Asian Journal of Case Reports in Surgery

Volume 16, Issue 2, Page 18-23, 2022; Article no.AJCRS.95140

Squamous Odontogenic Tumor Mimicking an Intra-Osseous Malignancy: A Rare Case Report

Garima Rawat [a++*] and Hema Malini Aiyer [a#]

[a] *Department of Pathology, Dharamshila Narayana Hospital, New Delhi, India.*

Authors' contributions

This work was carried out in collaboration between both authors. Both authors read and approved the final manuscript.

Article Information

Open Peer Review History:
This journal follows the Advanced Open Peer Review policy. Identity of the Reviewers, Editor(s) and additional Reviewers, peer review comments, different versions of the manuscript, comments of the editors, etc are available here: https://www.sdiarticle5.com/review-history/95140

Case Study

Received: 17/10/2022
Accepted: 24/12/2022
Published: 26/12/2022

ABSTRACT

Aim: Squamous Odontogenic Tumor (SOT) is a rare odontogenic epithelial neoplasm, first described by Pullon et al. This lesion was classified as a distinct pathological entity by the World Health Organization (WHO) in 1992. The pathogenesis is still controversial with possible origin from epithelial rests of Malassez or rests of Serres. It is a locally infiltrative and occasionally destructive lesion.
Case Presentation: We report a case of a 34-year-old male who was referred to our centre with complaint of fracture of mandible which was subsequently diagnosed as a Squamous odontogenic tumor.
Discussion: Histologically, the tumor was characterized by islands of bland terminally differentiated squamous epithelium with keratinization or calcifications. The patient underwent resection of the lesion and is under follow up since then.
Conclusion: An extensive literature review showed that less than 50 cases have been reported worldwide with minority of cases in Indian sub-continent.

[++] *Oral & Maxillofacial Pathology;*
[#] *Professor & Head;*
**Corresponding author: E-mail: garima3103@gmail.com;*

Keywords: Mandible; squamous; odontogenic; tumors; radiograph; case report.

1. INTRODUCTION

Squamous odontogenic tumor (SOT) is an extremely rare odontogenic epithelial neoplasm. This lesion was first described as a "radiolucent lesion causing bone destruction associated with the roots of teeth" [1,2]. It was classified as a distinct pathological entity in 1992 WHO classification as a "benign epithelial odontogenic tumor with cells showing terminal squamous differentiation" [2-4]. The pathogenesis of this neoplasm is controversial. SOTs are usually slow growing; locally infiltrative lesions that can extend into neighbouring anatomical structures [5]. We report a unique and uncommon case of squamous odontogenic tumor (SOT) in a 34-year-old male who presented with a fracture of the right mandible in the lower molar region mimicking an intra-osseous malignancy.

2. CASE REPORT

A 34-year-old male presented to the outpatient department of a tertiary care centre with the chief complaint of swelling and pain in right lower back tooth region for 2 weeks. On extra-oral examination, there was tenderness near the right angle of mandible with normal overlying skin. Intra-oral examination revealed a fracture in the mandibular molar region with associated grade I/II mobility of mandibular molars. Overlying mucosa appeared erythematous with no frank ulceration. The clinical history and findings pointed towards an intra-osseous pathology. Amongst the various differential diagnosis considered, first was aggressive odontogenic tumor followed by locally aggressive neoplasm or primary intra-osseous epithelial malignancy. The patient was advised a Computed Tomography scan (CT) which showed a 3.5 cm osteolytic lesion destroying the buccal and lingual cortices of the mandible with root resorption of associated teeth (arrow in Fig. 1A, B). An incisional biopsy done outside was reported as an epithelial malignancy. The slides/blocks of this biopsy were not reviewed in our department. The clinico-radiological findings were evocative of a malignant intra-osseous neoplasm or a primary intra-osseous epithelial malignancy. The case was presented to the multi-disciplinary team and was planned for surgical resection - right hemi-mandibulectomy along with ipsilateral supra-omohyoid neck dissection (level I-III) followed by reconstruction using local flap and reconstruction plate. The soft tissue and decalcified sections from lesional tissue showed tumor composed of benign appearing islands of squamous epithelium scattered randomly in a mature fibrous and occasionally myxoid connective tissue stroma (Fig. 1A, B).

