

# Squamous odontogenic tumor: report of a case of unusual involvement

## *Tumor odontogênico escamoso: relato de um caso de envolvimento incomum*

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### ABSTRACT

The squamous odontogenic tumor (SOT) is defined as a very rare benign neoplasm, locally infiltrative and can extend to neighboring structures. This study aimed to report a case of SOT unusual involvement. A female patient, 56-year-old, smoker showed mild swelling in the chin region in the portion between the left canine and right first premolar. Axial, coronal and sagittal images showed expansive hypodense lesion in the mandibular symphysis and parasymphysis. Partial removal of the lesion was performed, which led to the diagnosis of SOT. The patient presented no recurrence during 4 years and 2 months follow-up.

**Key words:** squamous odontogenic tumor; mandible; neoplasms.

### INTRODUCTION

Squamous odontogenic tumor (SOT) was described for the first time in 1975 by Pullon *et al.*<sup>(1)</sup>, who reported six cases of an oral lesion showing radiolucent area of bone destruction associated with the roots of adjacent teeth. However, it was only in 1992 that this lesion was described and classified as a distinct pathological entity by the World Health Organization (WHO).

Defined as a very rare benign neoplasm, SOT is locally infiltrative and can extend to neighboring structures such as the maxillary sinuses and nasal cavity. As observed during normal odontogenesis, SOT also arises from variable inductive interactions between the odontogenic epithelium and ectomesenchyme, and is the result of neoplastic transformation of epithelial cell rests of Malassez or rests of Serres, and its embryological origin derived from the dental lamina<sup>(2-4)</sup>.

Clinically, SOT appears as an asymptomatic swelling in the alveolar process and the affected teeth can exhibit discrete mobility and mild discomfort to percussion. In most cases, the tumor is accidentally discovered on routine dental radiographs, which reveal a generally well-defined, radiolucent, unilocular, triangular-shaped image between or along the roots of adjacent teeth, mimicking severe periodontal bone loss<sup>(4,5)</sup>.

Treatment of SOT consists of the surgical removal of the whole lesion, curettage and extraction of the affected teeth, if present. On the other hand, more extensive lesions infiltrating neighboring structures require more radical interventions such as en bloc resection<sup>(2,4-6)</sup>.

The objective of the present study was to describe the clinical, radiographic and histopathologic features of an uncommon case of SOT involving the mandible.

### CASE REPORT

A black female patient 56-year-old, smoker was seen at the Oral Maxillofacial Service complaining of maladjusted lower complete denture. Extraoral physical examination revealed mild swelling in the chin region which caused discrete facial asymmetry. The skin covering the region had normal color. Intraoral physical examination showed normal mucosa without signs of ulceration and the lesion was hard on palpation. The swelling and buccal cortical expansion occurred in the anterior region of the mandible, particularly in the portion between the left canine and right first premolar.

Axial, coronal and sagittal images (voxel size: 0.25 mm) were obtained from the mandibular arch. Analysis of the scans showed a

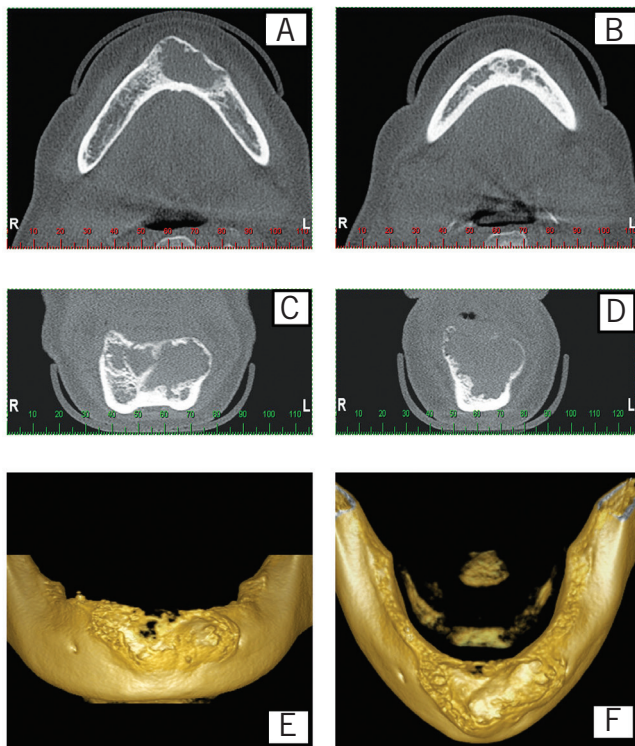
hyper dense image in the region of tooth 47 compatible with bone sclerosis, and an expansive hypodense lesion in the mandibular symphysis and parasymphysis, extending in the laterolateral direction until teeth 33 and 43 (**Figure 1A**). The image of the lesion was multilocular and there were no calcifications inside the lesion. Buccal cortical expansion and areas of bone destruction were also observed (**Figure 1B to 1F**). The remaining structures had a normal radiographic appearance. The lesion measured 16.75 mm (anteroposterior), 37.25 mm (laterolateral) and 20.51 mm (superior-inferior) in its major axes.

