ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at: www.ijdsir.com

Volume – 2, Issue – 2, March - April - 2019, Page No. : 317 - 321

Peripheral Desmoplastic Ameloblastoma: Case Report And Review of Literature.

[1]Dr. Kartik Poonja, Lecturer, Department of Oral and Maxillofacial Surgery, Mahatma Gandhi Mission's Dental College & Hospital, Navi Mumbai – 410209, India

[2] Dr. Janaki Iyer, Lecturer, Department of Oral and Maxillofacial Pathology, Mahatma Gandhi Mission's Dental College & Hospital, Navi Mumbai – 410209, India

[3] Dr. Niharika Swain, Lecturer, Department of Oral and Maxillofacial Pathology, Mahatma Gandhi Mission's Dental College & Hospital, Navi Mumbai – 410209, India

[4] Dr. Shilpa Patel, Professor and Head, Department of Oral and Maxillofacial Pathology, Mahatma Gandhi Mission's Dental College & Hospital, Navi Mumbai – 410209, India

Correspondence Author: Dr. Kartik Poonja, Department of Oral and Maxillofacial Surgery, Mahatma Gandhi Mission's Dental College & Hospital, Junction of NH - 4 and Sion Panvel Expressway, Sector - 1, Kamothe, Navi Mumbai – 410209. Maharashtra, India.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Sixty-seven percent of peripheral odontogenic tumors are ameloblastoma, however, among all ameloblastomas, the peripheral variant accounts to 2 – 10 %. Peripheral desmoplastic ameloblastoma is extremely rare variant of benign odontogenic tumours. In this report, we present a case of peripheral desmoplastic ameloblastoma, emphasizing the need for mandatory histopathologic evaluation of every lesion.

Keywords: Ameloblastoma, Desmoplastic, Peripheral

Introduction

Peripheral odontogenic tumors (POT) are less encountered lesions of the oral and maxillofacial region, accounting from 1.24% to 3.4% of all odontogenic tumors [1]**.** Sixty-seven percent of peripheral odontogenic tumors are ameloblastoma, however, among all ameloblastomas, the peripheral variant accounts to 2 – 10%[2]. Periphreal Ameloblastoma (PA) is a rare entity that was initially described by Eversole in 1984 [3, 4]. A histopathological variant of Peripheral Desmoplastic Ameloblastoma (PDA) is relatively rare with only four previously reported cases. Microscopically this variant presents as irregular compressed islands of odontogenic epithelium arranged in nests and thin cords embedded in a densely hypocellular collagenized stroma. In this case report, we describe an interesting case of a PDA in the mandibular anterior lingual gingiva.

Case Report

A 60 year old man presented with an asymptomatic, slow growing swelling in the lingual attached gingiva in relation to 42,41,31,32 of a year's duration. Clinical examination revealed a well defined, non tender, firm, non compressible, sessile growth of 2cm in diameter, with overlying intact and smooth mucosa. **(Fig 1)** Extra oral examination revealed no relevant abnormality. Medical and dental histories were unremarkable. An occlusal radiograph showed no evidence of bone resorption in the area. **(Fig 2).**

Such a pathology described on the mandibular anterior lingual gingiva could be a lesion of either developmental, hamartomatous, reactive, traumatic or neoplastic origin. Considering its benign nature, the lesion was excised with patient's due consent and the specimen was referred to our Department of Oral Pathology for histopathological evaluation. **(Fig 3)**

Microscopic examination with routine Hematoxylin and Eosin stain showed a lesion surfaced by parakeratinised hyperplastic stratified squamous epithelium, with a regular pattern of maturation. The underlying lamina propria presented irregular islands of odontogenic epithelium embedded in a densely collagenized stroma. The epithelial islands were arranged in nests, thin cords and strands of severely constricted and compressed odontogenic epithelium, interconnected at places. **(Fig 4, 5)** Columnar cells with reverse polarization were not prominent **(Fig 6)**. Some islands were hypercellular with central squamous metaplasia. Cystic degeneration was also observed in some of the islands. Though the stroma was densely collagenized, myxoid changes were found adjacent to epithelial islands. These features were suggestive of a peripheral desmoplastic ameloblastoma.