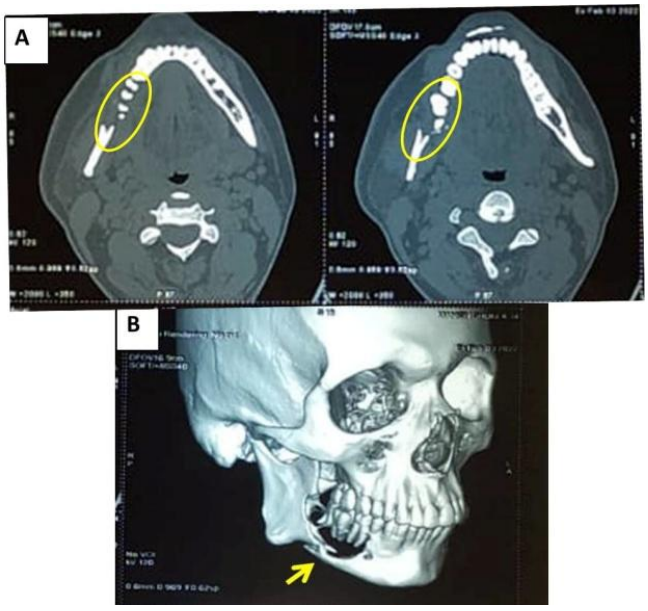

Fig. 1. A. Imaging showing radiolucency in the region of mandibular molars destroying mandible (circle); B. Reconstructed image showing destructive osteolytic lesion in the body of the mandible (arrow)

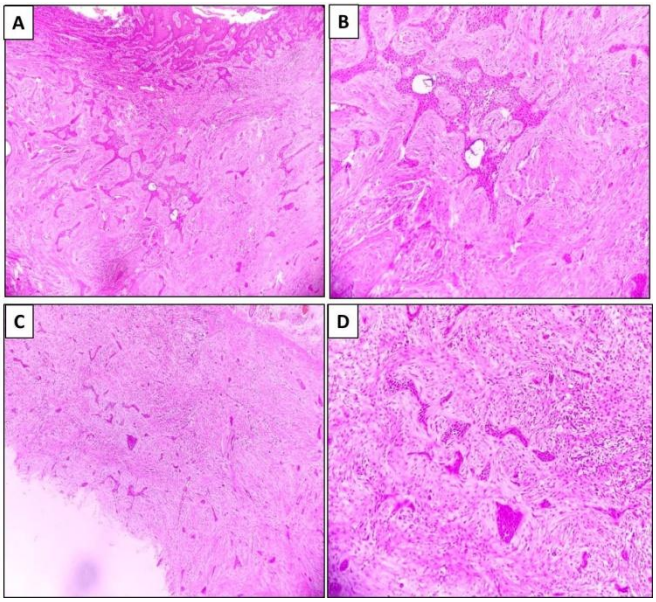

Fig. 2. A. Photomicrograph showing hyperplastic squamous epithelium with small infiltrative islands. (HE 40x); B. Photomicrograph showing islands of bland squamous epithelium with microcystic degeneration and keratinisation. (HE 100x); C. Photomicrograph showing small islands in a mature fibrous connective tissue stroma. (HE 40x); D. Photomicrograph showing variably sized islands of terminally differentiated squamous epithelium. (HE 400x)

The cellular morphology was bland with no mitotic Figs. or pleomorphism. The islands were of varying shapes and sizes with a peripheral layer of flattened cells accompanied by vacuolization and individual cell keratinization (Fig. 2A, B, C). The features were consistent with a Benign Odontogenic Epithelial tumor- favour Squamous Odontogenic Tumor. Thirty reactive lymph nodes were dissected from the neck dissection specimen. The patient was discharged uneventfully and is under follow up since then without any recurrence.

3. DISCUSSION

In the 1992 classification, WHO added Squamous Odontogenic tumor (SOT) as a distinct entity under the benign epithelial odontogenic tumor category. SOT was first reported by Pullon et al in their case series. These authors presented a previously unprecedented lesion occurring in close association with the dentition and radiographically presenting as a triangular or circular radiolucency adjacent to a tooth root [1-2].

