Recurrent squamous odontogenic tumor: A case report and review of the literature

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Received September 9, 2014; Accepted June 11, 2015

DOI: 10.3892/ol.2015.3632

Abstract. Squamous odontogenic tumors (SOTs) are benign, locally infiltrative neoplasms that localize to the periodontium. In total, <50 cases have been reported since the first description of SOTs in 1975. Although the exact etiology of SOTs is unknown, the tumors are considered to derive from the epithelial cell rests of Malassez. SOTs are characterized by radiological and clinical signs and symptoms, including pain with increased sensitivity in the affected area, bone expansion and increased tooth mobility. The present study describes the case of a patient that experienced numerous SOT recurrences and also discusses recommendations for treatment. A locally invasive mandibular SOT was identified in a Caucasian 41-year-old female patient. The treatment involved recommended conservative surgery, including local curettage. In addition, 49 cases published in the literature were reviewed to assess the treatment strategies. The present patient experienced two recurrences of the tumor during the 6-year follow-up period. Ultimately, the vitality of the adjacent teeth was compromised. An apicoectomy with a small amount of resection of the marginal bone was necessary. In >50% of the reported cases of SOT in the literature the adjacent teeth were extracted. The present case of SOT and the associated literature were also discussed. It was concluded that the treatment of choice appears to be a conservative surgical removal, but the successful management of SOTs often requires the removal of the adjacent teeth.

Introduction

The interest in odontogenic tumors has considerably grown since the first edition of the World Health Organization (WHO) tumor classification published in 1971 (1). A novel and completely revised WHO classification from 2005, which encompasses the histopathological and genetic criteria of SOTs, describes a group of epithelial odontogenic tumors that comprises the ameloblastoma family, consisting of solid/multicystic, extraosseous/peripheral, desmoplastic and unicystic ameloblastoma, and squamous, adenomatoid, calcifying and keratocystic odontogenic tumors (2).

Squamous odontogenic tumors (SOTs) are rare benign tumors of the periodontium that possess an unknown etiology and were first described in 1975 (3,4). In the literature, >50 cases have been reported (5). SOTs are slow-growing, locally infiltrating tumors with only a few clinical signs and symptoms. Indicators for the underlying tumor include mobility of the teeth, increased periodontal pocket depth, sensitivity, swelling and erythema of adjacent gingiva, swelling of the alveolar process, and moderate pain (5-7). SOTs have also been reported to occur in various age groups, yet mainly affect adults in the third decade of life (8). The male to female gender ratio is 1.4:1 (5). The mandible is affected more often than the maxilla, with a preferential occurrence in the posterior premolar and molar area. Maxillary SOTs are described to be primarily present in the anterior area, and appear to be more aggressive compared with SOTs in the mandibular area (9,10). Multifocal lesions have been reported to be more frequent in these regions compared with other odontogenic tumors (11).

SOTs are hypothesized to derive from the epithelial cell rests of Malassez. These tumor entities usually appear on the lateral root surface. The typical radiographic presentation is a triangular radiolucent defect involving the lateral root surface of erupted and vital teeth (12). The wide base of the radiolucency is localized between the diverging apices of the adjacent roots (13). The most common variant of SOTs is an intraosseous or central type. However, a rare peripheral variant has also been described (14-17).

SOTs consist of islands of well-differentiated squamous epithelial cells of varying sizes and shapes, surrounded by mature connective tissue (9). An epithelial hamartomatous proliferation may also be suspected. As a result, this lesion is often described as a benign epithelial odontogenic tumor, acanthomatous ameloblastoma, acanthomatous ameloblastic fibroma or occasionally, well-differentiated squamosus cell

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Key words: tumor of the periodontium, squamous odontogenic tumor, recurrence, treatment recommendations

carcinoma or pseudoephiteliomatous hyperplasia (7). A histopathological misinterpretation may therefore lead to either therapeutic over- or under-treatment. The prognosis of SOT therapy is good. Recurrence appears to be rare, and may occur due to incomplete tumor removal.

In the present study, the clinical, radiographic and histological characteristics of the squamous odontogenic tumor with locally invasive growth and two recurrences were reported for six years subsequent to the primary surgical removal. Subsequently, the known literature on treatment recommendations for SOT was critically reviewed.

