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An ambiguous asymptomatic swelling in the maxillary anterior region—A case report

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ABSTRACT

INTRODUCTION: Glandular odontogenic cyst is a rare and recently recognized type of developmental odontogenic cyst. Being odontogenic in origin, because of the pluripotentiality of the odontogenic epithelium it can show glandular or salivary features.
PRESENTATION OF A CASE: A 46 year old female patient was referred to the Oral and Maxillofacial Surgery department with chief complaint of painless swelling in the right anterior region of maxilla, radiographically associated with teeth 12, 13. Mucosa over the swelling was slightly bluish in colour and no associated palatal swelling was seen. No incidence of trauma was reported and involved teeth were not mobile.
DISCUSSION: Although we have many differential diagnoses, our working diagnosis was a periapical cyst, so conventional treatment of root canal treatment, cyst enucleation, and apicoectomy was planned.
CONCLUSION: Here we present a case which was initially misdiagnosed and mismanaged but on subsequent histopathologic examination revealed the final diagnosis.

1. Introduction

Cystic swellings appearing in the oral cavity can be due to many reasons and in most of the times these cystic swellings pose a challenge in its diagnosis. Here we report a case of cystic swelling in maxillary anterior region, which was initially misdiagnosed and mismanaged as periapical cyst and on histopathologic examination found to be a rare case of Glandular odontogenic cyst. This was not considered in the initial different diagnosis due to the extreme rarity of its appearance in the anterior maxillary region.

Glandular odontogenic cyst (GOC) is a rare, benign cystic lesion of jaws, which occurs frequently in the mandible, particularly in the anterior region. We present a rare case of Glandular odontogenic cyst and discuss the various differential diagnosis, clinical, radiologic and histopathological features and its management.

2. Case presentation

A 46 year old female patient was referred to the Oral and Maxillofacial Surgery Department with chief complaint of painless swelling in the right anterior region of maxilla, radiographically associated with teeth 12, 13. Clinical examination revealed a swelling at the mucogingival junction approximately of 1.0 × 1.5 cm in size, in relation to 12, 13 region. The swelling was oval shaped and diffuse extending from the distal aspect of 11 to the mesial aspect of 13 and superiorly extending into the sulcus (Fig. 1). The swelling was not fluctuant. History revealed that the patient noticed an asymptomatic swelling 6–7 months back, which gradually increased in size. Intra oral periapical radiograph revealed a unilocular radiolucency, in between the roots of right lateral incisor and right canine with well-defined sclerotic borders (Fig. 2). There was no resorption of roots of the involved teeth. Occlusal view shows well defined radiolucency involving the root apices of 12, 13 with well-defined sclerotic border (Fig. 3). The involved teeth 12, 13 were non vital. No incidence of trauma was reported and involved teeth were not mobile. Mucosa over the swelling was slightly bluish in colour and no associated palatal swelling was seen.

3. Differential diagnosis

Differential diagnosis of a well circumscribed, asymptomatic, radiolucent lesion of the right maxillary anterior region on the basis of its position, clinical features and radiological features comprises several pathologies, including Globulomaxillary cyst, Lateral periodontal cyst, Adenomatoid odontogenic tumour, Squamous odontogenic tumour, Dentinogenic ghost cell tumour and Periapical cyst [1].

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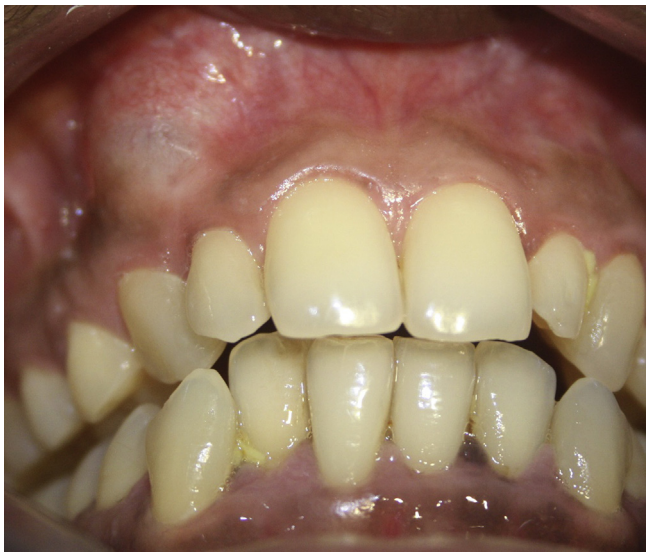

