Squamous Odontogenic Tumor: Literature Review Focusing on the Radiographic Features and Differential Diagnosis

Nilson do Rosário Mardones¹, Thiago de Oliveira Gamba², Isadora Luana Flores²,* Solange Maria de Almeida² and Sérgio Lúcio Pereira de Castro Lopes³

¹São Leopoldo Mandic Dental School – Brazil; ²Piracicaba Dental School, State University of Campinas – UNICAMP, Brazil; ³São José dos Campos Dental School, State University of São Paulo – UNESP, Brazil

Abstract: Since its first publication in 1975, the squamous odontogenic tumor remains the rarest odontogenic lesion, with around 50 cases in the English-language literature in which the microscopic characteristics are frequently very well demonstrated. However, articles which discuss the radiographic aspects are scarce, especially with emphasis on the differential diagnosis. The present treatise proposes an assessment of jaw lesions with the same radiographic characteristics of the squamous odontogenic tumor to clarify the main findings for dental clinicians during routine diagnosis.

Keywords: Differential diagnosis, non-odontogenic lesions, odontogenic lesions, radiographic aspects, squamous odontogenic tumor.

INTRODUCTION

Squamous odontogenic tumor (SOT) is a benign odontogenic tumor classified according to the World Health Organization (WHO) in 2005 as an epithelium odontogenic tumor with around 50 cases reported in the English-language literature at this time [1-3]. This rare entity was described for the first time in 1975 by Pullon et al. [4]; before this it was considered as an acanthomatous ameloblastoma or a squamous cell carcinoma. The pathogenesis of SOT is still unclear in which remnants of dental lamina (rests of Serres), epithelial rests of Malassez or gingival epithelium are the main suspected origin [2, 4]. The SOT presents well-defined histopathological aspects and previous studies discussed these aspects [3, 5] in which islands of squamous epithelium in a dense fibrous connective tissue stroma are the classical microscopic findings. Nevertheless, there are scarce articles that described radiographic features of SOT [6], and only one author included SOT in a list of possible diagnosis before the histopathological examination [7]. Therefore, we propose to discuss these aspects based on the clinical relevance of differential diagnosis with other lesions more frequently found in routine jaw radiographies.

LITERATURE REVIEW

Clinically, SOT can be presented as an asymptomatic, slow growing, intrabony lesion with few clinical signs and symptoms. Nevertheless, mobility and displacement of teeth, swelling of alveolar process, and mild to moderate pain are the main findings [2, 6, 8]. SOT occurs on average in the fourth decade of life with a slight predilection for males [2]. An equal distribution between maxilla and mandible with preference for posterior mandible and anterior maxilla is observed [2, 9]. Commonly, it is a central lesion with few cases occurring as peripheral lesions [3]. The most typical presentation of SOT detected in routine intraoral radiographs is an unilocular radiolucent defect with triangular or semicircular shape between or along the roots of adjacent vital teeth [3, 6-9]. Fig. (1) showed an interproximal lesion with these radiographic aspects. In these cases, a careful evaluation of all lesions found in the periodontal region should be performed, especially when a interproximal bone loss involves only one isolated area [7, 8]. This affirmation can be confirmed due to slow growing of SOT within a periodontal location, mimicking severe periodontal bone loss in a significant number of previous cases described in the English-language literature [2, 4, 6, 7, 9-20].

SOT can also present radiographic aspects that resemble odontogenic and non odontogenic lesions as cysts and tumors with emphasis on extensive lesions with unilocular or multilocular appearance involving the mandible and/or maxilla, pushing the maxillary sinus or in association with an impacted tooth [21]. A broad list of possible diagnoses include developmental or noninflammatory odontogenic cysts, such as lateral periodontal cyst, dentigerous cyst and glandular odontogenic cyst; inflammatory odontogenic cysts, such as radicular and residual cyst; odontogenic tumors, such as keratocystic odontogenic tumor, adenomatoid odontogenic tumor, central odontogenic fibroma, unicystic and multicystic ameloblastoma; hematological disorders, such as Langerhan’s cell histiocytosis and multiple myeloma and bone pathology, such as central giant cell lesion and metastasis.