An Immunohistochemical analysis of the specimen was done for cytokeratin (CK) 13, 14 and 19. Strongly positive cytokeratin-14 stain was observed in the basal and parabasal layer. **(Fig 7)** Whereas the central cells showed focal immunoreactivity for cytokeratin-13. **(Fig 8)** Negative immunoreactivity for cytokeratin-19 was observed. **(Fig 9)**

Discussion

Peripheral odontogenic tumours that have been reported to occur in the jaws include the soft tissue counterparts of the intraosseous odontogenic tumours. Sixty-seven percent of peripheral odontogenic tumors are ameloblastoma, however, among all ameloblastomas, the peripheral variant accounts to 2 – 10 % [4].

Desmoplastic ameloblastoma (DA) is considered a rare variant of ameloblastoma, since its original description by Eversole et al. [5]. Of all the reported cases of DA, only five (including the present case) can be considered as peripheral variant [1, 3, 5, 6].

The clinicopathological details of all the histopathologically diagnosed cases of peripheral DA are featured in **Table I**. The histological findings in our case were in accordance with previous reports. Though the histopathological features of DA i.e. compressed and interconnecting chords of odontogenic epithelium, with absence of reversal of polarity and stromal desmoplasia are pathognomic, the marked similarities with squamous odontogenic tumor (SOT), pose as a diagnostic dilemma [7, 8]. Hence minute microscopic details that aid in differentiation should be accounted for, as highlighted in **Table II**.

Although immunohistochemistry plays a limited role in the diagnosis, it can assist in determining the cell origin, differentiation and maturation, in odontogenic tumours. In the present case, the negative immunoreactivity for CK-19 and lack of continuity of the lesional tissue with surface epithelium, supports the origin of this neoplasm from remnants of dental lamina located in the gingival tissues, as suggested by Philipsen et al. [9]. Immunohistochemical expression of CK-14 in basal and suprabasal cells in the present case offered evidence of maturation of epithelial component [1]. Focal and weak staining for CK-13 in central cells of the tumour islands, suggested lack of prominent squamous differentiation, in present case.

Surgical excision is the treatment of choice in cases of peripheral DA. In general, the peripheral counterparts of the intraosseous odontogenic tumours are known to be less aggressive, but, the exact biological behavior of peripheral

DA cannot be adequately ascertained due to limited literature.

Figures

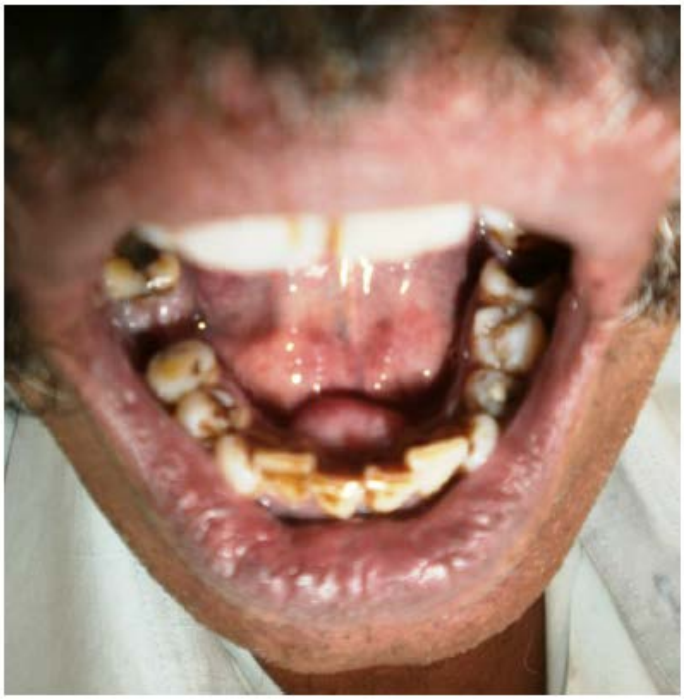

Fig 1: Well defined, solitary sessile growth on the lingual gingival aspect measuring approximately 2 x 2 cms in size, extending from lingual aspect of 32 to 42. Mucosa overlying the growth is normal (Intra oral view).