Fig. 1. Intraoral photograph showing swelling extending from distal aspect of 11 to mesial aspect of 13.

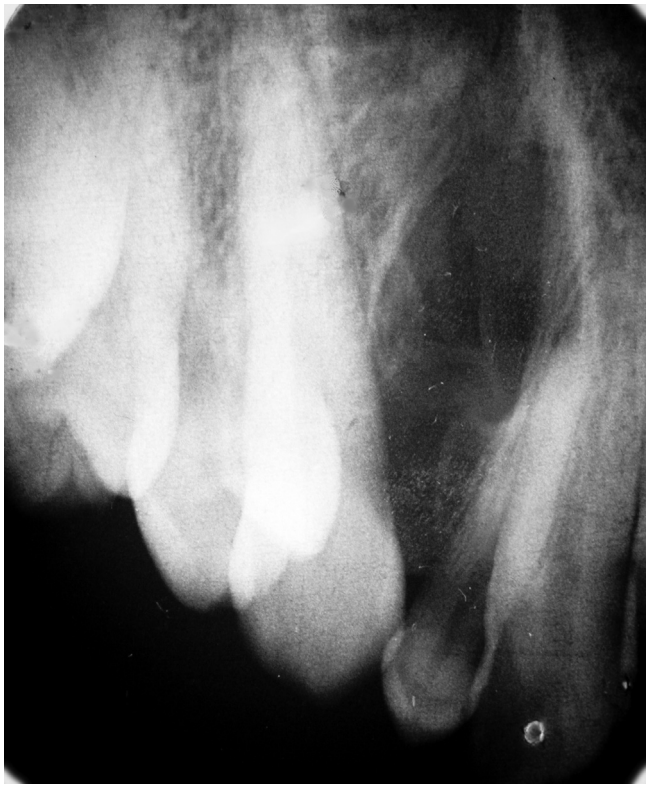

Fig. 2. IOPA showing unilocular radiolucency.

Lateral periodontal cyst is a developmental odontogenic cyst that is radiographically characterised by a well-defined unilocular or pear shaped radiolucency located in an inter-radicular location or lateral to the roots of erupted teeth. Postsurgical recurrence is not a feature of this cyst. Hence lateral periodontal cyst was considered in the differential diagnosis [2].

Adenomatoid odontogenic tumour has a remarkable tendency to occur in the anterior portion of the jaws, commonly in maxilla than in mandible. Females are affected twice as often as males. They are relatively small and seldom exceed 3 cm in greatest diameter. Peripheral form occurs as sessile masses usually of small size

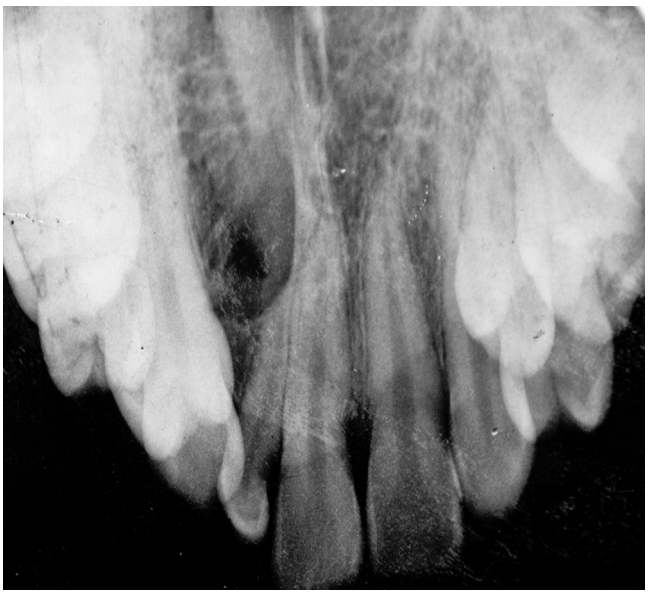

Fig. 3. Occlusal view showing well defined radiolucency with definite sclerotic border.

