Squamous Odontogenic Tumor: Diagnosis and Management

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The squamous odontogenic tumor (SOT) is a rare, benign, locally infiltrative neoplasm of the jaws that appears to originate from the rests of Malassez. It has been confused with other pathologic entities such as ameloblastomas, carcinomas, and fibromas and clinically may resemble localized periodontal disease. The tumor is often asymptomatic, although it can present with symptoms of pain and tooth mobility. A characteristic radiographic appearance is that of a triangular-shaped or semi-circular lucency associated with the roots of erupted teeth. Histologically, the tumor is characterized by the formation of variably sized nests and cords of uniform, benign-appearing, squamous epithelium with occasional vacuolization and keratinization. Treatment of SOT by conservative surgical excision is normally curative with rare episodes of recurrence reported. Since the clinical presentation of SOT may mimic more common pathologic entities, this case report reinforces the need for careful histologic evaluation of all lesions found in the periodontium. J Periodontol 2002;73:653-656.

KEY WORDS

Neoplasms, squamous cell; odontogenic tumors/diagnosis.

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The squamous odontogenic tumor (SOT) is a rare pathologic entity that was originally defined and named in 1975 by Pullon et al.[1] as a series of 6 previously undescribed lesions. Since that time, fewer than 50 cases have been described in the literature. The tumor typically arises as a solitary lesion in the third decade of life, however, multifocal cases have also been described.[1,2] Favored sites include maxillary canine and mandibular premolar regions of the jaw. The tumor exhibits indolent growth and is usually asymptomatic. Occasionally local pain, gingival swelling, and mobility of adjacent teeth have been reported. The maxillary lesions seem to be more aggressive in nature than tumors located in the mandible. No gender or racial predilection of this neoplasm has been described.

Radiographically, SOT often exhibits a characteristic unilocular and triangular-shaped radiolucency of the alveolar bone, with the wide base of the lucency localized between the diverging apices of the adjacent roots. Occasionally, scalloping and saucerization of the underlying bone is seen; however, this is regarded as a pressure phenomenon rather than tumor infiltration.[3]

In this case report, we document the clinical, radiographic, and histologic characteristics of a squamous odontogenic tumor lesion that closely resembled the clinical presentation of localized periodontal disease.

CASE REPORT

A 43-year-old African-American male patient was referred to The Ohio State University Section of Periodontology by his general dental practitioner for periodontal surgical care. The medical history of the patient was unremarkable. His dental history revealed restorative care as well as non-surgical periodontal therapy for a period of a year prior to examination in the periodontal graduate clinic. The patient reported removal of an unerupted tooth from the area of the upper left canine approximately 20 years earlier. Previous dental practitioners explained the radiographic lucency between teeth numbers 11 and 12 as an osseous defect which resulted from the surgical removal of the impacted tooth.

Clinical examination revealed gingival recession on the facial surfaces of teeth numbers 11, 12, and 13, with probing depths of 7 mm distobuccal and distolingual to tooth number 11. Transgingival probing revealed significant interproximal bone loss amounting to almost three-quarters of the buccal and and one-third of lingual bone between teeth numbers 11 and 12.

The gingiva exhibited a mild marginal erythema. There were no noticeable signs of soft tissue swelling or osseous expansion. The papilla in the interdental region between teeth numbers 11 and 12 had a slight nodular appearance and minor loss of papillary height was evident (Fig. 1). Bleeding on probing was present at the sites around these teeth and both teeth were responsive to electrical stimulation. No sensitivity to percussion was noted for teeth numbers 11 or 12.

Radiographic assessment showed a radiolucency with severe alveolar bone loss localized between teeth numbers 11 and 12, which was associated with markedly diverging roots (Fig. 2). Periapical radiographs from the previous year did not reveal any remarkable differences in the size or extent of the current radiolucent finding.