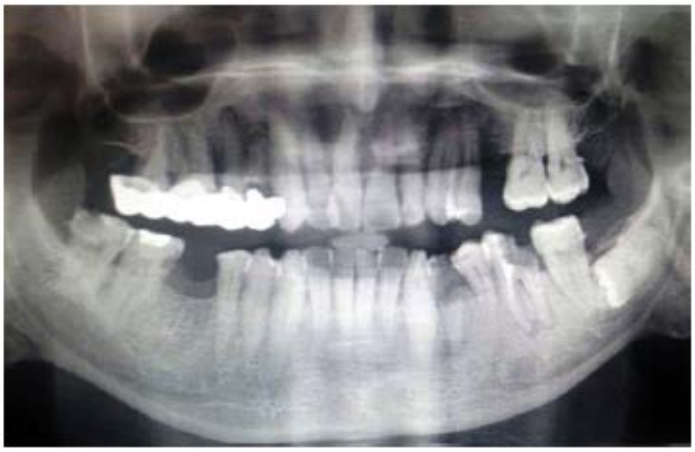

Fig 2: OPG showed generalised bone loss with no specific significant pathology with related area.

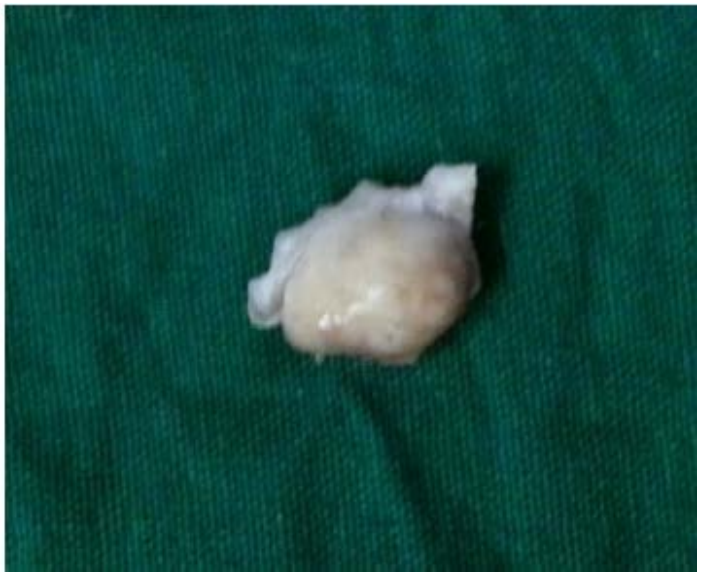

Fig 3: Excised specimen of soft tissue lingual growth.

