Case Report:

Squamous odontogenic tumor-exceedingly rare neoplasm

***Dr Hema Pant, Dr Sachin Pathak**

Name of the Institute/college: SRMS Institute of Medical Sciences, Bareilly, UP, India

***Corresponding author:** Dr Hema Pant : Email id : hemaufht@yahoo.com

Abstract :

Squamous odontogenic tumor is exceedingly rare, benign neoplasm of the jaws, originating from the rests of Malassez and less than 50 cases have been reported. Here we report this tumor in young female involving anterior part of mandible. Very few cases have been published till now in literature, here we present a case of squamous odontogenic tumor arising in the anterior portion of mandible, which itself is a rare site for this tumor. .

Keywords : Squamous Odontogenic tumor

Introduction

Squamous odontogenic tumor is a lesion which has been recognized as apparent entity for a number of years but had not been named or reported until 1975, when Pullon and a small group of other oral pathologists combined their cases and published six cases. It is thought to arise from a neoplastic change of epithelial rests of Malassez[7]. The most important aspect of this lesion is its mistaken histologic identification as an acanthomatous ameloblastoma or as a desmoplastic ameloblastoma[2] . Very few cases have been published till now in literature, here we present a case of squamous odontogenic tumor arising in the anterior portion of mandible, which itself is a rare site for this tumor. .

Case report

A 19 year old female reported at outpatient department of dentistry with chief complaint of swelling and pain in lower mandible of 3 months duration. The patient was moderately nourished with an unremarkable medical history.

Physical examination revealed a mild swelling over the left mandible. On radiograph, there was well defined radiolucency in mandible which was not crossing the midline. Laboratory investigation showed Hb 13.6% ,TLC-14,200/cumm, DLC , N-61%, L-32%, E07%, and all other parameters were within normal limits. A provisional diagnosis of fibro-osseous lesion was made.

Intraoperatively, a firm cystic swelling measuring about 2x3 cm was found, which was extending from the alveolar process to the half way down lower border of mandible superio-inferiorly and from left canine region to medial to the root of left mandibular first molar tooth anteroposteriorly. Expansion of the buccal cortical plate can be seen but the lingual cortical plate of mandible is not affected at all. On enucleation the lesion came out in toto leaving a bony cavity of about 2x3 cm with moderate bleeding and was sent to department of pathology. Received biopsy specimen measures 3.0cm x 2.0cm x 1.0cm, firm in consistency, irregular in shape and cut surface was lobulated. Histopathologic examination of the lesion showed

irregularly shaped islands, clusters, trabeculae and strands of monotonous squamous cells surrounded by abundant colllagenous stroma. Thus overall features were of squamous odontogenic tumor.

Discussion

Squamous odontogenic tumor is a rare lesion with few cases reported till date in the literature.

Pullon et al.[1] were the first to describe it. They reported six cases and established diagnostic criteria and surgical approaches that are still followed today. Very few cases have been reported in literature and most of these have been located within the bone, although a few peripheral cases have also been discussed. Before 1975, this lesion was probably believed to represent an atypical acanthomatous ameloblastoma or even a squamous cell carcinoma.[2] Histogenesis of squamous odontogenic tumor may be varied. Rests of Malassez are the epithelial proliferation for lesion that are associated with alveolar process adjacent to lateral root surface or the teeth, and dental lamina may be the source of the lesions that developed in association with the crown of unerupted or impacted tooth. Surface stratified squamous epithelium and rests of Serres have been cited as the sources of the extraosseous variant.[3]Squamous odontogenic tumors have been found in patients whose ages ranged from 8 to 74 years (average age 38)

From the limited number of published cases, however it is still possible to draw some conclusion regarding the usual demographic, clinical and radiographic presentation of the tumor. There is no apparent predilection for occurrence in either the mandible or in maxilla. However lesions of maxilla tend to occur more often in the anterior regions, while mandibular cases are more often found in the posterior areas. Males and females appear to be equally affected. [2] Maxillary lesions seem to grow more aggressively than do mandibular ones.[4]Squamous odontogenic tumor's clinical and radiographic features are neither unique nor sufficient for diagnosis, as this tumor may be confused with a number of other pathologies. Patients may present with an increase in the volume of the maxillae or mandible, tooth mobility, ulceration of the soft tissue, painful symptoms, and tooth displacement.[5] The squamous odontogenic tumor generally produces a readily recognizable pattern on histologic examination. The tumor consists of numerous islands of proliferative squamous epithelium dispersed uniformly in a connective tissue stroma. The islands are numerous and easily recognized, being sharply demarcated from the surrounding stroma by a flattened layer of cells at their periphery. The epithelium in these rounded islands often shows a swirling or "whirlpool" pattern to the central squamous cells. Areas of cystic change centrally within the epithelial islands are also a frequent finding and keratinization of the central cells may be present too.[9] In light of the relatively few reported cases, there are no consistently recorded clinical features of squamous odontogenic tumor and there is no gender or site predilection.[8] Diagnosis is based on recognition of the histopathologic features of squamous odontogenic tumor to obviate possible misdiagnosis. Misdiagnosis as peripheral ameloblastoma can also occurs occasionally.[10] In that lesion it is possible to observe palisaded columnar ameloblastic cells lining the epithelial islands and a stellate reticulum configuration of the central portion of the islands.