Lateral periodontal cyst (LPC) is an uncommon developmental odontogenic cyst that occurs in the adjacent or lateral area of a vital tooth [22]. LPC is asymptomatic and found in the incisor-canine-premolar region, especially in mandible, during a routine radiological examination [22-24].
A radiolucent interradicular triangular lesion associated or not with displacement of the teeth root and with sclerotic borders is the classical radiography aspect [24]. SOT can arise in the same area and also present characteristic circumscription with frequent root divergence; however, the margin may or may not be corticated as in LPC. Of all cases reviewed for SOT, at least 8 previous cases present similar aspects of LPC [2, 4, 7, 9, 10, 16, 17, 25] and considering radiological aspects, SOT should be included as a differential diagnosis of LPC.

Fig. (1). Periapical radiography showed an unilocular radiolucent defect with triangular shape between the roots of inferior left second premolar and the inferior left first molar. A located periodontal bone loss is the main differential diagnosis of SOT.

Dentigerous cyst (DC) is the most common developmental odontogenic cyst arising from the crowns of unerupted teeth in mandible and maxilla [26]. Mandibular third molars and maxillary canines are the most often involved teeth, followed by the mandibular premolars and the maxillary third molars [26, 27]. The classical radiographic aspect of DC appears as a well-defined unilocular radiolucent with sclerotic borders associated with the crown of an unerupted tooth [26-28]. Some cases of SOT also presented similar aspects to DC and involved mandible and maxillary third molars [4, 29-32]. Moreover, both lesions can be found only in routine radiographic examination [1,4, 26-32]. Glandular odontogenic cyst (GOC) is a rare developmental odontogenic cyst with aggressive behavior that frequently involves the anterior mandible [33]. GOC is now well accepted being odontogenic origin; however, it presents glandular or salivary features as mucus cells and ductal structures [33, 34]. An extensive unilocular or multilocular radiolucent lesion with well-defined scalloped borders is a common finding in radiographic exams [33]. Tatemoto et al. in 1989 described a case of SOT presenting as radiolucency in the apical area of the vital mandibular central incisors in which the differential diagnosis of GOC was considered [25].

Fig. (2). Periapical radiography showed an unilocular periradicular radiolucent defect associated with superior right central incisor. Although rare the SOT diagnosis also should be included.

Keratocystic odontogenic tumor (KOT) is a benign odontogenic lesion with aggressive and infiltrative behavior that frequently appears in the posterior mandible areas; however, it can affect any site of the jaws [40]. Radiographically, KOT presents as a well or poorly circumscribed uni- or multilocular radiolucent lesion with variable sizes and shapes [40] and mimicking several jaw lesions including SOT. Thirteen authors presented SOT cases in which KOT should be mentioned as a highly suspicious differential diagnosis [4, 10, 13, 16-18, 25, 38, 41-45].

Adenomatoid odontogenic tumor (AOT) is an epithelial odontogenic tumor with slow and progressive growth that commonly involves the anterior portion of maxilla; however, anterior portions of mandible can also be affected [46]. The follicular AOT is the most frequent type and is associated with a crown and root of an unerupted tooth, especially canines. Extrafollicular AOT is not associated with teeth and it can be found between the roots of erupted teeth. An asymptomatic well-defined unilocular radiolucent lesion with or without radiopaque foci, eventual teeth displacement, and cortical expansion is the radiographic aspect of intraosseous AOT [46]. SOT lesions can show similar findings to follicular and extrafollicular AOT [4, 9, 10, 18, 47].

