

Odontogenic Keratocyst with Diverse Variations: A Rare Case Report

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Abstract

Rationale: An odontogenic keratocyst (OKC) is a developmental odontogenic cyst lined by squamous epithelium having intrinsic growth potential. Hence, metaplastic changes such as the formation of mucous cells, ciliated cells, and hyaline bodies with ortho/para keratinisation have been known to create unusual histopathological variations. **Patient Concerns:** A 34-year-old male patient reported with swelling on the lower right side of the face and numbness on the overlying skin. **Diagnosis:** Based upon the histopathological findings, a final diagnosis of glandular odontogenic cyst with OKC was confirmed presenting mixed features of basal layer palisading squamous epithelium with goblet cells and satellite cysts appeared to be entrapped in the connective tissue wall. **Treatment:** Surgical enucleation of the cyst was done. **Outcomes:** No recurrence was reported in 1 year of follow-up. **Take-away Lessons:** Diverse variations appear within odontogenic cysts and tumours. The high recurrence rate and aggressive nature of the cyst, divulges appropriate treatment and long-term follow-up.

Keywords: Enucleation, glandular odontogenic cyst, mucous metaplasia, nonsyndromic odontogenic keratocysts, odontogenic keratocyst

INTRODUCTION

Diverse variations are seen with cysts and it is important to give accurate diagnosis with accurate treatment. Clinically, the lesions mimic each other, which cause a diagnostic dilemma for the clinicians. Histopathology remains the gold standard for diagnosing such lesions. The cystic lumen of odontogenic keratocyst (OKC) poses inbuilt pluripotency and multipotency which is the cause of variations and diversities.^[1] The various types of metaplasia and degeneration are observed in the OKC which include mucous metaplasia.^[2] We are presenting a rare case report of an OKC with mixed features of glandular epithelium highlighting the diverse variations with pertinent clinical, radiographic, and histopathological features as well as successful surgical management.

CASE REPORT

A 34-year-old male patient reported to the department with a chief complaint of pain and swelling on the lower right side of the face for a month. On eliciting history, the swelling was spontaneous in onset, gradually increased to the present size and was associated with dull aching pain. Extraoral examination revealed a diffuse swelling present on the right

side of the face roughly measuring 2 cm × 2 cm extending supero-inferiorly from 2 cm below the right commissure of the lip to the inferior border of the mandible and medio-laterally from the right commissure of the lip to 2 cm in front of the angle of the mandible. Overlying skin was normal. On palpation, swelling was firm in consistency and nontender to touch and afebrile with numbness present [Figure 1a]. Intraoral examination revealed restorations in 43 and 44 [Figure 1b]. A provision diagnosis of benign tumour extending from 44-46 was made.

A panoramic radiograph showed a well-defined multilocular radiolucency in the periapical area of 44, 45, and 46 extending

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from the distal root of 43 to the distal root of 46, and from the alveolar crest to 1 cm above the inferior border of the mandible, approximately 3 cm × 3 cm in dimensions. The internal structure was radiolucent with scalloped and corticated borders. A characteristic soap bubble appearance was seen. Erosion of the superior border of the inferior alveolar canal was appreciated [Figure 1c].

A cone beam computed tomography sagittal section revealed a well-defined multilocular osteolytic lesion with corticated structure and scalloped border in relation to 44, 45, and 46.

The internal structure was hypodense [Figure 1e] and axial section showed thinning and discontinuity of the buccal cortex [Figure 1d] and coronal section showed erosion on the superior border of the inferior alveolar canal [Figure 1f]. Radiographic differential diagnosis of OKC, ameloblastoma, and odontogenic myxoma was made.

Histopathology from an incision biopsy revealed that the epithelium was predominantly thin and nonkeratinised squamous with cuboidal or ciliated epithelium 2-3-4-5 layered at different locations. The superficial layer of the epithelium

