Squamous Odontogenic Tumor

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Abstract

The twenty-fifth reported case of squamous odontogenic tumor is presented. The patient was a 39-year-old Chinese Malaysian male. The lesion was asymptomatic and did not recur after excision. It is important to distinguish this type of lesion from squamous odontogenic tumor-like proliferation. The rest of Malassez is thought to be responsible for the histogenesis of the lesion. Better understanding will only be achieved when more cases are reported and studied.

Introduction

Squamous odontogenic tumor (SOT) is a term used to describe a rare tumor first characterized in 1975 by PULLON et al.^[1]. Together with their six cases, there are now 24 documented in the literature^[2-5]. The actual number is probably less than 24, as some authorities have cast doubt on the true nature of some of the reported cases^[2,6]. ERSEVAN et al.^[7] preferred to use the term squamous odontogenic fibroma instead of SOT for their lesion, since the stromal connective tissue was loose and reminiscent of a fibroma, with abundant stellate and fusiform connective tissue cells. Comprehensive reviews of the literature up to 1986 are available^[2,4,5,8]. SOT should not be confused with squamous odontogenic tumor-like proliferation in odontogenic cysts, as described by SWAN and McDANIEL^[9] and SIMON and JENSEN^[10]. This paper reports a case of SOT in a Malaysian Chinese, thus bringing the total number of documented cases of SOT to 25.

Case Report

The patient was a 39-year-old Chinese man who was referred by his general dental practitioner for a firm painless swelling measuring about 3 cm in diameter on the palate adjacent to the upper right cuspid and first bicuspid, which were both asymptomatic. He did not know how long the swelling had been present. Radiographically, there was a hemispherical area of decreased radiolucency with a sclerotic margin in the palatal bone around the roots of the upper right cuspid and first bicuspid.

An incisional biopsy was taken. Microscopically, the tissue was composed of numerous islands of well differentiated stratified squamous epithelium of various

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sizes distributed in a stroma of mature collagenous tissue (Fig. 1). The squamous epithelium appeared mature and benign and was bounded by a peripheral layer of flattened cells. Microcysts were seen within the islands (Fig. 2). Cleavage between the squamous epithelium and the peripheral cells was seen (Fig. 3). Larger cystic spaces within the islands were seen to contain variable amounts of capillaries (Fig. 4). Small calcified masses surrounded by keratin were present within the epithelial islands (Fig. 5). There was no connection of the tumor tissue with the surface epithelium, and there was no evidence of amyloid when stained with Congo red or crystal violet. The connective tissue stroma did not show any reactive signs in the vicinity of the epithelial islands except for the presence of slight chronic inflammation. The lesion was finally excised. Although the margins of the excised lesion were not free of tumor tissue, there has been no recurrence after almost four years.

Discussion

As this is the twenty-fifth case of SOT reported so far in the literature, and there have been extensive recent literature reviews, it is now only necessary to comment on this new case. SOT has frequently been mistaken for ameloblastoma. Terminologies like “epithelial odontogenic tumor”, “benign epithelial odontogenic tumor”, “acanthomatous ameloblastoma” and “acanthomatous ameloblastic fibroma” have been used to describe this lesion^[1]. The author is confident that the present lesion is in fact SOT, since its clinical, radiographic and histopathological appearances, together with its behavior, were consistent with those of other established SOT cases reported so far, incorporating the following features:

The average age of SOT patients was reported to be 37.4 years (range 11 to 67 years). The lesions were manifested as hemispherical or triangular radiolucent areas around the roots of the teeth. Histologically, SOT is different from ameloblastoma in that it lacks palisading peripheral columnar cells, polarization of the nuclei away from the basement membrane and cytoplasmic vacuolation. In addition, stellate reticulum-like cells, which are always present in ameloblastoma, are never present in SOT. Calcific bodies may be present in some cases of SOT^[5]. SOT does not characteristically recur, in contrast to ameloblastoma which invariably recurs, if inadequately removed^[8].

There was, however, one feature in the present case which has never been mentioned by previous authors, although a similar feature was seen in some reported cases^[7,9]. This was the presence of a plane of ready cleavage between the peripheral cuboidal cells and the central epithelial tissues (Fig. 3). This cleavage probably occurred because of junctional weakness between the two dissimilar structures. The constant presence of intercellular edema within the lesion^[11] and cystic areas may contribute to the weakening of cellular cohesion, which may finally result in the formation of a cleavage plane.

The origin of SOT is uncertain, although it has been speculated that it may arise from the rests of Serres^[11], the overlying gingival epithelium^[1], or more popularly, the rests of Malassez^[1-3,13-15]. The rests of Malassez are believed to be

responsible because SOT is always associated intimately with the roots of teeth. Although the present author is unable to offer any hypothesis about the histogenesis of SOT, the intimate association of the lesion with the roots of the right upper cuspid and the first bicuspid, coupled with the concurrent presence of several rests of Malassez adjacent to the tumor (Fig. 6), tends to lend support to the belief that the rests of Malassez may indeed be responsible. It was noted that the tumor tissue in the present case was not in contact with the overlying gingival epithelium when serial sections were taken. It is therefore highly unlikely that this tumor arose from the gingival epithelium. More information is needed about SOT, and until more cases are found and studied, many aspects of the lesion will remain speculative.

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Fig. 1 Islands of well differentiated stratified squamous epithelium of variable sizes distributed in a stroma of mature collagenous tissue (H & E x40)

Fig. 2 A higher-power view of an epithelial island composed of mature and benign stratified squamous epithelium showing no significant dysplasia. The peripheral layer of cells is more darkly stained and appears cubical or flattened. Many microcysts are seen within the island (H & E x4000)

Fig. 3 Cleavage between the cuboidal peripheral cells and the squamous epithelium of the island is evident at top right (H & E x400)

Fig. 4 Larger cystic areas containing variable amounts of capillaries (H & E x400)

Fig. 5 Two small calcified areas (arrow) surrounded by less darkly stained keratin (H & E x400)

Fig. 6 Epithelial rest of Malassez (arrow) adjacent to and outside the field of the squamous odontogenic tumor (H & E x400)