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Rare Case of Glandular Odontogenic Cyst in Maxila: Diagnosis and Treatment

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Abstract

The Glandular Odontogenic Cyst presents itself as a rare odontogenic cyst of development that affects in greater number of male individuals, with an average of 48 years of age. Having prevalence in the anterior region of the mandible, being able to cross the midline, it presents radiographically as a unilocular or multilocular, radiolucent lesion, with well-defined limits and sclerotic borders. Histopathologically, it is covered by squamous epithelium with cubic or columnar cells, usually containing mucicarminophilic inside the epithelium, may resemble low-grade intraosseous mucoepidermoid carcinoma. Thus, the article in question reports the case of a 34-year-old male patient who seeks dental care for oral rehabilitation with dental implants, on a radiographic examination it was observed radiolucent areas, unilocular, well-circumscribed in the region of elements 16 to 22, crossing the midline. The treatment of choice consisted of decompression of the lesion and subsequent enucleation. The patient was followed up and no signs of recurrence were observed. Thus, observed the importance of a correct treatment plan, valuing the care of the adjacent structures and reducing postoperative complications.

Keywords

Decompression; Mandible; Recurrence

Introduction

Odontogenic cysts originate from the epithelium present in the odontogenesis process and its embryonic remains. The cystic cavity is filled with liquid material and coated with epithelium [1]. Glandular Odontogenic Cyst (COG) is a rare type of cyst that was first described in 1987 as a "sialo-odontogenic" cyst by Padayache and Van Wynk. However, in 1988 its nomenclature was altered to Glandular Odontogenic Cyst by Gardner et al. , since its glandular origin was not evidenced and to have the odontogenic origin emphasized. It was recognized by the World Health Organization (WHO) in 1992, classifying itself as a developmental odontogenic cyst, because salivary glandular origin had not been confirmed and histologically its characteristics were indicative of origin [2]. It is a developmental cyst considered rare, which usually affects the anterior region of the mandible, being accepted as a cyst of odontogenic origin, also exhibiting glandular characteristics [3]. Three possibilities for the formation of the Glandular Odontogenic Cyst can be assumed: A glandular cyst may have originated from a salivary gland or epithelial remains differentiated into glandular epithelium. A primordial cyst may have undergone prosoplasia in its epithelial lining and originated glandular epithelium. A low-grade mucoepidermoid carcinoma originated a cystic space and not multicystic spaces [4]. A predilection for the female sex is observed, usually between the fifth or sixth decades of life, with slow growth, and may also be destructive and recurrent. It is difficult to diagnose, as it may resemble other cystic entities both radiographically and histologically. Even though none of the clinical or radiographic characteristics are pathognomonic, this type of lesion presents potentially aggressive behavior [4].

Dental elements close to the lesion usually have a positive response to pulp vitality tests, but dental inclination may be noted due to the expansion of the lesion. In occlusal radiographs, it is possible to perceive expansion of bone corticals, and may be accompanied by perforation [2]. Histologically, non-keratinized squamous epithelium with cuboid or superficial columnar cells is observed, and may be confused with a low-grade intraosseous mucoepidermoid carcinoma, multi-cut examination allows these to be differentiated injuries [3]. Radiographically, it usually presents as a radiolucent area, with well-defined limits and can be uni or multilocular [5]. Histologically resembles other maxillary cysts, such as botrioid cysts, toothcysts, mucoepidermoid carcinoma, so it is important to make

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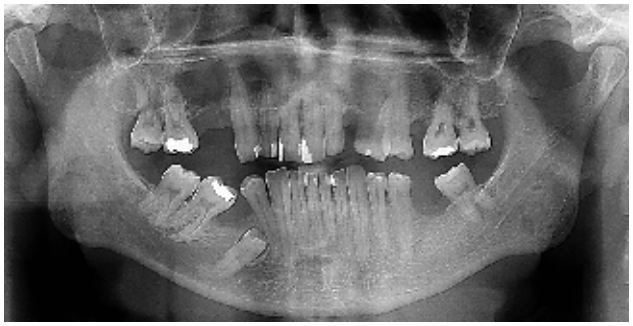

