

Squamous Odontogenic Tumor: A Rare Case Report

Pratibha Poudel¹, Barsha Bajracharya², Prof. S Bhattacharyya³, Ritesh Giri⁴, Dipshikha Bajracharya⁵

^{1,2}Resident, ^{3,4}Professor and Head, ⁵Assistant Professor

^{1,2,3,4}Department of Oral Pathology, Microbiology and Forensic Odontology, Kantipur Dental College Teaching Hospital and Research center

ABSTRACT

Squamous odontogenic tumor is a rare benign locally infiltrative neoplasm first described by Pullon et al. (1975). Since then, there have been less than 50 cases reported. The tumor is often asymptomatic, although it can present with symptoms of pain and tooth mobility. The characteristic radiographic appearance is that of a triangular-shaped unilocular radiolucency with divergent roots of the involved teeth. Histologically, the tumor is characterized by the proliferation of benign appearing, squamous epithelium in the connective tissue stroma with occasional vacuolization and keratinization. Here, we report a rare case of Squamous odontogenic tumor in anterior mandible of a 40 years old female patient.

Key words: odontogenic tumor, Squamous epithelium

INTRODUCTION

Squamous odontogenic tumor (SOT) is a rare benign neoplasm first described in 1975 by Pullon et al.¹ It arises from the neoplastic transformation of epithelial rests of Malassez. The World Health Organization (WHO) in 2005 classified it as an epithelial odontogenic tumor with around 50 cases reported in the literature.²

SOT is defined as a benign but locally infiltrative neoplasm. It affects a wide age range showing a slight male preponderance and occurs more frequently in the mandible with preference for posterior mandible and anterior maxilla.² Maxillary lesions appear to be more aggressive in biologic behavior than those occurring in mandible.³ Radiographically, SOT is seen as a unilocular radiolucent triangular shaped area whose base is localized at the apices of the diverging adjacent roots.⁴ Histologically the lesion consists of islands of squamous epithelium in a fibrous stroma. Cystic degeneration or calcification may occasionally be observed in the epithelial islands.⁵ Clinico- pathologically, three main types are identified: Intraosseous, Mural and Extraosseous forms.¹ 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.¹

CASE REPORTS

A 40 years old female patient reported with the complain of an asymptomatic swelling on the right side of mandible since 4 months. On clinical examination hard swelling of about 1.5 x1.5 cm was seen on buccal aspect of lower anterior region wrt 42, 43 and 44. The overlying mucosa was normal. On radiographic examination (Fig.1), areas of mixed radiolucency and radiopacities were seen wrt 42 and 43 and the roots of the same were divergent. On the basis of clinical and radiographical findings, clinical differential diagnosis were made of Ossifying fibroma or Calcifying epithelial odontogenic tumor. The incisional biopsy of the lesion was taken (Fig.2 & Fig.3) and sent for histopathological examination. Histological examination revealed proliferation of mature stratified squamous epithelial islands in a dense fibrous connective tissue stroma (Fig.4) These islands showed peripheral flat or cuboidal shaped cells and central squamous cells with intercellular bridges (Fig.5).

Correspondence: *Dr.Dipshikha Bajracharya*; e-mail: *drdipshikhabaj@gmail.com*

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Areas of cystic degeneration and keratinization were seen within these islands (Fig.6). Thus depending on clinical and histological features the final diagnosis of SOT was confirmed.

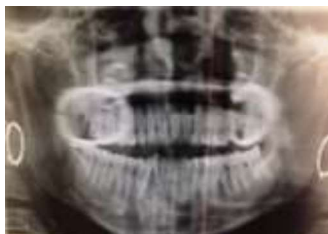


Fig 1: Panoramic radiograph reveals areas of mixed radiolucency and radiopacities involving roots of mandibular lateral incisor and canine.



