Case Report

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A Case Report of Asymptomatic Glandular Odontogenic Cyst Mimicking as a Nasopalatine Duct Cyst

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ABSTRACT

BACKGROUND AND OBJECTIVE: Glandular odontogenic cyst is a rare developmental odontogenic cyst which most frequently occures in mandible. According to low incidence and unusual shape and location of this case, we decided to report the case.

CASE REPORT: Here we present an asymptomatic case in anterior part of maxilla that radiographically was small and unilacular mimicking as a nasopalatine duct cyst. We have found it incidentally through radiographic evaluation before implant insertion. After enucleation and curettage, histopathologic evaluation revealed a glandular odontogenic cyst. In three months follow-up, no complication or recurrence was noticed, and the site of the lesion has filled with healthy bone.

CONCLUSION: In some cases enbloc resection is suggested for glandular odontogenic cysts, but according to the size and locularity, there was no need for aggressive treatment.

KEYWORDS: *Odontogenic Cyst, Glandular Odontogenic Cyst, Nasopalatine Duct Cyst.*

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Introduction

The glandular odontogenic cyst is a rare developmental odontogenic cyst that can show aggressive behavior. At first, oral pathologists discussed this lesion in 1984, but Padayachee and Vanwyk in 1987 reported two similar cases which had criteria of botryoid odontogenic cyst with a gland element. Hence they named it "sialo-odontogenic cyst" (1-4). Gardner et al. in 1988, showed the odontogenic nature of the epithelial lining of the cyst and coined the name "glandular odontogenic cyst" which was later accepted by WHO (5-7). High et al. in 1996 entitled it as "polymorphous odontogenic cyst" because of polymorphous nature of the cyst (8-10). The glandular odontogenic cyst occurs most commonly in middle-aged adults, and about three out of four cases are in mandible specifically in the anterior region. There is a wide variation in cyst size and clinical appearance from small asymptomatic lesions less than 1 cm in diameter to massive cysts with expansion, pain, or paresthesia.

Radiographically, the cyst may appear unilocular, but most cases are multilocular, and borders are usually well defined, sclerotic, and scalloped (1,2,11). Histopathologically, the glandular odontogenic cyst is lined by nonkeratinized squamous epithelium with focal thickenings. The epithelial cells contain cuboidal or columnar cells which show hobnail appearance. Mucin pools, gablet cells, and glandular or ductlike spaces are other findings (1,2).

Glandular odontogenic cysts are successfully treated with enucleation and curettage when they are small and unilocular with healthy marginal bone. Due to the recurrence rate of 30%, some authors recommend enbloc resection, particularly for large multiloculated lesions(1,2,11).

According to low incidence of this lesion and unusual shape and location of this case which was unilocular radiolucency in maxilla in contrast with multilocular radiolucency in mandible in more common cases, we decided to report the case.

Case-Report

A 53-year-old Iranian male referred to the outpatient section of oral and maxillofacial surgery department at the Babol University of Medical Science for placing dental implants in his edentulous areas of the maxilla. The patient had no history of medical disorders, and extraoral and intraoral examination did not show any swelling or unusual appearances. We observed some remained roots and central incisors with grade III mobility. CBCT was requested for accurate evaluation of the area, and unexpectedly we noticed a small unilocular radiolucency in the anterior part of maxilla between roots of central incisors. The labial border was intact, but perforation with bilateral extension developed on the palatal margin of the lesion. No root resorption or displacement of adjacent teeth was evident The location, size, and shape of the lesion were suggestive of a nasopalatine duct cyst (fig1).

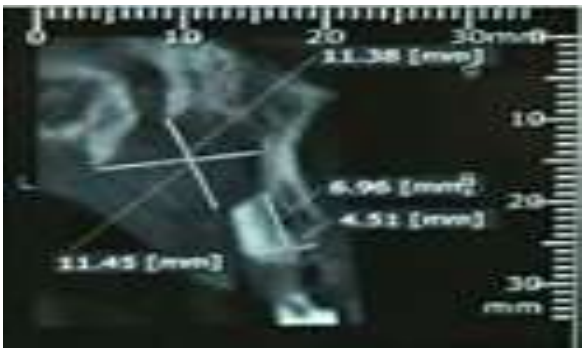

Figure 1. CBCT showing unilocular radiolucency in anterior part of maxilla with perforation of palatal cortex

The patient was consented and underwent the operation with local anesthesia using Lidocaine 2% and Epinephrine 1.100000. The crestal incision was made from left to right tuberosity with vertical releasing flap on both sides. Buccal and palatal mucoperiosteal flaps were elevated as well as extraction of remained roots. The cyst completely enucleated with a palatal approach. The total size of the lesion was about 2x1cm. No aspiration was done before surgical approach, but we did not notice any liquid discharge or unusual bleeding during the operation. Although enucleation of the cyst was very demanding because of sticky nature of cyst lining, we successfully separated it from the surrounding tissue and accessed to an intact bone (fig 2).

