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Surgical Treatment of A Rare Mandibular Glandular Odontogenic Cyst with 6-Year Follow Up: A Case Report

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Glandular odontogenic cysts (GOC) of the jaws are relatively rare cystic lesion, which poses a diagnostic challenge as well as a challenge in treatment. GOC can involve either jaws, but the anterior region of the mandible is the most affected area. It affects distinct age groups, with an average patient age of 50 years. Treatment by enucleation or curettage carries the highest risk for recurrence, especially in large and multilocular lesions. Peripheral osteoectomy or marginal resection can eliminate the recurrence risk.

We report a case of GOC in the left mandible in a 52-year-old man. The lesion was located in the mandibular left molar region spreading through the canine to mandibular ramus. Neither intaoral nor extraoral sweling as well as lymphadenopathy have been noted. Clinical and radiographic features were evaluated and curretage was performed.

The surgical specimen was investigated histopathologically and immunohistochemically. After histopathological analysis, the lesion was diagnosed as GOC. At follow up after 6 years, there were no signs of recurrence.

GOC is a rare and aggressive lesion with a high recurrence rate. Careful clinical and radiological evaluation must be carried out. Long term follow data of patients with GOC is required.

Clinical relevance

Complete surgical removal of the lesion with curettage along with long term follow-up can be recommended for treatment of GOC.

Key Words: *Glandular odontogenic cyst, surgical treatment, mandible, six years follow-up.*

Cerrahi Olarak Tedavi Edilen Mandibuler Glanduler Odontojenik Kistin 6 Yıllık Takibi: Olgu Sunumu

Glanduler odontojenik kistler (GOK), tedavisinde olduğu kadar teşhisinde de zorluklar yaşanabilen oldukça nadir gözlenen kistik lezyonlardır. GOK her iki çenede de gözlenmekle birlikte en çok mandibular anterior bölgede görülür. Her yaş grubunda görülür ancak hastalar ortalama 50 yaş civarındadır. Küretaj veya enükleasyonla yapılan cerrahi tedaviler sonrası nüks oranı yüksek olabilir. Ancak Periferal ostektomi veya marjinal rezeksiyon tekrarlayan nüksleri önleyebilir.

Bu makalede 52 yaşında erkek hastada sol mandibular molar bölgede kanin dişten mandibular ramusa kadar uzanan geniş çaplı GOK varlığı bildirilmektedir. Klinik muayenede herhangi bir ekstra- intraoral şişlik veya LAP varlığı gözlenmemiştir. Klinik ve radyolojik değerlendirmeler yapıldıktan sonra lezyon cerrahi olarak küretajla alınmıştır.

Elde edilen cerrahi materyal histopatolojik ve immünohistokimyasal olarak incelenmiş ve elde edilen bulgularla GOK tanısı konmuştur. Hastanın 6 yıllık takibinde herhangi bir nüks varlığı tespit edilmemiştir.

Nadir gözlenen agresiv bir lezyon olan GOK; yüksek oranda nüks potansiyeline sahiptir. Bu yüzden GOK tanısı konmuş hastalar klinik ve radyografik olarak dikkatli bir şekilde değerlendirilmeli ve uzun dönem takipleri yapılmalıdır.

Bu çalışma, GOK' in cerrahi olarak küretajının ardından hastanın uzun dönem takip edilmesinin GOK'in tedavisinde kullanılabilecek bir yöntem olduğunu göstermiştir.

Anahtar Kelimeler: *Glanduler odontojenik kist, cerrahi tedavi, mandibula, altı yıllık takip.*

Introduction

Glandular odontogenic cysts (GOC) is an uncommon jaw bone cyst of odontogenic origin, which was first described in 1988 by Gardner et al (1). It was listed in the second version of the World Health Organization (WHO) Histological Typing of Odontogenic Tumors (2) as a developmental odontogenic cyst under the terms GOC (or sialo-odontogenic cyst) (2-5), although the WHO 2005 no longer lists this cyst (6). To the best of our knowledge only 111 cases of GOC have been reported in English language journals(7) and another 10 in the Japanese literature (8).