Figure 1: Initial panoramic radiography

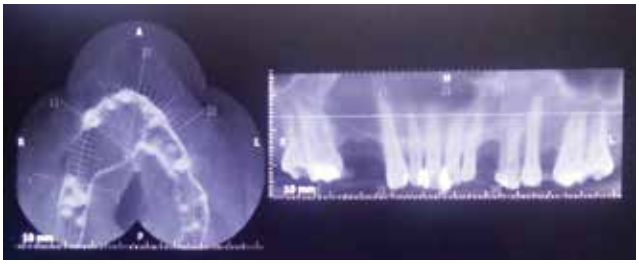

Figure 2: Tomography evidencing maxillary lesion

Figure 3: Intraoral aspect with increased volume in the right maxillary region

Figure 4: Aspirated lesion content

the differential diagnosis [5]. The most reported symptom is local edema, which may come accompanied by pain or not, depending on some factors, such as compression of nerve structures or secondary infections, depending on the site may also be observed paresthesia in some nerves [6]. Conservative treatment consisting of enucleation and curettage is preferred by many surgeons; however, there is a chance of recurrence, being more common in multilocular lesions, some opt for block resection, especially in multilocular lesions [3]. Dessa forma, o presente trabalho tem como objetivo relatar o diagnóstico e tratamento cirúrgico de um caso raro de cisto glandular em maxila através dos métodos de enucleação associada à descompressão.

Case Report

A 34-year-old male leukoderma patient randomly attended a private clinic specialized in oral rehabilitation to perform rehabilitation treatment with dental implants. During the initial evaluation for rehabilitation planning, panoramic radiography (Figure 1) and conical beam computed tomography (Figure 2), lesion in the right maxillary region, more precisely between elements 16 and 22, were detected in panoramic radiography (Figure 1) and computed tomography (Figure 2). On extraoral examination, no facial asymmetry was detected. On intraoral examination, volume alteration scans were detected in mucous/gingival tissue during palpation in the region of element 16 and a hard palate on the right side (Figure 3). The absence of dental elements 16, 15, 14, 23 and 26 was noted. On tomographic examination, one can observe a radiolucent area well circumscribed by radiopaque, unilocular line, covering an extensive area of the region of element 16 to element 22, crossing the midline. According to the clinical and radiographic characteristics, the diagnosis of a possible cystic lesion was raised. Therefore, as an auxiliary method of diagnosis it was decided to perform aspiration puncture, resulting in the presence of yellowish fluid with traces of blood, characteristics of cystic fluid with the presence of a mucous content (Figure 4). Clinical, radiographic and aspiration puncture findings helped us to choose the type of treatment, so cystic decompression was chosen with the aid of a drain (Figure 5) due to the extension of the lesion, with removal of the drain after 21 days, of this a panoramic radiography was requested and bone neoformation and reduction of the lesion were observed (Figure 6), then it was decided to perform total enucleation of the lesion (Figure 7). During the surgical act of decompression, incisional biopsy was performed for pathological analysis (Figure 8). Two fragments of brown and elastic tissue were collected, the largest measuring 1.5 x 1.5 cm, being

Figure 5: Drain positioned in cystic cavity.

