

Glandular Odontogenic Cyst Coexisting with a Dentigerous Cyst: Case Report

Assist.Prof.Dr. Serap Keskin Tunç¹, Dt. Erkan Feslihan¹, Dt. Zeynep Dilan Orhan¹,
Prof.Dr. Ömer Günhan²

¹ Van Yuzuncu Yil University, Faculty of Dentistry, Department of Oral and
Maxillofacial Surgery, Van, Turkey

² TOBB University of Economics and Technology, Department of Pathology,
Ankara, Turkey

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ABSTRACT: Glandular odontogenic cyst (GOC); is a painless, slow growing swelling that usually affects the anterior regions of the jaws. GOCs are benign lesions showing locally aggressive behavior and high recurrence rates. In this study; diagnosis and treatment of GOC, existing in the mandibular posterior region, is presented. The specificity of this case is the coexistence of GOC and dentigerous cyst in the multilocular cyst cavity. A 46-year-old male patient referred to us for the diagnosis and treatment of a multilocular radiolucent lesion at mandibular right posterior region which was recognized during routine radiographic examination. The lower third molar was extracted, the lesion was enucleated and the flap was closed primarily. Histopathologic examination revealed a combination of glandular odontogenic cyst and dentigerous cyst. No recurrence occurred during the 3-year follow-up period. Non-specific clinical and radiographic features of GOC require careful histopathological examination for definite diagnosis. Long-term follow-up of patients is recommended to detect possible recurrences.

Keywords: Dentigerous cyst, glandular odontogenic cyst, oral pathology

I. INTRODUCTION

Glandular odontogenic cyst (GOC) is a rare developmental cyst of the jaws. Initially, it was described by Padayachee and van Wyk in 1987 as "Sialo-odontogenic cyst" because of the histological similarity of the cyst epithelium to the salivary gland ducts. In 1988, Gardner et al. suggested the name "Glandular-Odontogenic Cyst" because the cyst epithelium was odontogenic and contained mucin elements with absence of salivary tissue (1,2).

GOC; is a painless, slow growing swelling that usually affects the anterior regions of the jaws. It generally occurs in the fourth and fifth decades of life and more common in male gender (3,4). Mandible is more affected than maxilla (4). The radiographic appearance is well-defined, unilocular or multilocular radiolucency. Loss of cortical integrity, root resorption, and impacted teeth in the cyst cavity can be observed (5). Histopathologically, it is characterized by eosinophilic cuboid and columnar cells, mucus cells, intraepithelial gland-like structures, surrounded with stratified non-keratinized squamous epithelium that shows nodular thickening and papillary projections. There is no

*Corresponding Author: Assist.Prof.Dr. Serap Keskin Tunç

Van Yuzuncu Yil University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Van,Turkey

inflammation in the subepithelial connective tissue (3,6). The differential diagnosis should be made with lateral periodontal cyst, botryoid odontogenic cyst and central mucoepidermoid carcinoma which have similar histological features (7). GOCs are benign lesions showing locally aggressive behavior and high recurrence rates (5).

The treatment of GOC is still controversial. There are different treatment options ranging from enucleation and curettage to extensive surgical resection. However, most researchers prefer marginal resection due to tendency to recur after curettage and enucleation (7).

In this study, diagnosis and treatment of GOC, existing in the mandibular posterior region, is presented. The specificity of this case is the coexistence of GOC and dentigerous cyst in the multilocular cyst cavity.

II. CASE

A 46-year-old male patient referred to Yüzüncü Yıl University Faculty of Dentistry, Department of Oral and Maxillofacial Surgery for the diagnosis and treatment of a multilocular radiolucent lesion at mandibular right posterior region which was recognized during routine radiographic examination. Intraoral examination revealed no cortical expansions, displaced teeth or pain-like symptoms on palpation in the affected area. Teeth 46 and 47 responded positively to pulp vitality tests. Radiographically, a well-defined two-compartment radiolucent lesion extending from first molar to the ascending ramus was observed. In the posterior part of the lesion, the lower third molar was impacted in the invert position while the anterior parts included the apices of the teeth 46 and 47 (figure 1). As a result of clinical and radiographical examination, it was decided to remove the lesion totally with the impacted tooth.

Under local anesthesia, the mucoperiosteal flap was raised and a suitable bone window was created with surgical drills to access the lesion. The lower third molar was extracted, the lesion was enucleated and the flap was closed primarily. The biopsy specimens including two different patterns were sent to histopathologic examination. Histopathologic examination revealed a combination of glandular odontogenic cyst and dentigerous cyst.

Microscopically, the first specimen was a cystic lesion lined by non-keratinized stratified squamous epithelium. plaque-like focal thickenings of the epithelium and de-epitheliasied areas due to seperation from underlying connective tissue were detected. The superficial layer of the epithelial lining consisted of cuboidal and in some areas columnar cells with cilia (figure 2). Small microcysts and goblet cells were present within the epithelium (figure 3). So, it was diagnosed as “Glandular Odontogenic Cyst”. The second specimen was associated with “Dentigerous Cyst” because there were transparent cytoplasmic alteration and ciliated layer in non-keratinized stratified squamous cyst epithelium (figure 4). The lesion healed without any complication and no recurrence was observed during the 3-year follow-up period (figure 5).