Figure 2. Intraoperative view

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Then drilling and implant insertion were done on 6432 | 2356 sites. The cavity was filled with biomaterial and covered by resorbable membrane and operation was fulfilled by suturing in a continuous pattern and vertical mattress.

Antibiotic (Amoxicillin 500mg TDS) and analgesic (Novafen TDS) and mouthwash (CHX 2%) were prescribed for the patient, and he was discharged. At one-week recall appointment, we noticed proper healing process with no complication.

The gross specimen was placed in formalin and sent for histopathological evaluation which revealed a cystic lesion lined by stratified squamous epithelium with variable thickness.

The superficial layer of the epithelium was cuboidal to columnar, and papillary surface with hobnail appearance was also present (fig 3). In some parts of epithelium, mucin pools, gablet cells, and cilia were seen. Fibrotic wall of the cyst showed a mild infiltration of inflammatory cells (fig 4). According to all these findings, the final diagnosis was confirmed as a "glandular odontogenic cyst". After three months follow-up, no complication or recurrence was noticed, and the site of the lesion has filled with healthy bone (fig 5).

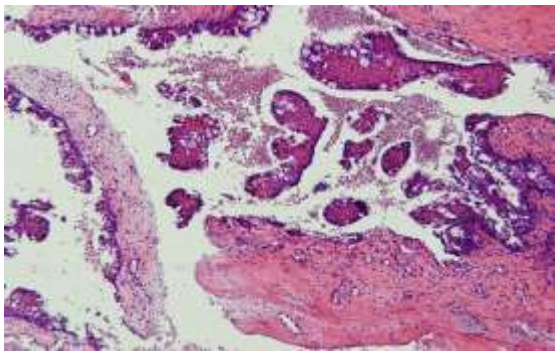

Figure 3. Stratified squamous epithelium showing hobnail appearance (H&E 10x)

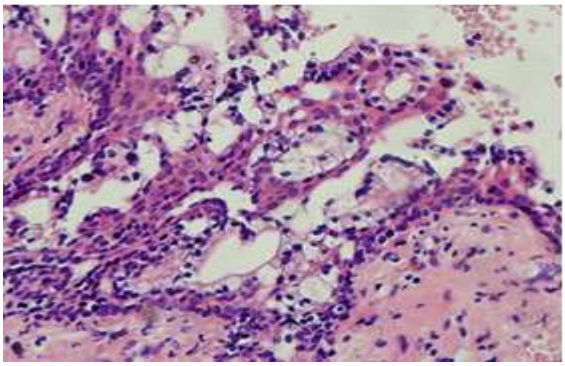

Figure 4. Epithelium showing mucin pools and gablet cells (H&E 40x)

Figure 5. Panoramic view after 3 monthes

Discussion

The glandular odontogenic cyst is a rare developmental cyst which usually shows multilocular radiolucency in the mandible. The case we presented here, involved the anterior part of maxilla which is an unusual region of this lesion; and we found it incidentally by CBCT evaluation for implant treatment as a small unilocular radiolucency. It was totally asymptomatic like what Akkas et al. presented previously (10).

The site and feature of the lesion simulated a nasopalatine duct cyst, but histopathologic examination revealed the criteria of a glandular odontogenic cyst. During operation, the sticky wall of the cyst made the enucleation very challenging, and the reason seemed to be high viscosity fluid or presence of ciliated epithelium of the cyst (11). Other literatures suggest that the low viscose and colorless fluid can help us clinically diagnose a glandular odontogenic cyst (12,13).

As other authors previously stated, only histopathological evaluation can make a definite diagnosis of a glandular odontogenic cyst, and recognition of this cyst purely based on clinical and radiographic examination is thoroughly impossible (8,14). CT or CBCT scans are appropriate for providing accurate information about the size, locularity, borders, and extension or expansion of the lesions. Moreover, any perforation and involvement of adjacent soft tissue may be detectable (7).

Complete enucleation was performed for the patient based on the recommendation of some authors that conservative treatment would be enough for small unilocular cysts because of the association of recurrence rate is with size and locularity of cysts (10,15). Marginal resection or partial jaw resection is recommended for larger cysts (16). The recurrence rate

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of small cysts was stated 14.4% in contrast to 85.6% of large lesions (17). Hence we hope that we will not detect any sign of recurrence in early and late future. Definite diagnosis of this cyst is possible just by histopathologic evaluation. There are different treatment plans for glandular odontogenic cyst, but in this case according to the size and locularity there was no need for aggressive treatment.

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