Case report

A 41-year-old woman presented to a general dentist (Frechen, North Rhine-Westphalia, Germany) with unusual sensitivity and slight pressure on the left premolar side of the mandible in December 2006. The first panoramic radiograph did not reveal any changes in the bone. However, the patient repeatedly presented with the same complaint. Subsequent examination with a small periapical radiograph revealed a radiolucent lesion located between the roots of the canine and the first premolar in the left mandible. The patient was then referred from a general dentist to the Department of Oral Surgery at University Hospital Bonn (Bonn, Germany) for additional evaluation in January 2006.

Intraoral examination revealed small hard tissue swelling, termed osseous expansion, on the lingual aspect of teeth 21 and 22 (Fig. 1A). No hypoesthesia, hyperesthesia or anesthesia of the left inferior alveolar nerve was present. No signs of tenderness, purulence or hemorrhage were observed, and the patient did not suffer from any pain. The teeth were evaluated for vitality using refrigerant spray, consisting of a butane, propane and isobutane mixture (Kältespray PluLine 200 ml Ds; Pluradent, Offenbach, Germany) and were deemed vital. The previous medical and surgical histories of the patient were completely negative; no tumors had been identified previously and the patient had not undergone any surgical procedures. Furthermore, the adjacent teeth were healthy. A panoramic and periapical radiograph revealed a triangular radiolucent lesion between the lower left canine and the first premolar, with the base of the radiolucency localized between the diverging apices of the adjacent roots (Fig. 1B). Based on these findings, the clinical diagnosis for the present patient was an odontogenic cyst, with the differential diagnosis being an epidermoid cyst or a keratocyst.

At the time of the procedure, visible soft swelling from the buccal side of teeth 21 and 22 was observed, and therefore, a vestibular approach to the lesion was used. The aim of the surgical procedure was to completely remove the lesion and perform curettage of the bony margins under local anesthesia. Shortly following the creation of marginal incisions and reflection of a mucoperiosteal flap, a vestibular bony wall perforation was identified, with the lesion visible underneath (Fig. 1C). The lesion was firm. Complete enucleation of the lesion was performed while preserving the lingual bony wall of the mandible. The root apices of the adjacent teeth extending to the lesion cavity were irrigated with sterile saline. The post-operative cavity was filled with a collagen sponge (TissuFleece E; Resorba GmbH, Nuremberg, Germany) and Penicillin G in powder form (Hydracillin; GlaxoSmithKline Biologicals, Dresden, Germany) to prevent additional collapse of the buccal area. The patient also received Phenoxymethylpenicillin at a dose of 1.5 million international units three times a day orally for one week (Penicillin V 1.5 Mega; Heumann Pharma GmbH, Nuremberg, Germany).

Table I. Post-operative intervals between follow-ups in the present patient and the corresponding clinical procedures and treatments for recurrence of SOT.

Post-operative interval, months	Procedure or examination	Diagnosis or treatment
0	1st surgery	SOT removal
6	Routine examination	NAD
12	Routine examination	NAD
20	2nd surgery	Removal of SOT recurrence
26	Routine examination	NAD
32	Routine examination	NAD
38	Routine examination	NAD
44	Routine examination	NAD
50	CBCT, 3rd surgery	No pathological abnormality
56	Routine examination	NAD
62	4th surgery	Removal of SOT recurrence
65	Apicoectomy	Pulpitis treatment
72	Routine examination	NAD

NAD, no abnormality detected.

Histopathological examination was performed on the specimen, and the presence of fragments of a cystic lesion with a multilayered and variable well-differentiated epithelium was revealed. These demonstrated a tendency for flat squamous/epithelial differentiation. In wide regions the peripheral layer exhibited a cylindroepithelial configuration with adenoid segments. Smaller sections were two or three-layered, with evidence of calcified material (Figs. 2 and 3). The diagnosis of SOT was confirmed by the Bone Tumor Reference Center at the Institute of Pathology, University Hospital Basel (Basel, Switzerland).

