Peripheral squamous odontogenic tumor masquerading as oral squamous cell carcinoma in an elderly female patient: A rare entity

Shikha Saxena¹, Sankalp Mittal², Sonal Priya², Krishna S. Sundaragiri₁, Chandni Shekhawat³, Akshay Bhargava₁, Bharat Sankhla₁

Departments of ¹Oral Pathology, ²Oral Surgery and ³Oral Medicine, RUHS College of Dental Sciences, Jaipur, Rajasthan, India

ABSTRACT

Squamous odontogenic tumor is a rare epithelial odontogenic tumor. Only a few cases have been reported in the literature, out of which central cases occurring within the jawbones are more common, and very rarely peripheral cases have been reported. Here, we report a rare case of a peripheral squamous odontogenic tumor occurring in the left lower gingivobuccal sulcus region in a 63-year-old female patient and presented as a firm and fibrotic swelling clinically. Radiographic examination revealed no evidence of a central lesion in the bone. Excisional biopsy of the lesion revealed the characteristic histopathological picture of peripheral squamous odontogenic tumor (PSOT). The healing was found to be satisfactory upon periodic evaluation of the patient done to study the healing and for any recurrence of the lesion.

Keywords: Benign, epithelial, odontogenic tumor, peripheral

Introduction

Odontogenic tumors (OTs) consist of benign and malignant odontogenic neoplasms, which can be epithelial, mesenchymal, and of mixed origin. They are majorly central occurring within the jawbones but some may involve the soft tissues.[³] Pullon et al.[²] in 1975 first described squamous odontogenic tumor (SOT). SOT is considered a rare, benign locally infiltrative, epithelial odontogenic tumor.[³]

Clinically, SOT can present as an asymptomatic, slow-growing, intrabony lesion to a painful bony expansion, with mobility of teeth, swelling of the alveolar process; maxillary lesions have been suggested to be more aggressive in clinical presentation. More commonly, it occurs as a central lesion, and only very few cases of peripheral lesions have been reported. A characteristic unilocular radiolucency of triangular or semicircular shape occurs between the diverging apices of the adjacent roots with occasional scalloping and saucerization of the underlying bone. Extensive SOTs may also present as multilocularity, while peripheral variant may present as saucerization due to mainly a pressure effect on the underlying alveolar bone.[⁴]

Clinicopathologically, three main types of SOT exist: the intraosseous, mural, and extraosseous variants. However, clinically, two main forms exist: central SOT and peripheral SOT (PSOT).[³] The central variant is more common with around 50 cases reported; however, the peripheral variant is rare. The purpose of this paper is to report a clinical case of a patient...
with a peripheral squamous odontogenic tumor with an unusual location and the paucity of reported cases.

Case Report

A 63-year-old female patient presented with a mass in the left lower cheek area. History revealed that she consulted a dentist but it was ignored for 6–7 months. The lesion was initially small and gradually increased in size. On examination, intraorally the mucosa appeared normal. There was obliteration of the left gingivobuccal sulcus. Bimanual palpation revealed a hard mass between mucosa and the cheek. The growth measured approximately 3 cm × 5 cm and had a smooth surface [Figure 1].

Radiographic examination revealed no bony changes. Preoperative computed tomography (CT) scans showed evidence of large, heterogeneously enhancing soft tissue lesion measuring 45 mm × 24 mm × 35 mm seen in the left lower gingivobuccal sulcus associated with erosion of mandibular alveolus in the molar region [Figure 2]. The growth involved buccal mucosa laterally and retromolar trigone medially with loss of fat plane with masseter muscle. No evidence of cervical lymphadenopathy was noted. Fine-needle aspiration cytology of the mass showed no evidence of malignant cells. Incisional biopsy was suggestive of an odontogenic tumor. The lesion was removed in toto using extra-submandibular incision. Marginal mandibulectomy was also done, keeping a safe margin. The patient recovered uneventfully.

The biopsy revealed peripheral stratified squamous epithelium. The islands and nests of well-differentiated squamous epithelial cells can be seen in underlying mature connective tissue stroma. The islands were lined by flat to cuboidal cells and exhibited vacuolization, microcyst formation, and squamous differentiation. The lesion was diagnosed as a peripheral squamous odontogenic tumor [Figure 3].

Discussion

Squamous odontogenic tumor is a rare lesion with few cases reported to date in the literature. A review of 36 cases was done by Philipsen et al. in 1996 [6]. We are tabulating the various cases of PSOT mentioned in the literature [Table 1] [5,7-12]. As only very few cases of PSOT have been reported in the literature, the incidence is thus unknown; however, studies on odontogenic tumors as a whole have reported a relative frequency of SOT to range from approximately 0% to 2% among different odontogenic tumors [13].

SOT is a slow-growing neoplasm, presenting with few clinical signs and symptoms, the most common being mobility of teeth adjacent to the tumor, swelling of the gum, and moderate pain. The most common locations for the development include the anterior maxilla and posterior mandible. The tumor has been found in patients in the age range of 8–74 years and shows a definite peak in the third decade of life. Males and females appear to be equally affected [6].

Radiography of the central variant of PSOT shows a well-defined unilocular radiolucency of triangular shape in the alveolar process present between the roots of the adjacent teeth. The peripheral variant may cause some sauceration of the underlying bone, which can be explained to be occurring because of pressure phenomenon rather than because of true tumor infiltration. Histopathologically, the tumor consists of numerous islands and nests of squamous epithelial cells dispersed in a mature connective tissue stroma. The islands are numerous and are sharply demarcated from the surrounding stroma by a flattened
layer of cells at their periphery. They tend to vary in shape, with the presence of round-to-oval epithelial islands. The epithelium in these rounded islands often shows a swirling or “whirlpool” pattern to the central squamous cells. Within the epithelial islands, cystic changes can be seen; keratinization of the central cells may be present too.\textsuperscript{[11]}

The differential diagnosis of squamous odontogenic tumor includes acanthomatous and desmoplastic variants of ameloblastoma, well-differentiated squamous cell carcinoma, and SOT-like proliferations that occur in the wall of odontogenic cysts such as dentigerous cyst and apical periodontal cyst.\textsuperscript{[12]}

Various immunohistochemical studies have been done on SOT. Siar et al.\textsuperscript{[13]} suggested that ameloblastoma differs from SOT in the degree of differentiation of the lesions, depending on the results of their immunohistochemical stain of the two lesions with Notch 1, 3, and 4, Jagged 1, and Delta 1.

Treatment includes enucleation, curettage, and local excision in cases of PSOT. Tumors located in the maxilla require more radical treatment because they show more aggressive biological behavior. Very rarely, malignant transformation is reported.\textsuperscript{[13]}

\textbf{Conclusion}

SOT is a rare, benign, epithelial odontogenic tumor of the jawbones. It is composed of mature connective tissue stroma with islands and nests of varying sizes and shapes and consisting of bland appearing well-differentiated epithelial cells. Because of histological similarity, SOT has often been overdiagnosed as ameloblastoma and squamous cell carcinoma. It thus becomes important to understand the clinical, radiologic, histopathologic, and treatment characteristics of this tumor.

\textbf{Declaration of patient consent}

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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\textbf{Conflicts of interest}

There are no conflicts of interest.

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