SOTs have been reported in a wide age range with most cases affecting young to middle-aged individuals [4]. Slight male predilection has been observed in the past literature [2, 5]. Our patient was also a 34-year-old male similar to previously established data.

Amongst the previously published less than 50 cases, SOTs occur with almost equal frequency in the posterior mandible and anterior maxilla. Some case reports have reported multifocal lesions also. The maxillary lesions have a higher tendency for invading adjacent structures [1, 5-6]. Though the lesions are mostly asymptomatic, a common feature is the mobility of the involved teeth with pain and tenderness. Radiographically, this neoplasm presents as well defined unilocular, triangular radiolucency between the roots of adjacent teeth, seldom causing root resorption [2-4]. Pardhe et al reported an aggressive destructive lesion in the mandible which showed extremely rare malignant transformation in SOT [7]. In the present case also similar to Pardhe et al, there was an osteolytic lesion in the posterior mandible region associated with a fracture of the mandible. Such radiographic presentations mimicking a malignant process should be evaluated with caution.

The pathogenesis of this lesion is still ambiguous with varied possibilities of origin taken into account. The most plausible origins include from

remnants of the dental lamina (rests of Serres), and epithelial rests of Malassez (EROM) of the gingival epithelium [2, 3]. Several authors have suggested the EROM as the most cogent origin for SOT [1, 5, 6]. Goldblatt et al proposed that the EROM persist even in adults in all areas of the periodontal ligament, most abundantly in the cervical half of the roots where they can even undergo proliferation and form structures similar to the rests of Serres. This supports the incidence of occurrence of SOT even in edentulous areas [6, 8]. According to an extensive literature search and our experience EROM is the most believable origin for this rare neoplasm.

Microscopically SOTs were first as a "tumor composed of mature collagenous connective tissue with numerous small islands of mature and benign squamous epithelium. The connective tissue stroma shows moderate number of plump ovoid to spindle-shaped fibroblasts. Focal areas of cystic degeneration and masses of laminar calcification can be seen [2]. The Ayoub-Shklar stain can demonstrate prekeratin making hyaline cytoplasmic masses and that the calcified bodies were surrounded by a band of mature keratin. Other author has also observed similar histopathological findings [5, 6, 9]. Few authors have reported squamous odontogenic tumor-like proliferations (SOT-LP) arising from odontogenic cysts (SOT-LPOC) [8, 10]. In the case being discussed here, a similar microscopic picture comprising of small islands of terminally differentiated squamous epithelium in a mature connective tissue stroma was observed. The islands showed microcystic degeneration and individual cell keratinization. No association with odontogenic tumor was seen in our case.

Table 1. Literature review (modified Table by Lucio et al.) [3, 5]

S.no	Author	Year	Number of cases
1.	Pullon et al	1975	6
2.	Mardones et al	2015	1
3.	Lucio et al	2015	1
4.	Mohr et al	2015	1
5.	Badni et al	2012	1
6.	Goldblatt et al	1982	5
7.	Pardhe et al	2021	1
8.	Doyle et al	1977	2
9.	McNeill et al	1980	1
10.	Hopper et al	1980	1
11.	Carr et al	1981	1
12.	Leventon et al	1981	1
13.	Kangvonkit et al	1981	1
14.	Cataldo et al	1983	1
15.	Swan and McDaniel et al	1983	1
16.	Norris et al	1984	1
17.	Kristensen et al	1985	1
18	Monteil and Terestri et al	1985	1
19	Warnock et al	1985	1
20	Mills et al	1990	1
21	Tatemoto et al	1989	2
22	Leider et al	1989	2
23	Yacoob et al	1990	1
24	Reichart and Philipsen	1996	1
25	Schwartz‐Arad et al	1990	1
26	Baden et al	1993	3
27	Saxby et al	1993	3
28	Haghighat et al	2002	1
29	Barrios et al	2004	1
30	Ruhin et al	2007	1
31	Jwa Young et al	2007	1
32	King kim et al	2007	2
	Present case	**2022**	**1**

Treatment options for SOT include conservative surgical removal or resection of the affected segment depending upon the destruction caused [2-4]. In our case, extensive destruction of the mandible bone along with a fracture of the inferior border clinically and radiographically warranted resection of the destroyed segment of the mandible. Recurrences have been infrequently reported except in two cases. Our patient is under regular follow up and there is no recurrence.