Follow-up was performed every 6 months up to 72 months (Table I) and consisted of oral examinations and periapical radiographs. The X-rays revealed gradual bone recovery. Teeth 21 and 22 each remained vital, with light disturbance of sensitivity in the innervations of the left inferior alveolar nerve for ~3 weeks. The patient exhibited no symptoms, with the exception of a small painless osseous expansion on the lingual aspect of teeth 21 and 22.

However, 1 year and 8 months subsequent to primary surgical treatment, routine X-ray examination revealed tumor recurrence (Fig. 4). At approximately the same time, the patient started to complain of pressure in the surgically resected region.

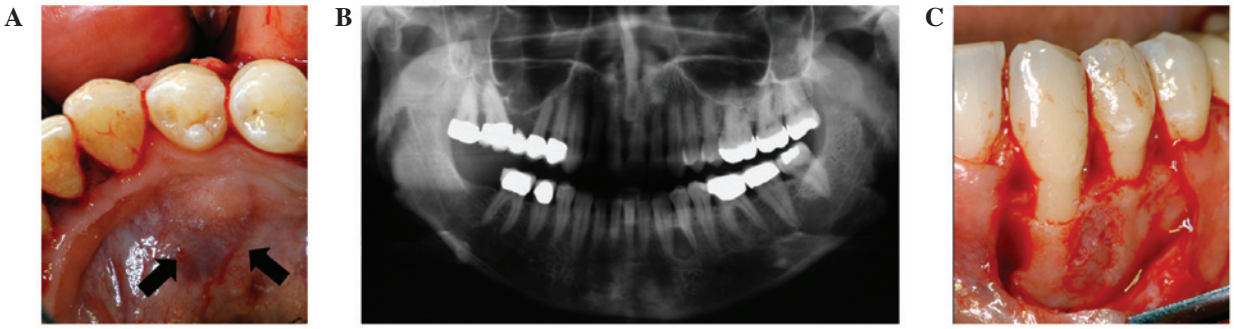

Figure 1. (A) Lingual osseous expansion in the lingual aspect of teeth 21 and 22. (B) Panoramic radiograph revealing the triangular radiolucent lesion between the roots of teeth 21 and 22, with the base of the radiolucency localized between the diverging apices of the adjacent roots. (C) Visible perforation of the vestibular bony wall subsequent to reflecting the flap.

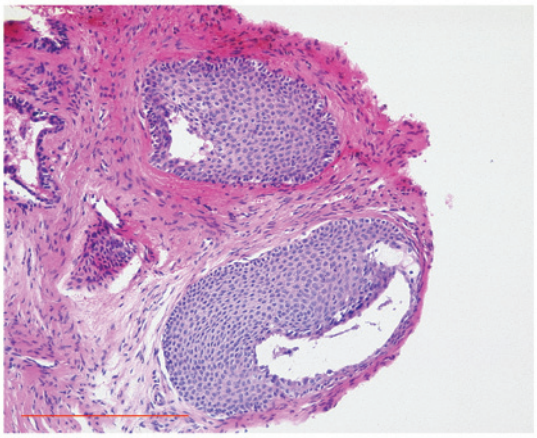

Figure 2. First image of the histopathological examination of the squamous odontogenic tumor. The examination revealed highly differentiated odontogenic epithelial cells forming oval islands, with a peripheral layer of low cuboidal epithelial cells and signs of microcystic degeneration (stain, hematoxylin and eosin; magnification, x200; scale bar, 200 μm).

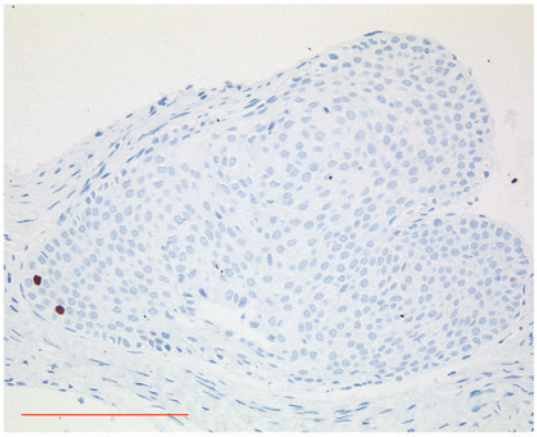

Figure 3. Second image of the histopathological examination, revealing low level expression of Ki67 in a small number of tumor cells, indicating minor mitotic activity (stain, Ki67; magnification, x400; scale bar, 100 μm).