Based on the clinical findings, the differential diagnosis included peripheral giant cell lesion, fibro-osseous lesion, localized severe chronic periodontitis, residual odontogenic cyst, squamous odontogenic tumor, ameloblastoma, lateral periodontal cyst, and radicular cyst.

Surgical Treatment

Due to the radiographic appearance, arrangement was made for a biopsy of the interdental lesion between teeth numbers 11 and 12. Intrasulcular incisions were made and a palatal mucoperiosteal flap was reflected for access to the lesion. Gently packing ribbon-gauze against the palatal bone, the lesion was displaced facially while still attached to the buccal gingiva (Fig. 3). The neoplasm was subsequently dissected from the buccal mucogingival flap with relative ease and the lesion enucleated *in toto*. The lesion was firm and rubbery in consistency and no fluid was aspirated. The roots of the adjacent teeth were root-planed, irrigated with saline, and the mucoperiosteal flap apically positioned and closure achieved using interrupted 4/0 silk sutures. Due to the osseous defect observed (horizontal defect with no bony walls), regeneration of the bone was not considered. The surgical site was monitored for 18 months postoperatively with no sign of a recurrence (Fig. 4). Although a mild marginal gingival inflammation persists, all probing depths for teeth numbers 11 and 12 were less than 4 mm 18 months postoperatively.

Histopathology

Macroscopically, the specimen was a tan-gray, soft tissue measuring 18 × 12 × 8 mm (Fig. 5). Micro-

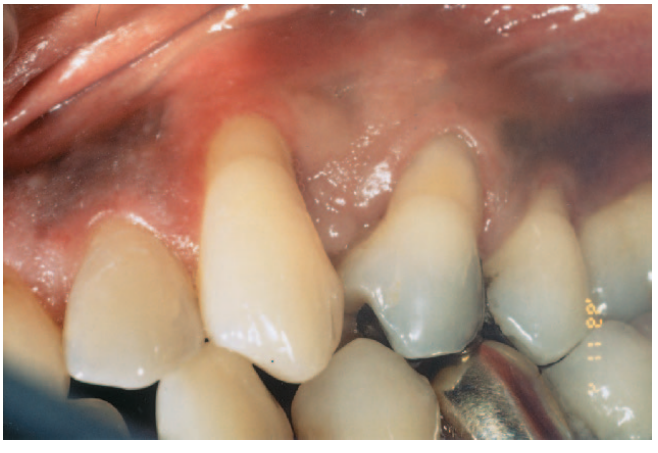

Figure 1.
Preoperative view of teeth numbers 10 through 13.

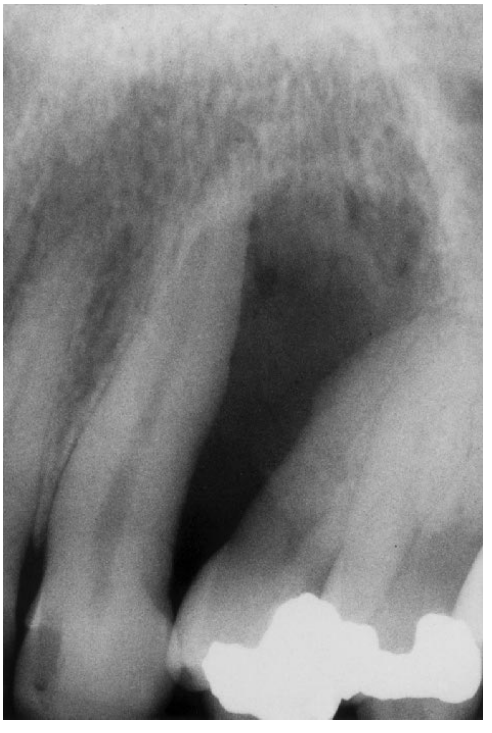

Figure 2.
Periapical radiograph of teeth numbers 11 and 12.

