



Case report

An extensive swelling in the anterior mandible – A case report



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HIGHLIGHTS

- GOC is an extremely rare, benign, cystic lesion of jaw with high rate of recurrence.
- Various differential diagnoses were present and we confirmed GOC by incisional biopsy.
- GOC has a definite histopathological criteria put forth by Kaplan.
- Managed according to proposed treatment protocol.
- We recommend incisional biopsy whenever possible to confirm lesion and modify treatment accordingly.

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ABSTRACT

Introduction: Glandular odontogenic cyst is a rare developmental odontogenic cyst, which often pose a challenge to diagnose it clinically.

Presentation of a case: A 32 year old female patient was referred to the oral and maxillofacial surgery department with a chief complaint of a painless swelling in the anterior mandible, extending from mandibular left premolar to right first molar region, with fluctuancy and egg shell crackling at right premolar region. The associated teeth were firm. Radio graphically a large radiolucent lesion was seen extending from mandibular left premolar to right first molar region.

Discussion: We had many differential diagnoses including keratocystic odontogenic tumour, ameloblastoma and radicular cyst. Incisional biopsy was taken from the most fluctuant area, which was histopathologically suggestive of glandular odontogenic cyst. Enucleation of the cyst, peripheral ostectomy, extraction of teeth and Carnoy's solution application were done under general anaesthesia.

Conclusion: We often neglect to include uncommon lesions in the differential diagnosis, which may lead to inadequacy in the management protocol. Whenever possible, incisional biopsy should be performed to confirm the lesion before surgical enucleation. Here we present a case in which we were fortunate enough to diagnose the lesion by an incisional biopsy and managed according to the standard protocol.

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1. Introduction

Intra oral lesions most of the times pose a challenge in its diagnosis. Among the various differential diagnoses we often neglect to include uncommon lesions. This may lead to pitfalls in the management protocol. One such uncommon lesion which we encountered recently is the glandular odontogenic cyst. Glandular odontogenic cyst is a rare, benign cystic lesion of jaw characterised histologically by epithelial lining with cuboidal or columnar cells,

both at surface and lining, with crypts or cyst like spaces within the thickness of the epithelium [1].

Glandular odontogenic cyst was first reported by Padayachee and Wyk in 1987 and misnamed as sialo odontogenic cyst [2]. Gardener et al. suggested the term glandular odontogenic cyst due to the lack of evidence of salivary gland origin [3]. Later glandular odontogenic cyst was classified under developmental odontogenic cyst by WHO (World Health Organisation) [1]. Here we report a rare lesion with an attempt to discuss the various differential diagnoses, clinical, radiological and histopathological features and its management. This case has been reported in line with SCARE (Surgical case report) criteria [4].

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2. Presentation of a case

A 32 year old female patient reported to our department with a swelling in relation to the anterior mandible which was noticed 6 months back and gradually increased to the present size. Three years back she had undergone extraction of mandibular left first molar due to caries. No other contributing medical or dental history noted. Intra orally vestibular obliteration was seen from second premolar on left side to first molar on right side with a swelling more prominent on the anterior mandibular region (Fig. 1). Swelling was firm on palpation with fluctuancy and egg shell crackling noted in relation to first premolar to first molar region. No punctum, discharge or sinus noted. Teeth were firm and no dental abnormalities were present other than generalised periodontitis. Vitality testing showed that all the teeth were vital. Panoramic view revealed a large radiolucent lesion extending from mesial root of first molar on right side to distal aspect of second premolar on left side with irregular ragged borders and extending in between the root apices (Fig. 2). Septae are seen extending into the lesion. No root resorption was noticed. Differential diagnosis for periapical radiolucencies like radicular cyst, keratocystic odontogenic tumour and ameloblastoma were derived. Ameloblastoma was ruled out since there was no root resorption. Provisional diagnosis was more in favour of keratocystic odontogenic tumour.