A second surgical procedure revealed that the recurrent tumor was smaller, located only intraradicularly, and that the lesion did not extend to the root apices.

The procedure was performed under local anesthesia. A vestibular approach to the lesion was chosen. Two specimens were excised by curettage, consisting of the recurrent tumor (Fig. 5A) and a small cementoma-like tissue (Fig. 5B). The cavity was subsequently filled using a TissuFleece E collagen sponge. The vestibular defect was then covered with a BioGide membrane (Geistlich Pharma AG, Wolhusen, Switzerland) to prevent additional collapse of the buccal region.

The histological features observed were similar to those found in the sample, revealing segments of sclerotic stroma and segments of cystic structure, the previous composed of multilayered epithelium, with signs of squamous/flat-epithelial differentiation, or in other segments, cylindroepithelial differentiation. Numerous sections were formed by cuboidal epithelial cells. The second specimen revealed a small cementoma.

The patient underwent regular follow-ups every 6 months, with no clinical symptoms and no complaint about any pressure or sensitivity disturbances in the affected area (Table I) (4,6,8,10,12-49). The two teeth 21 and 22 remained vital. A periapical radiograph revealed radiolucent changes in the surgical region 2 years and 6 months subsequent to the first recurrence. A cone beam computed tomography (CBCT) was performed shortly thereafter (Fig. 6).

According to the precise CBCT assessment, the vestibular bone between teeth 21 and 22 had been completely rebuilt. A round shaped radiolucent area ~3 mm at the level of the middle third of tooth 33. Lingual bone expansion caused by the tumor was visible.

The surgical procedure was planned with an approach from the lingual aspect being considered. The aim was for a radical procedure that retained the vital teeth. The lingual osteotomy of teeth 21 and 22 revealed an empty cavity void of fluids or tissues. No pathological specimens were excised. Any superficial irregularities of the bone were subsequently removed. The surgical region was sutured.

The follow-up 6 months later revealed no clinical or radiological abnormalities. However, 12 months subsequent to the third procedure, the patient complained of pressure in the surgical region again. The teeth remained vital.

A novel lesion ~7x7 mm in size, identified on an X-ray performed at the general dentist, was removed from the vestibular side, followed by curettage of the bone according to recommendation from the Bone Tumor Reference Center at the Institute of Pathology, University Hospital Basel. The

Table II. Summary of reported cases of SOT ('Solid form').

First author, year (Ref.)	Age, years	Gender	Site	Therapy		Follow up, months	Recurrence
				Tumor	Teeth		
Pullon *et al*, 1975 (4)	23	F	Multicentric maxilla/mandible	Excision	Removal	5	Numerous
				Excision	Removal of all remaining teeth	7	None
	11	M	Maxilla	Curettage	Removal	60	None
	19	M	Mandible	Curettage	(No contact)	144	None
	31	F	Maxilla	Excision	Removal	12	None
	42	F	Maxilla	Excision	Not stated	60	None
	29	M	Mandible	Excision	Removal	216	None
Doyle *et al*, 1977 (23)	26	M	Maxilla	Enbloc resection	Removal	0	-
	65	M	Maxilla	Partial maxillectomy	Removal	7	None
[a]Doyle *et al*, 1977 (23)							
Mc Neill *et al*, 1980 (20)	26	F	Multicentric maxilla/mandible	Removal of lesions	Removal	12	None
[a]Van der Waal *et al*, 1980 (21)							
Hopper *et al*, 1980 (29)	22	F	Multicentric maxilla	Modified hemimaxillectomy	Removal	3	None
			Mandible	Excision	Removal	3	None
Carr *et al*, 1981 (36)	66	F	Maxilla	Curettage	Removal	0	-
Leventon *et al*, 1981 (24)	59	M	Mandible	Extirpation	Not stated		-
Goldblatt *et al*, 1982 (28)	60	F	Mandible	Curettage	Removal	0	-
	29	M	Mandible	Enucleation	Not stated	0	-
	30	F	Maxilla	Curettage	Removal	0	-
	26	F	Mandible	Curettage	Not stated	0	-
	67	F	Not stated	Excision	Not stated	0	-
[b]Anneroth & Hansen, 1982 (18)							
Cataldo *et al*, 1983 (34)	24	F	Mandible	Curettage	Root-planing	8	None
[b]Swan and McDaniel, 1983 (l22)							
Norris *et al*, 1984 (37)	26	M	Maxilla bilateral	Excision	Removal	0	-
Kristensen *et al*, 1985 (32)	61	M	Multicentric maxilla	Partial maxillectomy	(tame)	84	None

Table II. Continued.