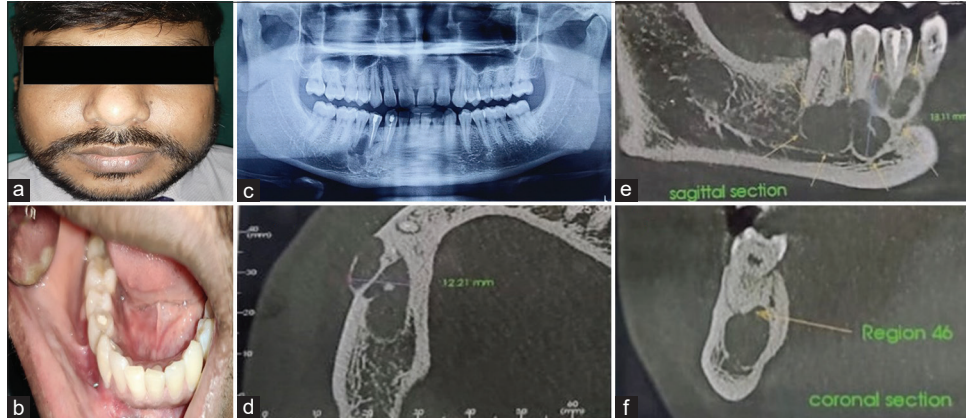


Figure 1: (a and b) Extraoral image showing swelling on the right side of the face with reduced mouth opening. (c) OPG shows a well-defined multilocular radiolucency in periapical area of 44, 45, and 46 with scalloped and corticated borders with soap bubble appearance. (d) CBCT in Axial Section shows well-defined hypodense multilocular osteolytic area with corticated structure and scalloped border. (e) CBCT in the sagittal section shows thinning and discontinuity of buccal cortical plate. (f) CBCT in coronal section shows erosion of the superior border of inferior alveolar canal. CBCT: Cone-beam computed tomography

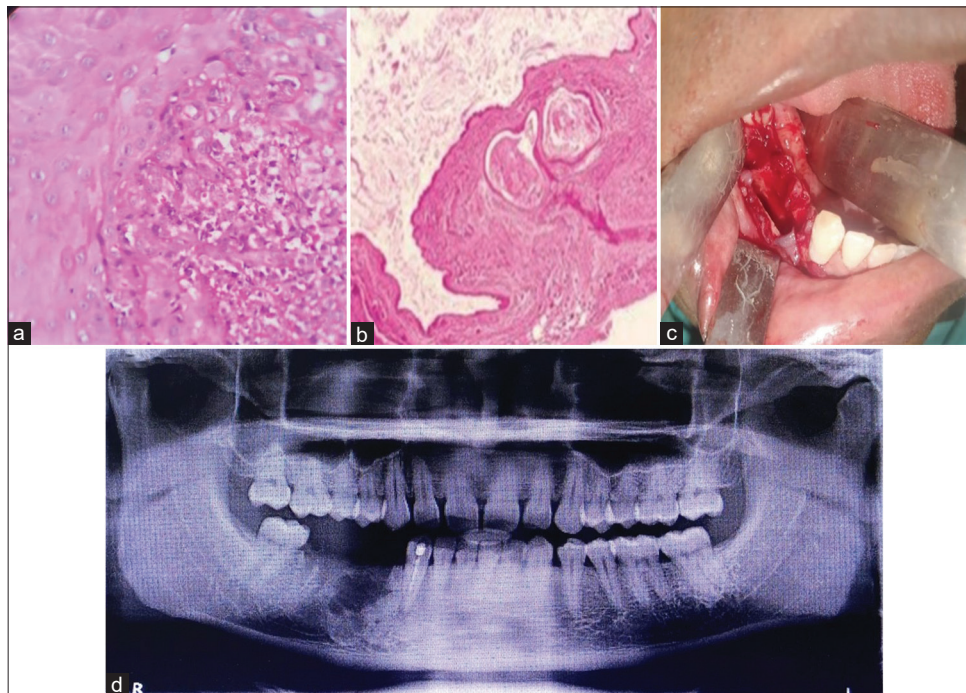


Figure 2: (a) H and E section shows mixed features of the basal layer palisading squamous epithelium with corrugated appearance and resembling reduced enamel epithelium and goblet cells. (b) H and E section shows intraepithelial crypts indicative of glandular epithelium within connective tissue stroma along with entrapped satellite cyst resembling OKC. (c) Surgical enucleation of the cyst. (d) Postoperative OPG on follow-up

showed goblet cells indicative of glandular odontogenic cyst (GOC). Some areas of the lining epithelium were composed of a uniform layer of stratified squamous epithelium, with a hyperchromatic, palisaded basal cell layer and a corrugated parakeratotic surface. Small satellite cysts appeared to be entrapped in the connective tissue wall suggestive of OKC [Figure 2a and b]. Based on this, a final diagnosis of GOC with features of OKC with 44, 45, and 46 was made.