GOC occurs mostly in middle-aged adults (4, 7, 9). There does not apeear to be strong evidence of a male or female sex predilection, but the GOC may be slightly more

common in men (10). Although it may be found in both jaws, most are in the mandible (3, 4, 7, 9-11) The majority of cases involve the mandible but, in contrast to many odontogenic lesions, the GOC is more common in the anterior portion of the jaws. When in the maxilla, the lesion seems to affect mainly the tuberosity region (5). Clinically, GOC's present as an asymptomatic slow growing pattern (1) and radiographically as well-defined radiolucencies with unilocular, or more commonly multilocular displacement with a well-defined border (3, 4, 12). Other radiographic characteristics of GOC include root displacement and resorption of the teeth involved as well as tapering, erosion and perforation of the cortical bone (12). GOC can be at a spectrum of dimensions ranging between 0,5-12 cm, the multilocular lesions are generally larger (7). Histopathologically, the cyst wall of GOC is lined by non-keratinized epitelium, with papillary projections, nodular thickening, mucous filled clefts, and 'mucous lakes'. The superficial layer of the epitelium consist of cuboidal cells that are sometimes ciliated (4). Other characteristics include mucin, mucous cells and hyaline bodies, within the thickest section of the epitelium and in the lumen of the cyst (12).

Although it is relatively rare, correct diagnosis is of major clinical importance, since GOC has an aggressive potential, a high incidence of cortical perforation and a relatively high rate of recurrence, especially in cases (13). Treatment of GOC is controversial, varying from curettage, enucleation, marsupialization to block resection (3, 4). The most frequent types of treatment is enucleation or curretage (7).

In the present report a case of GOC is described. The current case of GOC was located in retromolar region, and associated with an impacted third molar of the mandible, mimicking a lesion of a dentigerous cyst. The diagnosis of GOC was confirmed with histopathological examination.

Case Report

During a routine radiographic examination, a 52-year-old male patient presented with an asymptomatic intrabony lesion. The patient was referred to our clinic for the evaluation of asymptomatic radiolucent lesion. The patient's medical history was unremarkable. Neither intaoral nor extraoral sweling as well as lymphadenopathy have not been noted. The lesion was located in the mandibular left molar region spreading through the canine to mandibular ramus. The panoramic radiography revealed a well-defined margins and radiopaque sclerotic edges around the impacted tooth. In vitality test while mandibular left canine, first and second premolar teeth were defined as to be vital, first and second molar teeth were devital. In panoramic radiography, an unilocular radiolucent lesion extending from canine tooth to ramus at the left side of mandibula was observed. It was noted that third molar was completely impacted in the lesion and that root reabsorbtion was present in the molar tooth adjacent to the lesion (Fig.1). The axial CT revealed that the lesion was measured 2.5 x 2.0 x 3.5 cm in size and lingual fenestration has been demonstrated while the vestibular bone was intact (Fig.2). According to clinical and radiographic findings, patient was prepared for surgery for the excison of the lesion. During surgery, cyst epithelium was completely removed and adjacent teeth with resorbed roots as well as the neighbouring impacted third molar have been removed simultanously. The bone defect was filled with mineralised allogen bone graft containing BMP in order to accelerate to healing.

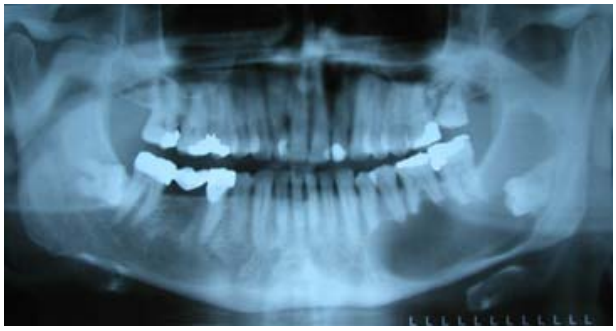

Figure 1. Initial panoramic radiography demonstrating a unilocular radiolucency with well-defined borders.