First author, year (Ref.)	Age, years	Gender	Site	Therapy		Follow up, months	Recurrence
				Tumor	Teeth		
Monteil et al, 1985 (38)	51	F	Mandible	Excision	Removal, canal treatment, apical resection	9	None
Warnrock *et al*, 1985 (39)	19	M	Mandible	Curettage	Not stated	0	-
[a]Hietanen *et al*, 1985 (14)							
Mills *et al*, 1986 (30)	26	M	Multicentric maxilla	Excision	Removal	4	None
			Mandible	En-bloc resection	Removal	24	None
Tatemoto *et al*, 1989 (40)	41	M	Mandible	Excision	Removal	0	-
	56	M	Maxilla	Partial resection	Removal	0	-
Leider *et al*, 1989 (19)	29	M	Multicentric maxilla	Curettage	Removal Root planing	-	-
	25	M	Multicentric maxilla	Curettage	Removal	48	None
[b]Leider *et al*, 1989 (19)							
Yaacob, 1990 (41)	39	M	Maxilla	Excision	Not stated	48	None
Reichart and Philipsen, 1990 (42)	56	F	Mandible	En-bloc resection	Removal	0	-
Schwarz-Arad *et al*, 1990 (43)	8	M	Mandible	Excision	Removal	30	None
Baden *et al*, 1993 (15)	46	M	Maxilla	Excision	Not stated	84	None
	39	M	Mandible	En-bloc resection	Edentulous area	12	None
[a]Baden *et al*, 1993 (15)							
Saxby *et al*, 1993 (6)	59	M	Mandible	Curettage	Not stated	12	None
Favia *et al*, 1997 (27)	56	Not stated	Maxilla	Enucleation	Not stated	60	None
	25	F	Mandible	Enucleation	Not stated	72	None
Kusama *et al*, 1998 (44)	42	F	Maxilla	Excision	Removal	20	None
[a]Ide *et al*, 1999 (25)							
Haghighat *et al*, 2002 (13)	43	M	Maxilla	Enucleation	Root planing	18	None
Barrios *et al*, 2004 (45)	11	M	Maxilla	Excision	Curettage	28	None
Cillo *et al*, 2005 (12)	45	F	Mandible	Exision	Removal	4	None
[a]Ide *et al*, 2005 (16)							
Ruhin *et al*, 2007 (10)	9	M	Maxilla	Curettage	Removal	12	Yes
				Radical surgery		84	None

Table II. Continued.

First author, year (Ref.)	Age, years	Gender	Site	Therapy		Follow up, months	Recurrence
				Tumor	Teeth		
Kim *et al*, 2007 (8)	15	F	Mandible	Curettage	Hemisection	12	None
	27	F	Mandible	Extirpation	Curettage of root tip	6	None
Oliveira *et al*, 2007 (33)	28	F	Mandible	Enucleation		11	Yes
				En-bloc resection		132	None
Siar *et al*, 2010 (35)	10	F	Mandible	Excision	Not stated	60	None
Agostini *et al*, 2011 (46)	64	M	Mandible	Radical excision	Removal	84	None
Jones *et al*, 2011 (47)	24	M	Maxilla	Exision	Removal	0	None
[a]Malathi *et al*, 2012 (17)							
[a]Tarsitano *et al*, 2012 (26)							
Badni *et al*, 2012 (31)	58	M	Mandible	Not stated	Not stated	0	-
Bansal and Joshi, 2013 (48)	26	F	Mandible	Excision	Curettage	12	None
Pant and Pathak, 2013 (49)	19	F	Mandible	Enucleation	Not stated	0	-

[a]Case not included in present study (see text). [b]Case previously reported (see text). M, male; F, female; CBCT, cone beam computed tomography.

