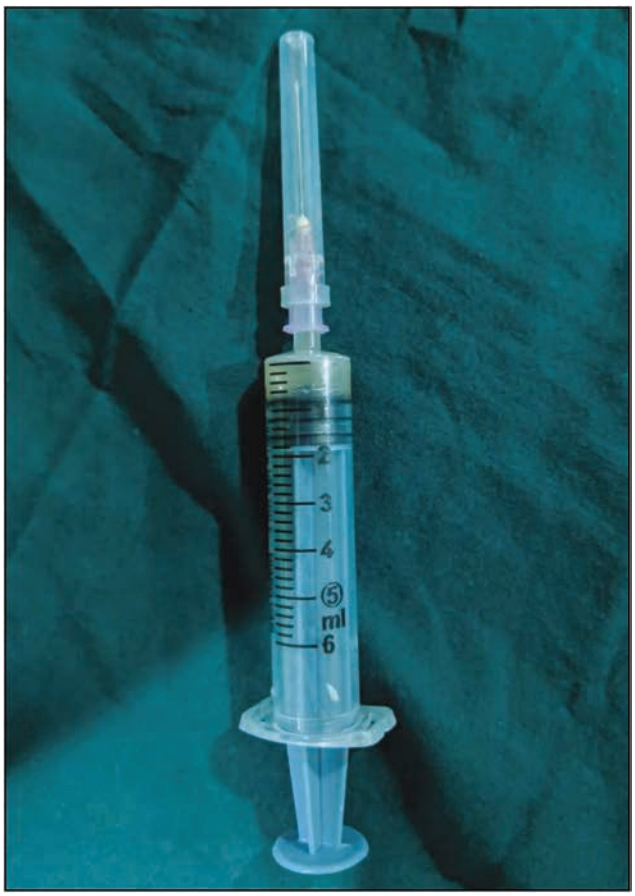

Figure 3: Pale yellow fluid aspirated from the lesion

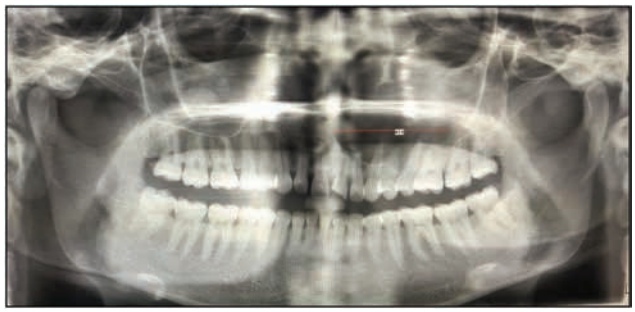

Figure 4: Panoramic radiograph showing extension into the antrum

The patient was on regular follow-up for 2 years due to its 30% recurrence rates chances and was doing well without any difficulty, with no signs and symptoms of disease.

Discussion

GOC is a very rare developmental cyst with aggressive growth potential. The term "GOC" can be attributed to the work by Gardner *et al.*[3] It was primarily described by Padayachee and Van Wyk[4] in 1987. They noticed that GOC shared clinical and histologic characters with botryoid cyst and mucoepidermoid carcinoma (MEC) and named the lesion as "sialo-odontogenic cyst" as they originally attributed the presence of mucin to salivary gland tissues.

This case posed a great deal of diagnostic dilemma for us. The clinical feature of the case was consistent with inflammatory cysts, such as a radicular cyst, as there were a few non-vital teeth present. Panoramic radiography showed the radiolucency centered around the non-vital tooth with extension into the maxillary antrum. Literature discusses about mucoceles of maxillary antrum with presentation similar to our case. Mucocele of the paranasal sinus is an epithelial-lined, mucus-containing sac that can fill the sinus completely. It is believed to be formed following obstruction of the sinus ostia. As mucus continues to be produced within the mucocele, it expands gradually. This results in remodeling and/or erosion of the surrounding bone. Mucocele is defined in studies as a completely opacified maxillary sinus on CT scan with an evidence of expansion or bone erosion, sometimes presenting with a palatal swelling.[5]