Central odontogenic fibroma (COT) is a rare odontogenic tumor with benign behavior and classified as a fibroblastic circular or ovoid radiolucent lesion with sclerotic borders and, frequently, associated with destruction of periradicular tissues and loss of lamina dura [36]. A lesion located near periapical or lateral region superimposed on the root completes the classical radiographic findings [35, 36]. SOT occurring in the same circumstances was described by at least 14 authors [4, 5, 7, 10-14, 16-18, 22, 37, 38]. Fig. (2) showed a radiolucent periradicular lesion with similar findings. Residual cyst (RC) is considered a retained radicular cyst from one tooth that was previously removed [34]. A radiolucent lesion usually asymptomatic involving an edentulous area, and discovered during a routine radiographic examination is the main aspect of RC [39]. One author described a case of SOT with radiographic characteristics of a residual cyst [14].
neoplasm that contains a wide amount of inactive odontogenic epithelium [1, 48]. COT presents as a slow and progressive lesion found in maxilla and mandible involving frequently periradicular region [48, 49]. Favia et al. 1997 described a case of SOT involving the apex of a first superior molar resembling this frequent radiographic appearance of COT [38]. However, some lesions can be found as a nonspecific well-defined unilocular radiolucency between erupted teeth causing root displacement or become associated with the crown of a unerupted tooth [48, 49]. In these cases, LPC, DC and ameloblastomas are some lesions that should be included as differential diagnosis of COT and, therefore, also of SOT [2, 4, 8, 9-11, 16, 17, 25, 29-32, 50, 51].

Ameloblastoma is a benign epithelial odontogenic tumor with two quite different intraosseous biologic variants [52]. The multicystic ameloblastoma (MA) is the most frequent type presenting aggressive and destructive characteristics with the involvement of posterior areas of jaws and impacted third molars in some cases [53]. Radiographically, MA shows as a radiolucent multilocular lesion with a “soap-bubbles” aspect associated with expansion and disruption of bone cortical [52, 53]. A unicystic ameloblastoma (UA) is less aggressive and commonly mimics odontogenic cysts frequently related with teeth in the area, especially, mandible third molars. A well-defined unilocular radiolucent lesion is the classical radiographic finding of the UA [52, 53]. Eleven cases of SOT were described with aspects that resemble uni- or multicystic ameloblastomas variants [2, 7-9, 11, 16, 17, 25, 38, 50, 51].

Langerhan’s cell histiocytosis (LCH) involves a rare group of hematological disorders originating from Langerhans cells that may affect the oral cavity [54]. Periodontal tissues are frequently involved and appear as located or generalized angular bone loss mimicking radiographic characteristics of an advanced periodontitis, such also is found in SOT [2, 4, 6, 7, 9-20, 54, 55]. Therefore, LCH and SOT should be considered as differential diagnosis when a severe periodontitis is present in the x-ray findings and no improvement is reached after periodontal treatment.

Multiple myeloma is a hematologic malignancy characterized by proliferation of plasma cells and nonfunctional monoclonal immunoglobulin in which medullary involvement through radiolucent osteolytic lesions is the most frequent presentation [56]. Nevertheless, a localized ill-defined radiolucency involving roots of teeth with lamina dura loss is also found in solitary plasmacytomas, and these myelomatous lesions could be misdiagnosed as periodontitis [56]. Thus, considering that the main radiographic aspect of SOT also mimics severe periodontitis, it should be included as a differential diagnosis of multiple myeloma and solitary plasmacytoma [2, 4, 7, 9, 11-16, 30, 41, 42, 50, 51].

Central giant cell lesion (CGCL) is considered a benign jaw lesion composed of osteoclast-like giant cells and commonly found in the mandible [57]. CGCL is more accepted as a reactive lesion presenting aggressive and non-aggressive behavior. The radiographic presentation is a well-defined non-corticated unilocular radiolucency in the small lesions until a multilocular aspect associated with ondulate septae in the bigger cases [57]. Some authors described SOT cases with x-ray findings suggestive of CGCL [2, 4, 8-14, 16, 25, 41, 42, 50, 51, 58, 59]. Finally, distant metastasis affecting oral cavity is quite uncommon; however, it can involve soft and bone tissues and requires a careful diagnostic process [60]. An osteolytic radiolucent lesion with irregular borders mimicks other jaw pathologies, since severe periodontitis until tumor process is the most frequent radiographic charac-

![Schematic chart showing the differential diagnosis of SOT based on radiographic aspects.](Image)
CONCLUSION

Although, all SOT cases in the literature present histopathological aspects which are well described, the variety of radiography findings of SOT mimicking odontogenic and non odontogenic jaw lesions is not well elucidated. Given the limitations of our approach, a review of the main radiographic presentations of SOT was proposed as a clinical diagnosis exercise for practicing clinicians.