In view of this clinical presentation, the initial management adopted consisted of partial removal of the lesion (incisional biopsy), which led to the diagnosis of SOT. Thus, total resection of the tumor and curettage were performed, followed by the placement and adaptation of a reconstruction plate for mandibular reconstruction.

Microscopic analysis revealed the presence of epithelial islands of variables size and shape, which consisted of well-defined squamous epithelium with benign features (**Figure 2A and B**). The epithelial islands were immersed in a stroma of cellular and

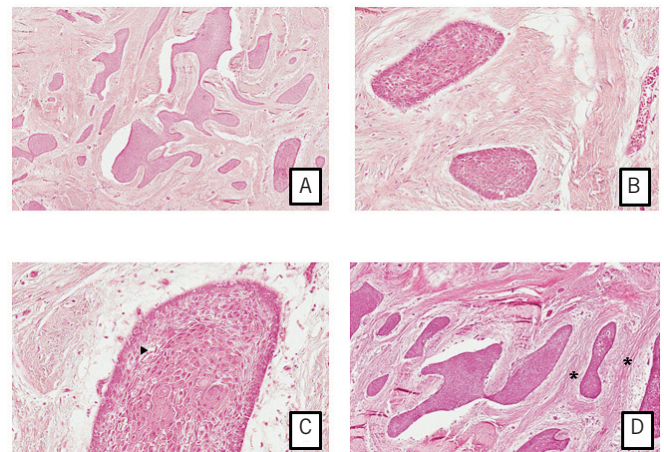
fibrous connective tissues and exhibited a peripheral layer of cuboidal cells without evidence of a palisade-like arrangement or nuclear polarization (**Figure 2C**). Intraepithelial microcystic degeneration (Figure 2C – arrow) and circular areas of fibrous condensation (**Figure 2D** – asterisk) were identified around some epithelial islands, suggesting a reaction of the connective tissue stroma to epithelial proliferation. On the basis of these histopathologic features, the definitive diagnosis was SOT.

The patient presented no postoperative complications and surgery provided good outcomes, with no recurrence during four years and two months follow-up.



**FIGURE 1** – Axial, coronal, sagittal and 3D images

Expansive hypodense lesion in the mandibular symphysis and parasymphysis (A); multilocular image, buccal cortical expansion and areas of bone destruction (B, E and F) – posterior region (C) and anterior region (D).



**FIGURE 2** – Epithelial islands of well-defined squamous epithelium with benign features

A and B) cellular stroma, peripheral layer of cuboidal cells; C) intraepithelial microcystic degeneration (arrow); D) circular areas of fibrous condensation (asterisk) (HE, 100× and 400×).

HE: hematoxylin and eosin.

## DISCUSSION

Odontogenic tumors arise from epithelial tissue, mesenchymal tissue, or both, which are involved in teeth development. The etiopathogenesis of these tumors and their causes or triggers are not well understood. Clinically, most of these tumors are asymptomatic, but tooth mobility, expansion and bone loss can be observed<sup>(2, 5, 6)</sup>.

Squamous odontogenic tumor is a very rare benign tumor that arises from remnants of the dental lamina and occurs equally in the mandible and maxilla. Tumors located in the anterior region of the mandible are uncommon. Despite the bone involvement and benign nature of SOT, this tumor can infiltrate adjacent soft tissues, causing major esthetic and functional impairment. In addition, SOT can appear as a single tumor or affect multiple sites at the same anatomic location (multicentric)<sup>(2-6)</sup>.

Characterized for the first time in 1975, SOT become largely known by the dissemination of a series of six cases described by Pullon *et al.*<sup>(1)</sup>. Until that time, some harmless lesions had been diagnosed and treated in a mutilating and aggressive manner since they were believed to be ameloblastomas or squamous cell carcinomas based on their histopathologic features. Since this first report, diagnostic criteria and a surgical approach have been established and some of these criteria are used until today<sup>(2, 4)</sup>.

