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Glandular odontogenic cyst of maxilla: A case report

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Abstract

The glandular odontogenic cyst (GOC's) is an uncommon developmental odontogenic cyst described as a distinct entity by Gardner *et al.* in 1988. The Glandular odontogenic cyst occurs more commonly in middle-aged people and has a predilection for the mandible. This article presents a case of glandular odontogenic cyst in a 16-year-old male patient in the left posterior region of the maxilla, which is quite rare.

Keywords: Odontogenic cysts, glandular, maxilla

Introduction

Glandular odontogenic cyst (GOC) is an uncommon developmental cyst. Frequency rate of GOC is 0.012% to 1.3% of all the jaw cysts and its prevalence is 0.17³. In 1987, Padayachee and Van Wyk reported the first two cases of this cyst as a "sialo-odontogenic cyst". This cyst has been established as a distinct entity by Gardner *et al.* [1].

The glandular odontogenic cyst appeared in the most recent WHO classification of odontogenic tumors and lesions from 1992 as an individual disease entity. It was placed in the subgroup of the developmental cysts as the epithelial odontogenic cyst [2].

Clinically, the most common site of occurrence is the anterior mandible. GOC has a slight male predilection and occurs primarily in middle-aged patients [3].

Radiologically, these cysts may be unilocular or multilocular with a well-defined border. Histologically, GOC is characterized by a cyst wall lining of non-keratinized epithelium, with papillary projections, nodular thickenings, mucous filled clefts, and "mucous lakes." It also includes cuboidal basalcells, sometimes vacuolated [4] Treatment of GOC can be classified as Enucleation, curettage, marsupialization, and other conservative procedures were categorized as "minor surgery," and peripheral ostectomy, marginal resection, and partial jaw resection as "major surgery" [5]. The risk of the recurrence fluctuates according to various sources in the range of 20–30% [2].

Case Report

A 16yr old patient reported to the Department of Oral & Maxillofacial Surgery Al Badar dental college & Hospital Gulbarga with chief complaint of swelling in left lower half of face since 3 months which was initially of smaller size gradually increased to present size. Swelling was associated with pain since 1 month. On examination solitary diffuse swelling seen on the left side of face approximately 3 x 3 cm roughly round in shape anteroposterior extending from ala of the nose to 2 cm anterior to ramus and super inferiorly from orbital margin to 1 cm inferior to ala tragus line. Intraorally Solitary swelling in left side of the palate approximately 2 x 3 cm oval in shape extending anteroposterior from marginal gingival of 21 22 up to hard palate. Mediolaterally from midline till the attached gingival of 24. On palpation swelling was soft in consistency and tender on palpation (figure 1). Radiographs revealed a 5 x 3-cm unilocular radiolucency in the left maxilla extending from 11 to 24. (figure 2 & 3) With displacement of 23 and 24. Under general anesthesia, the cyst was enucleated, 21 22 were extracted, peripheral ostectomy was done using round bur under copious saline irrigation and the bony cavity was fixed with Carnoy's solution (figure 4). The wound was closed primarily and healing was uneventful



Fig 1: Pre-Operative

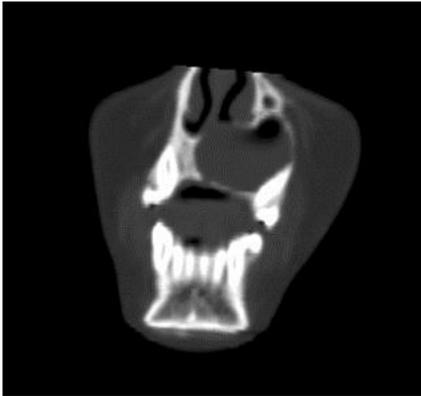


Fig 2: Pre OP CT scan



Fig 3: Pre OP OPG



Fig 4: Intra Operative

Result

Histopathologic examination of the biopsy tissue showed a cystic lesion with luminal epithelium and surrounding connective tissue. The cyst was lined by nonkeratinized stratified squamous epithelium.

Epithelial thickness varied between 4 to 6 cells, along with papillary projections into the cyst's lumen the epithelial superficial layer showed eosinophilic cuboidal and ciliated columnar cells Focal epithelial thickenings (plaques) were also observed.

