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(CASE STUDY)

Case report: A 62-year-old woman with glandular odontogenic cyst

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Abstract

Glandular odontogenic cyst (GOC) is an uncommon developmental and aggressive cyst of jaws with unpredictable behaviour. Generally, it is seen in middle-aged adults and involves the anterior region of the jaws, especially the mandible, with a tendency to recur. The case described in this report regards a 62-year-old female with a presentation of a glandular odontogenic cyst, without any anamnestic pathological history.

Keywords: Odontogenic cyst; Aggressive cyst; Jaws

1. Introduction

Glandular odontogenic cyst (GOC) is a rare jaw lesion (0.0012% to 1.3% of all jaw cyst) with odontogenic origin [1]. It was first described by Gardner et al. as a well-defined pathology in 1988 [2] suggesting the name "Glandular odontogenic cyst" because the cyst wall epithelium was odontogenic and contained mucin elements with absence of salivary tissue [3]. GOC has also been described as sialo-odontogenic cyst, mucoepidermoid odontogenic cyst, and polymorphous odontogenic cyst [3].

In 1992, the World Health Organization (WHO) described GOC as "a cyst arising in the tooth-bearing areas of the jaws and characterized by an epithelial lining spaces within the thickness of the epithelium" [4]. GOC usually occurs most commonly in males over 40 years of age, with an anterior region of mandible predilection [5, 6]. Its importance relates to its morphological and histological similarities to central mucoepidermoid carcinoma (CMEC) [7]. Thus, it is really hard to make definitive diagnosis [8], and only histopathological examination allows for a certain diagnosis [9]. Radiographically, it presents an intra-osseous lesion with well-defined margins with or without root resorption [10]. The objective of the present study consists in reporting an unusual case of a glandular odontogenic cyst, which was diagnosed in a 62-year-old female, who presents an intra-osseous lesion affecting the ascendant branch of the mandible.

2. Case report

A 62-year-old female, without any linkable pathologies, reported a diffuse swelling over the left side of the mandible, with no pain in palpation.

Panoramic radiography showed a well-defined oval-shaped structure (figure 1).

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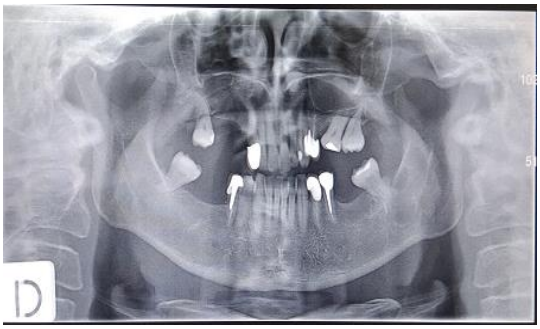

Figure 1 Panoramic radiography showed a well-defined oval-shaped structure

CT scan revealed a roundish, thick, cystic and radiolucent lesion, measuring around 1,5 x 0,8 cm, at the left mandibular angle (figure 2).

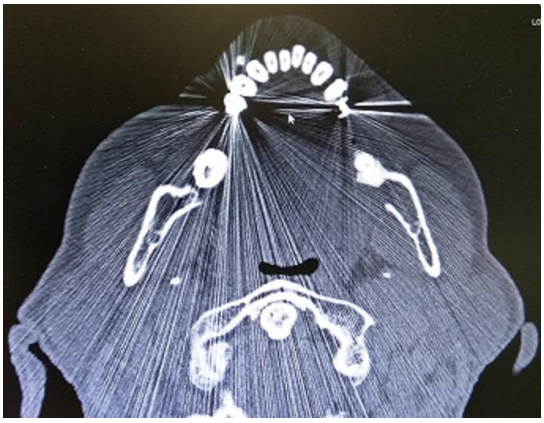

Figure 2 CT scan revealing a roundish, thick, cystic and radiolucent lesion at the left mandibular angle

