



Squamous Odontogenic Tumor of Posterior Maxilla- A Report of a New Unusual Case & Literature Review

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Study

ABSTRACT

Squamous odontogenic tumor is a benign epithelial odontogenic tumor that is very unusual in the maxilla & mandible. Mostly, these are present as single lesions, but rarely they can be multifocal lesions. The nature of maxillary lesions is more aggressive. Because of their benign nature, these lesions are frequently treated with conservative surgical techniques that include curettage and surgical enucleation. We will discuss the instance of a 29-year-old lady who was misdiagnosed and treated conservatively by us. This case was examined in the light of current knowledge of the prevalence, genesis, diagnosis, and treatment of squamous odontogenic tumors, as well as a literature review.

Keywords: *Odontogenic tumor; posterior maxilla; squamous cells.*

1. INTRODUCTION

Squamous odontogenic tumor (SOT) is a rare, benign, slow-growing, locally infiltrative tumor that arises from the odontogenic epithelium.

The WHO classifies it as a benign epithelial odontogenic tumor, with only approximately 60 occurrences known. It was initially identified by Pullon et al in 1975 [1,2, 3].

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SOT can affect people of any age. However, it is most commonly diagnosed in their forties. Its often associated with erupting and vital teeth. Although all gnathic bones are affected, there is a preference for the anterior maxilla and posterior mandible [4]. SOT usually presents as a single intraosseous lesion, but multifocal and peripheral tumors have also been reported. Asymptomatic edema with distinct movement of the corresponding teeth, as well as periodontal bone loss, are some of the common clinical findings. Many documented cases, on the other hand, are the result of a routine radiographic examination. Although a few have presented as growing, multilocular lesions, the majority of lesions are present as unilocular radiolucency. SOT appears as a triangular radiolucency between the roots of teeth, with the Triangulum base pointing towards the root apices. Local surgical excision with enucleation and curettage is the preferred treatment option.

2. CLINICAL FINDINGS

A female patient of 29-years old reported to Dershan clinic for further investigation of pain and swelling in the upper left posterior region. The past medical records were non-contributory. She had been experiencing pressure in the affected location for the preceding 8 months, but there was no evidence of left infraorbital nerve hypesthesia. Intra-orally, revealed an erythematous, firm swelling in the vestibular mucosa, gingiva & interdental papillae that extended from the left canine to the left first premolar region. It was soft & rubbery in consistency and the tenderness was appreciated. There had been no previous discharges.

On vitality testing of the upper teeth, there was evidence of positive sensitivity. Deep palatal periodontal pockets were identified on both canine and first premolar in maxilla. A triangular radiolucency was present within the anteroposterior maxilla stretching from the upper left second premolar to the upper left canine was seen on the panoramic radiograph [Fig: 1]. Although there was no root resorption.

At the patients initial visit to our clinic, an incisional biopsy of the lesion within the maxilla was performed under local anesthesia. [Fig:2] In the pathology report, multiple proliferations of squamous epithelium surrounded by moderately cellular connective tissue stroma were seen. These epithelial

islands were distinct from the stroma, and their size and shape varied. Few islands have demonstrated cystic degeneration. Foci of calcification were also evident. There was no evidence of palisaded basal cells or columnar cells or stellate reticulum. [Fig: 4] Overall, there had been no histopathologic presentation of any malignancy.

As a result of these observations, the pathologist proposed the diagnosis of SOT. Under local anesthetic, surgical enucleation and thorough curettage of the lesion in the maxilla were performed after gaining informed consent. [Fig: 3] Healing was uneventful in the first, second, and third weeks after surgery, as well as a few months later. The diagnosis of multifocal SOT within the maxilla was confirmed by correlating clinical, radiological, intraoperative data and histopathological report after resection.

3. DISCUSSION

SOT is a benign odontogenic tumor, according to WHO's 2017 classification of head and neck tumors. [3] Its a locally infiltrative neoplasm with islands of well-differentiated squamous epithelium inside the fibrous stroma, according to the description. [3,5] SOT was first described and named by Pullon et al. in 1975. [6] There have been around 50 cases of this strange entity cited in the literature. [1,2] SOT is more common in patients between the age group of 8 to 74 years (mean, 38 years). Overall, 1.8:1.3 is the male: female ratio. Even though both the gnathic bones are equally affected by SOTs, the mandible has a slight preference [1]. Mandibular posterior and maxillary anterior areas are mostly affected by SOT, but unfortunately, in our case, it was found in the maxillary posterior area, which makes it a very rare case in literature. However, maxillary SOT is more aggressive as compared to mandibular SOT because the maxillary bone is more porous than the mandible. [7,8,9,10,11] Mandibular bone can withstand tumor expansion better as it is denser than maxillary bone.[4].