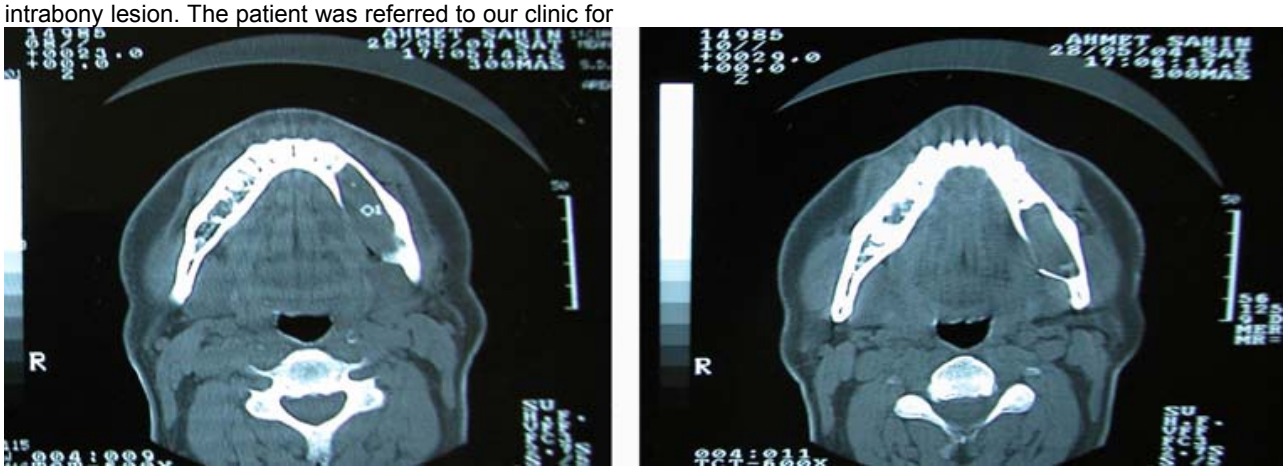

Figure 2. Computed Tomography images. Axial CT scans showing a large, destructive unilocular lesion with cystic pattern of the posterior mandible. There is a moderate expansion, particularly on the lingual aspect of the mandible.

Histologically, the cyst is lined by 3-5 rows of non-keratinizing cubic epithelium with cilia at the luminal surface was observed. Cyst epithelium was composed partly of multilayered epithelial plaques. In addition the epithelium had a glandular structure with intra-epithelial crypts lined by ciliated and partly with mucous nature (goblet) cells (Fig.3). They contained a structureless eosinophilic material which gives a positive mucicarmine reaction (Fig.4). The connective tissue wall of the cyst was composed of cellularised fibrous conjunctive tissue containing focal chronic inflammatory cell infiltration. These histological findings confirmed the diagnosis glandular odontogenic cysts.

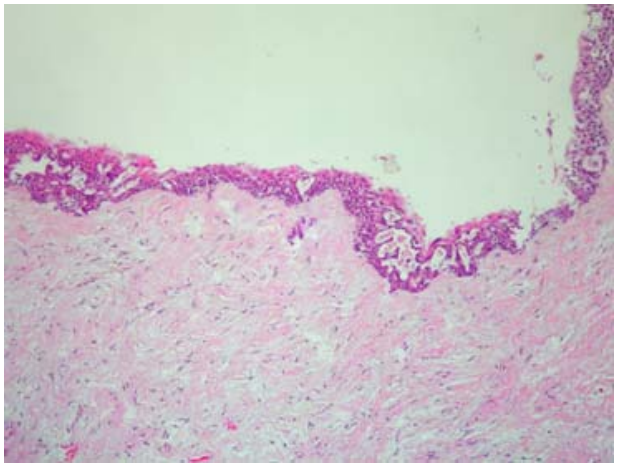

Figure 3. High power view of the cystic lining. The cystic spaces were lined by non-keratinised stratified epithelium having irregular surface. Some intra-epithelial glandular structures were recognized (Hematoxylin-Eosin x100).

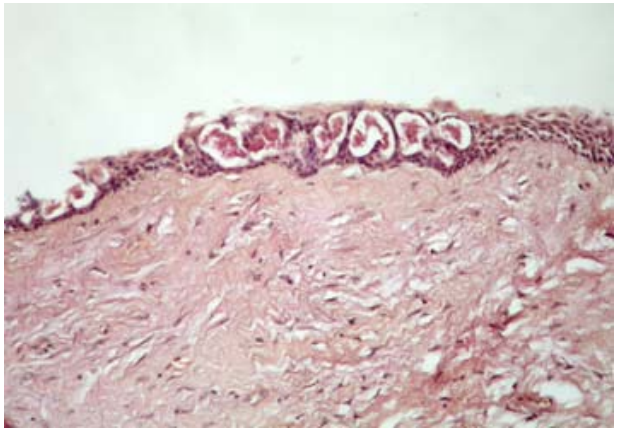

Figure 4. Intra-epithelial microcystic/glandular structures contained mucinous materials (Musicarmine Meyer x200).

The patient's follow up has been carried out for six years and no reccurence was noted either clinically or radiologically (Fig.5).

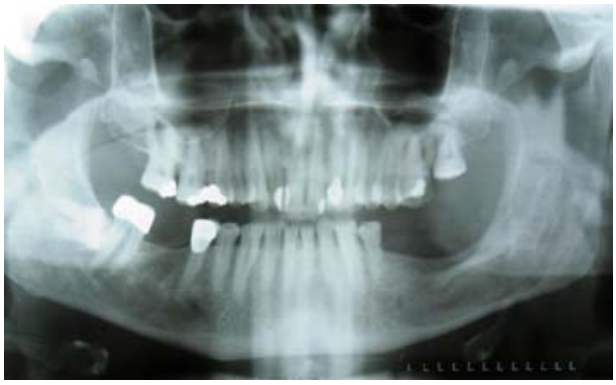

Figure 5. Panoramic radiograph 6 years later showing complete filling of the cavity by bone.

Discussion

The GOC is a rare cyst of all the jaw cysts hereby presented and its prevalance is <1% (7).