Cholesterol granuloma is a foreign body reaction to the presence of cholesterol crystals and an element of the granulation tissue formed during an inflammatory

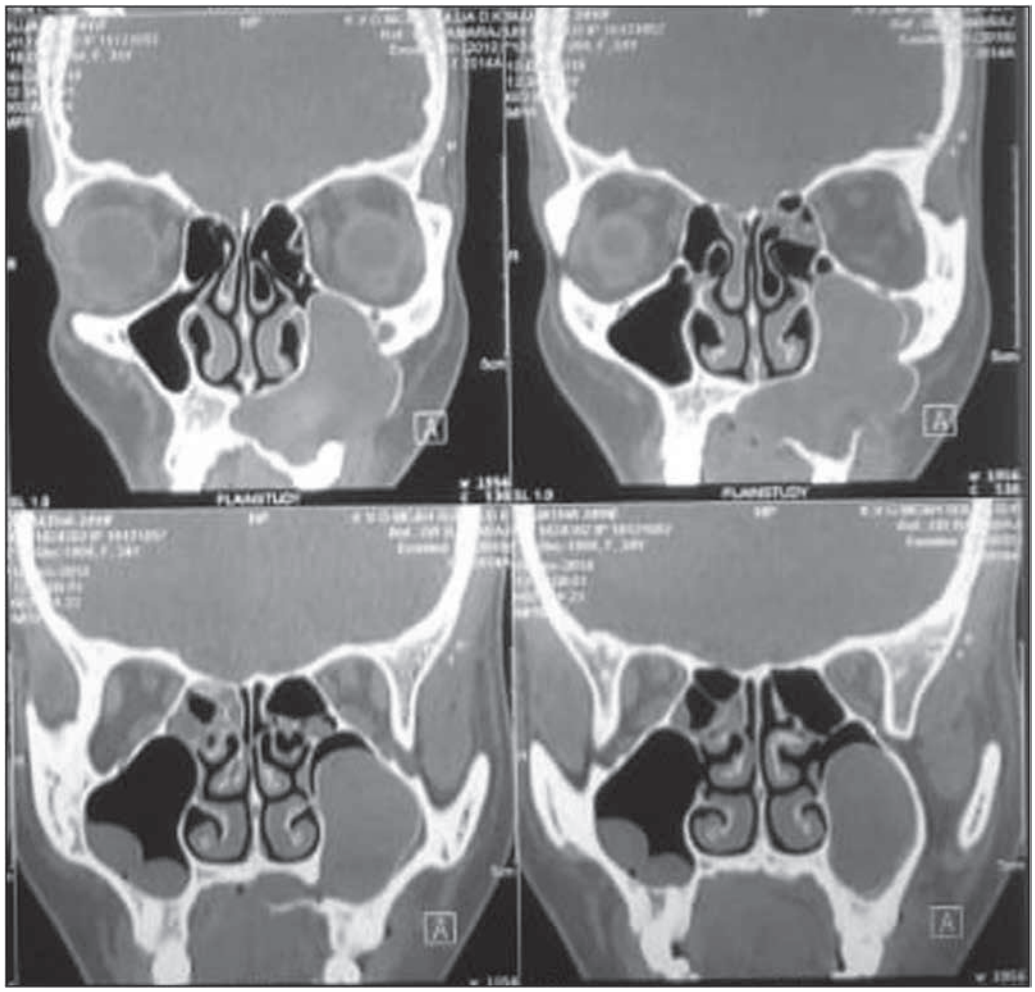

Figure 5: Computed tomography PNS shows a radiopaque lesion extending into the antrum along with the erosion of the anterolateral wall of maxillary sinus and palatal roof. PNS = paranasal sinus view

process. Cholesterol granuloma of the maxillary sinus presents with similar clinical and radiographic features. Bony erosions were described in a patient who had a known palatal swelling for six weeks and nasal obstruction for one year.[6] Almada *et al.*[7] deduced that cholesterol granuloma must be included in differential diagnosis of diseases that cause opacification on the paranasal sinuses, especially sinusitis, and cystic and tumoral lesions.

