

CASE REPORT

Glandular odontogenic cyst of the mandible: A case report

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يعتبر الكيس السني الغدي من الأكياس النادرة سريريا ومن الناحية التشريحية المرضية. ومع انه لا يوجد قبولا عاما بوجود هذا المرض كأفة منفصلة فإنه لا يزال هناك بعض الاختلافات بالرأي فيما يتعلق بالطبيعة الحيوية والتطور السريري لهذا المرض مقارنة بطريقة التعامل السريري. في هذه المقالة يتم وصف سريري وإشعاعي والصفات التشريحية المرضية لكيس غدي سني كبير أصاب الجزء الأمامي والجانب من الفك السفلي في مريض ذكر يبلغ 63 عاما من العمر. وتم مناقشة الحالة بالمقارنة مع ما ذكر في الأدب الطبي.

The glandular odontogenic cyst is a clinically rare and histopathologically unusual type of odontogenic cyst. While there is now general acceptance of the existence of this lesion as a separate entity, there is still some controversy regarding its biological nature and clinical behavior relative to the issue of clinical management. In this paper, the clinical, radiographic and histopathological features of an extensive glandular odontogenic cyst occurring in the anterior and lateral mandible of a 63 year old male are described. The features of the case are discussed relative to the pertinent literature.

Introduction

Odontogenic cysts are relatively common clinical lesions and frequent accessions in diagnostic pathology laboratories. The most commonly encountered jaw cysts are usually radicular (periapical) cysts and dentigerous cysts. Other less frequently encountered lesions include the odontogenic keratocyst, paradental cyst and residual cyst. This report describes a case of a rare, and occasionally controversial, jaw cyst - the glandular odontogenic cyst (sialo-odontogenic cyst).

Since it was first described by Padayachee and Van Wyk¹ in 1987, a number of cases of glandular odontogenic cyst (GOC) have been reported in the literature. Two recent and comprehensive literature reviews of this unique lesion have been published. In 1997, Ramer *et al.*² identified 39 cases, while a later study by Koppang *et al.*³ reported 47 cases. Since that time several additional cases of the glandular odontogenic cyst have been reported including those published by de Sousa *et al.*⁴ Chavez and Richter,⁵ and Bhatt *et al.*⁶ In this paper, a new case of glandular odontogenic cyst that clinically presented as an extensive, loculated radiolucency in the anterior mandible is described.

Case Report

The patient, a 63 year old male, was referred to the Oral and Maxillofacial Surgery Department of the Adelaide Dental Hospital for treatment of a large cystic lesion in the anterior mandible. The lesion was initially detected by a dental

practitioner as an incidental finding on an orthopantomogram view. Radiographically, the lesion presented as a well defined, loculated radiolucency in the left mandible extending from the midline to the mesial root of the second permanent molar tooth (Figure 1). The superior aspect of the lesion extended to include the superficial alveolus, while the base extended to the inferior border. The first premolar tooth on the affected side was missing and there was some displacement of the canine and second premolar teeth by the lesion. A CT scan showed that the lesion had caused buccal and lingual expansion of the mandibular cortex, but no perforation was evident. The patient had reportedly experienced no symptoms from the lesion.

A provisional differential diagnosis of odontogenic keratocyst, ameloblastoma, myxoma or other odontogenic pathology was established on the basis of clinical and radiographic assessment.



Fig. 1. Orthopantomogram showing an extensive, loculated radiolucency in the anterior mandible. The lesion extends from the midline to the mesial root of the second permanent molar tooth.

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Histopathology

An incisional biopsy was performed and submitted for histopathological examination. Sections showed several, variably sized epithelial-lined cystic cavities separated by fibrous connective tissue stroma (Figure 2). The epithelial lining of the cystic spaces ranged in type from thin cuboidal to thin squamous through to pseudostratified focally ciliated columnar epithelium (Figure 3). PAS-D stained sections confirmed the presence of occasional mucous-secreting cells within the epithelium. Other epithelial features present included duct or gland-like spaces, sometimes containing PAS-D positive material and occasional larger cystic spaces containing eosinophilic coagulum. In some areas the epithelium assumed a more solid rather than cystic architecture. Features of odontogenic keratocyst and ameloblastoma were not seen. A provisional histopathological diagnosis of glandular odontogenic cyst was made with the comment that histopathological distinction between this lesion, muco-epidermoid carcinoma and botryoid variant of periodontal cyst can be difficult and that a final diagnosis in this case would probably only be possible after examination of the surgical specimen. The lesion was treated surgically by removal of associated teeth and careful enucleation. The patient specifically requested conservative treatment.