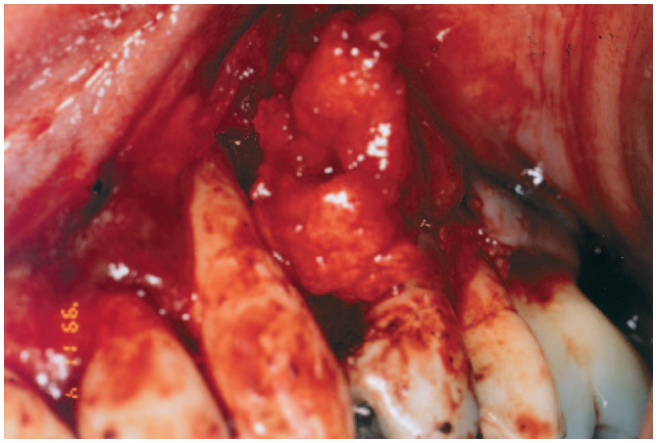

Figure 3.
Neoplasm has been displaced facially while maintaining attachment to the buccal mucoperiosteal flap.

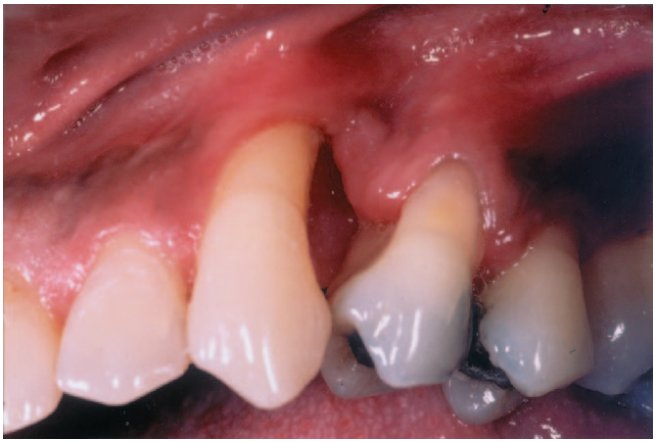

Figure 4.
Postoperative view of teeth numbers 10 through 13 eighteen months following surgical excision.

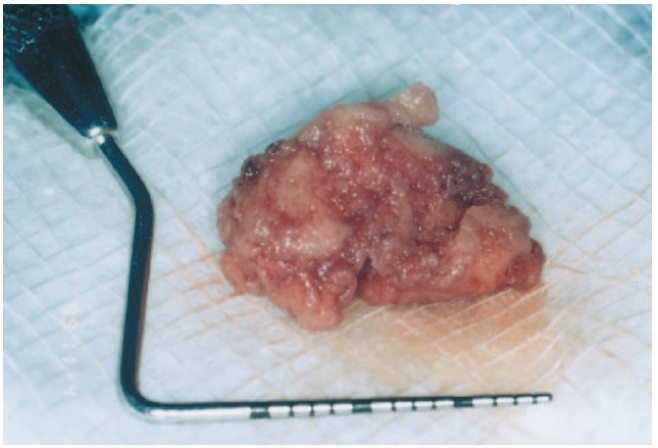

Figure 5.
SOT measuring 18 × 12 × 8 mm.

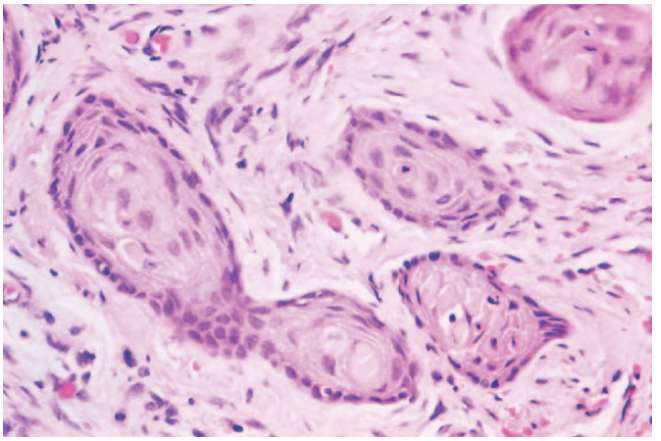

Figure 6.
Histophotomicrograph of SOT (hemotoxylin and eosin; original magnification ×40).