The majority of SOTs are solitary lesions that develop in the maxilla or mandible, while some multifocal variants are also described in the literature. Elmuradi et al. [2] looked at a total of 8 instances and discovered that the lesions were predominantly seen in African-Americans and that the patients were in their younger age

groups. Another example of SOT with localization within the maxilla is presented here. SOTs are frequently classified as central

or peripheral [12]. The central SOT is found within the jaw bones in the majority of instances.



Fig. 1. Triangular radiolucency can be appreciated between 23 and 24 involving 25 also



Fig. 2. Tumor mass can be appreciated from distal of 23 till mesial of 25



Fig. 3. Complete excision of SOT was done

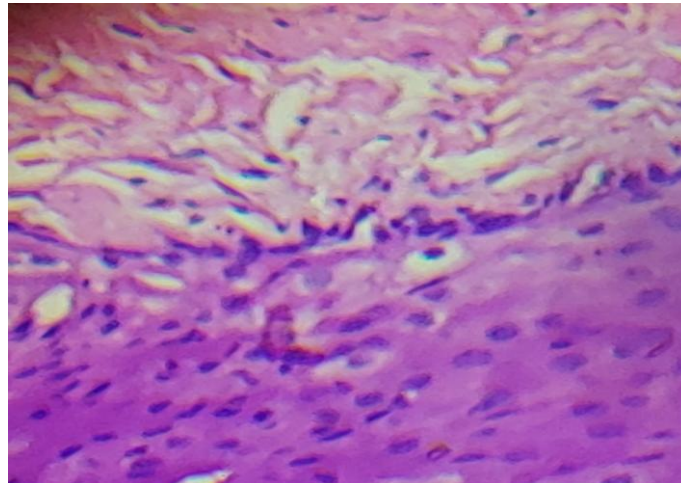


Fig. 4. The microphotograph shows multiple squamous epithelial proliferations surrounded by relatively cellular connective tissue stroma. Islands of epithelial are well demarcated and are of variable size and shape. Few islands demonstrated cystic degeneration. Foci of calcification are also evident

This type of SOT is caused by Epithelial Rests of Malassez (ERMS) [2,8,12] The Hertwig epithelial root sheath, which is a part of ERMS, plays an important role in embryonic dental expansion. ERMs are found in the periodontal ligament and continue to exist even after tooth formation. They proliferate when activated and play an important role in the formation of odontogenic cysts and tumors such as radicular cysts and, in this case, SOT. Although genetic and immunohistochemical approaches are being used to research the pathways that lead to SOT formation, the triggers that cause the pathological transformation of ERMs remain unknown. [13,14] Rare peripheral SOTs are caused by remnants of the dental lamina and gingival epithelium and are usually associated with tooth germs or impacted teeth. [10,15].

SOTs can present with symptoms or as an incidentaloma. Erythema of the overlying gingiva, increased periodontal pocket depth, swelling of the alveolar process, mobile teeth, tooth displacement, erosion of the alveolar bone, and mild to moderate pain are some of the symptoms. [4,16,17] These symptoms, which vary from case, to case may cause origin of lesion.

SOT appears on radiographs as a unilocular radiolucent region with a triangular or semi-circular shape between or along the roots of neighboring teeth. The defect may also have sclerotic boundaries. [16] It should be emphasized that the radiological results are

often regarded as vague. A radicular cyst or a localized periodontal bone loss to a dentigerous cyst, an odontogenic keratocyst, an ameloblastoma, or even a hematological disease such as Langerhans cell histiocytosis, might be the differential diagnosis on radiography. [16,18]

The sole gold standard for SOT diagnosis is the correct histological interpretation. A SOT comprises of well-defined Squamous Epithelial islands with mature fibrous connective tissue. The islands are scattered uniformly and are easily distinguished from the surrounding stream, and may vary in their size and shape. Vacuolization, calcification, and expansion of microcysts also occur [3,5]. SOT with oral squamous cell carcinoma and acanthomatous or desmoplastic ameloblastoma variations are commonly misdiagnosed [2,15,17,18, 19].