This paper suggested a scheme to enhance the differential diagnosis hypotheses considering the routine image findings. Thus, this concise approach can help the clinicians to outline feasible diagnostic possibilities in front of the wide spectrum of odontogenic and non-odontogenic lesions. Fig. (3) showed a schematic chart based on the radiographic aspects of SOT. Nevertheless, due to the rarity of SOT and range of radiographic aspects for this entity, a careful microscopic examination should be performed before definitive diagnosis can be reached.

CONFLICT OF INTEREST

The authors confirm that this article content has no conflict of interest.

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REFERENCES

[1] Barnes L, Eversoll JW, Reichart P, Sidransky D, Eds. World Health Organization Classification of Tumors. Pathology and Genetics of Head and Neck Tumours, Lyon: IARC Press 2005.
[2] Badm I, Nagaura A, Kamath V. Squamous odontogenic tumor: a case report and review of literature. J Oral Maxillofac Pathol 2012; 16: 113-7.
[3] Bansal S, Joshi SK. Squamous odontogenic tumor with unusual localization and appearance: a rare case report. Case Rep Med 2013; 2013: 407967.
[4] Pullon PA, Shafer WG, Elzay RP, Kerr DA, Corio RL. Squamous odontogenic tumor. Report of six cases of a previously undescribed lesion. Oral Surg Oral Med Oral Pathol 1975; 40: 616-30.
[5] Carr RF, Carlson DM Jr, Marks RB. Squamous odontogenic tumor: report of case. J Oral Surg 1981; 39: 297-8.
[6] Agostini T, Sacco R, Bertolai R, Accocella A, Colafranceschi M, Lazzeri D. Peri-implant squamous odontogenic tumor. J Craniomaxillofac Surg 2011; 22: 1151-7.
[7] Haghhighat K, Kalmar JR, Mariotti AJ. Squamous odontogenic tumor: diagnosis and management. J Periodontol 2002; 73: 653-6.
[8] Barrios TJ, Sudol JC, Cleveland DB. Squamous odontogenic tumor associated with an erupting maxillary canine: case report. J Oral Maxillofac Surg 2004; 62: 742-4.
[9] Jones BE, Sarathy AP, Ramos MB, Foss RD. Squamous odontogenic tumor. Head Neck Pathol 2011; 5: 17-9.
[10] Cataldo E, Less WC, Giunta JL. Squamous odontogenic tumor. A lesion of the periodontium. J Periodontol 1983; 54: 733-5.
[11] Doyle JL, Grodjesk JE, Dolinsky HB, Rafel SS. Squamous odontogenic tumor: report of three cases. J Oral Surg 1977; 35: 994-6.
[12] McNeill J, Price HM, Stoker NG. Squamous odontogenic tumor: report of case with long-term history. J Oral Surg 1980; 38: 466-71.
[13] Hopper TL, Sadeghi EM, Pricco DF. Squamous odontogenic tumor. Report of a case with multiple lesions. Oral Surg Oral Med Oral Pathol 1980; 50: 404-10.