Fig 2: Intraoperative view



Fig 3: Incisional biopsy of the lesion



Fig 4: Photomicrograph showing scattered islands of squamous epithelium in dense fibrous connective tissue stroma (4x magnification)

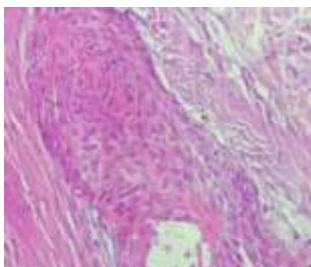


Fig 5: Photomicrograph showing peripheral flat or cuboidal shaped cells and central squamous cells with intercellular bridges (40x magnification)

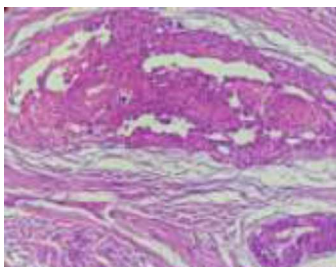


Fig 6: Photomicrograph showing microcystic degeneration and keratinization (10x magnification)

DISCUSSION

Squamous odontogenic tumor is a rare benign neoplasm with very few cases reported in literature till date. Pullon et al in 1975 reported six cases and described SOT for the first time. They established the diagnostic criteria and surgical approaches that are still

followed today.⁶ Before 1975, this lesion was believed to represent an atypical acanthomatous ameloblastoma or even a squamous cell carcinoma.⁷

Most of the cases reported in literature have been found to be located within the bone (intraosseous), although a few peripheral cases have also been discussed.⁸ The central or intraosseous variety is thought to arise from the rests of Malassez or derived from the periodontal ligament whereas the peripheral variety is thought to arise from the gingival surface epithelium or the rests of Serres.⁸

SOT has been found in wide age range, from first decade to eighth decade of life, with mean age of occurrence being 38.2 years. Badni et al reviewed 44 cases of SOT and found slightly more predilection among males compared to females (F: M=1:1.8).⁴ Considering the reported cases, the most common location for development of SOT in the maxilla is anterior region and in case of mandible is posterior region.⁹ In our case the lesion was located in the anterior region of mandible which is in consistency with the finding of Bansal et al.⁵ 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.^{9,10}

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.¹¹

Radiographically, common central variant of SOT shows a well-defined unilocular, triangular radiolucency between the roots of adjacent teeth.¹² but in our case areas of mixed radiolucency and radiopacity was seen. 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.³

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} Sometimes the morphologic

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appearance of the islands of odontogenic epithelium in SOT is similar to the follicular pattern of ameloblastoma, which may lead to the misdiagnosis of the tumor. However, lack of polarization of peripheral cells in the epithelial islands, which is typical of ameloblastoma, favors the diagnosis of SOT.¹⁷ In our case, squamous epithelial islands in mature connective tissue stroma, presence of intercellular bridges and areas of microcystic degeneration and absence of ameloblasts like cells at periphery was of major importance in establishing the diagnosis. Newer diagnostic techniques such as immunohistochemistry has shown expression of keratin and Notch receptor (Notch 1,3,4) and its ligand (jagged 1 and delta 1) in SOT.¹ However, routine eosin and hematoxylin technique remains the gold standard for the diagnosis of SOT.

SOT can be managed conservatively by local excision, curettage, enucleation and scaling of the teeth. In case of maxilla, since the lesion is more aggressive, the lesion has also been treated surgically by en bloc excision and hemimaxillectomy.^{8,18,19}

Recurrence of the lesion is rare and only a single case of recurrence has been reported in the literature, which may be due to incomplete removal of the lesion. Local curettage or extraction of adjacent teeth was found to be effective in management of recurrences.¹⁹

Ide *et al.* in 1999 reported malignant transformation of SOT such as Intraosseous Squamous Cell Carcinoma.²⁰ Kim *et al.* in 2007 reported two cases of SOT that led to erosion of lingual cortical plate of mandible, which may impact the therapeutic modalities and management of these lesions. This indicates that these lesions do not always behave similarly.²¹

CONCLUSION

Rare as it may seem since its first description till date SOT has always been an enigma in the field of dentistry. Hence analysis of each case clinically, radiographically and histologically is important in diagnosing the disease and choosing the right treatment modality.