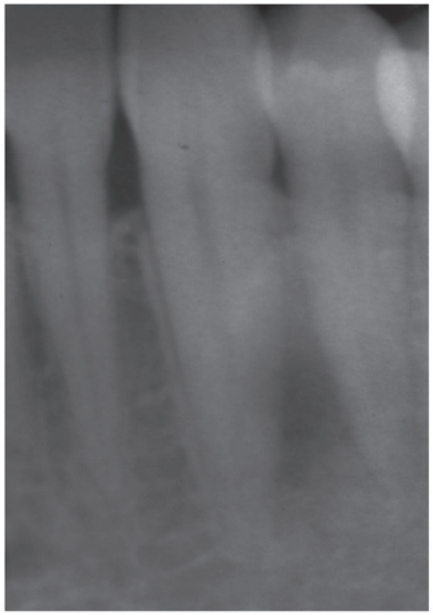

Figure 4. Periapical radiograph performed subsequent to the first recurrence revealing an interradicular osteolysis.

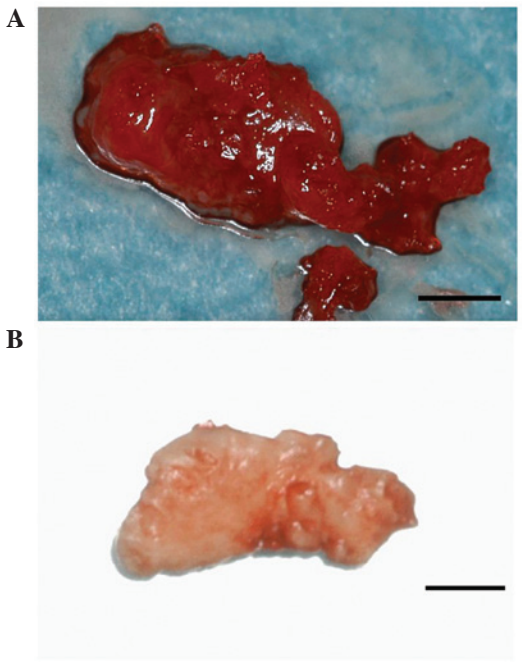

Figure 5. Two different tissues from the same lesion. (A) Squamous odontogenic tumor tissue and (B) cementoma-like sample (scale bars, 2 mm).

patient began to exhibit disturbances and pulpitis-like symptoms 12 weeks subsequent to the fourth procedure that affected teeth 21 and 22. An apicoectomy with minimal resection of the marginal bone was then performed. The histological examination did not reveal any indication of recurrence of SOT. No squamous epithelium was observed. The sample consisted mainly of collagen connective tissue in addition to bony fragments. The healing of the wound was without complication, and the follow-up was performed every 6 months until May 2013, and revealed no pathological findings.

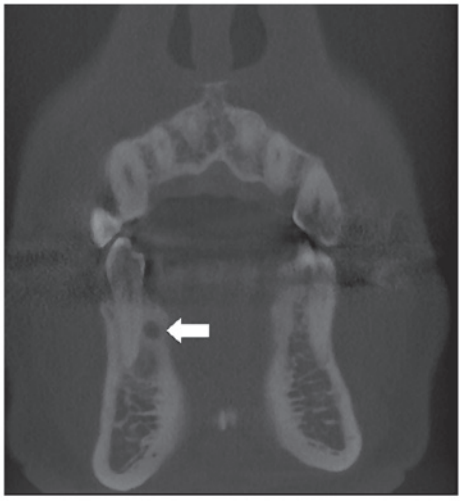

Figure 6. Cone beam computed tomography examination. The arrow indicates a lingual osteolytic space.