[14] Goldblatt LJ, Brannon RB, Ellis GL. Squamous odontogenic tumor. Report of five cases and review of the literature. Oral Surg Oral Med Oral Pathol 1982; 54: 187-96.
[15] Swan RH, McDaniel RK. Squamous odontogenic proliferation with probable origin from the rests of Malassez (early squamous odontogenic tumor?). J Periodontol 1983; 54: 493-6.
[16] Kim K, Mintz SM, Stevens J. Squamous odontogenic tumor causing erosion of the lingual cortical plate in the mandible: a report of 2 cases. J Oral Maxillofac Surg 2007; 65: 1227-31.
[17] Warnock GR, Pierce GL, Correll RW, Baker DA. Triangular-shaped radiolucent area between roots of the mandibular right canine and first premolar. J Am Dent Assoc 1985; 110: 945-6.
[18] Saxby MS, Repin JW, Sheron JE. Case report: squamous odontogenic tumor of the gingiva. J Periodontol 1993; 64: 1250-2.
[19] Yaacob HB. Squamous odontogenic tumor. J Nihon Univ Sch Dent 1990; 32: 187-91.
[20] Kangvonkit P, Sirichitra V, Hansasuta C. Squamous odontogenic tumor (report of a case and review of the literature). J Dent Assoc Thai 1981; 31: 25-33.
[21] Philipson HP, Reichart PA. Squamous odontogenic tumor (SOT): a benign neoplasm of the periodontium. A review of 36 reported cases. J Clin Periodontol 1996; 23: 922-6.
[22] Dubey KN, Garg S, Atri R. Diagnosis and osseous healing of a lateral periodontal cyst mimicking a deep unusual interdental socket in a young patient. Contemp Clin Dent 2010; 1: 47-50.
[23] Kumuda Arvind Rao HT, SHetty SR, Babu S. Unusual clinicora-diographic presentation of a lateral periodontal cyst. J Dent (Te-hran) 2012; 9: 265-9.
[24] DiFiore PM, Hartwell GR. Median mandibular lateral periodontal cyst. Oral Surg Oral Med Oral Pathol 1987; 63: 545-50.
[25] Tatometo Y, Okada Y, Morii M. Squamous odontogenic tumor: immunohistochemical identification of keratin. Oral Surg Oral Med Oral Pathol 1989; 67: 63-7.
[26] Di Pasquale P, Shermatero C. Endoscopic removal of a denigerous cyst producing unilateral maxillary sinus opacification on computed tomography. Ear Nose Throat J 2006; 85: 747-8.
[27] Jones AV, Craig GT, Franklin CD. Range and demographics of odontogenic cysts diagnosed in a UK population over a 30-year period. J Oral Pathol Med 2006; 35: 500-7.
[28] Kasat VO, Karjodkar FR, Laddha RS. Denigerous cyst associated with an ectopic third molar in the maxillary sinus: a case report and review of literature. Contemp Clin Dent 2012; 3: 373-6.
[29] Wright JM Jr. Squamous odontogenic tumorlike proliferations in odontogenic cysts. Oral Surg Oral Med Oral Pathol 1979; 47: 354-8.
[30] Norris LH, Baghaeri-Rad M, Maloney PL, Simpson G, Guinta J. Bilateral maxillary squamous odontogenic tumors and the malignant transformation of a mandibular radiolucent lesion. J Oral Maxillofac Surg 1984; 42: 827-34.
[31] Cillo JE Jr, Ellis E 3rd, Kessler HP. Pericondial squamous odontogenic tumor associated with an impacted mandibular third molar: a case report. J Oral Maxillofac Surg 2005; 63: 413-6.
[32] Ide F, Shimoyama T, Horie N, Shimizu S. Intraosseous squamous cell carcinoma arising in association with a squamous odontogenic tumour of the mandible. Oral Oncol 1999; 35: 431-4.
[33] Fowler CB, Brannon RB, Kessler HP, Castle JT, Kahn MA. Glan-dular odontogenic cyst: analysis of 46 cases with special emphasis on microscopic criteria for diagnosis. Head Neck Pathol 2011; 5: 364-75.
[34] Shafer WG, Hine MK, Levy BM, Rajendran R, Sivapathasun-haram B. Shafer’s Textbook of Oral Pathology. 6th ed. New York: Elsevier 2009.
[35] Ricucci D, Pascon EA, Ford TR, Langeland K. Epithelium and bacteria in periapical lesions. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006; 101: 239-49.
[36] Ricucci D, Mannocci F, Ford TR. A study of periapical lesions correlating the presence of a radiopaque lamina with histological findings. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006; 101: 389-94.
[37] Unal T, Gomel M, Gunel O. Squamous odontogenic tumor-like islands in a radicular cyst: report of a case. J Oral Maxillofac Surg 1987; 45: 346-9.
[38] Favia GI, Di Alberti L, Scaran0 A, Piattelli A. Squamous odonto-genic tumour: report of two cases. Oral Oncol 1997; 33: 451-3.
[39] Jamdade A, Nair GR, Kapoor M, Sharma N, Kundendu A. Localization of a Peripheral Residual Cyst: diagnostic role of CT scan. Case Rep Dent 2012; 2012: 760571.