on the facial gingiva of the maxilla. Clinically it is difficult to differentiate adenomatoid odontogenic tumour from gingival fibrous lesions. They are frequently asymptomatic and are discovered during the course of routine radiographic examination. Less often the adenomatoid odontogenic tumour is a well delineated unilocular radiolucency that is not related to an unerupted tooth but rather is located between the roots of erupted teeth [1].

Squamous odontogenic tumour is haphazardly distributed throughout the alveolar process of both maxilla and mandible, found in age groups ranging from 8 to 74 years has no sex or site predilection. It appears as a painless or slightly painful gingival swelling along with the mobility of involved teeth. Radiographic findings are not specific or diagnostic and consist of a triangular radiolucent defect lateral to the root or roots of teeth. In some instances this suggests vertical periodontal bone loss. It seldom exceeds 1.5 cm in diameter [1].

We also thought about Dentinogenic ghost cell tumour as we have recently reported a case of an asymptomatic swelling in anterior maxilla with similar radiological findings [3]. Dentinogenic ghost cell tumour is a locally invasive rare neoplasm, seen in any age group from 10 to 90 years, and there is no significant difference between genders. It is predominantly an intra-osseous lesion. Both intra osseous and extra osseous forms occur with about equal frequency in the maxilla and mandible. 55% of cases are found in incisor-canine area. Dentinogenic ghost cell tumour is usually unilocular, well defined radiolucency, although the lesion may occasionally appear multilocular. Most Dentinogenic ghost cell tumour is between 2 and 4 cm in greatest diameter [1]. Case reports are there where DGCT has appeared as an asymptomatic swelling in anterior maxilla with radiolucency between anteriors [3].

Since this cystic lesion was asymptomatic and the involved teeth were non vital, presumptive diagnosis of periapical cyst was also made. The tooth from which the periapical cyst originates usually does not respond to thermal and electric pulp testing. There will be loss of lamina dura along the adjacent root, and a rounded radiolucency encircles the affected tooth apex. Root resorption is common. Significant growth is possible, and lesions occupying an entire quadrant have been noted. Root canal therapy is advised on those non vital teeth [1].

The globulomaxillary cyst (GMC) was thought to be a fissural cyst originating from epithelial inclusions at the line of fusion

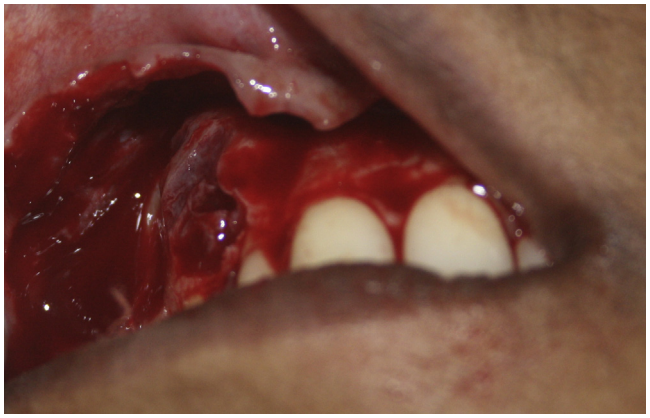

Fig. 4. Intraoperative view showing the evident cystic lining.

between the medial nasal process and the maxillary process [4]. Globulomaxillary cyst has a developmental origin. Although occasionally globulomaxillary cysts have been reported between the central incisors and lateral incisors, it is classically seen between the roots of lateral incisor and cuspid teeth [5]. Classical radiographic appearance of globulomaxillary cyst is a well-circumscribed unilocular, inverted pear or tear shaped radiolucency between the teeth. As the lesion expands, tipping of the tooth roots may occur. Now the so called GMC, a fissural cyst which is caused by entrapped epithelium between the nasal and maxillary process, is no longer considered for its own entity [6]. Because a fissural cyst in this region probably does not exist, the term GMC is no longer used. When radiolucency between maxillary lateral incisor and canine is encountered, the clinician should first consider an odontogenic origin for the lesion [1].

