



Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp



Case report

Decompression of an extensive Keratocystic Odontogenic Tumor in an elderly patient

Clenivaldo Alves Caixeta^{a,*}, Cíntia Magalhães Ribeiro^b, Marina Lara de Carli^c,
Felipe Fornias Sperandio^d, Alessandro Antônio Costa Pereira^e,
João Adolfo Costa Hanemann^f

^a Department of Clinic and Surgery, School of Dentistry, Federal University of Alfenas, Brazil

^b Department of Clinic and Surgery, School of Dentistry, Federal University of Alfenas, Brazil

^c Department of Clinic and Surgery, School of Dentistry, Federal University of Alfenas, Brazil

^d Department of Pathology and Parasitology, Institute of Biomedical Sciences, Federal University of Alfenas, Brazil

^e Department of Pathology and Parasitology, Institute of Biomedical Sciences, Federal University of Alfenas, Brazil

^f Department of Clinic and Surgery, School of Dentistry, Federal University of Alfenas, Brazil

ARTICLE INFO

Article history:

Received 14 September 2016

Received in revised form 19 October 2016

Accepted 1 November 2016

Available online xxx

Keywords:

Odontogenic keratocyst

Decompression

Mandible

Surgical management

ABSTRACT

The Keratocystic Odontogenic Tumor (KCOT) is a benign intraosseous neoplasm, usually found between the second and fourth decades of life, and with a large predilection for the mandible, especially the ramus and angle regions. The KCOT is characterized by a high rate of recurrence and its treatment choices are still very controversial, usually falling into conservative or aggressive approaches. Here we report a 65-year-old patient with an extensive KCOT involving the body, angle and ramus of the mandible. The treatment consisted only in decompression and, the lesion disappeared completely along with facial contour restoration; five years of follow-up show no evidence of recurrence. Thus, we emphasize the importance of a conservative treatment for extensive cases and situations that involve elderly patients or children, once conservative techniques are simpler, induce lower morbidity and allow for good success rates.

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1. Introduction

The Keratocystic Odontogenic Tumor (KCOT), previously known as Odontogenic Keratocyst (OKC), was first described in 1956 [1,2]. Ever since then and until, it was considered to be an odontogenic development cyst; in 2005, though, the term KCOT was introduced due to its aggressive clinical behavior and, high recurrence rates that better resemble a neoplastic nature [3]. Such classification change is also based on the conspicuous presence of tumor islands, microcysts within the fibrous walls of the cyst [4] and mitotic figures present in the above-basal layers [5].

KCOT has a predilection for males (2:1) [1,2,6] and, can affect patients from the first to ninth decades of life [2], although it has a higher prevalence between the second and fourth decades [2,6,7]. Radiographically, it appears as a radiolucent uni or multilocular image, often surrounded by a sclerotic well defined border [3,6]. Mainly affects the posterior region of the mandible and maxilla in when these lesions tend to be smaller [3]. Clinical growth is more pronounced through the anterior-posterior direction without expanding the cortical [8], and thus KCOT has the potential to achieve large sizes without getting noticed, only being discovered after routine panoramic radiographic examinations [6,9].

In fact, KCOT has an aggressive clinical behavior with high recurrence rate that can range from 0 to 62% [4,6,10]. This high recurrence has been explained by the presence of satellite cysts or epithelial islands in the mucosa [4], the type of treatment performed, follow up period [10], and eventually a new KCOT developed in any area and misinterpreted as relapse [4].

Several treatment modalities have been proposed, and are usually classified as conservative or aggressive [6,7]. In order to choose from such therapies various factors such as the patient's age, size and location of the lesion, soft tissue involvement and history of

* AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

* * Corresponding author at: Department of Clinic and Surgery, School of Dentistry, Federal University of Alfenas, Rua Gabriel Monteiro da Silva, 700 – Alfenas, MG 37130–000, Brazil.

E-mail address: clenivaldodontista@yahoo.com.br (C.A. Caixeta).

<http://dx.doi.org/10.1016/j.ajoms.2016.11.001>

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Fig. 1. Extra-oral clinical aspect demonstrating facial asymmetry caused by tumor in the left parotid region.

recurrence need to be considered [2,11]. In that way, the aim of this study is to report a 65-year-old patient who presented an extensive KCOT that involved the mandible from its middle portion of the body to its coronoid process and who was treated only with decompression. After five years of follow-up, the patient presented total regression of the lesion and absence of recurrence.