[40] Titinchi F, Nortje CJ. Keratocystic odontogenic tumor: a recurrence analysis of clinical and radiographic parameters. Oral Surg Oral Med Oral Pathol Oral Radiol 2012; 114: 136-42.

[41] Mills WP, Davila MA, Beuttenmuller EA, Keeferle BM. Squamous odontogenic tumor. Report of a case with lesions in three quadrants. Oral Surg Oral Med Oral Pathol 1986; 61: 557-63.

[42] Leider AS, Jonker LA, Cook HE. Multicentric familial squamous odontogenic tumor. Oral Surg Oral Med Oral Pathol 1989; 68: 175-81.

[43] Philipsen HP, Reichart PA, Siar CH, et al. An updated clinical and epidemiological profile of the adenomatoid odontogenic tumour: a collaborative retrospective study. J Oral Pathol Med 2007; 36: 383-93.

[44] Leventon GS, Happonen RP, Newland JR. Squamous odontogenic tumor. Am J Surg Pathol 1981; 15: 671-7.

[45] Kim JY, Kim JC, Cho BO, Kim SG, Yang BE, Rataru H. Squamous odontogenic tumor: A case report and review of literature. J Korean Assoc Oral Maxillofac Surg 2007; 33: 59-62.

[46] Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumor of the mandible: review of the literature and report of a rare case. Head Face Med 2005; 1: 3.

[47] Monteil RA, Terestri P. Squamous odontogenic tumor related to an unerupted lower canine. J Oral Maxillofac Surg 1985; 43: 888-95.

[48] Mosqueda-Taylor A, Martínez-Mata G, Carlos-Bregni R, et al. Central odontogenic fibroma: new findings and report of a multicentric collaborative study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011; 112: 349-58.

[49] de Matos FR, de Moraes M, Neto AC, Miguel MC, da Silveira EJ. Central odontogenic fibroma. Ann Diagn Pathol 2011; 15: 481-4.

[50] Kristensen S, Andersen J, Jacobsen P. Squamous odontogenic tumour: review of the literature and a new case. J Laryngol Otol 1985; 99: 919-24.

[51] Baden E, Doyle J, Mesa M, Fabié M, Lederman D, Eichen M. Squamous odontogenic tumor. Report of three cases including the first extraosseous case. Oral Surg Oral Med Oral Pathol 1993; 75: 733-8.

[52] More C, Tailor M, Patel HJ, Asrani M, Thakkar K, Adalja C. Radiographic analysis of ameloblastoma: a retrospective study. Indian J Dent Res 2012; 23: 698.

[53] Chawla R, Ramalingam K, Sarkar A, Muddiah S. Ninety-one cases of ameloblastoma in an Indian population: a comprehensive review. J Nat Sci Biol Med 2013; 4: 310-5.

[54] Aruna DR, Pushpalatha G, Galgali S, Prashanthy, Langerhans cell histiocytosis. J Indian Soc Periodontol 2011; 15: 276-9.

[55] Artzi Z, Grosky M, Raviv M. Periodontal manifestations of adult onset of histiocytosis X. J Periodontol 1989; 60: 57-66.

[56] Cardoso RC, Gengross PJ, Hofstede TM, Weber DM, Chambers MS. The multiple oral presentations of multiple myeloma. Support Care Cancer 2014; 22(1): 259-67.

[57] Triantafillidou K, Venetis G, Karakinaris G, Iordanidis F. Central giant cell granuloma of the jaws: a clinical study of 17 cases and a review of the literature. Ann Otol Rhinol Laryngol 2011; 120: 167-74.

[58] Schwartz-Arad D, Lustmann J, Ulmansky M. Squamous odontogenic tumor. Review of the literature and case report. Int J Oral Maxillofac Surg 1990; 19: 327-30.

[59] Ruhin B, Raoul G, Kolb F, et al. Aggressive maxillary squamous odontogenic tumour in a child: histological dilemma and adaptive surgical behaviour. Int J Oral Maxillofac Surg 2007; 36: 864-6.

[60] Ogütcen-Toller M, Metin M, Yildiz L. Metastatic breast carcinoma mimicking periodontal disease on radiographs. J Clin Periodontol 2002; 29: 269-71.