4. Management

Although we have many differential diagnoses, our working diagnosis was a periapical cyst, so conventional treatment constituted by root canal treatment, cyst enucleation, and apicoectomy was planned. With proper anaesthesia, a two sided full thickness mucoperiosteal flap was elevated within the area from tooth 14 to 21. The lesion was identified in relation to 12, 13. Clinically labial cortical plate overlying the lesion was perforated at some levels. Once the overlying bone was removed, cystic lining was evident (Fig. 4). Following cystic enucleation, surgical bed demonstrated bone loss at the mesial margin of 13 (Fig. 5). Later root apical end resection and retrograde filling was done in relation to 12, 13. The excised specimen was send for histopathological examination.

5. Diagnosis

Histopathological findings of the excised soft tissue section show a cystic lumen lined by non-keratinized epithelium of varying thickness with a flat epithelial connective tissue interface. Epithelium exhibits pseudo stratified columnar appearance with areas of plaque like thickening. Superficial cells of the epithelium are either cuboidal or columnar with some showing filiform extensions of cytoplasm. Within the pseudo stratified ciliated columnar epithelium there were goblet cells (Fig. 6). Intraepithelial micro cysts surrounded by cuboidal cells and containing eosinophilic material is seen (Fig. 7). Underlying connective tissue is moderately collagenous with focal collection of chronic inflammatory cell infiltrate and haemorrhage. Periodic Acid Schiff staining also showed micro cysts and goblet cells (Fig. 8). The histopathologic picture gave an impression of Glandular odontogenic cyst (GOC). Immunohistochemical staining was done with CK19 and Ki 67. Ki67 showed

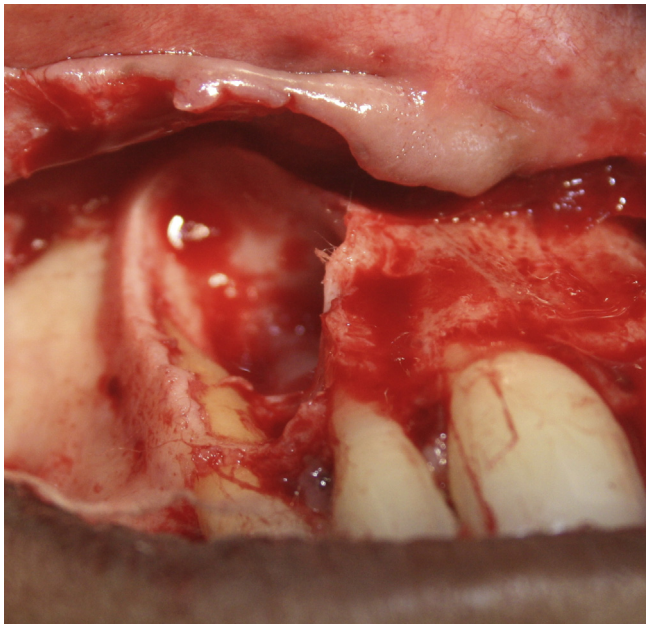

Fig. 5. Surgical bed demonstrating bone loss mesial to 13.

