RELATO DE CASO

Squamous odontogenic tumor: with recurrence and 12 years of follow-up

Tumor odontogênico escamoso: com recidiva e doze anos de acompanhamento

Márcia Gaiger de OLIVEIRA[1]
Vinícius Coelho CARRARD[2]
Cristiane Cademartori DANESI[3]
Pantelis Varvaki RADOS[1]
Manoel SANT´ANA FILHO[1]

ABSTRACT

Squamous odontogenic tumor is a rare benign neoplasm and may affect multiple sites in the mouth. The authors review the clinical, radiographic and histopathological features of Squamous odontogenic tumor and report a case of a patient with recurrent Squamous odontogenic tumor and 12 years of follow-up and discuss diagnostic criteria and therapeutic approaches.

Indexing terms: *Diagnosis. Recurrence. Odontogenic tumors.*

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[1] Universidade Federal do Rio Grande do Sul, Faculdade de Odontologia, Programa de Pós-Graduação em Odontologia. R. Ramiro Barcelos, 2492, sala 503, 90035-003, Porto Alegre, RS, Brasil. Correspondência para/Correspondence to: M. SANT´ANA FILHO. *E-mail*: <manoel@ufrgs.br>.

[2] Doutorando em Patologia Bucal, Universidade Federal do Rio Grande do Sul, Programa de Pós-Graduação em Odontologia, Porto Alegre, RS.

[3] Universidade Federal de Santa Maria, Departamento de Patologia. Santa Maria, RS, Brasil.

RESUMO

O tumor odontogênico escamoso é uma neoplasia benigna rara que pode ser única ou múltipla. Os autores apresentam uma revisão das características clínicas, radiográficas e histopatológicas do tumor odontogênico escamoso e relatam um caso de um paciente com recidiva com acompanhamento de doze anos e discutem os critérios de diagnóstico e abordagens terapêuticas.

Termos de indexação: Diagnóstico. Recidiva. Tumores odontogênicos.

INTRODUCTION

Squamous odontogenic tumor (SOT) is a rare benign neoplasm first described in 1975 and now classified as an independent entity. It is thought to arise from a neoplastic change of epithelial rests of Malassez[1-3].

Squamous odontogenic tumor occurs mainly in the third decade of life and is approximately equally distributed between the maxilla and the mandible. In some cases, it may affect multiple sites in the mouth[1,2,4-5]. Leider et al.[6] reported three cases of SOTs in siblings, which suggests a possible familial pattern in the occurrence of this lesion. Radiographic features of SOT are non-specific and consist of a triangular-shaped radiolucent lesion adjacent to the roots of teeth[2]. The lesion is usually central, but sometimes it may be peripheral[7,8].

Its typical microscopic appearance justifies the name "squamous odontogenic tumor": a stroma of mature connective tissue with islands of odontogenic epithelium. These islands have a purely squamous pattern, and the peripheral cells do not show the typical pre-ameloblast polarization seen in ameloblastomas. Cystic degeneration in the center of the islands is a frequent finding. Pre-keratin is found in some epithelial cells, and laminated calcifications may be seen inside keratin pearls[1,2,4-7,9-14].

Different therapeutic approaches, ranging from conservative, such as curettage, to radical surgery, such as hemimandibulectomy, have been adopted[15,16].

The purposes of this paper are to report a clinical case of a patient with a recurrent SOT and a 12-years follow-up, and to discuss diagnostic criteria and treatment approaches.

Case report

A 28-year-old white man presented a swelling in the first and second molar region of the left side of mandible. The swelling had been observed three years before consultation, but did not cause any symptomatology. Tooth 36 and 37 were missing. The patient reported that a biopsy was performed one year before consultation, but the diagnosis was inconclusive.

Radiographs revealed a unilocular radiolucency with undefined borders between the roots of teeth 35 and 38 (Figure 1).

An incisional biopsy was performed, and microscopic examination revealed proliferation of

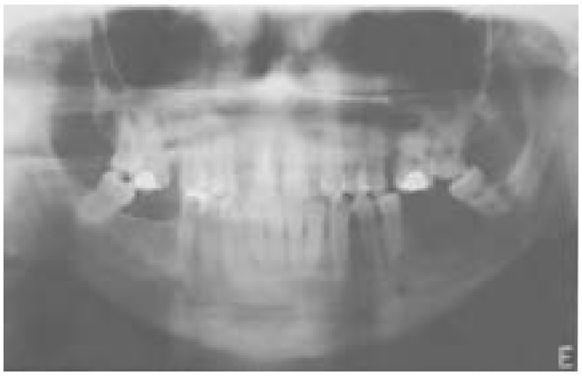

Figure 1. Initial panoramic radiograph showing radiolucent area with undefined borders.

odontogenic epithelium with islands of squamous epithelial cells surrounded by peripheral layers of flat or low cuboidal cells (Figure 2). This pattern was repeated in the regions where the lesion was homogeneous and had a high degree of cellularity. In some areas, the odontogenic epithelium infiltrated the fibrous connective tissue. No atypical mitoses or cellular pleomorphism were observed. The histopathological diagnosis was squamous odontogenic tumor.

The tumor was enucleated, but 11 months later, when the patient returned for follow-up, radiographs revealed a larger multilocular lesion and tumor recurrence was observed. The histological features observed were similar to those found in the previous study, showing islands of odontogenic epithelium exhibiting peripheral cells without polarization. An en bloc resection was performed and a metal reconstruction plate was used.