REFERENCES

1. Siar H, H K. Squamous odontogenic tumor of the mandible: a case report demonstrating immunoexpression of notch1, 3, 4, jagged1 and delta1. *Eur J Med Res.* 2010;15:180–4.

2. Mardones NDR, Gamba TDO, Flores IL, de Almeida SM, Lopes SLPDC. Squamous Odontogenic Tumor: Literature Review Focusing on the Radiographic Features and Differential Diagnosis. *Open Dent J.* 2015 Jan;9:154–8.
3. Reichart PA, Philipsen HP. Squamous odontogenic tumor. *J oral Pathol Med.* 1990;19(5):226–8.
4. Badni M, Nagaraja A, Kamath V. Squamous odontogenic tumor: A case report and review of literature. *J Oral Maxillofac Pathol.* 2012 Jan;16(1):113–7.
5. Bansal S, Joshi SK. Case Report Squamous Odontogenic Tumor with Unusual Localization and Appearance : A Rare Case Report. *Case Rep Med.* 2013;2013:1–4.
6. Pullon PA, Shafer WG, Elzay RP, Kerr DA, Corio RL. Squamous odontogenic tumor: report of six cases of a previously undescribed lesion. *Oral Surgery, Oral Med Oral Pathol.* 1975;40(5):616–30.
7. Neville BW, Damm DD, Allen CM, Bouquet JE. Odontogenic cysts and tumors. *Oral Maxillofac Pathol.* 2002;3:683–7.
8. 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.* 2014 Jul 15;31(2):129–34.
9. Moussa M, ElShafei MM. Squamous odontogenic tumor: A case report. *Int J Case Reports Images.* 2013;4(11):607.
10. Pant H, Pathak S. Case Report : Squamous odontogenic tumor-exceedingly rare neoplasm. *Indian Journal of Basic & Applied Medical Research.* 2013;(8):1023–6.
11. Odontologia F De, Alegre P, Federal U, Maria DS. Squamous odontogenic tumor : with recurrence and 12 years of follow-up. *Rev Ciênc Méd, Campinas.* 2007;16(1):51–6.
12. Barrios TJ, Sudol JC, Cleveland DB. Squamous odontogenic tumor associated with an erupting maxillary canine: case report. *J oral Maxillofac Surg.* 2004;62(6):742–4.
13. Favia GF, Di Alberti L, Scarano A, Piattelli A. Squamous odontogenic tumour: report of two cases. *Oral Oncol.* 1997;33(6):451–3.

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14. Doyle JL, Grodjesk JE, Dolinsky HB, Rafel SS. Squamous odontogenic tumor: report of three cases. *J oral Surg.* 1977;35(12):994.
15. Goldblatt LI, Brannon RB, Ellis GL. Squamous odontogenic tumor: report of five cases and review of the literature. *Oral Surgery, Oral Med Oral Pathol.* 1982;54(2):187–96.
16. Haghghat K, Kalmar JR, Mariotti AJ. Squamous odontogenic tumor: diagnosis and management. *J Periodontol. Am Acad Periodontology;* 2002;73(6):653–6.
17. Philipsen HP, Reichart PA. Squamous odontogenic tumor (SOT): a benign neoplasm of the periodontium. *J Clin Periodontol.* 1996;23(10):922–6.
18. Kristensen S, Andersen J, Jacobsen P. Squamous odontogenic tumour: review of the literature and a new case. *J Laryngol Otol.* Cambridge Univ Press; 1985;99(09):919–24.
19. Mills WP, Davila MA, Beuttenmuller EA, Koudelka BM. Squamous odontogenic tumor: report of a case with lesions in three quadrants. *Oral Surgery, Oral Med Oral Pathol.* 1986;61(6):557–63.
20. 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(4):431–4.
21. Kim J-Y, Kim J-C, Cho B-O, Kim S-G, Yang B-E, Rataru H. Squamous odontogenic tumor: a case report and review of literatures. *J Korean Assoc Oral Maxillofac Surg.* 2007;33(1):59–62.