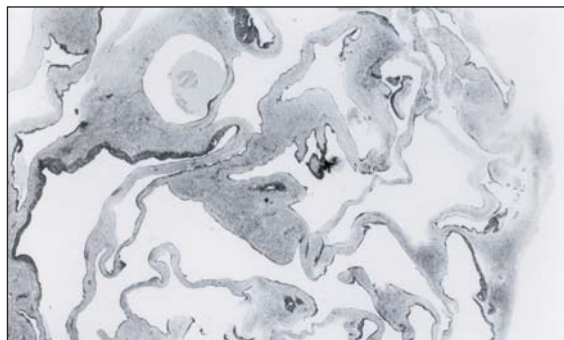


Fig. 2. Low power photomicrograph, illustrating the multicystic nature of the lesion. (Original magnification x20)

Discussion

The glandular odontogenic cyst is a very rare cystic lesion of the jawbones. In their review of 47 cases of glandular odontogenic cyst, Koppang *et al.*³ found that it occurred more frequently in adult patients (average age 47 years in males and 50 years in females) and that the anterior mandible

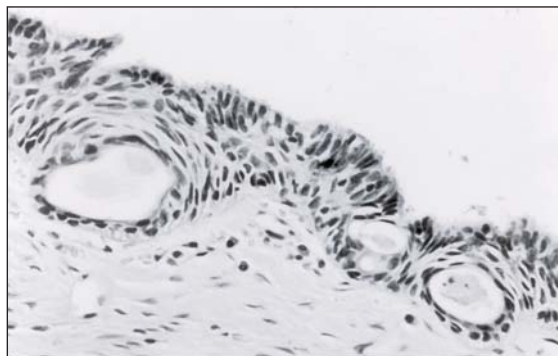


Fig. 3. Photomicrograph showing the glandular epithelial architecture observed in some areas of the epithelial cyst lining. Duct-like spaces and superficial, columnar ciliated epithelial cells are seen. (Original magnification x200)

region was the most commonly reported site. The exact relationship between the glandular odontogenic cyst and the botryoid odontogenic and lateral periodontal cysts remains unclear. While all lesions share some histopathologic features, their clinical and behavioural characteristics are more distinctive.^{7,8} For example, it is recognised that both the botryoid and glandular odontogenic cysts have a higher recurrence potential than the lateral periodontal cyst. Because of its sometimes prominent glandular features, the glandular odontogenic cyst has also been linked to the central mucoepidermoid tumour^{1,9} especially in the context of histopathologic differential diagnosis.

The histogenesis of the glandular odontogenic cyst is also uncertain, although recent immunohistochemical studies^{3,10} have concluded that it is of odontogenic origin, hence the general acceptance of the term glandular odontogenic cyst. Recent studies such as those of Barreto *et al.*¹¹ and Tosios *et al.*¹² have begun to address the molecular biology of these lesions by investigating such activities as PTCH gene expression and regulation of apoptosis and cell growth using immunohistochemical markers such as bcl-2, Ki-67 and p53. Such studies may eventually provide a more satisfactory description of the biological nature of the glandular odontogenic cyst per se and when compared to other types of odontogenic cysts and tumors.

It is recognized that the recurrence rate for glandular odontogenic cyst is relatively high. In their extensive review of reported cases of this lesion, Koppang *et al.*³ found that for the 38 lesions where follow-up was carried out, 6 cases (21%) recurred between 2-8 years after the initial

surgery. They also found that two cysts recurred twice. The possibility of recurrence is an important consideration in the overall management of the glandular odontogenic cyst. Careful surgery ranging from thorough enucleation to marginal resection, patient counseling and regular clinical and radiographic review, are essential elements in the management of this rare odontogenic lesion. However, the observation by Bhatt *et al.*⁶ provides an interesting management perspective, based on the clinical observations of a single case, that perhaps these lesions may not always require aggressive surgery.

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