Under general anaesthesia, mucoperiosteal incision was given and the flap was raised. The cyst was enucleated and was fixed with Carnoy's solution along with extraction of 44, 45, and 46 [Figure 2c]. The flap was then replaced and the wound was closed primarily. The patient was kept on Amoxicillin 500 mg twice a day for 5 days and Ibuprofen twice a day for 5 days to prevent infection, inflammation, and pain postsurgery. On the 7th day, sutures were removed and healing was uneventful. The patient was recalled after 1 month and was clinically and radiologically examined [Figure 2d]. No recurrence has been noted after 1 year of follow-up.

DISCUSSIONS

GOC is an uncommon cyst of odontogenic origin. Gurler *et al.* in 2017 stated that the prevalence of GOC varies from 0.012% to 1.3% of all jaw cysts, with the mean being 0.17%. Clinically, the most common site of occurrence is the mandible anterior region, in middle-aged males,^[3] but in the present case, although the patient was a 34-year-old male, the site of interest was the mandibular posterior region. The lesion is painless, slow growing and can vary in dimensions. In the present case, patient had pain with swelling due to slow bone expansion of approximately 3 cm × 3 cm in size. Radiologically, it may present as unilocular or multilocular radiolucent lesion, usually with well-defined borders.^[5] Present case showed a well-defined multilocular radiolucency with scalloped border.

Kaplan *et al.* have divided certain histological characteristic of GOC into major and minor categories,^[4] as shown in Table 1.

Table 1: Histological characteristic of GOC (Kaplan *et al.*, 2005)

Major criteria	Minor criteria
1. Squamous epithelial lining, flat interface	1. Papillary projections
2. Variations in thickness of the lining with or without epithelial "spheres" or "whorl", no palisades	2. Ciliated cells
3. Cuboidal eosinophilic cells or "hobnail" cells	3. Multicystic or multiluminal architecture
4. Mucous "goblet" cells with interepithelial mucous pools with or without crypts lined by mucous producing cells	4. Clear or vacuolated cells in basal or spinous layer
5. Interepithelial glandular micro cystic or duct like structures	

The histological features of the present case were hobnail cells and goblet cells with crypts. The cyst described in this report mainly exhibited features of GOC along with the presence of a prominent histological component of an OKC. According to the literature, the epithelial lining of a GOC may be able to induce an ameloblastomatous and squamous odontogenic tumours-like proliferation in the connective tissue wall.^[5] High *et al.*^[6] reported a similar case, where one small area of keratinisation was observed, but the associated basal layer was cuboidal without any polarity of the nuclei. An OKC may possibly undergo glandular metaplasia from a stratified squamous to a more highly differentiated ciliated columnar or glandular type. The likely pathogenic mechanism of the present case would appear to be a reflection of the pluripotential character of the odontogenic epithelium. Odontogenic epithelial rests are the remnants of the dental lamina, which may show squamous metaplasia that differentiate into the glandular or stratified squamous epithelium.^[2,7,8] Our case indicates that the multipotential odontogenic epithelial tissue has the ability to develop diverse differentiation.^[7,10] OKCs may occur in two different forms, either as solitary (non-syndromic OKCs) or as multiple OKCs (syndromic OKCs). Multiple OKCs usually occur as one of the findings in Gorlin–Goltz syndrome, but the present case was nonsyndromic/solitary type.^[8] The prognosis and biological behavior in the present case will probably be as expected for a GOC. Treatment recommendations for GOC vary from enucleation, marsupialisation, and curettage with adjuvant Carnoy solution to a more aggressive approach including marginal resection or partial jaw resection. In the present case, enucleation along with fixation by Carnoy's solution followed by extraction of 44, 45, and 46 was done. As reported in the literature, the range for recurrence rate for GOC lies between 21% and 55% suggesting its aggressive nature. Hence, complete surgical removal of the cyst with periodic follow-up plays a dynamic role in the success of the treatment.^[9,10]

CONCLUSION

Diverse variations are seen with odontogenic cysts and tumours, although they have common embryonic derivatives. The clinical and radiological findings may overlap creating a diagnostic dilemma for clinicians. Through histopathological investigations, precise diagnosis with appropriate treatment and long term follow-up serves as the foundation in the management of such cases.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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