Incisional biopsy was planned and was taken from mandibular right premolar region which was the area of suspected cortical perforation (Fig. 3). Consistency of the lining was thick unlike that of KCOT which aroused the suspicion of a different lesion. Histopathological analysis revealed a rare lesion – glandular odontogenic cyst. Treatment plan has to be modified and a more radical approach was taken under general anaesthesia. Intraoperatively, the lining mucosa was seen unusually thick (Fig. 4). Severe bone loss and cortical bone perforation was noticed in relation to mandibular anterior region (Fig. 5). The lining was adherent to the roots in these regions, so an intraoperative decision was taken to extract the involved teeth. The root apices of left canine and premolar and right second molar involved in the lesion were removed and the root canal treatment of these teeth was done at a later date. The entire lesion was surgically enucleated and peripheral osteotomy of 2–3 mm was done. As an additional precaution Carnoy's solution was applied as per the protocol for treatment of keratocystic odontogenic tumour. Wound was primarily closed after proper haemostasis. Histopathological examination of the enucleated soft tissue specimen followed the Kaplan's criteria and diagnosis was confirmed with glandular odontogenic cyst (Fig. 6). Postoperative period was uneventful (Fig. 7). At 2 weeks post op period a good healing was noted (Fig. 8). 6 months follow up radiograph revealed no evidence of recurrence and there was



Fig. 1. Intraoral photograph showing swelling extending from distal aspect of 35 to mesial aspect of 46.

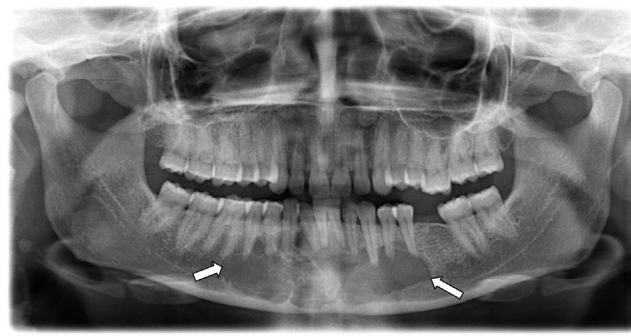


Fig. 2. Panoramic view showing large radiolucent lesion extending from distal aspect of 35 to mesial root of 46.



Fig. 3. Incisional biopsy from 44 to 46 region.



Fig. 4. Intraoperative view showing thick cystic lining.

initiation of bone formation.

3. Discussion

Glandular odontogenic cyst commonly occur in mean age group of 45–50 [5], rarely occurs before the age of 20. Commonest site of preference is anterior region in mandibular lesion. Size can range from as small as 1 cm to large lesions involving most of the jaw. Clinical feature relies mostly on size of lesion. Small lesion may be asymptomatic whereas large lesions may cause expansion which



Fig. 5. Surgical bed after Carnoy's solution application.

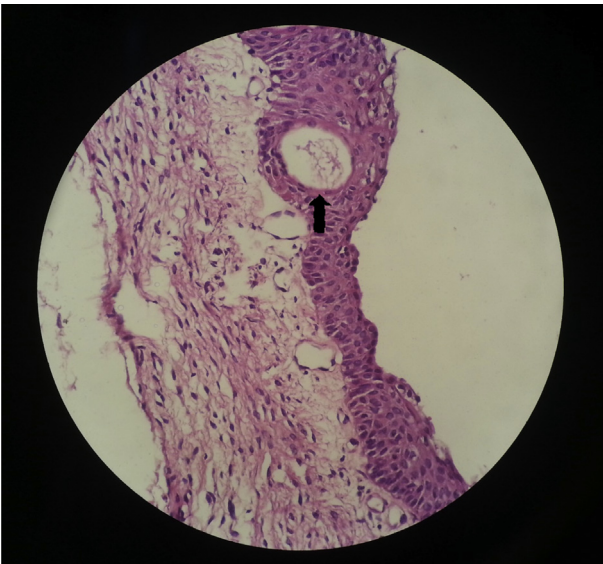


Fig. 6. Epithelium demonstrating microcysts and goblet cells [H&E, 40x].



Fig. 7. Surgical site on 2nd post-operative week.



Fig. 8. Post-operative Panoramic view.

which is much younger than the proposed age group.