As there may be some degree of pleomorphism, hyperchromatism and occasionally an increased

number of mitotic figures, these lesions have mistakenly been diagnosed as squamous cell carcinoma by some pathologists unfamiliar with this neoplasm.[11] In squamous cell carcinoma, the epithelial connective tissue interface is ruptured, numerous epithelial islands exhibiting dysplastic features and surrounded by chronic inflammatory cells can be seen**.** But in our case such features were not visible.

Ameloblastic carcinoma shows irregular masses with interdigitating cords of epithelial cells having palisading pattern around the periphery. The tissue in the centre of the cellular islands is composed of stellate reticulum. Other areas show islands of keratinizing well-differentiated squamous cell carcinoma infiltrating the adjacent bone. In our case there was no such palisading pattern of epithelial cells was noted. Thus ameloblastic carcinoma was ruled out.[1,12,13]

In our lesion, semi-serial sections revealed a typical pattern of squamous odontogenic tumor in most parts. Thus, the most appropriate interpretation for this lesion as squamous odontogenic tumor was made. Squamous odontogenic tumors have some invasive capacity and infrequently recur after conservative therapy. Curettage or excision is the treatment of choice.[5]

Conclusion

Squamous odontogenic tumors is a rare odontogenic tumor with very few cases have been reported in the literature. Histopathlogically it resembles Desmoplastic Ameloblastoma and Acanthomatous Ameloblastoma and hence must be differentiated from same.

References :

1. Pullon PA, Shafer WG, Elzay RP. Squamousodontogenic tumor. Report of six cases of a previously undescribed lesion. Oral Surg Oral Med Oral Pathol.1975; 40(5):617-30.

2. Neville and Damn, Saunders, Oral and Maxillofacial Pathology, 2nd edition, , 625-626

3. Squamous odontogenic tumor: Review of literature and a new case; The Journal of Laryngology & Otology (1985), 99: 919 -924

4. Olivera,Carrard, Danesi; Rev Cierc. Squamous odontogenic tumor: with recurrence and 12 years follow up; Med., Campinas; 2007 Jan/fev.,
16(1); 51- 56,

5. Regezi, Sciubba, Jordan; Saunders (Elsevier); Oral Pathology, Clinical Pathologic Correlations; 277

6 . ; Reichart, Philipsen; Odontogenic tumors and allied lesionsQuintessance Publishing co; Pg 90 -91.

7 . Odukoya O.;Odontogenic tumors: analysis of 289Nigerian cases. J Oral Pathol Med. 1995; 24(10):454-6

8. Ruhin B, Raoul G, Kolb F, Casiraghi O, Lecomte-Houcke M, Ghoul S, et al. Aggressive maxillary squamous odontogenic tumour in a child: Histological dilemma and adaptive surgical behaviour. Int J Oral Maxillofac Surg 2007;36:8646.

9 King Kim, Sheldon M. Mintz, Jeffrey Stevens. Squamous odontogenic tumor causing erosion of the lingual cortical plate in the mandible: a report of 2 cases. J Oral Maxillofac Surg 2007;65:1227-31.

10 Barrios Tomas J., Sudol Jonathan C., Cleveland Deborah B. Squamous odontogenic tumor associated with an erupting maxillary canine: case report. Oral Maxillofac Surg 2004;62:742-4.

11 Favia GF, Di Alberti L, Scarano ZA, Piattelli A. Squamous odontogenic tumour: report of two cases. Oral Oncology 1997;33(6):451-3.

12 Ide F, Shimoyama T., Horie N, Shimizu S. Intra-osseous squamous cell carcinoma arising in association with a squamous odontogenic tumour of the mandible. Oral Oncology 1999;35:431-4.

13 Arotiba JT, Mohammed AZ, Adebola RA T Arotiba, DS Adeola, SO Ajike, AH Rafindadi: Ameloblastic carcinoma: Report of a Case. Nigerian J Surg Res 2005;7(1-2):22

Date of submission: 4 June 2013	Date of Provisional acceptance: 25 June 2013
Date of Final acceptance: 27 July 2013	Date of Publication: 04 September 2013
Source of support: Nil	Conflict of Interest: Nil