The patient was followed up every year for two years and no recurrence was observed. However, three years after the second surgery, radiographic control showed a radiolucent area in the base of the mandible. The patient was kept under strict annual control. After eight years, the radiographic image has not changed, and has been interpreted as a healing pattern (Figure 3). In spite of that, annual clinical and radiographic follow-up is planned in order to identify early any possible sign of recurrence.

DISCUSSION

Squamous odontogenic tumor is a rare lesion with few cases reported to date in the literature. Pullon et al.[1] were the first to describe it. They reported six cases and established diagnostic criteria and surgical approaches that are still followed today.

Squamous odontogenic tumor clinical and radiographic features are neither unique nor sufficient for diagnosis, as this tumor may be confused with a number of other pathologies. Patients may present with an increase in the volume of the maxilla or mandible, tooth mobility, ulceration of the oral soft tissue, painful symptoms, and tooth displacement. Our patient did not present with tooth mobility, but two teeth in the area affected by the tumor had been lost due to unknown causes.

According to Hopper et al.[4], the type of radiographic border may help define the type of treatment to be adopted since a more aggressive lesion has poorly defined radiographic borders. Therefore, a definitive diagnosis requires that a more extensive biopsy be performed to obtain a larger portion of the lesion. In the case reported here the lesion had an initial unilocular appearance that was later identified as multilocular with poorly defined borders, which may explain recurrence in less than one year.

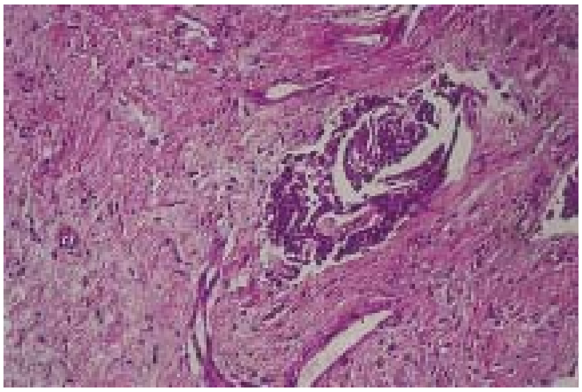

Figure 2. Photomicrograph showing island of squamous odontogenic epithelial cells surrounded by peripheral layers of low cuboidal cells in a mature connective tissue stroma (Hematoxylin and eosin stain. Magnification X100).

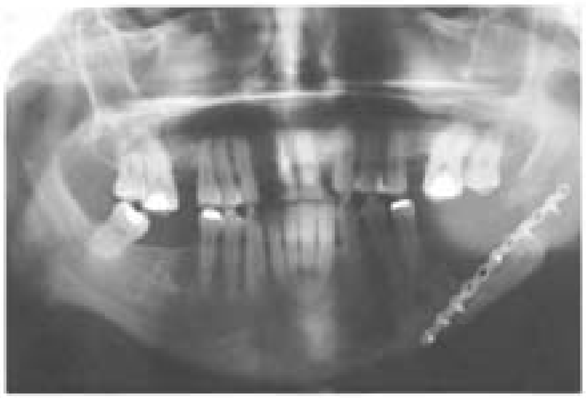

Figure 3. Last radiographic control 11 years after the second surgery showing radiolucent area interpreted as a healing pattern.

Although surgical excision is successful in most cases, studies in literature report two cases of recurrence in a short period[1,7]. This may suggest that these lesions do not always behave similarly. Many cases treated with excision were followed up for a relatively short period[1,5,7,13]. The use of en bloc resection for all cases, as proposed by Doyle et al.[9], may be an excessively radical approach. Practical experience with ameloblastomas has shown that some odontogenic tumors may have late recurrence, and this is particularly important in cases of SOT, as most cases reported in literature have been followed up for a short period.

The morphologic appearance of the islands of odontogenic epithelium in SOT is sometimes similar to the follicular pattern of ameloblastomas, which may lead pathologists to misclassify it as a benign odontogenic epithelial tumor, desmoplastic ameloblastoma, or ameloblastic fibroma, as have occurred in some cases[16]. However, lack of polarization of peripheral cells in the epithelial islands, which is typical of ameloblastomas, is a differential criterion in favor of SOT.

In our case, no intraepithelial calcifications or intracellular keratinization was observed. The appearance of the squamous epithelial islands, with peripheral cell do not exhibiting the polarization characteristics of ameloblastoma, was of major importance in establishing the diagnosis. Mostly, they showed the typical arrangement described by Pullon et al.[1], but sometimes had an invasive pattern, mixing with fibrous connective tissue, a feature already reported by McNeill et al.[5]. This may suggest an infiltrative growth similar to that of ameloblastomas, which emphasizes the importance of life-long follow up. In spite of that, no atypia, pleomorphism or mitoses were observed, which ruled out primary intraosseous carcinoma. These microscopic features, as well as the surgeon's information about the difficulty in separating the lesion from the bone, and the radiographic image with poorly defined borders may explain recurrence.

Our little experience with this type of lesion and the limited number of cases published to date, as well as the features of the case reported here and the analysis of the literature, led us to the conclusion that an individual analysis of each case is important in choosing treatment. Microscopic investigation alone does not provide the variables required to support a decision. As reported by Hopper et al.[4] it is essential that the radiographic borders of the lesion be observed. In the case reported here, this was only taken into consideration when the tumor recurred, which then led us to a broader approach than the one adopted during the first intervention.

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