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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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. It's 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 Anterior Maxilla | Loose teeth | Irregular RL, 4.0 cm (L mandible) | Excision, extraction Excision, complete Odontectomy | Numerous | 57 |  |
| 2.   | 11         | M/C         | Anterior Maxilla | Painless swelling, unerupted canine, expansion of buccal and palatal cortical plates | Loose teeth | Root divergence & missing labial Plate | Surgical excision | 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 |  |
| 6.   | 65         | M/AA        | R posterior Maxilla All 4 quadrants, extending into soft tissues Painless swelling | | Multiocular RL | Partial maxillectomy, Extraction | None | 7 | Doyle et al (1977) |
| 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) |
| 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 teeth | 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 3rd molars, Constant intense pain | Well-defined RL | Surgical excision, extraction | NR | Died 1yr later 84 | Norris et al (1984) |
| 12.  | 61         | M/NR        | Anterior maxilla | Constant intense pain | Osteolytic lesion involving anterior 2/3rd palate | Hemi-maxillectomy | None | 28      | 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 | Mils et al (1986) |
| 14.  | 29         | M/AA        | Multifocal maxilla & mandible | Severe bone loss | NR | Thorough curettage, osseous recontouring, extraction | NR | NR | |
| 15.  | 25         | M/AA        | Multifocal maxilla & mandible | Slightly expansile RL, 2.0cm | Slightly expansile | Curettage, total maxillary odontectomy | None | 48      | Leider et al (1989) |
| 16.  | 39         | M/A         | R maxilla | Asymptomatic, firm swelling on palate | Hemispherical RL with a sclerotic border | Excision | None | 48      | Yaacob (1990) |
| 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 | Unilocular RL | Surgical excision, extraction Enucleation | None | 20      | Baden et al [13] Kusama et al (1998) Haghighat et al [12] |
| 19.  | 43         | M/AA        | L maxilla | Associated with impacted canine | Associated with impacted canine | Excision | None | 18      | |
| 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 | 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 | 12      | Ruhin et al [22] |
| 23.  | 24         | M/NR        | R maxilla | Peri-radicular associated | Circumscribed | Through curettage | None | 84      | NR | Jones et al |
| Case | Age (yrs.) | Gender/Race | Site | Features associated | Radiographic Presentations | Treatment plan | Recurrence | FU (mo) | Reference (year) |
|------|------------|-------------|------|---------------------|----------------------------|----------------|------------|---------|-----------------|
| 24.  | 15         | F/NR        | R anterior maxilla | canine | triangular RL root divergence III-defined area | extraction | NR         | NR      | NR              |
| 25.  | 43         | M/AA        | Multifocal All 4 quadrants | Loose teeth, mild expansion | Ill-defined area | NR          | NR         | Lost to FU | Elmuradi et al [2] |
| 26.  | 29         | F/NR        | Multifocal Ant maxilla, R ant mandible | Impacted 2nd and 3rd molars | 2 Multilocular & 4 Unilocular RL, 2 wedge-shaped RL, corticated Margins | Excision and extensive curettage, peripheral ostectomy, extraction | None       | 6       | Verhelst et al (2017) |
| 27.  | 29         | F/A         | POSTERIOR MAXILLA | Painless swelling, pressure sensation in mandible | Triangle RL, root divergence, erosion of cortical bone, hypoesthesia of right mental region | Initial surgical enucleation & aggressive curettage | None       | None    | None (mandible)   |

*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.

REFERENCES

1. Mohr B, Winter J, Wahl G, Janska E: Recurrent squamous odontogenic tumor: A case report and review of the literature. Oncol Lett. 2015;10:2713.
2. Elmuradi S, Mair Y, Suresh L, DeSantis J, Neiders M, Aguirre A: Multicentric Squamous Odontogenic Tumor: A Case Report and Review of the Literature. Head Neck Pathol 2017;11:168.
3. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ. World health organization classification of head and neck tumours. 4th edition.
4. Moussa M, EIShafei MM: Squamous odontogenic tumor: A case report. Int J Case Reports Images. 2013;4:607.
5. 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.
6. Slootweg PJ. Dental pathology: A practical introduction. 1st ed. (Schröder G, ed.). Heidelberg: Springer Berlin Heidelberg;2007.
7. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, eds. Geneva: WHO Press, 2017.
8. 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.
9. Pullon PA, Shafer WG, Elzay RP, Kerr DA, Corio RL: Squamous odontogenic tumor. Oral Surgery, Oral Med Oral Pathol. 1975;40:616.
10. Emmanuel RV, Neelakantan S, Nair PP, Thomas S, Saxena V, Yadav NS: A harmless but confusing tumour on the anterior maxilla. BMJ Case Rep. 2012;2012: bcr20120015394.
11. Barrios TJ, Sudol JC, Cleveland DB: Squamous odontogenic tumor associated with an erupting maxillary canine: case report. J Oral Maxillofac Surg. 2004;62:742.
12. Haghhighat K, Kalmar JR, Mariotti AJ. Squamous Odontogenic Tumor:
13. Baden E, Doyle J, Mesa M, Fabié M, Lederman D, Eichen M: Squamous odontogenic tumor. Oral Surgery, Oral Med Oral Pathol. 1993;75:733.

14. Keinan D, Cohen RE: The significance of epithelial rests of malassez in the periodontal ligament. J Endod. 2013;39:582.

15. Siar C, Nakano K, Ng K, Tomida M, Nagatsuaka H, Kawakami T: Squamous odontogenic tumor of the mandible: a case report demonstrating immune expression of Notch1, 3, 4, Jagged1 and delta1. Eur J Med Res. 2010;15:180.

16. Singh A, Agarwal N, Sinha A, Singh G, Srivastava S, Prasad RK: Squamous odontogenic tumor of the maxilla: a case report and review of the literature. Oral Radiol. 2015;31:129.

17. Mardones N do R, Gamba T de O, Flores IL, Almeida SM de, Lopes SLP de C: Squamous Odontogenic Tumor: Literature Review Focusing on the Radiographic Features and Differential Diagnosis. Open Dent J. 2015;9:154.

18. Lin Y-L, White DK: Squamous odontogenic tumor. Oral Maxillofac Surg Clin North Am. 2004;16:355.

19. Jones BE, Sarathy AP, Belinda Ramos M, Foss RD: Squamous Odontogenic Tumor. Head Neck Pathol. 2011;5:17.

20. Upadhyaya JD, Banasser A, Cohen DM, Kashtwary D, Bhattacharyya I, Islam MN: Squamous Odontogenic Tumor: Review of the Literature and Report of a New Case. J Oral Maxillofac Surg. 2021;79(1):164-176.

21. Goldblatt LI, 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.

22. Ruhin B, Raoul G, Kolb F, Casiraghi O, Lecomte-Houcke M, Ghoul S, Auriol M, Ferri J: Aggressive maxillary squamous odontogenic tumour in a child: histological dilemma and adaptive surgical behaviour. Int J Oral Maxillofac Surg. 2007;36:864.

23. Favia GF, Alberti LD, Scarano A, Piattelli A: Squamous odontogenic tumour: Report of two cases. Oral Oncol. 1997;33:451.