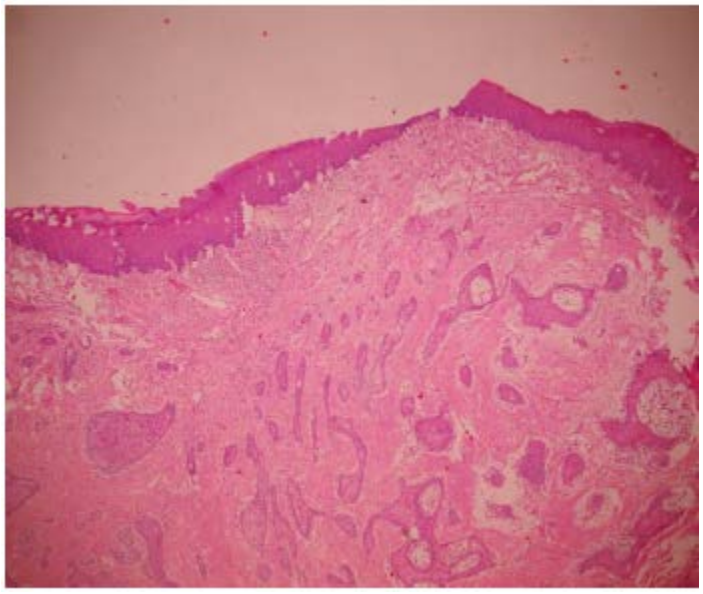

Fig 4: Parakeratinised squamous epithelium of normal thickness. The underlying collagenised stroma shows irregular islands which are arranged in nests and narrow/ broad cords. (H & E, 40x).

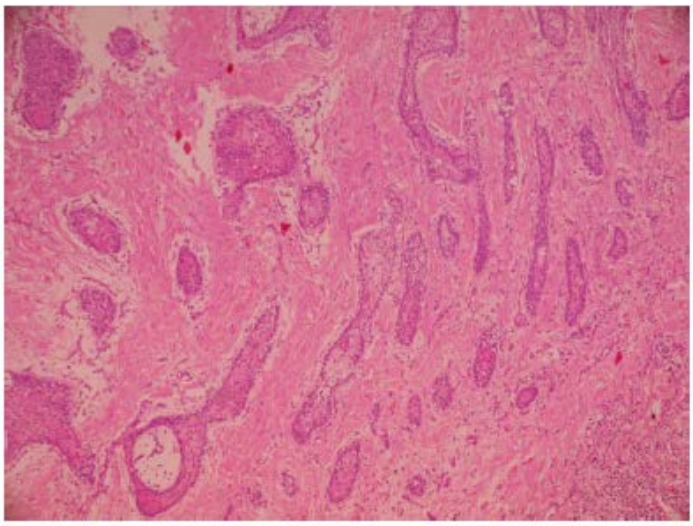

Fig 5: Odontogenic epithelial islands of various shapes and sizes dispersed in a collagenous stroma. (H & E, 100x).

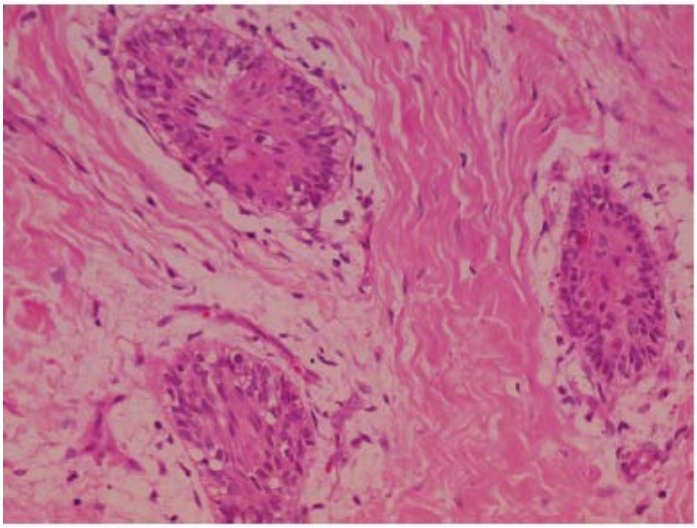

Fig 6: Islands show peripheral layer with flattened to cuboidal to columnar cells with reverse polarity. The central areas of the islands show spindled or squamatoid cells. Juxtaepithelial myxoid degeneration evident. (H & E,400x).