Glandular-like structures and mucous cells were seen throughout the lining of the epithelium. The underlying connective tissue consisted of densely fibrous tissue without inflammation. (Figure 5)

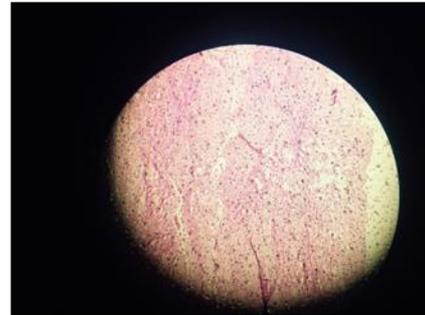


Fig 5: Histopathology

Discussion

GOC is an uncommon developmental cyst. Frequency rate of GOC is 0.012% to 1.3% of all the jaw cysts and its prevalence is 0.17%. Clinically, the most common site of occurrence is the mandible (85%), especially in the anterior region, followed by the anterior region of maxilla [3] GOC does not display specific or pathognomonic radiographical features. It may present as a multilocular or unilocular radiolucency with welldefined borders [3].

Most authors conclude that there is no radiological feature distinctive for GOC largest GOC were multilocular and resembled ameloblastoma radiologically. Radiological features which may be helpful in distinguishing multilocular GOC's from ameloblastomas include irregular loculations and a partially sclerotic border with foci of perforation [1].

The distinction between low-grade central mucoepidermoid carcinoma and GOC is difficult, if not impossible. Both are reported to be unilocular or multilocular and may infiltrate and destroy bone. Microscopically, the lining of the cystic spaces of both exhibit squamous-, cylindrical- and cuboidal epithelium and mucus producing cells arranged in papillary folds. Within the epithelial lining of both mucus containing crypts (or gland-like structures) are found. The only feature which has not been reported in low-grade central mucoepidermoid carcinoma and which may justify the existence of GOC as a separate entity is occasional presence of epithelial plaques, similar to those seen in lateral periodontal cysts [1].

Several studies indicate that GOC is a relatively aggressive lesion with a high tendency for erosion or perforation of the cortical plates as well as a high recurrence rate. The aggressive biologic behavior of GOC and its propensity for recurrence might be associated with cell kinetics in the lining epithelium. Certain studies have reported an increased Ki-67 index and decreased P53 positivity suggesting that GOC lining displays increased proliferation, but not malignant transformation potential. Tosios *et al.* demonstrated increased Bcl2 which is an anti-apoptotic protein suggesting that the biological behavior of GOC is associated with dysregulation of cell death in lining epithelium [6].

But in contrast, few studies based on immunostaining using P53, Ki-67, and Bcl2 have suggested that biologic behavior of GOC was not associated with cell proliferation and high recurrence rate is due to its multilocular nature and the tendency of the epithelium to separate from the connective tissue [6].

The treatment for GOC remains controversial because of the small number of reported cases. The lesions vary considerably in size and aggressiveness, and treatment ranges from simple curettage to marginal resection [1].

Some of the authors recommend radical surgical treatment taking into consideration partial resection of the bone around

the lesion. The methods applied include cryotherapy, classical surgery used in the cyst treatment and additionally, as in the case of the keratocyst, the application of the Carnoy's solution in the place of the removal of the lesion.

The risk of the recurrence fluctuates according to various sources in the range of 20–30%, and it seems to be lower with the appliance of the more radical surgical methods, however, on the basis of the relatively small amount of cases described in the available literature it should not be considered a conclusion [2].

According to Ficarra [7] the treatment of choice is complete enucleation and fixation of the surrounding bone with Carnoy's solution. Hussain *et al.* [8] suggested local en bloc excision with primary reconstruction. Bhatt *et al.* [9] advised that more conservative methods (ie, excision, enucleation, curettage, thorough extirpation, and cryotherapy should be applied.

In our case report we found no recurrence on 2 year follow up with enucleation, peripheral ostectomy and fixing cavity with canroys solution. We suggest large sample to be studied with this modality of treatment to assess recurrence and success rate.

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