scopically, the sections showed a soft tissue specimen, which was consistent of a tumor of epithelial origin within a mature collagenous stroma. The lesional cells were characterized by formation of variably sized nests and cords of uniform, benign-appearing, squamous epithelium (Fig. 6). The individual cellular islands revealed a lack of peripheral cellular palisading (Fig. 6). Occasional vacuolization and keratinization were noted (Fig. 6). The microscopic diagnosis was SOT.

DISCUSSION

Although an extremely rare lesion of the periodontium, periodontists and general dentists should be aware of SOT because of its ability to closely mimic periodontitis. SOT can share many of the characteristic features of periodontitis including gingival inflammation, deep probing depths, and radiographic evidence of bone loss. The principal method to clinically differentiate SOT from periodontitis is the appearance of a unilocular, triangle-shaped radiolucency that may sometimes also be located between the roots of teeth. There were no clinical signs or symptoms associated with teeth numbers 11 and 12 that would suggest the presence of SOT except for the incidental finding on radiographic assessment.

Histologically, SOT contains multiple, small islands

of squamous epithelium with a moderately cuboidal or flattened peripheral basal cell layer in a collagenous fibrous connective tissue stroma. Cystic degeneration of the islands has been reported and some have been shown to contain both prekeratin and laminar calcified masses.[1,4] Clear cells show a positive PAS reaction which indicates the presence of glycogen. The lack of nuclear palisading by the peripheral epithelial cell layer of the tumor islands should help rule out the diagnosis of ameloblastoma. Similarly, the bland histologic features of the epithelial nests should exclude consideration of carcinoma.

The histogenesis of SOT remains controversial. Some researchers have suggested that these tumors originate from gingival epithelium[5] or gingival rests of Serres;[6] however, most investigators support the theory that SOT develops from the rests of Malassez.[1,4,7] Supporting this concept is the observation that SOT is often associated with the roots of erupted vital teeth, although impacted teeth can also be affected.[1] In this case, no expansion of the alveolar bone or swelling of the gingiva was present. Furthermore, the tight adherence of the tumor to the canine root and the ease of separation from the buccal soft tissues suggest that the origin of the SOT is most likely the rests of Malassez. The rests of Malassez have long been thought as having no function; however, recently they have been implicated in having pathologic potential.[8,9] The past history of an extraction from the site of SOT in this case and the presence of inflammation may have been triggering factors in development of neoplastic growth from rests of Malassez.

While considered a benign neoplasm, SOT has been known to infiltrate into adjacent tissues, with resorption of alveolar bone and invasion of the overlying gingiva and oral mucosa. This is especially true in the maxilla, where occasionally invasion of adjacent structures such as the sinus and nasal cavity has necessitated wide en bloc resection, including hemimaxillectomy. As seen in this case, the tumor is slow-growing and typical conservative surgical intervention including enucleation and curettage has often proved curative. It should be noted that, depending on the amount of bony destruction, the possibility for regeneration of the periodontium should be considered. In this case, the destruction of osseous tissue did not provide adequate architecture for regeneration to be considered. In the original description of the lesion by Pullon et al.[1] only one case had a recurrence which required more extensive surgical excision. In our case, no sign of a recurrence has been evident 18 months postoperatively. Although it is considered a benign tumor, recent observations suggest the possibility of carcinomatous transformation.[10] A rare case of intraosseous squamous cell carcinoma arising in association with and presumed to be a malignant variant of a SOT lesion emphasizes the need for histopathological assessment of all oral pathology specimens.

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