

Case report of a rare glandular odontogenic cyst in a child: A diagnostic dilemma

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ABSTRACT

This paper reports a case of glandular odontogenic cyst (GOC) in a 12-year-old male patient. GOC is a rare entity occurring in the jaw bones. Known for its aggressive behavior and nonspecific clinical features, it may mimic other clinicopathological lesions such as botryoid odontogenic cyst and central mucoepidermoid tumor making the diagnosis challenging. It is more prevalent in the middle age adult population, and very few cases have been reported in children.

Keywords: Child, glandular odontogenic cyst, maxilla, odontogenic cyst, sialo-odontogenic cyst

Introduction

The glandular odontogenic cyst (GOC) is an unusual jaw bone tumor of odontogenic origin. It was initiated and explained by Padayachee and Van Wyk^[1] in 1987 where they reported two cases that were related to two types of cysts, however, with a glandular element then termed it as sialo-odontogenic cyst. Eight cases were reported in 1988 Gardner *et al.*^[2] with similar histopathologic features. They termed the lesion as GOC. It has high recurrence potential, aggressive nature, and rare occurrence, especially in children.^[3,4]

Case Report

A 12-year-old male patient reported to the Department of Paediatric and Preventive Dentistry in SDM College of Dental Sciences and Hospital, Dharwad, Karnataka, with a chief complaint of swelling in the upper front region of the jaw since 2 months. The patient gave a history of trauma 2 months back while playing. Extraorally a diffused swelling was present on the left side of the face extending from the left infraorbital margin to the corner of the mouth and left ala of the nose [Figure 1a]. Intraoral examination revealed a solitary swelling of 5 × 4 cm with diffused margins in anterior one-third of the palate.

Anteroposterior extent was from the palatal aspect of 11 to the distal aspect of 25. Mediolateral extent was from the region of midpalatal suture involving the palatal gingival margin of 21–25 [Figure 1b]. The swelling was firm during the palpation, bony hard in uniformity, whereas, it was not at all tender and pulsatile when there was no discharge. Grade I mobility was present with 21–25. The involved teeth when subjected to cold test and electronic pulp testing were found to be vital.

Orthopantomogram (OPG), maxillary occlusal radiograph and digital volumetric tomography (DVT) were performed. The orthopantomography revealed a unilocular radiolucency of 3 × 4 cm with well-defined borders, prolonging since, and mesial feature of 11 toward the distal feature of 25. Authoritatively lengthening up to the nasal cavity and maxillary sinus and inferiorly extending up to the middle third of roots of 21–25. The radiolucency revealed the presence of an impacted supernumerary tooth. Similar findings were observed with DVT [Figure 2a and b].

Fine-needle aspiration cytology was done, and 2 ml of blood tinged and sticky fluid was withdrawn. The cytosmear revealed a mixed inflammatory lesion predominantly of chronic inflammatory cells. Based on clinical, radiographic and cytologic

features a provisional diagnosis of infected dentigerous cyst was made. Endodontic treatment was done for the involved teeth followed by their semi-rigid splinting. Due to the close proximity of the lesion to vital structures, enucleation and curettage were done under general anaesthesia. Complete excision of the lesion was done. Apicectomy was done for the involved teeth followed by placement of platelet-rich fibrin. The gross excised tissue was brown in color and 2.5×2.7 cm in size with a supernumerary tooth attached to the specimen [Figure 1c].

Histopathologically, on processing specimens from different areas of the excised lesion the cystic lining consisting of non-keratinized stratified squamous epithelium with flat interface [Figure 3a] showed predominant uniform thickness with focal luminal proliferation called as epithelial plaque [Figure 3b], through a plane epithelial, connective tissue boundary without palisading the basal cell. Vacuolated/clear cells in the basal/spinous layer and periodic acid-Schiff (PAS)-positive

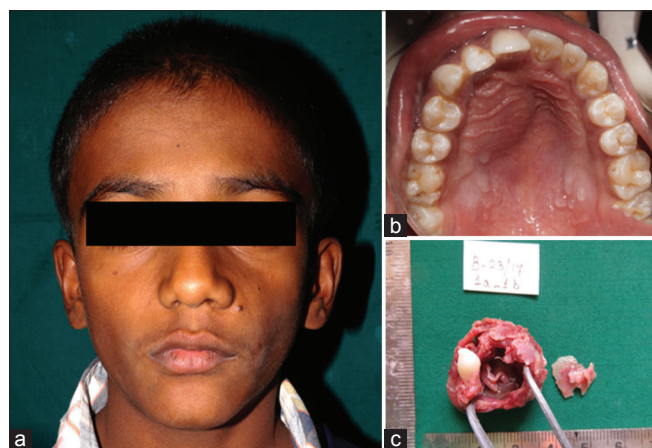


Figure 1: (a) Pre-operative view of extraoral diffused swelling on the left side of the face, (b) Pre-operative view of intraoral solitary swelling with diffused margins in anterior one-third of the palate, (c) Gross specimen of the excised lesion with the impacted supernumerary tooth