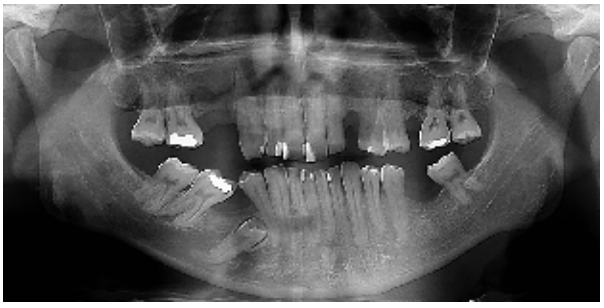

Figure 6: Radiography showing bone neoformation and decreased lesion

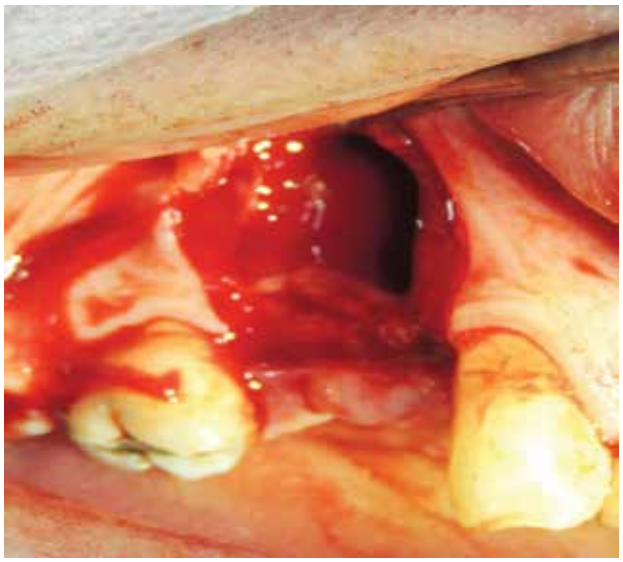

Figure 7: Enucleation of the lesion

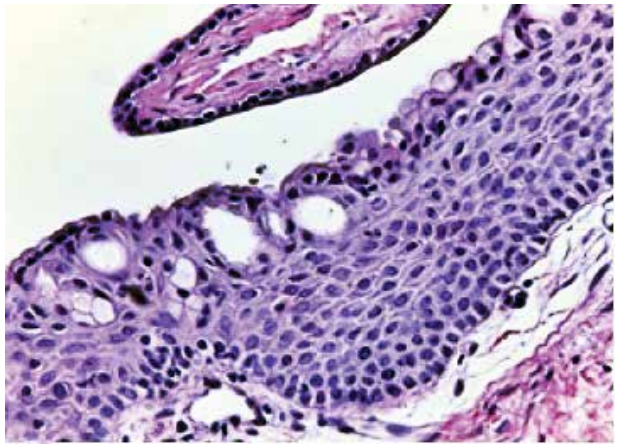

Figure 8: Blade presenting epithelium with cuboid cells

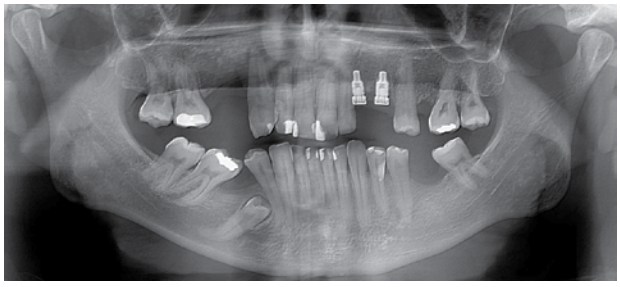

Figure 9: Radiographic aspect after six months of enucleation

fixed in 10% formaldehyde. The pathological examination confirmed the diagnosis of cystic lesion, compatible with Glandr Odontogenic Cyst (GOC). The postoperative aspect of 06 months is shown in a panoramic image, where bone neoformation is noted in the residual cavity and absence of recurrences (Figure 9).

Discussion

The glandular odontogenic cyst presents as a rare pathology, which presents aggressive growth and high recurrence rate, is often misdiagnosed due to the fact that it resembles other lesions of the maxillofacial complex. The prevalence of this lesion is reported in the anterior region of the mandible [7]. In the present study, the lesion is in the maxillary region, going from the posterior limit to the anterior, crossing the midline. According to Shah, et al. [8], lesions such as residual cysts, keratocysts, botrioid cysts, central mucoepidermoid carcinoma, ameloblastoma should be included in the differential diagnosis. Which have similarities with cog. Radiographically, it usually presents as a uni or multilocular radiolucent lesion, with well-defined sclerotic edges [9]. The case in question presented a unilocular radiolucent lesion, having well-defined edges and limits, with no root resorption of the teeth involved. Histologically, it presents squamous epithelium coating with cuboid or columnar cells. It may resemble otherlesions, such as botrióidecystand low-grade mucoepidermous carcinoma, thus hindering the definitive diagnosis [3]. The histogenesis of COG is uncertain, but many authors believe that it derives from the odontogenic epithelium, due to its morphology and the fact that it originates in areas close to the teeth. At the same time salivary components were also observed [10]. Aspiration puncture may be useful in the diagnosis of glandular odontogenic cyst, which is performed before the surgical procedure. Fluids of different aspect are present in the literature [11]. Several treatment modalities are present in the literature, this range from conservative methods to more invasive methods. Conservative methods consist of enucleation with or without previous decompression, which are usually followed by some auxiliary method, to decrease the chances of recurrence, such as curettage, peripheral osteotomy, cauterization, or cryocautery. More invasive methods consist of resection of the lesion with safety margin [9].

The most commonly used form of treatment consists of enucleation and curettage, however, due to the high degree of recurrence, which is more prevalent in multilocular lesions, many authors opt for the block resection method [12]. The treatment used in the present case consisted of previous decompression followed by enucleation. According to Chandra, et al. [13], this lesion is often confused with others because it has nonspecific characteristics. Thus, careful evaluation is extremely important to plan the appropriate form of treatment. Small diameter unilocular lesions have enucleation as the treatment of choice. While unilocular lesions of larger diameter have as treatment enucleation with peripheral osteotomy, thetreatment of multilocular lesions includes partial or marginal resection [14]. The recurrence rate of COG is directly related to the size of the lesion, so the treatments are different for large and small lesions. Presenting more aggressive treatment for the larger ones, these should also be monitored for a long period of time [15].

Conclusion

It can be concluded that the diagnosis should be made through the association of clinical, histological and imaging examinations, since such lesion may resemble other lesions of the maxillofacial complex. The diagnosis and treatment of pathologies in the oral cavity are extremely important, thus minimizing the morbidity rate and damage to the patient's health. The development of an appropriate treatment plan is essential for a good prognosis. It is also extremely important the periodic follow-up of the patient, thus minimizing greater complications due to possible cases of recurrence.

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