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SQUAMOUS ODONTOGENIC TUMOR: A RARE PRESENTATION

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ABSTRACT

Squamous odontogenic tumor, ever since it has been first reported in the literature, remains to be a very rare odontogenic lesion. Pullon.et.al was the first to report a case of Squamous odontogenic tumor in 1975. Squamous odontogenic tumor is defined as a very rare benign neoplasm, locally infiltrative lesion which can extend to neighboring structures. The tumor generally presents itself as painless lesion but there cases where it has been seen associated with pain and tooth mobility. Squamous odontogenic tumor is usually manifested in anterior maxilla and posterior mandible. Here wepresent an atypical case of squamous odontogenic tumor of the posterior region of maxilla in a young female patient.

KEYWORDS: Odontogenic, Rare, Semicircular, Tumor, Unilocular.

INTRODUCTION

Squamous odontogenic tumor was first reported in the literature when Pullon and his co workers published six of cases in 1975.[1] World Health Organization in 2005 classified Squamous odontogenic tumor as an epithelial odontogenic tumor with around 50 cases

reported in the English-language literature till date.[2,3] Before this it was considered as an atopic acantomatous ameloblastoma or a squamous cell carcinoma. The pathogenesis of SOT is still unclear. Remnants of dental lamina (rests of Serres), epithelial rests of Malassez or gingival epithelium are the main suspected origin.[2,4]

Here we are reporting one such case of Squamous odontogenic tumor with a rare clinical presentation and also a brief outlook of review of literature.

CASE REPORT

A 18 year old female patient reported to our department of Oral Medicine and Radiology with the complaint of bleeding gums in upper left back tooth region for past 2 weeks. Patient noticed bleeding 2 weeks back. Bleeding was sudden in onset, intermittent in nature and was associated with dull aching, intermittent, localized type of pain. No aggravating and relieving factors were reported. No history of any trauma, swelling, fever or any such previous episodes. Patient consulted dentist in her locality for her complaint. Radiograph was advised & scaling was done. On radiographic examination the dentist noticed the extensive bone loss and advised her graft for which she has consulted our hospital for a second opinion. The patient's medical history was non contributory. Her sibling was diagnosed to have Squamous cell carcinoma of tongue 3 months back.

Patient's general and extra-oral examination was not contributory. On intra-oral examination a bluish discolouration was seen on the attached gingiva of palatal aspect of 25 "Figure 1". No evidence of any caries was present. On palpation gingiva was soft and edematous. Periodontal pocket of approximately 6mm was present. Tenderness on palpation was present in relation to 25. Tenderness could also be elicited in relation to the interdental aspect of 24 and 25. There was no mobility of associated teeth.

Taking into account the history and clinical examination a provisional diagnosis of of Localised periodontitis and hematoma of 25 was given. As part of investigation IOPAR and RVG of left maxillary posterior region was taken. IOPAR of 24, 25 reveals severe interdental bone loss and displaced teeth "Figure 2". RVG revealed a unicystic, semicircular completely radiolucent region superimposed in the interradicular region of 24 & 25 "Figure 3". CBCT reveals a semi-circular hypodense area of size approximately 1×1 cm in inter radicular aspect of 24 and 25 "Figure 4". Sagittal section reveals that hypodensearea was seen to diverge the teeth "Figure 5". Based upon the radiographic findings a radiographic differential diagnosis

of Lateral periodontal cyst, Lateral radicular cyst and lately Squamous odontogenic tumor were considered “Figure 6”.

The lesion was completely excised and sent for histopathological examination. Histopathological report revealed cellular fibrous connective tissue stroma showing areas of hyalinization. Numerous islands of odontogenic epithelium (odontogenic rests) were noticed within the connective tissue stroma. Larger islands of predominantly squamous cells with rich eosinophilic cytoplasm were also present, which suggested SOT “Figure 7”.
Thus on the basis of histopathological report, a diagnosis of Squamous Odontogenic tumor was given. The healing was uneventful without any swellings or other complications. The patient is under evaluation and one month post operative clinical and radiograph has been included “Figure 8 & 9”.

DISCUSSION

Squamous odontogenic tumor is a rare benign entity of jaws with only few cases reported in literature. There has been only 50 cases of Squamous odontogenic tumor published in literature.[5] Squamous odontogenic tumors have been found in patients whose ages ranged from 8 to 74 years (average age 38). Badni.et.al found that the gender ratio among 44 cases is 1: 1.8 (F :M) showing slightly a male predilection.[2] This is not in correlation with our case where the patient was female and in her second decade. Leider et al. reported three cases of SOTs in siblings, which suggests a possible familial pattern in the occurrence of this lesion.[6] Present case showed no familial occurrence.

Considering the reported cases, the most common location for development of SOT is the maxillary anterior region andmandibularposterior region.[7] In our case the lesion was located in the posterior region of maxilla which is a rare site of occurrence with only approximately four to five cases published in literature to the best of our knowledge. It has been stated that SOTs occurring in maxilla were found to be more aggressive than in mandible which is mainly due to the anatomy and porous nature of bone.[7,8] But in the present case the lesion was not seen invading into the sinus may be due to it’s early detection.

Clinically, SOT appears as a slow growing lesion, which leads to an increase in the volume of the maxilla or mandible, tooth mobility, ulceration of the soft tissue, painful symptoms, and tooth displacement. However, lesion may sometimes be asymptomatic and detected only on routine radiographic investigations.[9] This stands very true for our case as patient’s only

complaint was bleeding gums and dull pain. The bluish discolouration could be associated with the fish bone injury.

Radiographically, common variant of SOT shows a well-defined unilocular, triangular radiolucency between the roots of adjacent teeth which can be appreciated in this case.[10] This is not pathognomonic and can mimic severe periodontal bone loss. Circumscription is characteristic, but the margin may or may not be corticated. An ill defined radiographic margin can suggest a more aggressive process.[11] In the present case there was a well defined border which explains the reason why a conservative treatment was done. The peripheral variant may show saucerization of underlying bone which is likely to be a pressure phenomenon rather than the result of true tumor infiltration.[12]

Histologically, the lesion usually presents as islands of benign squamous epithelium in mature connective tissue stroma without the evidence of peripheral columnar cells, palisading nuclei, or stellate reticulum.[13] Cystic degeneration in the center of the islands is a frequent finding. In some epithelial cells prekeratin is present and laminated calcifications may be seen inside keratin pearls.[14,15,16] Clinico- pathologically, three main types are identified: Intraosseous, Mural and Extraosseous forms. Intraosseous or central type is more common. Mural type presents as SOT like proliferation in the wall of cyst. Extraosseous or peripheral type is rare and only one case has been reported in the literature.[17]

Treatment of SOT consists of conservative surgical removal (local excision), enucleation and/or complete curettage.[16] However, more aggressive intervention may be necessary in the case of tumors located in the maxilla due to the aggressive potential of the lesion at this site. Complete excision of lesion was done and a aggressive approach was not considered as it was not in need. Recurrence is rarely reported in the literature and is attributed to incomplete removal of the initial tumor. Only one case of recurrence was observed among the six cases evaluated by Pullon et al.[16] To avoid such circumstances in our case the patient is under regular evaluation till date.

CONCLUSION

Even after three decades since it has been reported Squamous odontogenic tumor still remains to be rare benign tumor due to the paucity of cases reported. With this case we would like to report a unique presentation of Squamous odontogenic tumor due to it's site and

gender predilection. This in turn emphasizes more the fact that each patient is unique requiring inimitable analysis.

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