Discussion

Review of the literature for the solid-form of the SOT was performed using PubMed (National Center for Biotechnology Information, US National Library of Medicine, Bethesda, MD, USA) with the search combination 'squamous odontogenic tumor and recurrence'. The search was limited to the English literature. In total, 49 cases of this tumor were reported in English language journals (Table II), 7 of which were excluded for the following reasons: Anneroth and Hansen (18), previously reported by Pullon *et al* (4); Leider *et al* (19), previously reported by McNeill *et al* (20); Van der Waal *et al* (21), lesion diagnosed as a 'possible SOT'; Swan and McDaniel (22), the diagnosis was questionable; Doyle *et al* (23), reported SOT-like proliferations in odontogenic cysts; Leventon *et al* (24), reported SOT-like proliferations in odontogenic cysts; Ide *et al* (25), reported an intraosseous cell carcinoma arising in association with a SOT; and Tarsitano *et al* (26), reported a multifocal epithelial odontogenic tumor associated with a SOT.

In the literature, 4 studies reported examples of peripheral SOT (14-17). The age at the time of initial diagnosis ranged between 8 and 67 years, with a mean age of 36.3 years. The majority of SOTs become visible in patients aged 20-29, with 17 patients (35%) in this age group.

The gender ratio from the 49 patients reported in the literature for whom this information was provided, including the present patient, is 1:1.2 (female:male). The study by Favia included no gender information (27). SOT occurred individuals of Caucasian, African and Asian descent, and predominance in a specific ethnicity was not observed.

The mandible was involved in 57.1% of all cases and the maxilla in only 38.8%. In total, 8 patients exhibited multicentric lesions of the tumor, 4 of which involved maxillary and mandibular sites and 4 of which involved only a maxillary site. In 1 study the site was not stated (28).

At present, the treatment of 50 patients with solid SOTs has been described, including the present patient. The varying clinical behavior of the sites of multicentric lesions has also been reported (29,30). In 1 study, information on the treatment

administered was not included (31). In summary, 43 lesions were treated with conservative surgery, consisting of curettage, excision and enucleation, and 8 lesions were treated with radical surgery, consisting of en-bloc resection, modified hemimaxillectomy and partial maxilloectomy (Table II).

In total, 3 studies reported lesions detected in edentulous areas (4,15,32). In all previous studies in the literature, the tumor was in contact with the surface of a tooth or teeth. In >50% of cases, the adjacent teeth were extracted (Table II). In 1 case, a hemisection of the remaining tooth was performed (8).

Substantial follow-up data was not available in the literature. Follow-up times were reported in 33 studies for 49 patients, and ranged between a few weeks and a maximum of 216 months (Table II).

In total, 4 cases of recurrence have been reported in the literature (4,10,28,33). Recurrence developed over a short period (7-12 months) in 2 cases subsequent to simple conservative treatment, without the removal of teeth (10,33). Recurrence developed subsequent to surgery with incomplete removal of the adjacent teeth in 1 case (4). A second surgical procedure was performed in all cases, which included the removal of all remaining teeth. No additional recurrence was reported in any of these cases. Goldblatt *et al* (28) also reported the development of a recurrent lesion, with well-demarcated triangular radiolucency of the molar roots. Only a simple curettage was performed for this patient and no follow-up time was provided.

According to the present literature review, the incidence of SOT is low. The female to male ratio (ratio, 1:1.2) in the present study was higher compared to the ratio reported in the study by Reichart (ratio, 1:1.4) (5). The tumor usually grows slowly and often demonstrates a lack of symptoms for a long time. The clinical and radiographic features of SOT are neither unique nor sufficient for diagnosis and this type of tumor may be confused with a number of other pathologies (33). Therefore, distinctive clinical, radiological and histological aspects are necessary for avoiding a misdiagnosis that may result in serious negative implications for the patient (25).

SOT may occur at any age, with predominance in the third decade. The youngest patient reported in the literature was a 9-year-old boy with maxillary SOT that was treated with local surgical tumorectomy. However, 10 months subsequent to the procedure, an extremely aggressive recurrence had to be treated by radical surgery (10). The maxilla appears to be involved more often in the region of the incisors, whereas the premolar and molar areas appear to be more involved in the mandible.