Forty-seven cases of SOT have so far been reported in the English literature (PubMed Database). Adults are affected more frequently,

but the cases reported comprise a wide age-range (8-74 years), with a mean age of 36.1 years. There is no gender preference for SOT, the male to female ratio is 1:1.6. Squamous odontogenic tumors tend to arise in the canine-premolar area of the maxilla and in the molar region of the mandible, but it can occur along the jaws (**Table**). However, SOTs located in the maxilla show a more aggressive behavior due to the natural porous and medullary anatomy of this bone<sup>(2, 6-8)</sup>. In the present case, the tumor involved the mandibular anterior region, causing esthetic impairment but no painful symptoms. Among the 47 cases analyzed (Table), 10 involved the mandibular anterior region.

**TABLE – Cases published of SOT between 1975-2015 years (PubMed Database)**

Reference	Number of cases	Sex	Age (years)	Site	Radiographic findings
Pullon <i>et al.</i> (1975) <sup>(1)</sup>	6	M (3) F (3)	23/11/19/31/42/29	Multicentric (1) Maxilla-anterior (3) Mandible-posterior (2)	Radiolucent area; alveolar bone loss
Doyle <i>et al.</i> (1977) <sup>(14)</sup>	2	M	26/65	Maxilla-anterior e posterior	Radiolucent area; multilocular
McNeill <i>et al.</i> (1980) <sup>(15)</sup>	1	F	26	Multicentric	Radiolucency; bone loss
Hopper <i>et al.</i> (1980) <sup>(16)</sup>	1	F	22	Maxilla and mandible	Radiolucent area
Kangvonkit <i>et al.</i> (1981) <sup>(17)</sup>	1	M	32	Maxilla-anterior	Well-defined radiolucency
Carr <i>et al.</i> (1981) <sup>(18)</sup>	1	F	66	Maxilla-posterior	Triangular-shaped radiolucent area
Leventon <i>et al.</i> (1981) <sup>(19)</sup>	1	M	59	Mandible-posterior	Well-defined radiolucent area
Goldblatt <i>et al.</i> (1982) <sup>(20)</sup>	5	M (1) F (4)	60/29/30/26/67	Mandible-posterior (3) Maxilla-posterior (1) Not stated (1)	Well-defined; triangular-shaped radiolucent area
Swan, McDaniel (1983) <sup>(21)</sup>	1	M	32	Maxilla-posterior	Not stated
Cataldo <i>et al.</i> (1983) <sup>(22)</sup>	1	F	24	Mandible-anterior	Radiolucent area between roots of right canine and first premolar
Norris <i>et al.</i> (1984) <sup>(23)</sup>	1	M	26	Maxilla-bilateral	Radiolucent area around crowns impacted third molars
Warnock <i>et al.</i> (1985) <sup>(24)</sup>	1	M	19	Mandible-anterior	Well-defined radiolucent area
Monteil, Teresti (1985) <sup>(25)</sup>	1	F	51	Mandible-anterior	Well-defined radiolucent area
Mills <i>et al.</i> (1986) <sup>(26)</sup>	1	M	26	Maxilla and mandible	Poorly defined radiolucent area
Tatemoto <i>et al.</i> (1989) <sup>(27)</sup>	2	M	41/56	Mandible-anterior Maxilla-anterior	Poorly defined radiolucency
Leider <i>et al.</i> (1989) <sup>(28)</sup>	2	M	29/25	Multicentric	Triangular radiolucency; bone loss
Reichart, Philipsen (1990) <sup>(29)</sup>	1	M	56	Mandible-anterior	Multilocular radiolucency
Yaacob (1990) <sup>(30)</sup>	1	M	39	Maxilla-anterior	Well-defined radiolucent area
Schwartz-Arad <i>et al.</i> (1990) <sup>(10)</sup>	1	M	8	Mandible-anterior	Radiolucent area between canine and first molar
Baden <i>et al.</i> (1993) <sup>(7)</sup>	3	M	46/39/74	Maxilla- posterior Mandible-posterior Extra-osseous	Multilocular radiolucency in edentulous area
Saxby <i>et al.</i> (1993) <sup>(31)</sup>	2	M	59	Mandible-anterior	Radiolucent area; bone loss
Kusama <i>et al.</i> (1998) <sup>(32)</sup>	1	F	42	Maxilla-posterior	Radiolucency associated with impacted tooth
Haghighat <i>et al.</i> (2002) <sup>(8)</sup>	1	M	43	Maxilla-anterior	Triangular radiolucency
Barrios <i>et al.</i> (2004) <sup>(11)</sup>	1	M	11	Maxilla-anterior	Triangular radiolucency
Ruhim <i>et al.</i> (2007) <sup>(13)</sup>	1	M	9	Maxilla-anterior	Radiolucent area, unilocular, well-defined
Kim <i>et al.</i> (2007) <sup>(12)</sup>	2	F	15/27	Maxilla-anterior	Triangular radiolucency
Siar <i>et al.</i> (2010) <sup>(3)</sup>	1	F	10	Mandible-anterior	Not stated
Jones <i>et al.</i> (2011) <sup>(6)</sup>	1	M	24	Maxilla-anterior	Radiolucent area, triangular, well-defined
Badni <i>et al.</i> (2012) <sup>(5)</sup>	1	M	58	Mandible-anterior	Triangular radiolucency; bone loss
Malathi <i>et al.</i> (2012) <sup>(33)</sup>	1	F	60	Mandible-posterior	Radiolucent area; bone loss
Bansal <i>et al.</i> (2013) <sup>(2)</sup>	1	F	26	Mandible-anterior	Radiolucent area; bone loss