GOC is often misdiagnosed.[8,9] Lee *et al.*[10] reported misdiagnosis of a case of GOC as MEC, for which they carried out a marginal mandibulectomy.

GOC was the end most diagnosis, which we had in our mind owing to its rare presence and predominant mandibular occurrence.

The incidence of GOC is said to be 1.3%.[11] Even so, nearly 100 cases are reported in English literature. Anand *et al.*[12] suggested that GOC is more common in the mandible. In maxilla, a preference for the anterior region is reported. Even though GOC is discussed in the literature, to the best of our ability, we could find that in India, less than five cases affecting the maxillary sinus is ever reported, with none explaining about such a huge unilocular cyst with well-defined border that has encompassed the whole of the ipsilateral maxillary sinus with perforation of the palatal cortex and expansion of the buccal cortical plate. Manor *et al.*[13] reported that 52% of the lesions were unilocular and 48% were multilocular. Well-defined borders were seen in 94.5% of their cases. It is commonly seen in middle-aged individuals,[13] whereas in our case, it was a young adult. Kaplan *et al.*[1] reported a slight male predilection.

Kaplan *et al.*[1] noted that GOC can be of dimensions ranging between 0.5 and 12 cm with a mean of 4.9 cm with only one of their seven cases having a palatal extension. One of their cases reported with an infraorbital paresthesia, whereas no such complaint was given by our patient.

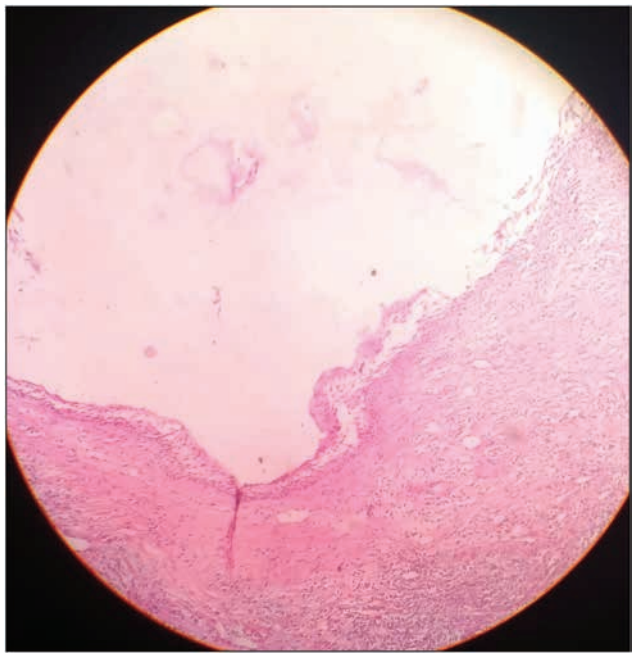

Figure 6: Microscopic picture suggestive of glandular odontogenic cyst (×20 magnification)

