

Case Report

Recurrent Glandular Odontogenic Cyst of Maxilla: a Case Report

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ABSTRACT

The glandular odontogenic cyst (GOC) is a rare lesion with odontogenic origin. It shows a propensity for recurrence revealed in 30% of all case. This investigation reports a case of recurrent GOC in a 35-year-old female in the anterior region of the maxilla, which is uncommon and discusses about IHC finding, surgical methods, and differential diagnosis. Under general anesthesia, peripheral bone ostectomy via large round bur for removal of remaining epithelium of the cyst wall was done. Finally liquid nitrogen was used to remaining bone. This article recommends that soft tissue adjacent to the cortical bone perforation should be excised, as well as peripheral bone ostectomy by large round bur for removal of remaining epithelium of the cyst and liquid nitrogen application to the bony cavity. Because of high recurrence rate of the lesion close follow up of the patients is needed.

Keywords: Odontogenic Cyst, Maxilla, Recurrence, Iran

Introduction

Glandular odontogenic cyst (GOC) is a rare lesion which has been reported between 0.012% to 1.3% of all jaw cysts (1, 2). Frequency rate of GOC was reported 19

cases in 1995 (3), 54 cases in 2002(4), and 113 cases until 2009(5). GOC also known as a sialo-odontogenic cyst (6), mucocyst, and polymorphous odontogenic cyst (7-9). GOC is accepted as an odontogenic cyst and occurs

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most commonly in middle-aged adults and has a predilection for anterior region of the mandible. Radiographically, GOC shows a unilocular or multilocular radiolucency (5). The radiolucency usually has a well-defined and often sclerotic rim. GOC is lined by squamous epithelium of varying thickness. The superficial epithelial cells are cuboidal to columnar with cilia. Gland like structure and cluster of mucous cells are present in epithelium. GOC have been treated with enucleation but a high recurrence rate of 30% has been noted, so this study suggests more aggressive treatments for recurrent lesions (10). This article reported a case of recurrent glandular odontogenic cyst in a 35-year-old female in the anterior region of the maxilla, which is uncommon and discussed about IHC findings, surgical methods, and differential diagnosis.

Case Report

A 35 year old female was referred to Department of Oral and maxillofacial Surgery, School of Dentistry, University of Mashhad, Iran with a history of swelling in buccal sulcus of right maxillary quadrant. There was no pain in palpation. Aspirated liquid was sticky and clear. Vitality tests showed that the teeth were vital. Pantomograph view showed a well-defined unilateral radiolucent lesion.

The lesion was not associated with an impacted tooth. Root resorption or displacement of teeth was not present. Coronal and axial CT revealed large and unilateral lesion with well defined borders, which extends from the right central incisor to the right second premolar of the maxilla with buccal Cortical plate Perforation. (Fig.1). Under local anesthesia, cyst wall was enucleated and cystic bony cavity was curetted. Soft tissue adjacent to the perforation was excised and electrically cauterized.

Histopathologic examination of the lesion revealed a cystic lesion with thin epithelium and surrounding connective tissue. The cyst was lined with stratified squamous epithelium that was variable in thickness between 4 to 6 cells, as well

as papillary projections into the cyst's lumen. In epithelial superficial layer a row of cuboidal to columnar cells was seen, with the presence of cilia. Focal epithelial nodular thickenings were also relived. Gland like spaces, periodic acid-Schiff (PAS) positive mucous cells were seen (Fig. 2). Calcification and cystic space lined with thin epithelium were seen throughout the connective tissue of cyst (Fig. 3). CK 7 expressed in epithelial superficial layer (Fig. 4).

Three years after initial surgery, the patient returned with right maxillary buccal swelling and panoramic radiograph showed well-defined multilocular radiolucency in the area of the preexisting lesion. Comparison with previous available OPG there was slight apical root resorption of right maxillary canine and because of traumatic episode, root canal therapy of central incisor was seen. The teeth in surgical field did not response to thermal and electric pulp testing and endodontic therapy requested. Under general anesthesia, peripheral bone ostectomy with large round bur for removal of remaining epithelium of cyst and liquid nitrogen application to remaining bony cavity was done (Fig. 5). One year after surgery, there was no sign of recurrence in clinical and radiographic examination.