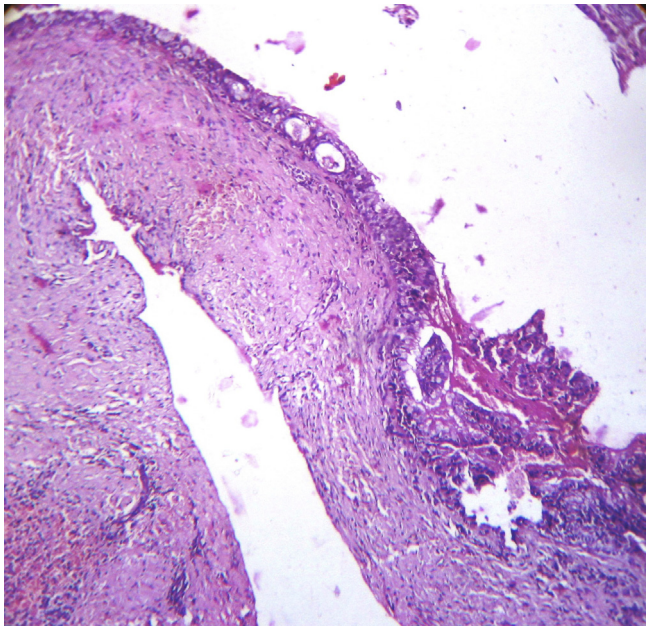

Fig. 6. Intraepithelial microcysts showing eosinophilic material (H&E, 10×).

negative/less than 1% staining (Fig. 9). The epithelium showed strong positivity for CK19 (Fig. 10). The asymptomatic swelling was diagnosed as Glandular odontogenic cyst.

6. Discussion

As Glandular odontogenic cyst is a rare and recently recognized type of developmental odontogenic cyst, it was not included in our differential diagnosis. The report of GOC was quite surprising as it was rare and never came in our discussion. Although it is accepted as of being odontogenic origin, because of the pluripotentiality of the odontogenic epithelium it can show glandular or salivary features [1].

Glandular odontogenic cyst was first suggested by Padayachee and Van Wyk in 1987 by reporting two cases that were similar to

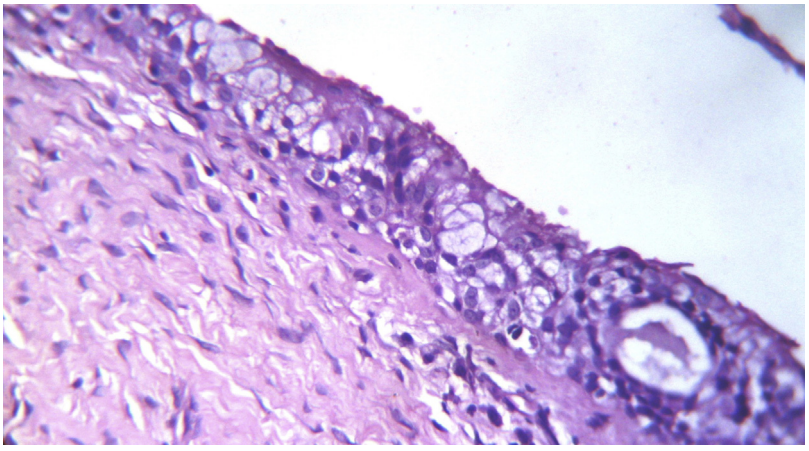

Fig. 7. Epithelium showing microcysts and goblet cells (H&E, 40×).