At the medial side, the cortical bone appears partially interrupted. At the left maxillary sinus floor it can be observed an inflammatory mucosal thickening, measuring around 2 x 1.7 cm. Histopathologically, the lesion was characterized by fibrous connective tissue with lympho-plasmocytic chronic inflammation. There were also glandular cystic dilated structures consisting of transitional epithelium (figure 3), confirmed through immunohistochemical positivity for low molecular weight cheratin (figure 4), squamous in basal and parabasal localization, and glandular in apical localization the latter consisted of both mucin and columnar ciliated elements, with absence of atypical elements and mitosis. It was performed an angle-mandibular cystectomy and second molar surgical extraction. Antibiotic Rifocin irrigation and U-shape suture were performed.

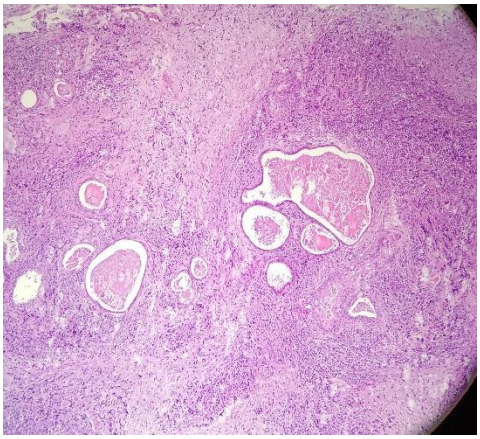

Figure 3 Microscopic picture of lesion with presence of glandular structures (E.E. 4X)

Figure 4 Detail of CK 7 immunohistochemical positivity in transitional epithelium (10X)

3. Discussion

GOC is a local aggressive cyst from odontogenic origin, with a frequency of 0.012%-1.3% of all the jaw cyst and its prevalence is 0.17% [11]. The mandible is the most common site for development, especially the anterior region [11].

According to a study that included the description of 169 cases, the lesions are commonly associated with bone expansion (73%), tooth displacement or un-erupted tooth (30,9%), cortical bone perforation (26%), presence of clinical symptoms (24,3%), root resorption (13,9%) [12].

GOC shows certain histological characteristics, which have been divided into major and minor categories by Kaplan et al. Major criteria include squamous epithelial lining with flat interface with the connective tissue wall, variations in the thickness of the lining, cuboidal eosinophilic cells, mucous goblet cells with intra-epithelial mucous pods with or without crypts lined by mucous-producing cells, inter-epithelial glandular microcystic or duct-like structures. Minor criteria include ciliated cells, multicystic or multiluminal architecture, clear or vacuolated basal cells [13].

Histopathologically, GOC should be differentiated from LPC, a developmental odontogenic cyst lined by thin non-keratinized epithelium. LPC is a commonly considered mimicker of GOC because it also exhibits focal epithelial thickenings and glycogen-rich epithelial cells, similar to those found in GOC.

The presented case shows most of the characteristic features of GOC describes above. The microscopic features of GOC have been found to resemble a number of lesions having a wide clinico-pathologic spectrum ranging from other odontogenic cysts, as lateral periodontal cyst, to malignant neoplasms such as mucoepidermoid carcinoma. Hence careful and detailed microscopic examination becomes essential for a correct differential diagnosis [14, 15].

The exact treatment of choice of this lesion remains controversial, and varies from enucleation and curettage to local block excision depending on the size and aggressiveness.

4. Conclusion

In conclusion GOC is a rare jaw cyst of odontogenic origin. It has a potentially aggressive behavior and high recurrence rate, so definite diagnosis is very important and should be carefully determined by correlating the microscopic criteria with clinical and radiographical findings. Furthermore, long term follow-up is mandatory not to overlook any recurrence.

Compliance with ethical standards

Disclosure of conflict of interest

None to declare.

Statement of informed consent

Informed consent was obtained from the patient included in the study.

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