Fig.1: Destruction of the buccal cortical plate and cortical plate bone thinning in coronal CT scan

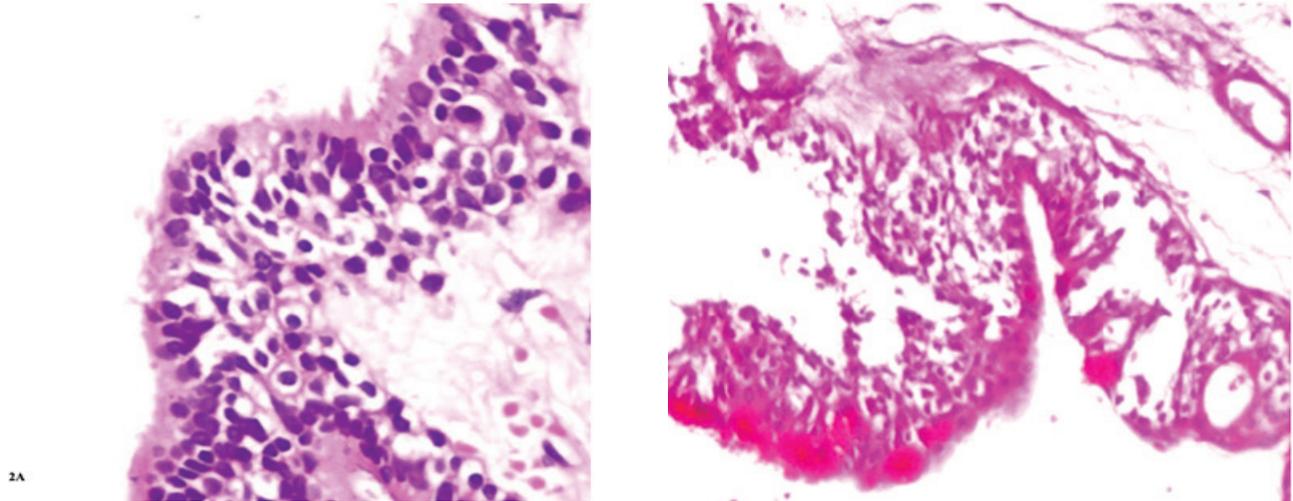


Fig. 2: **A)** The cyst is lined by stratified squamous epithelium that exhibits surface columnar cell with cilia (H&E, original magnification 400×); **B)** pseudo glandular structures, mucous pools and mucicarmine-positive mucous are observed (H&E, original magnification ×100)

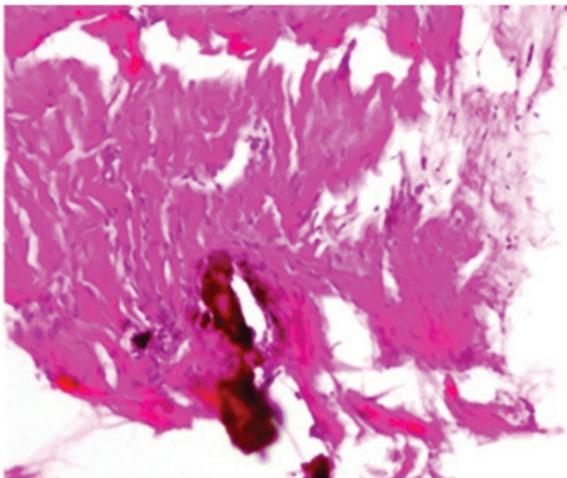


Fig.3: Focal calcifications in the cyst wall (connective tissue) is shown. This criteria mentioned by Gardner as a 7th criteria but seldom seen in reported cases so in major and minor criteria of Kaplan, this two specificities is not discussed (H&E, original magnification ×400)

Discussion

Increasing reported cases of GOC in recent years are obtained from increasing knowledge of pathologists and surgeons about this entity. GOC occurs more commonly in middle-aged adult and has a predilection for the anterior mandible and much more often will cross the midline, followed by the anterior region of maxilla (11), and the posterior area of maxilla is a rare place for GOC (12). The clinicopathological features of 64 cases of GOC in the English language literature from

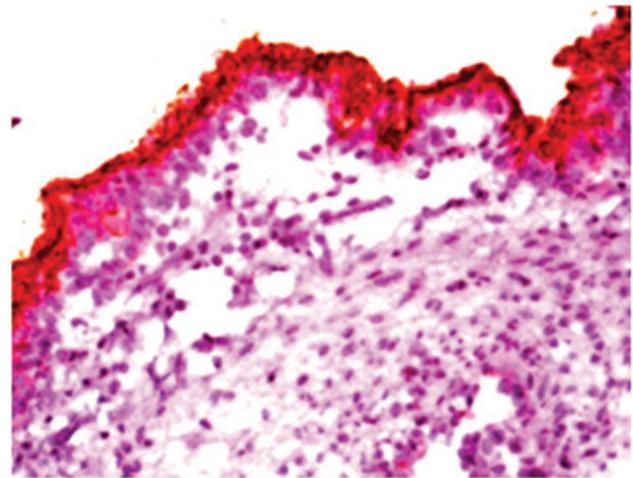


Fig.4: Immunohistochemical staining for CK7 which stains surface layer (H&E, original magnification ×100)

1996 to 2006 were reviewed by Sittitavornwong *et al.* thirteen cases occurred in the maxilla, while twenty eight of them were identified in the anterior maxilla (13). In the study by Craig *et al.* among 45 evaluated glandular odontogenic cyst cases, 20% were located in the maxilla among them 88.9% were in the anterior region (14). The size of GOC is variable from small lesion to large destructive lesions. Small lesions are often asymptomatic, but large cysts usually produce clinical expansion, that may be associated with pain or paresthesia (7).