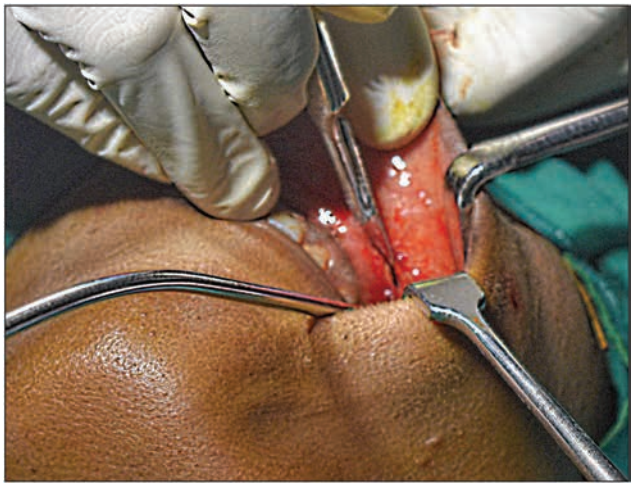

Figure 7: Intraoperative photograph showing the Caldwell-Luc approach

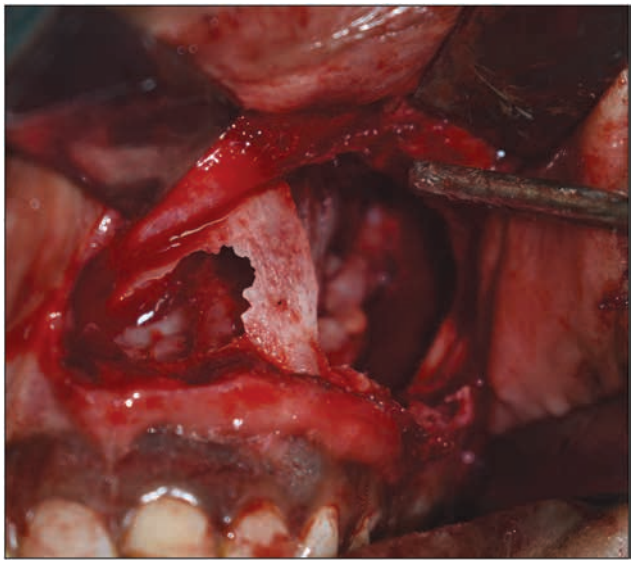

Figure 8: Defect after removal of the lesion

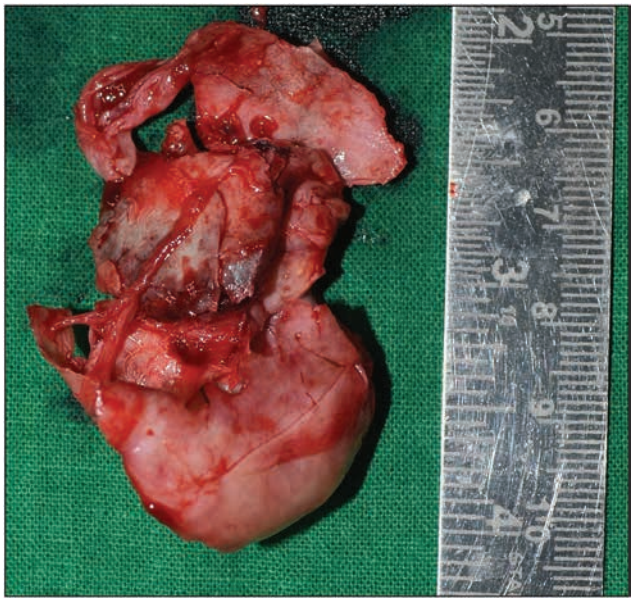

Figure 9: Removed cystic wall along with the maxillary sinus lining mucosa

There is a marked difference in the microscopic features reported by different authors. The cyst may be lined in parts by a non-keratinized squamous epithelium of variable thickness, with a chronic inflammatory infiltration of connective tissue wall. The diagnosis is made when the superficial layer of the epithelial lining consists of columnar or cuboidal cells, referred to as hobnail, occasionally with cilia or filiform extensions of the cytoplasm. Clear or vacuolated cells, which contain clear cytoplasm may be present in the basal and/or parabasal layers. The presence of glandular structures and goblet cells can be noted. Islands of odontogenic epithelium, irregular calcifications, and even microcysts may be present in the connective tissue wall of the cyst. One of the variations reported by Ide *et al.*[14] is the presence of hyaline bodies in thickened epithelium and in the lumen of an otherwise typical specimen. In our case, the cystic cavity was seen to be lined by nonkeratinized stratified epithelium with a thickness of three to four layers of cuboidal cells, with numerous mucous cells and stellate reticulum-like cells. Supporting connective tissue consisted of loosely arranged collagen fibers moderately infiltrated with chronic inflammatory cells. Though there were no conflicting findings in our case