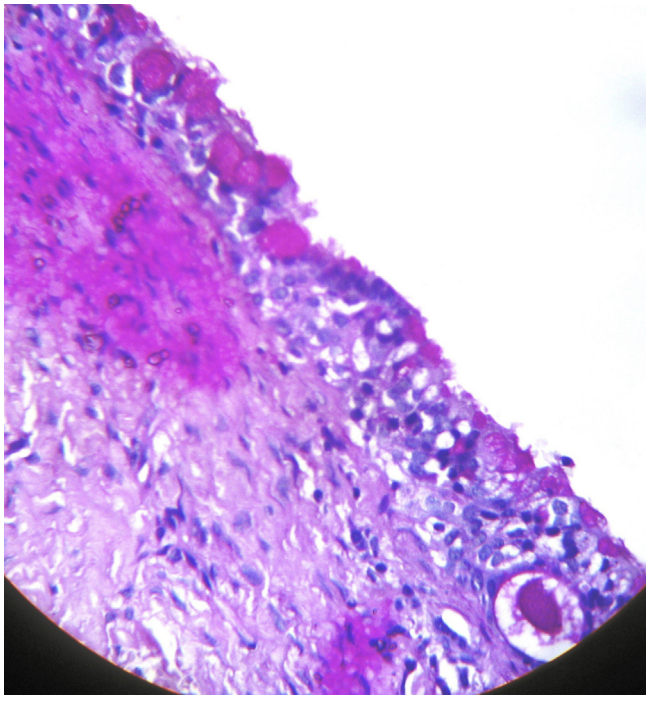

Fig. 8. Epithelium showing microcysts and goblet cells showing positivity for PAS (PAS, 40×).

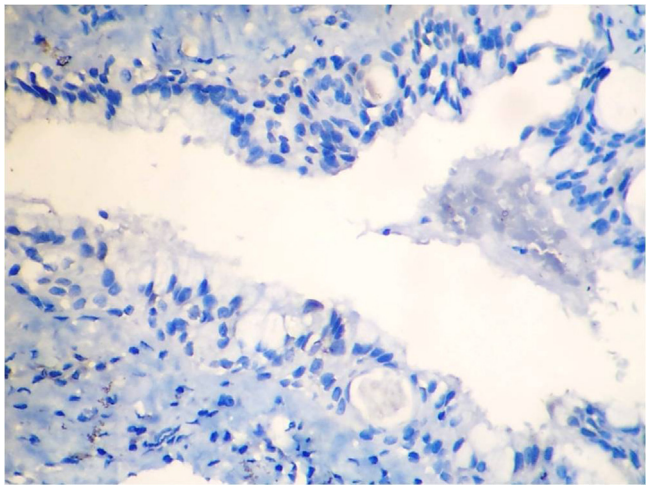

Fig. 9. Ki-67 negative 1%; (IHC 40×).

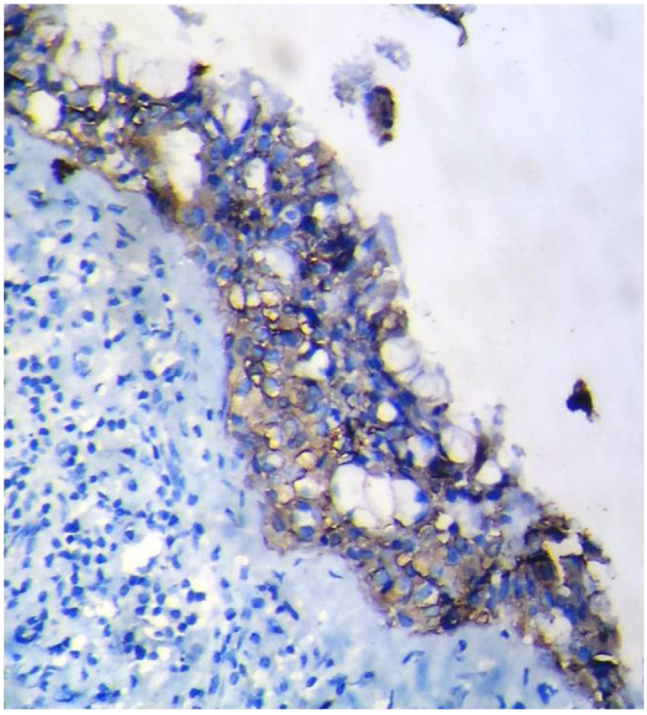

Fig. 10. CK 19 positive (IHC 40×).

the botyroid odontogenic cyst, but with a gland element and suggested the name "sialo odontogenic cyst" [7]. Later it was described in detail by Gardner et al. in 1988 [8]. High et al. in 1996 described the polymorphous nature of glandular odontogenic cyst and coined it as polymorphous odontogenic cyst [9]. This cyst was recently listed by the World Health Organization as a developmental odontogenic epithelial cyst. The GOC occurs commonly in middle aged adults with a mean age of onset of 49 years at the time of diagnosis; rarely does it occur before the age of 20. GOC have a preference for the anterior regions, especially in maxillary lesions. The size of the cyst can vary from small lesions less than 1 cm in diameter to large destructive lesions that may involve most of the jaw. Small cysts may be asymptomatic; however large cysts often produce clinical expansion, which sometimes can be associated with pain or paraesthesia.