Squamous odontogenic tumors have been recognised as a distinct entity in the near past compared to other odontogenic lesions mainly attributable to the lack of published literature and experience of specialists leading to diagnostic pitfalls. These lesions have been erroneously diagnosed as acanthomatous ameloblastomas or well-differentiated squamous cell carcinoma [3, 5]. Pindborg et al described two cases of ameloblastoma with squamous metaplasia, keratinization and onion-like calcifications within keratin pearls. The keratin pearls were observed within islands of ameloblastoma where squamous metaplasia was present. In the squamous odontogenic tumor, the islands are squamous only and the peripheral layer of tall columnar polarized cells characteristic of acanthomatous ameloblastoma are lacking. The keratin formation seen in SOT apparently arises from keratohyaline granules [1, 4, 9]. We found less than 50 published cases of SOT with an extensive literature search and the contribution from the Indian subcontinent has been very less (Table 1).

4. CONCLUSION

Squamous Odontogenic Tumors are exceedingly rare neoplasms which have been speciously reported as malignant neoplasms leading to aggressive treatment approaches. The potential for recurrence in even benign SOTs may depend on the accessibility of the SOTs for surgical treatment and the biological behaviour of the SOTs. In our present study, we recommend an individualized treatment plan to respond to the biological reaction of the SOT rather than to the histopathology of the tumor.

CONSENT

As per international standard or university standard, patient(s) written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Pullon PA, Shafer WG, Elzay RP, Kerr DA, Corio RL. Squamous odontogenic tumor. Report of six cases of a previously undescribed lesion. Oral Surg Oral Med Oral Pathol. 1975;40:616-30.
2. Mardones Ndo R, Gamba Tde O, Flores IL, de Almeida SM, Lopes SL. Squamous Odontogenic Tumor: Literature Review Focusing on the Radiographic Features and Differential Diagnosis. Open Dent J. 2015;9:154-158.
3. Priscilla SC. Lúcio Marcelo Augusto O. Sales Gustavo P. Godoy Rivadávio FB. Amorim. Squamous odontogenic tumor: Report of a case of unusual involvement. Bras Patol Med Lab. 2015;1:427-431.
4. Mohr B, Winter J, Wahl G, Janska E. Recurrent squamous odontogenic tumor: A case report and review of the literature. Oncol Lett. 2015;10(5):2713-2722.
5. Badni M, Nagaraja A, Kamath V. Squamous odontogenic tumor: A case report and review of the literature. J Oral Maxillofac Pathol. 2012;16(1):113-7.
6. Upadhyaya JD, Banasser A, Cohen DM, Kashtwari 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.
DOI: 10.1016/j.joms.2020.06.031
7. Laungani N, Hengen S, Nester C, Smith MH. Pericoronal radiolucency surrounding an impacted mandibular molar. Oral Surg Oral Med Oral Pathol Oral Radiol. 2020; 129(4):286-289.
DOI: 10.1016/j.oooo.2019.01.070
8. Chrcanovic BR, Gomez RS. Squamous odontogenic tumor and squamous odontogenic tumor-like proliferations in odontogenic cysts: An updated analysis of 170 cases reported in the literature. J

Craniomaxillofac Surg. 2018;46(3):504-510.
DOI: 10.1016/j.jcms.2017.12.023.

9. 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(2):168-174.
DOI: 10.1007/s12105-016-0753-2

10. Barbeiro CO, Barbeiro RH, da Silveira HA, de Almeida LY, León JE, Bufalino A. Maxillary dentigerous cyst showing squamous odontogenic tumor-like proliferation: Surgical approach and literature review. Autops Case Rep. 2021; 11: e2021302.
DOI:10.4322/acr.2021.302.

Peer-review history:
The peer review history for this paper can be accessed here:
https://www.sdiarticle5.com/review-history/95140