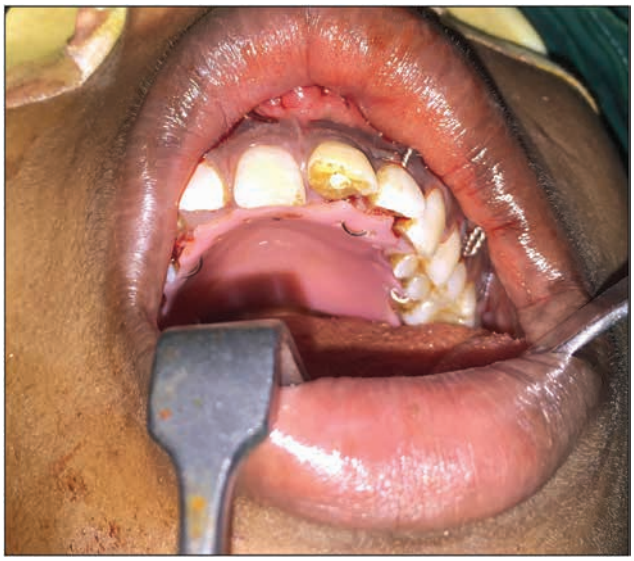

Figure 10: Acrylic plate used to cover the defect

intraepithelial hemosiderin could be evaluated in cases with such histopathologic findings.[15]

Immunohistochemistry could be a potential aid in diagnosis, as it is reported by Mascitti *et al.*[16] that there were differences in the expression of cytokeratins (CK) in GOC. The cyst expresses CK-7, 13, 14, and 19. The minimal or lack of expression of markers such as epithelial membrane antigen in the areas of glandular structures strongly suggests the odontogenic nature of GOC, and thereby rejecting the theory of glandular origin. As compared to central MEC, GOC showed decreased p-53 positivity and increased ki-67 index, which put forward the fact that GOC lining displays increased proliferation and not a potential for malignant transformation potential.[17] B-cell lymphoma 2, an antiapoptotic protein, was shown to be increased by Tosios *et al.*,[18] suggesting that the biological behavior of GOC is associated with dysregulation of cell death in the lining epithelium.

Mastermind Like Transcriptional Coactivator 2(MAML2) rearrangement analysis is a potential tool, which could identify central MEC from GOC.[19] Fluorescent in situ Hybridization (FISH) analysis for GOC in a study indicated that 10 of the 11 recurrent GOCs did not possess MAML2 gene rearrangements, which could help investigators in differentiating between intraosseous mucoepidermoid carcinoma (IMEC).[20] Reddy *et al.*[19] suggested that MAML2 rearrangement is often not conclusive in identifying IMEC from GOC.[21]

Podoplanin in GOC could be evaluated to be negative and is stated to be not related to tumor growth factor-beta expression.[22]

A wide array of treatment modalities is available from marsupialization to enucleation, primary closure or packing open with adjuvant therapy such as cryotherapy or Carnoy's solution, marginal or radical resection.[23] In their case series, Kaplan *et al.*[24] concluded that 30% of their cases recurred. Enucleation or curettage was the most frequent treatment modality they chose, reported in 83.5% of the cases. Shear[25] recommended biopsy of larger lesions. However, the success of the treatment depends mainly on the site involved, size of the lesion, the proximity of the vital structures, and appropriate surgical procedure with a regular clinical follow-up.[26] Shear[25] stated that the risk of recurrence is less in unilocular lesions and simpler treatment modalities such as enucleation is advised, whereas multilocular lesions are stated to be more aggressive, and the need for aggressive treatment is outlined. Our case was unilocular in nature, extending solely buccally, not involving any vital structures posteriorly we chose to enucleate the lesion. As our case was unilocular in nature, we chose to enucleate the lesion.

Conclusion

Gardner's Cyst or GOC is a rare entity, aggressive with relatively high recurrence rate. It requires careful clinical, radiological, and histopathologic evaluation. Diagnosis of GOC should be arrived upon after carefully evaluating the locularity of the lesion, cortical integrity, expansion and extent of the lesion, and involvement of the contiguous soft tissue. Correct diagnosis leads to correct execution of treatment. Rare pathologies of maxillofacial region compulsorily need extra intervention than usual cases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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