Simultaneous Occurrence of Central Giant Cell Granuloma and Odontogenic Keratocyst in Mandible

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Abstract

Odontogenic keratocysts and central giant cell granulomas are distinct entities of separate origin that may appear simultaneously in the jaw, making them difficult to differentiate. The objectives of this paper are to report a case of a 54-year-old man presenting with a large lesion in the right mandible and to describe the histological characteristics of that lesion. Imaging revealed tumefaction and multilobulation. The case was surgically managed. Histopathological examination revealed that the lesion was a hybrid of an odontogenic keratocyst and a central giant cell granuloma. It remains unclear as to whether this lesion represented a coincidental or related injury. Since both types of lesion involved have a high rate of recurrence, however, long-term follow-up is scheduled.

Key words: Central giant cell granuloma — Odontogenic keratocyst — Jaw — Mandible

Introduction

Odontogenic tumors originate in epithelial, ectomesenchymal, or mesenchymal tissues involved in the formation of the dental apparatus. According to the World Health Organization, such tumors are found exclusively in the maxillofacial region and may occur at any age⁵. The odontogenic keratocyst, one of the most studied odontogenic lesions, usually affects males in the second, third, or fourth decades of life, and posterior mandible involvement is particularly common⁵. The central giant cell granuloma (CGCG), a distinct entity, is a benign tumor of non-odontogenic origin that exhibits locally destructive behavior affecting the bones of the craniomaxillofacial complex⁶. This pathology represents approximately 7% of benign lesions in the maxillomandibular complex.
and mainly affects women between the first and third decades of life. Histologically, it is composed of fibrous connective tissue with foci of hemorrhage and hemosiderin deposits, osteoclast-like giant cells, and reactive bone \(^8\). However, only a few cases of a hybrid lesion containing characteristics of both odontogenic keratocyst and CGCG have been reported, indicating that the present case is rare and therefore of importance.

**Case Presentation**

The patient was a 54-year-old Caucasian man who was referred to the Department of Oral and Maxillofacial Surgery at the Erasto Gaertner Hospital by his dentist for a bone tumor detected on a routine imaging examination. The only comorbidity was systemic arterial hypertension, which was being controlled with medication. An oral examination revealed a 3-cm swelling in the right mandible. The lesion showed bone consistency and was covered with healthy mucosa (Fig. 1). Cone Beam CT revealed a lytic, multilobulated lesion approximately 30×20 mm in size (Fig. 2). Some areas presented a thick septum, giving the impression of separation between the wells (Fig. 3).

**Clinical Procedures and Outcomes**

The treatment option was enucleation under general anesthesia (Fig. 4). As seen in the CT image, 3 separate wells with narrow connective areas were observed intraoperatively. Histopathological examination revealed two different lesions. The material harvested from the posterior area comprised a fibrous cellular stroma with multiple multinucleated giant cells. Meanwhile, the anterior area of the lesion comprised a cystic lesion with thin epithelium containing cuboidal palisaded cells in the basal layer, and hyperchromatic and corrugated parakeratin layers in the luminal surface. Abundant desquamated keratin was observed in the cyst lumen (Fig. 5). The histopathological diagnosis was odontogenic keratocyst and CGCG. An immunohistochemical analysis for CD68 showed positive staining for giant cells (Fig. 6). Regular follow-up is being continued, and no recurrence has been observed at one year postoperatively (Fig. 7). The patient is scheduled to return to the hospital every 6 months for a panoramic X-ray of both lesions in accordance with the protocol of this institute.
Fig. 3 CT in axial view revealed thick septum separating lesion's wells

Fig. 4 Intraoperative aspect after osteotomy showing pathological cavities

Fig. 5 Histological blades left side, fibrous cellular stroma with multiple multinucleated giant cells (arrow)
Right side, cystic lesion with thin epithelium containing cuboidal palisaded cells in basal layer, and hyperchromatic and corrugated parakeratin layers in luminal surface.

Fig. 6 Immunohistochemical analysis showed positive staining for CD68, highlighting giant cells (arrow)

Fig. 7 Panoramic X-ray obtained at 1 year postoperatively revealed bone formation in progress
Discussion

Hybrid lesions, or lesions that occur simultaneously at the same site, have been described in the literature, and there are some reports of an odontogenic keratocyst found in association with cartilage chondroma and others presenting cartilage on histological examination. Other authors have also reported simultaneous cases of odontogenic keratocyst and traumatic bone cyst.

Some reports have described CGCGs occurring simultaneously with other types of lesion, such as odontogenic fibroma and ameloblastoma. A consensus remains to be reached, however, on whether these cases represent true hybrid injuries or only a collision, meaning two independent lesions forming and developing side by side.

To our knowledge, only 3 cases of an odontogenic keratocyst occurring simultaneously with a CGCG lesion have been reported to date. Yoon et al., who described the first such case in the left posterior mandible of a 10-year-old boy, believes that it might be an odontogenic tumor where there was a reactive component of giant cells, but does not rule out the possibility of the injury being independent and only coincidental. Yoon et al. emphasize that the CGCG-like lesion concerned may have resulted from a reactive osteoclastic process already destroying bone in the odontogenic keratocyst region. The second and third cases, published respectively by Adyanthaya and Jose and Ravi et al., described a similar case in the left posterior mandible, both involving 29-year-old men. Both authors agree that their cases probably involved two lesions coalescing, as the CGCG-like area was observed mainly in only one part of the lesion. Ravi et al. noted that root resorption was unusual in odontogenic keratocysts, with occurrence varying from between only 4.4 to 11%. In the case they reported, however, root resorption was severe, affecting approximately half the length of the root in teeth #34, 35, 36, and 37. On the other hand, CGCG is reported to be aggressive, with root resorption being a common feature and with varying degrees of expansion or cortical plates. The features of the present case appear to agree with some of these earlier findings, as slight root resorption in tooth #45 and expansion of cortical bone were observed in the same region, which is where the major CGCG component was located.

It remains unclear whether the present case represents coincidental or related injury, mainly because there was only a small distance between the two types of lesion. On the initial examination, the multilobulated aspect of the lesion suggested a concomitant pathology, indicating how such a morphology can confuse a diagnosis. The treatment option was surgical enucleation of the lesion under general anesthesia. Histopathological analysis after enucleation confirmed the diagnosis of concomitant lesions. As both lesions have a high rate of recurrence, long-term follow-up has been scheduled.

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