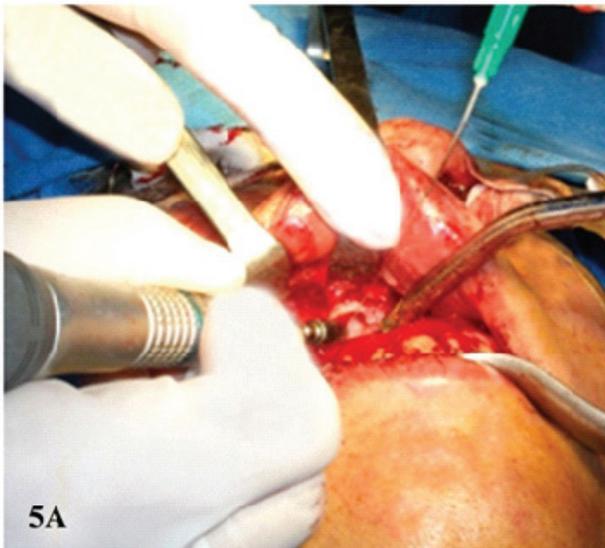


Fig.5: a) and b. Peripheral bone osteotomy with large round bur for removal of remaining epithelium of cyst and liquid nitrogen application to remaining bony cavity

In radiographic features multilocular lucency with well-defined margins (50%), slight root resorption (22%) or tooth displacement (24.4%), cortical expansion (87%) and perforation (50%) as well as absence of impacted tooth in many cases are reported (4). This case poses all seven microscopic criteria which described by Gardner *et al.* (15).

This criteria include: thin nonkeratinized stratified squamous epithelium with variable thickness covered via cuboidal or columnar cells with cilia on luminal surface (criteria I, II) through thickness of epithelium there is mucicarminophilic pools (III), foamy mucous cells (IV) and focal spherical epitheloid plaques (V) papillary projections into lumen and folding of cyst epithelium, like an intraepithelial microcysts, interepithelial glandular microcystic or duct like structures. Hyperchromatic basal cells and calcification within connective tissue also present (VI, VII).

Subepithelial calcification and satellite cysts were present in this case. Immunohistochemical staining with cytokeratines (CKs.7, 8, 10, 14, 19) and Bcl2, P53, PCNA, and Ki67 are helpful for definitive diagnosis (16-18). There is similarity between microscopic findings of the GOC with cystic intraosseous mucoepidermoid carcinoma and dentigerous cyst because mucosa cells exist in these lesions. Cellular atypia and solid

microcystic epithelial proliferation are pathologic findings in cystic intraosseous mucoepidermoid carcinoma but not present in GOC (8, 3), and the dentigerous cyst does not exhibit cuboidal surface cells, nodular thickening, and gland like spaces. Buttoroid odontogenic cysts and lateral periodontal cyst, both contain epithelial spheres and should be considered in differential diagnosis (8,6). However, the lateral periodontal cyst would lack the cuboidal cells and mucin pools.

GOC have been treated with range from a conservative approach such as enucleation, marsupialization, curettage with or without peripheral osteotomy, curettage with adjuvant Carnoy's solution, or cryotherapy to more aggressive treatments like marginal or segmental resection due to high recurrence rate after conservative treatment (12, 19). Application of the liquid nitrogen is not widely used in treatment of glandular odontogenic cysts. It is a form of cryosurgery that can be used as an adjuvant method in treatment of this entity especially when the lesion is located in vicinity of vital structures of the jaws (20).

The recurrence rate increases in multilocular lesions (17). The most cases that experienced recurrent disease had large multilocular cysts with cortical perforations, as similar the presented case while in smaller unilocular lesions conservative treatments appeared to be more responsive (19).

Conclusion

This study recommends that soft tissue adjacent to the cortical bone perforation should be excised, as well as peripheral bone ostectomy by large round bur for removal of remaining epithelium of the cyst and liquid nitrogen application to remaining bony cavity. Because of high recurrence rate of the lesion close follow up of the patients is needed.

Acknowledgements

The authors declare that there is no conflict of interests.

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