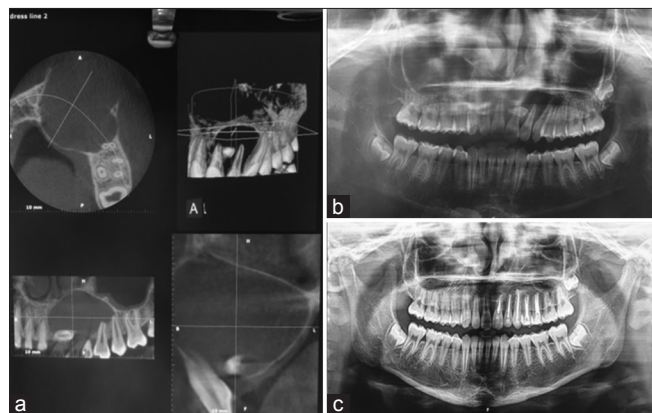


Figure 2: (a) Pre-operative digital volumetric tomography image, (b) Pre-operative orthopantomogram (OPG) reveal a unilocular radiolucency with well-defined borders, associated with an impacted supernumerary tooth, (c) Post-operative OPG on 1-year follow-up revealing new bone formation at the site of the cystic lesion

metaplastic numerous mucous goblet cells with occasional ciliated cells in the superficial cell layer were also present in the cystic lining [Figure 3d]. Based on the histopathologic findings a final diagnosis of GOC was confirmed.

So far, on 1 year follow-up no recurrence was observed and OPG revealed new bone formation at the site of the cystic lesion [Figure 2c]. A long-term follow-up of 5 years is advised.

Discussion

GOC is an uncommon odontogenic lesion occurring as 0.012–1.3% of all jaw bone cysts.^[3] So far, 181 cases have been reported globally since it was first described in 1987.^[4] It has male predilection and is more common in the mandible. In maxilla, a preference for the anterior region is reported. In the present case also, the lesion occurred in anterior maxilla in a male patient. GOC occurs over a wide age range with most cases being reported in the middle-aged adult population.^[5-7] Previously only two cases of GOC have been reported in patients below the mean age group by Noffke *et al.*^[8] and Faisal *et al.*^[4] in 11-year-old patients involving mandible. Radiographically, it may present as a unilocular or multilocular radiolucency; however, the unilocular pattern is the most common form, similar to the finding of our case. GOC is seldom related to unerupted teeth. Qin *et al.*^[9] reported five cases of GOC in maxilla associated with the unerupted tooth. In the present case, the lesion was associated with an impacted supernumerary tooth.

Kaplan *et al.*^[5] have categorized the microscopic features into major and minor criteria. Accordingly, major criteria must be present for diagnosis; however, the presence of minor criteria is supportive of diagnosis but not mandatory. However, Fowler *et al.*^[6] proposed that a necessary diagnosis of GOC is a combination of specific microscopic features and all the major criteria may not be present for a definitive diagnosis of GOC such that presence of seven or more microscopic parameters was highly predictive of a diagnosis of GOC, while the presence of five or less microscopic parameters was highly predictive of a non-GOC diagnosis. Thus, the present case is highly predictive of GOC as it exhibits the following features:

1. Squamous epithelial lining, with a flat interface with the connective tissue wall, lacking basal palisading [Figure 3a].
2. Epithelium exhibiting variations in thickness along the cystic lining with or without epithelial “spheres” or “whorls” or focal luminal proliferation [Figure 3b].
3. Apocrine snouting of hobnail cells [Figure 3c].
4. Mucous (goblet) cells with intraepithelial mucous pools, with or without crypts ruled by mucous-producing cells [Figure 3d].
5. Ciliated cells.
6. Multicystic or multiluminal architecture.
7. Clear or vacuolated cells in the basal or spinous layers.

All the below mentioned and on the basis of exclusively

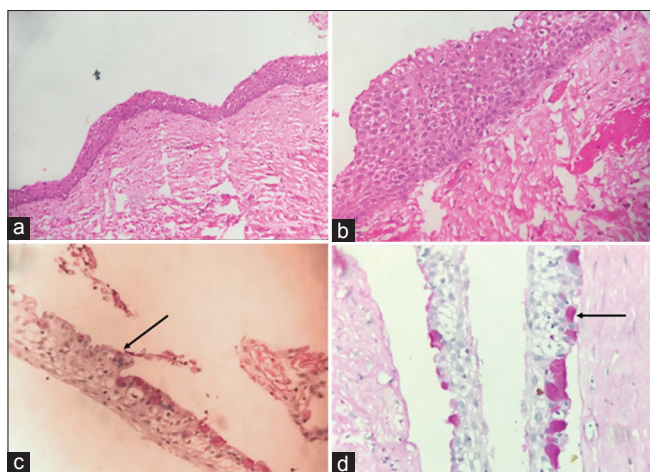


Figure 3: Photomicrograph showing (a) Cystic lining consisting of nonkeratinized stratified squamous epithelium with a flat interface, (b) Cystic epithelial lining with focal luminal proliferation representing “epithelial plaques,” (c) Vague “pinching-off” such as areas or hobnail cells (Hematoxylin and Eosin stain, $\times 10$), (d) PAS-stained section showing cystic epithelial lining with few mucous cells and vacuolated cells

features mentioned above the differential diagnosis of the lateral periodontal cyst, botryoid odontogenic cysts, mucoepidermoid carcinoma, globulomaxillary cyst, and the dentigerous cyst can be excluded.

GOC poses aggressive behavior with the recurrence rate ranging from 21 to 25%. It may be attributed to cell kinetics in the lining epithelium or a conservative treatment approach.^[5,10] To the best of our knowledge, this is the third case to be reported globally in a child below the mean age group, out of which this is the only case reported in anterior maxilla and also associated with an impacted supernumerary tooth, contributing to the rarity of the case.

Conclusion

Although rare in children with more cases of this kind being reported in literature, it should be considered as one of the

differential diagnosis while treating children. As it is aggressive in nature, a long-term follow-up is desirable.

Patient's consent

Written informed consent has been taken from the studied subject.

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