Figure 1. Preoperative panoramic radiograph; a well-defined, multilocular radiolucent lesion extending from mandibular right first molar to the ascending ramus. The right lower third molar was also impacted and associated with the lesion.



Figure 2. Histopathological section showing cuboidal and columnar cells in non-keratinized stratified squamous cystic epithelium. Plaque-like focal thickenings of the epithelium and de-epitheliasied areas due to seperation from underlying connective tissue were detected. (Hematoxylin and eosin stain (HE), original magnification x100)

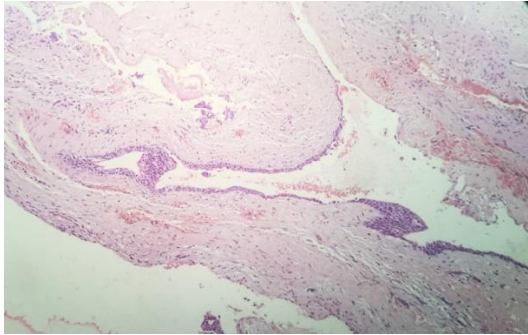


Figure 3. Photomicrograph showing small microcysts and goblet cells within the epithelium which are characteristics of Glandular Odontogenic Cyst.

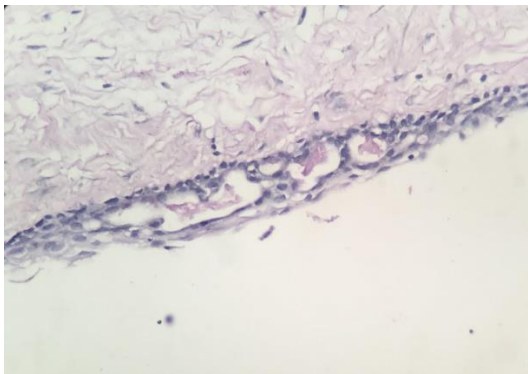


Figure 4. Histopathological section showing transparent cytoplasmic alteration and ciliated layer in non-keratinized stratified squamous cyst epithelium. (HE, original magnification x 400)

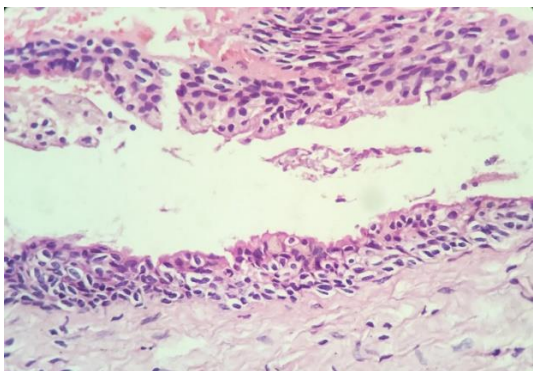


Figure 5. Panoramic radiograph 3 years after surgery. The lesion healed completely without any complication.



III. DISCUSSION

GOC is a rare developmental cyst with an incidence rate of 0.012% - 1.3% in all jaw cysts. It usually occurs in middle-aged men after the fourth decade of life. It is four times more common in the mandible than maxilla and affect mostly anterior region of the jaws (1). This case was consistent with the literature in terms of age and gender criteria but it was observed in the mandibular posterior region.

There is no specific pantognomonic panoramic image of GOC (8). Therefore, most clinicians evaluate these radiolucent lesions as dentigerous cyst or radicular cyst at first examination (9). In addition; Manor et al. reported that there are almost equal numbers of unilocular and multilocular lesions in the literature (8). Multilocular lesions should be distinguished clinically and histopathologically from keratocystic odontogenic tumor, central mucoepidermoid tumor, central giant cell granuloma and ameloblastoma (10). In this case, biopsy specimen was sent to histopathological examination with prediagnosis of dentigerous cyst and keratocystic odontogenic tumor and finally the definitive diagnosis was established as GOC.

Characteristic histologic features of GOC were defined by Kaplan et al. as major and minor criteria according to incidence of these criterias (2). Major criterias were deterministic and minor criterias were supportive for definitive diagnosis. Major criteria include cyst epithelium with variable thickness, cuboidal eosinophilic cells, intraepithelial goblet cells and secretory gland-like structures. The papillary epithelial projections in the cyst epithelium, multicystic appearance and the presence of the siliated cells within the epithelium were accepted as minor criterias (6).

Because of aggressive behavior and high recurrence rates of GOC; different treatment methods ranging from conservative approaches such as enucleation, marsupialization, peripheral osteotomy with curettage, Carnoy solution with curettage or cryotherapy to marginal or segmental jaw resections are preferred by surgeons (7). Long-term follow-up of GOCs treated with enucleation or curettage should be done. In the literature, recurrent cases have been reported 7 years after original treatment (3). Kaplan et al. recommended that patients treated with GOC diagnosis should be followed for 3-7 years (6). In this case, no recurrence occurred during the 3-year follow-up period.

IV. CONCLUSION

Non-specific clinical and radiographic features of GOC require careful histopathological examination for definite diagnosis. Long-term follow-up of patients is recommended to detect possible recurrences.

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