SOT: squamous odontogenic tumor; M: male; F: female; anterior: anterior to the distal surface of the canine tooth; posterior: posterior to the distal surface of the canine tooth; multicentric: multiple sites at the same anatomical location.

Most cases of SOT arise and develop in the periodontium of the permanent dentition. Pullon *et al.*<sup>(1)</sup> reported a case associated with the deciduous dentition and cases involving edentulous areas<sup>(1, 7-10)</sup>. The present case is the 48<sup>th</sup> case of SOT reported in the English literature. The location of SOT in an edentulous region is uncommon, since the tumor is more frequent in areas with teeth. In addition, involvement of the mandibular anterior region and the multilocular radiographic appearance are also uncommon findings<sup>(2)</sup>.

Although not pathognomonic, most SOTs are detected during routine radiographic examination as a radiolucent, unilocular, triangular-shaped image associated with the roots of adjacent teeth. The tumor may be misdiagnosed from the time when the lesion is referred to as a severe periodontal defect. Some rare cases exhibit a multilocular radiolucency, circumscription of the tumor with a cortical border or, in more aggressive cases, lack of definition of tumor margins. In addition to advanced periodontal bone loss, Langerhans cell disease, lateral periodontal cyst, keratocystic odontogenic tumor, central odontogenic fibroma and other odontogenic tumors should be included in the radiographic differential diagnosis<sup>(2-6, 11-13)</sup>. Although computed tomography permits to delimit the tumor, the present case exhibited an uncommon radiographic feature, i.e., the presence of a multilocular radiolucent image.

Microscopically, SOT is characterized by multiple proliferations of squamous epithelium surrounded by a moderately cellular fibrous-collagenous connective tissue stroma. The epithelial islands, which can be smoothly contoured and demarcated from

the stroma and vary in size and shape, occasionally exhibit foci of cystic degeneration and central calcification. The harmless appearance of the pavement epithelium characterizes the lesion, since ameloblastic nuclear polarization and a palisade-like arrangement of peripheral columnar cells are absent in SOTs. Furthermore, the squamous odontogenic tumor-like proliferations seen in the walls of inflammatory odontogenic cysts are not considered a manifestation of SOT<sup>(2-6, 11-13)</sup>. Therefore, the histopathological differential diagnoses for SOT include the acanthomatous and desmoplastic ameloblastoma and gingival squamous cell carcinoma; however, SOT shows no microscopic evidence of malignancy<sup>(2, 3, 5)</sup>.

Treatment of SOT consists of conservative surgical removal (local excision), enucleation and/or complete curettage. However, more aggressive intervention may be necessary in the case of tumors located in the maxilla due to the aggressive potential of the lesion at this site. Recurrence is rarely reported in the literature and is attributed to incomplete removal of the initial tumor. In these cases, the extraction of adjacent teeth involved in the tumor is indicated<sup>(2, 4-6)</sup>. Only one case of recurrence was observed among the six cases evaluated by Pullon *et al.*<sup>(1)</sup>.

The present results demonstrate the need for a comprehensive diagnostic evaluation considering all clinical, radiographic and especially histopathologic features in order to guarantee the success of treatment indicated in each case, promoting quality of life for patients with SOT. In addition, the present case highlights the importance of individual analysis of each case.

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## RESUMO

*O tumor odontogênico escamoso (TOE) é definido como uma neoplasia benigna rara, localmente infiltrativa, podendo estender-se a estruturas vizinhas. Objetivou-se relatar um caso de TOE de acometimento incomum. Paciente do sexo feminino, 56 anos de idade, fumante, demonstrou leve aumento de volume na região do mento na porção compreendida entre o canino esquerdo e o primeiro pré-molar direito. Imagens axial, coronal e sagital demonstraram lesão expansiva hipodensa na sínfise e na linha média mandibular. Fez-se a remoção parcial da lesão, o que levou ao diagnóstico de TOE. A paciente não apresentou recidiva durante quatro anos e dois meses de acompanhamento.*

*Unitermos: tumor odontogênico escamoso; mandíbula; neoplasias.*

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