Cellular atypia, mitosis, and keratin production are not observed in SOT but are prevalent in SCC, [17] and are frequently used to distinguish SOT from squamous cell carcinoma. The peripheral cells of the islands should be thoroughly studied in order to differentiate ameloblastoma from SOT. The peripheral cell layer with tightly packed columnar cells and nuclei polarization from the basement membrane is palisaded in the ameloblastoma. This peripheral cell layer is not present in SOT, and it is generally flattened [2,7,17].

Table 1. Total 27 reported cases of Squamous Odontogenic Tumor (SOT) of maxilla in literature [20]

Case	Age (yrs.)	Gender/Race	Site	Features associated	Radiographic Presentations	Treatment plan	Recurrence	FU (mo)	Reference (year)
1.	23	F/AA	Multifocal maxilla & mandible	Loose teeth	Irregular RL, 4.0 cm (L mandible)	Excision, extraction Excision, complete Odontectomy	Numerous None	57	
2.	11	M/C	Anterior Maxilla	Painless swelling, unerupted canine, expansion of buccal and palatal cortical plates Loose teeth	RL with indistinct borders, 4.0 cm	Excision & curettage, extraction	None	60	Pullon et al [9]
3.	31	F/C	Anterior Maxilla	Loose teeth	Root divergence & missing labial Plate	Surgical excision	None	12	
4.	42	F/C	Anterior Maxilla	Loose teeth, severe periodontal bone loss	NR	Surgical excision	None	60	
5.	26	M/AA	Anterior Maxilla	Loose teeth, pain Osteolytic lesion with destruction of labial cortical plate	NR	En bloc resection, Extraction	None	Lost to FUP	Doyle et al (1977)
6.	65	M/AA	R posterior Maxilla	Painless swelling	Multilocular RL	Partial maxillectomy, Extraction	None	7	
7.	26	F/AA	All 4 quadrants, extending into soft tissues	Pain, mobility and sensitivity to percussion, destruction of facial alveolar bone	Severe bone loss in all four quadrants	Excision with radical alveolectomy and alveoloplasty, total odontectomy	Stable lesions	12	McNeill et al (1980)t
8.	22	F/C	R maxilla, L posterior Mandible	Loose teeth, bone loss, pressure sensation, destruction of cortical plates	Large, diffuse RL with indistinct borders	Modified hemi maxillectomy. Excision and enucleation of mandibular lesion, extraction	None	3	Hopper et al (1980)
9.	66	F/C	L maxilla	Painless swelling, loose teeth, perforation of buccal plate	Large triangular RL extending to maxillary antrum, root resorption	Local excision, Extraction	None	NR	Carr et al (1981)
10.	30	F/C	R posterior maxilla	Loose tooth	RL, 2.0 x 3.0 cm	Curettage, extraction	NR	NR	Goldblatt et al [21]

Case	Age (yrs.)	Gender/Race	Site	Features associated	Radiographic Presentations	Treatment plan	Recurrence	FU (mo)	Reference (year)
11.	26	M/AA	Bilateral maxilla	Acute pain & swelling, impacted 3 rd molars	Well-defined RL	Surgical excision, extraction	NR	Died 1yr later	Norris et al (1984)
12.	61	M/NR	Anterior maxilla	Constant intense pain	Osteolytic lesion involving anterior 2/3 rd palate	Hemi-maxillectomy	None	84	Kristensen et al (1985)
13.	26	M/AA	L posterior mandible, Maxilla	Asymptomatic	Ill-defined RL 1.0 x 1.2cm	Mandible: en bloc resection, extraction Maxilla: Surgical excision, extraction	None	28	Mils et al (1986)
14.	29	M/AA	Multifocal maxilla & mandible	Severe bone loss	NR	Thorough curettage, osseous recontouring, extraction	NR	NR	Leider et al (1989)
15.	25	M/AA	Multifocal maxilla & mandible	Slightly expansile	RL, 2.0cm	Curettage, total maxillary odontectomy	None	48	Yaacob (1990)
16.	39	M/A	R maxilla	Asymptomatic, firm swelling on palate	Hemispherical RL with a sclerotic border	Excision	None	48	
17.	46	M/NR	R maxilla	Asymptomatic Enlargement of maxilla, loose teeth	Poorly-defined RL	Excision	None	84	
18.	42	F/A	L maxilla	Painful swelling, unerupted premolar	Well-defined Unilocular RL	Surgical excision, extraction	None	20	Baden et al [13] Kusama et al (1998)
19.	43	M/AA	L maxilla	Associated with impacted canine	RL with severe bone loss, root divergence	Enucleation	None	18	Haghighat et al [12]
20.	15	M/NR	L Anterior Maxilla	Painless swelling, impacted permanent canine	Large, well-circumscribed, unilocular triangular RL	Enucleation, Extraction	None	6	Krithika et al (2007)
21.	37	F/NR	L Anterior Maxilla	Painless swelling	Large, well-circumscribed, unilocular triangular RL	Local excision	NR	NR	Tamgadge et al (2007)
22.	9	M/NR	L maxilla	Painful swelling of gingivae, loose teeth	Well-defined unilocular RL, 2.5cm	Initial curettage Chemotherapy & radical surgery for recurrent lesion	Yes None	12 84	Ruhin et al [22]
23.	24	M/NR	R maxilla	Peri-radicular associated	Circumscribed	Through curettage,	NR	NR	Jones et al