Clinically, the most common site of occurrence is mandibular anterior region (3,10,12), but in the current case report, extension from the left mandibular premolar to molar region was reported. It has been shown in the literature that there is a male predilection and lesion occurs mostly in middle aged patients (3,5,7). In the present case, patient was a middle aged man. This corroborates the reports in the literature. Radiographically, GOC is usually localized intraosseous and may appear as a unilocular or multilocular radiolucent lesion with well defined borders (4,10). Sometimes presenting as peripheral osteosclerosis and scalloping, root resorption and displacement of tooth are also noted. The most common symptom is asymptomatic slow growing swelling (4,5). Most authors agree that the clinical and radiographic features are not pathognomic of GOC (4,5,9). The differential diagnosis is made with keratocysts, central mucoepidermoid carcinoma and ameloblastoma (9,10).

In the present case, lesion detected incidentally in routine panoramic radiological examination appeared as a well defined unlilocular radiolucency in the region extending from the distal of left lower canine teeth to the ramus of the mandibula and root resorption was seen in lower mandibular first and second molar.

The CT scans are recommended because they provide accurate information about locularity of the lesion, cortical integrity, expansion of the lesion and involvement of the contiguous sft tissue. The CT scan revealed a multilocular lesion and cortical bone perforation, validating the need for the multiple plan images in cases of GOC. The CT clarified the limits of the lesion, cortical bone perforation, the involvement of the adjacent tissues and proved helpful in treatment planning (3). In this case, CT examination suggests that vestibular bone is at normal density, destruction is present in lingual bone and that lesion develops expansively rather than by making infiltration.

An incisional biopsy, fine needle aspiration and exfoliative cytologic examination can help to differantiate between GOC and other similar lesions. But their true nature can only be determined by histopathologic examination (5). In the present study, the initial diagnostic hypothesis was of dentigerous cyst, so that we decided to perform the complete surgical removel of the lesion with curretage. The specimen sent to the oral pathology laboratory to certain histopathological analysis.

Histopathologically GOC should be differentiated from lateral periodontal cyst (LPC), botryoid odontogenic cyst (BOC) and central mucoepidermoid carcinoma. LPC is a developmental odontogenic cyst lined by thin non-keratinized epithelium and also exhibits focal epithelial thickenings and glycogen rich epithelial cells. BOC is a locally aggressive polycystic variant of LPC and it consists of epithelial plaques and areas of glycogen rich clear cells (14). However, the identification of ciliated epithelium and duct like spaces with mucous cells specifically differentiated from LPC and BOC. The differentiation of low grade central mucoepidermoid carcinoma (LGCMEC) from GOC especially its multicystic variant is more important and difficult. Significant histological overlap exists between GOC and LGCMEC. However, superficial cuboidal cells, epithelial whorls, ciliated cells, and intraepithelial microcyst or duct like structures are not typical for LGCMEC and their presence or absence can help in establishing a definitive diagnosis (15). In our case histopathologically consisted of certain characteristic features of GOC like non-keratinized stratified epithelium, epithelial whorls or spheres within the lining, eosinophilic cuboidal or columnar cells which are occasionally ciliated and presence of mucous cells with microcystic areas. These findings helped us to confirmed the final diagnosis of GOC.

Various treatments ranging from curretage, enucleation to marginal resection and segmental resection are present in GOC (3,5). Minor procedures such as enucleation or curretage are the most frequent types of treatment are reported in literature (6). The recurrence mechanism may be partially related to the thinness of the cyst wall and the presence of microcysts that hamper the complete removal of the lesion, as well as the surgical technique employed for treatment (16). The aggressive nature of GOC is also cited as a possible cause of recurrence (17). As a result, a local block excision is suggested as the best treatment option (16). In the present study, based on the patient's decision, the surgeon prefered conservative treatment with careful curettage to avoid damage to the inferior alveolar nerve. During surgery the lesion was easily removed and there were not any distruption of the cyst wall. Long term follow up was performed by periodical radiographic examinations for six years.

The aggressive nature of the lesion was evident, especially because of the recurrence and the significant increase in size since the first diagnosis (3). Thus GOC cases need a long follow-up period for the recurrence rate to be evaluated (6). In the present case, follow up was made for six years due to the high recurrence rate of GOC and no recurrence was observed.

In conclusion GOC is a rare and aggressive lesion with a high recurrence rate. Careful clinical and radiological evaluation must be carried out. The increased recurrence rates can be due to multilocularity of the cyst and incomplete removal of the lining following conservative treatment. Long term followed-up of patients with diagnosis of GOC is required.

Conflicts of interest: The authors declare that they have no conflict of interest. We have not contacted any company with regard to this article.

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