Even though SOT is a benign lesion, it should be considered as semi-malignant in certain cases, particularly in the maxilla, where SOTs demonstrate increased aggressiveness (10). The 41-year old female patient in the present study was first treated with enucleation of the tumor and surgical curettage while maintaining the involved vital teeth. Follow-up performed every 6 months revealed early stage small recurrences that could be immediately surgically treated. SOT is a slow growing tumor. The treatment recommendation of the WHO is conservative surgical intervention (5). However, the present study also revealed that a conservative approach, such as enucleation and simple curettage with the intent of preserving the vitality of involved teeth, may not be sufficient to prevent recurrence, but a more aggressive treatment is required. The patient returned for a regular follow-up 2 years and 6 months subsequent to the second surgical treatment, and did not exhibit any clinical symptoms, although CBCT revealed an abnormality lingual of tooth 22 (Fig. 5). However, no second recurrence was detected intraoperatively. One year later, a third lesion was removed followed by apicoectomy. Additional recurrence may lead to the decision of a radical surgical procedure with loss of the adjacent teeth. Compromised surgical therapy was performed due to the desire of the patient to preserve the teeth (Table I). The data from the literature has also indicated that curettage and extraction of the adjacent teeth has acted as an adequate therapy. Pullon *et al* (4) also described numerous recurrences following the completion of simple curettage with incomplete removal of the adjacent teeth. Goldblatt *et al* (28) reported a recurrent lesion with well-demarcated triangular radiolucency of the roots of the first and second molars. Although this previous study concluded that the excision and extraction of the involved teeth is an adequate treatment in the majority of cases, a simple curettage was performed. Follow-up data was not provided. Ruhin *et al* (10) and de Oliveira *et al* (33) described the development of recurrence following simple curettage. Subsequent to the removal of the remaining teeth, the patients were tumor-free during a long follow-up period. However, there are two cases in the literature where the post-operative radiograph revealed similar appearances, of triangular-shaped radiolucency, between the remaining teeth compared with the pre-operative film (6,34). Therefore, these studies indicate that tumor recurrence cannot be excluded based on radiograph results alone.

It may be of clinical relevance to establish individual treatment plans to adequately respond to the biological behavior of these rare tumors.

SOT presents as a locally infiltrative neoplasm and has been known to infiltrate into adjacent tissues, with resorption of the alveolar bone and invasion of the overlying gingival and oral mucosa (2). In 2007, Kim *et al* (8) initially described a lesion associated with the erosion of the lingual cortical plate in the mandible in two cases of female patients aged 15 and 27 years. Until then, the perforation of the cortical bone has only been presented in the maxilla, where the bone is much thinner compared with the mandible and less resistant to erosion. In the present study, the case of a patient with buccal bone erosion of the mandible that was uncovered intraoperatively is reported. The lingual bone possessed a complete cortical layer, but with visible bone expansion.

The etiology of SOT has yet to be elucidated. However, immunohistochemical evaluation performed in a previous study revealed positive reactivity of varying intensity in the neoplastic epithelial cells for the Notch1, Notch2 and Notch3 transmembrane receptors and their ligands. These findings suggest that these receptors play a role in the cytodifferentiation of SOTs (35).

Although SOTs are considered to be benign neoplasms, the behavior and local expansiveness of the tumor indicates the possibility of carcinomatous transformation. In 1999, Ide *et al* (25) first reported a rare occurrence of intraosseous squamous cell carcinoma arising in association with squamous odontogenic tumors. The enucleated specimen of the tumor, which was attached to an impacted third molar of the mandible, revealed a characteristic pattern of SOT. However,

within 2 months, aggressive bone destruction exhibiting the typical findings of intraosseous squamous carcinoma was identified. It is questionable whether in the aforementioned case the squamous carcinoma arose *de novo* or presented a misleadingly benign appearance. However, the extensive assessment of specimens provided support to the hypothesis of the malignant alteration of SOT. The present study revealed that an odontogenic tumor, which is difficult to access, is prone to the development of recurrence (Table I). This may be due to the inability of the surgical treatment to fully remove all tumor cells from the tooth.

In summary, the potential for recurrence developing in even benign SOTs may depend on the accessibility of the SOTs for surgical treatment and the biological behavior of the SOTs. The present study recommends an individualized treatment plan in order to respond to the biological reaction of the SOT rather than to the histopathology of the tumor. The present knowledge of treatment is based on a total of 49 cases. Treatment of the solid form of SOT associated with the roots of the teeth by local curettage with removal of the adjacent teeth appears to be effective for the prevention of recurrence.

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