Between 1977 and 1995, Magnusson et al. [10] analysed 5800 biopsies of jaw cysts and observed that only seven cases fulfilled GOC criteria, which comprised of 0.012% of total. In 2012 de

Morais et al. stated that approximately 114 cases of GOC have been reported in the literature by 2010 [11].

Radiographically, the lesion may appear as a unilocular, or more commonly, a multilocular radiolucency with a well-defined sclerotic rim or as a perifollicular radiolucency. Radiographic findings of this lesion play a major role in diagnosis as there is lack of consistency in the clinical manifestations and the intra osseous development of these lesions. Gardner et al. proposed the histopathological characteristics of GOC [8] and Kaplan et al. proposed a list of microscopic criteria for GOC that includes non-keratinized stratified squamous epithelium, eosinophilic cuboidal or columnar cells, epithelial whorls or spheres within the lining, which are occasionally ciliated and presence of mucous cells with microcystic areas [12]. The sub-epithelial connective tissue is usually free of inflammation. This case satisfied all the characteristic features of GOC. The recurrence rate of GOC ranges between 21% and 55% [13]. The recurrence nature of GOC might be associated with cell kinetics in the lining epithelium [12].

There are several articles in which Root canal treatment has been done due to misdiagnosis, on a pre-assumption that it was a periapical cyst. Similarly we also did RCT in the presumption that it is a periapical cyst [8].

Retrospectively we feel that GOC is underdiagnosed because the strict criteria put forwarded by Gardener is not followed. Moreover GOC are always mismanaged as in our case because we initially thought it was a periapical cyst. Similar reports are there, where GOC was mismanaged as periapical cyst.

Different treatment modalities of GOC have been recommended. Ficarra et al., proposed the treatment of GOC as complete enucleation and fixation of the surrounding bone with Carnoy's solution [14]. According to Hussain et al. it was local en bloc excision with primary reconstruction because of its aggressive nature and tendency for recurrence [15]. Bhatt et al. proposed that conservative treatment is enough for GOC [16]. However in our case the diagnosis of GOC come only in excisional biopsy. So we had done only a conservative enucleation. Since there are conflicting reports in the literature, regarding the treatment modality and since only few cases are reported in the world literature, we have opted to observe the patient periodically. She is on routine follow up and on 9 month review, there is no evidence of any recurrence.

7. Conclusion

Hence this is a unique case which appeared as a radiolucent lesion in relation to 12, 13 which was initially misdiagnosed and mismanaged as periapical cyst and on histopathologic examination found to be a rare case of Glandular odontogenic cyst. It is time to rethink whether the previously neglected periapical cyst were also GOC?

Conflict of interest

None declared.

Funding

None.

Ethical approval

Although we have many differential diagnoses, our working diagnosis was a periapical cyst, so conventional treatment of root canal treatment, cyst enucleation, and apicoectomy was planned. The treatment plan was approved in the joint discussion by the maxillofacial surgeons, and oral pathologist who are co authors for this paper.

Author contribution

The lead surgeons were Dr. Surej Kumar, and Dr. Suvy Manuel. This case was assisted by Dr Vinod Nair S. Dr. Bindu J Nair examined the histopathologic slides for the case. In addition, Dr. Surej Kumar is responsible for concept and definition of intellectual content. Dr. Vinod Nair S was responsible for literature search and manuscript preparation. Manuscript editing and review were carried out by Dr Suvy Manuel.

Guarantor

Dr. Vinod Nair S.

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