2. Case report

A 65-year-old female patient was referred to the Stomatology Clinic for evaluation and treatment of an extensive lesion on the left side of the face. The patient reported that the lesion was present for several months and that showed continuous growth. She also reported to be hypertensive and to cardiopathy. The extra-oral physical exam revealed facial asymmetry, which was caused by an diffuse volumetric increase in the left parotid region (Fig. 1). Intra-orally, we noted a swelling area, of imprecise limits, covered by normal colored and consistent flabby mucosa and causing deletion of the left oral vestibule. A conventional panoramic radiographic exam showed a multiloculated radiolucent lesion, expanding and destroying the bone cortical, extending from the middle portion of the left body until the coronoid process (Fig. 2). The tomographic exam revealed the extent of the damage and the almost total

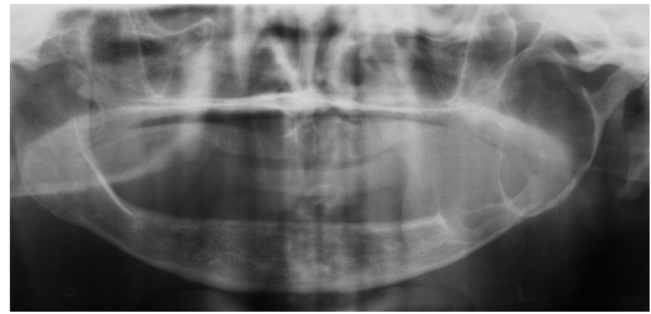


Fig. 2. Conventional panoramic radiograph revealing multilocular osteolytic lesion causing destruction of the body and ramus of the mandible.

impairment of the body region and ramus of the mandible on the left side (Fig. 3A, B). On the basis of clinical and radiographic aspects, it was suggested as diagnostic hypotheses keratocystic odontogenic tumor, ameloblastoma, ameloblastic fibroma, and residual cyst. Puncture aspiration was performed, which was positive for a white-yellowish content. Then, an incisional biopsy was performed and the fragment of tissue was referred to histopathological analysis, following the installation of a drain for decompression of the lesion; the drain stayed put for 7 days (Figs. 4A, B). The histopathological analysis revealed fibrous connective tissue composing a cystic capsule, while lacking inflammation and lined by thin, uniform, stratified cuboid/squamous epithelium, corrugated at its surface layers of parakeratin. Hyperchromatic basal cells in palisade, besides free fragments of keratin within the cystic lumen and also flat epithelial-fibrous tissue junction were noted. Based on those histopathological findings we established the diagnosis of KCOT (Fig. 5). Five years after decompression, the local site shows bone neoformation, facial contour restoration and no clinical or radiographic signs of recurrence (Fig. 6).

3. Discussion

KCOT is a benign odontogenic neoplasm that mainly affects male individuals, usually more frequent between the second and fourth decades of life, and with a wide predilection for the mandible, especially the ramus and angle regions [6,7,12,13]. In this report, we present a KCOT diagnosed in a 65-year-old female patient, what diverges from what is more frequently reported in the literature. However, regarding the location, the lesion arose from the posterior region of the mandible, which is among the most regular

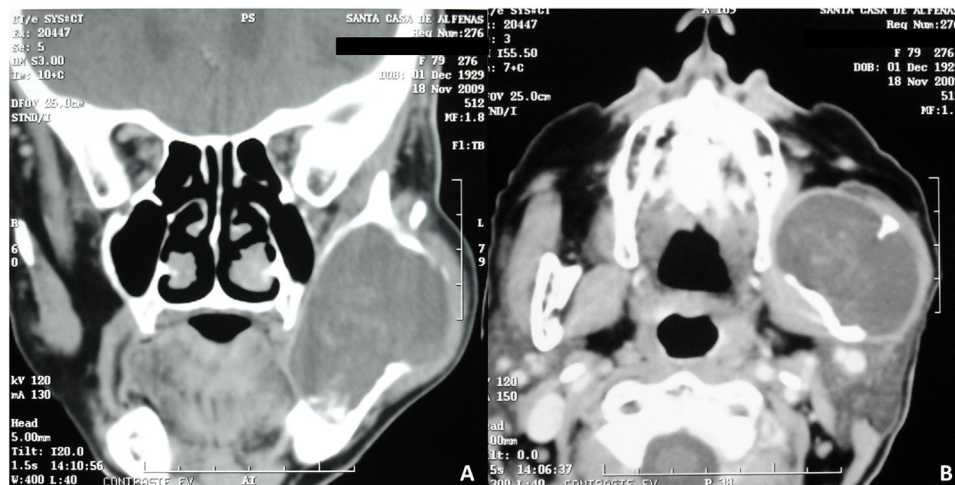


Fig. 3. A e B Tomographic coronal and axial cuts showing expansion and destruction of the vestibular and lingual cortical and increased volume of soft tissue.

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