Case	Age (yrs.)	Gender/Race	Site	Features associated	Radiographic Presentations	Treatment plan	Recurrence	FU (mo)	Reference (year)
				canine	triangular RL root divergence	extraction			[19]
24.	15	F/NR	R anterior maxilla	Loose teeth, mild expansion	Ill-defined area	NR	NR	NR	Moussa & ElShafei [4]
25.	43	M/AA	Multifocal All 4 quadrants	Impacted 2 nd and 3 rd molars	2 Multilocular & 4 Unilocular RL, 2 wedge-shaped RL, corticated Margins	Excision and extensive curettage, peripheral ostectomy, extraction	-	Lost to FU	Elmuradi et al [2]
26.	29	F/NR	Multifocal Ant maxilla, R ant mandible	Painless swelling, pressure sensation in mandible	Triangular RL, root divergence, erosion of cortical bone, hypoesthesia of right mental region	Initial surgical enucleation & aggressive curettage Resection of mandible alveolar process & segmental osteotomy, curettage	Yes (mandible)	6	Verhelst et al (2017)
27.	29	F/A	POSTERIOR MAXILLA	Painful swelling, pressure sensation in mandible	Triangular RL, Well-defined unilocular	Excision and extensive curettage.	None None	NR 24	Present case

*AA, African-American; A, Asian; C, Caucasian; FU, follow up; L, left; NR, not reported; R, right; RL, radiolucency; RO, radiopaque; T, Turkish; TTP, tender to percussion

Conservative surgical excision is recommended as a treatment for SOT and includes curettage, local excision, or enucleation. Removal of tooth along with x-ray translucency gives enough access to excise the full lesion. Most authors and the WHO state that the low chance of recurrence, especially when dealing with mandibular lesions, might also lead to conservative operative abduction. [1-3,11,17,18] In addition, certain SOTs are aggressive in nature. The cortical bone degradation is seen in the mandible or in a large part of the maxilla. [4,5,9,10,23] Early recurrence should even be regarded as an aggressive feature. Literature has described cases of malignant transition into intraosseous squamous cell carcinoma. 6 It is advisable to use a more radically-blocked strategy for excision and rebuilding in recurring instances, or in lesions of initial aggression.

In our case, we have approached more conservatively, preserving the tooth and infraorbital nerve. Complete local surgical excision with thorough curettage was performed under local anesthesia, making it more economical and comfortable for the patient. With no post-operative complications as seen after general anesthesia. The majority of cases were performed under general anaesthesia, which is more costly, unpleasant, and burdensome for the patients and family members, according to the literature. The Patient is under regular follow up till date and no evidence of recurrence has been found.

4. CONCLUSION

SOT can be a rare benign, variable-like epithelial odontogenic tumor, ranging from asymptomatic to severe bone damage. Conservative surgery, conventional treatment of SOT and its benign biological nature, has been advocated traditionally for the minimal chance of recurrence. SOT can also display aggressive biological characteristics in wide-ranging or multifocal instances.

CONSENT AND ETHICAL APPROVAL

As per international standard guideline participant consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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