Study by Magnousson et al. who analysed 5800 biopsies of jaw cysts and observed that only 7 cases fulfilled the glandular odontogenic cyst criteria [7]. A literature review by Morais et al. states that only 114 cases of glandular odontogenic cyst have been reported, of which 70% occurred in mandible [8].

Radiographically it may appear either as a unilocular or a multilocular lesion with a well-defined sclerotic rim or may appear as perifollicular radiolucency [9]. Radiographic finding is very important in diagnosis due to lack of proper clinical manifestations and intra osseous development of these lesions.

Differential diagnosis of such lesion is often confusing and taken very lightly because of clinical behaviour and radiological sign.

In order to avoid such clinical misdiagnosis and confusion, Gardner et al. proposed certain histopathological characteristics of glandular odontogenic cyst [3]. Later Kaplan et al. put forth certain major and minor criteria for diagnosis of glandular odontogenic cyst [10]. The major criteria include non-keratinized squamous epithelial lining with a flat interface, presence of spherules or whorls or focal luminal proliferations, epithelial lining exhibits surface cuboidal eosinophilic cells, mucous/goblet cells with intraepithelial mucous pools present with or without crypts lined by mucous producing cells, intraepithelial glandular microcystic or duct like (pseudo glandular) structures. Minor criteria include papillary proliferation, ciliated cells, multicystic or multiluminal arch and clear or vacuolated cells in spinous layer.

Diagnosis of glandular odontogenic cyst was reached after an incisional biopsy, which followed the criteria proposed by Gardner as well as Kaplan. Histopathological analysis of the specimen showed stratified squamous lining epithelium with papillary projections into lumen, plaque like thickening, microcystic spaces and mucous cells which met Kaplan's criteria.

Glandular odontogenic cyst are often misdiagnosed and treated as benign lesion. Surgical options ranging from peripheral osteotomy to marginal resection has been proposed for glandular odontogenic cyst. High rate of recurrence has been proposed which might be associated with cell kinetics in lining epithelium like infoldings, microcysts and plaque which are suggestive of actual cell proliferation [11].

The preservation of dentition is questionable and lacking in evidence. There has been report of cases where conservative root canal therapy was done which met with larger incidence of recurrence. Since the lesion was diagnosed on incisional biopsy, a more radical enucleation was done including extraction of affected teeth, peripheral osteotomy and tooth extraction.

Application of Carnoy's solution to reduce the recurrence rates has been reported by Ficarra et al. which we also advocated [12]. A regular follow up has to be maintained for evidence of recurrence

may be associated with pain, paraesthesia and secondary infection [6]. In the current case report, the patient presented with a slow growing swelling in the anterior mandible. She is of 32 years of age,

[13]. As per Kaplan's treatment protocol, large unilocular or multilocular lesion should be biopsied to establish diagnosis before treatment decision and all patients should be followed up for 3–7 years.

4. Conclusion

Hence, no lesion should be taken lightly. Whenever possible, histopathological diagnosis should be made to avoid chances of recurrence and morbidity to patients. We were fortunate enough to diagnose it on an incisional biopsy and managed according to standard treatment protocol established by Kaplan. Such patient should be kept under regular follow up. We also feel that in earlier days we might have misdiagnosed many such lesions as peri apical cyst and a re-evaluation of the slides will give more insight into such undiagnosed lesions.

Ethical approval

Although we had many differential diagnoses, our working diagnosis was glandular odontogenic cyst as suggested by incisional biopsy result. The treatment plan was approved in the joint discussion by the maxillofacial surgeons, the oral medicine –radiologist and the oral pathologist.

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None.

Author contribution

The lead surgeons were Dr Nikhil M Kurien and Dr.Surej Kumar. This case was assisted by Dr Uma P B. Radiological examination and proposal of differential diagnoses were done by Dr Vivek V. Dr. Anna P Joseph examined the histopathological slides for the case. In addition, Dr.Surej Kumar is responsible for concept and definition of intellectual content. Dr. Uma P B is responsible for literature search and manuscript preparation. Manuscript editing and review were carried out by Dr Surej Kumar.

Conflicts of interest

None declared.

Guarantor

Dr Uma P B.

Consent

All appropriate patient consent forms are obtained. The patient understand that his/her identity will not be revealed.

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