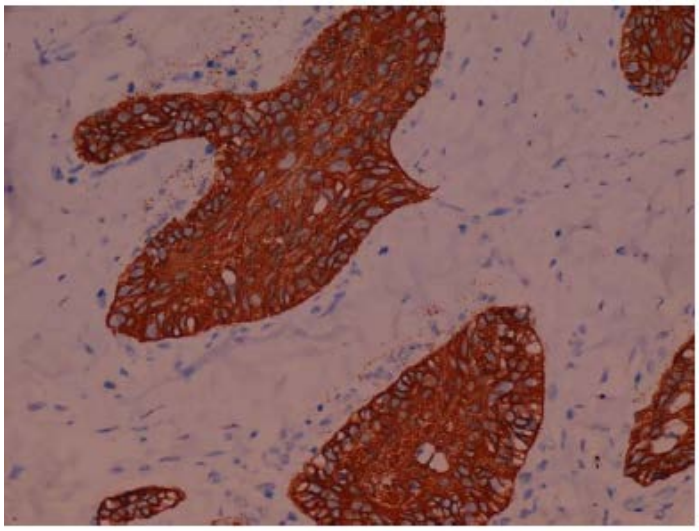

Fig 7: Tumour islands showing strong positivity for CK-14 (400x) .

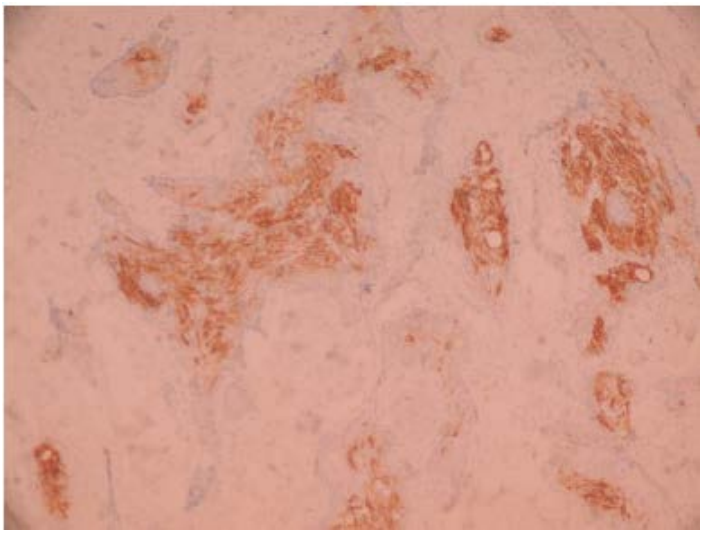

Fig 8: Central cells of the tumour islands are immoreactive for CK- 13 (400x).

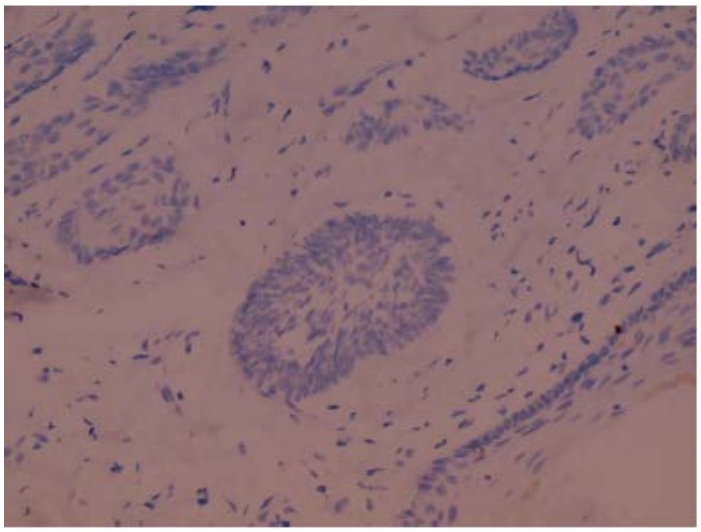

Fig 9: Tumour islands are non reactive for CK -19. (400x)

Conclusion

- Extremely rare lesion, as only four previous similar cases have been reported in literature.
- Oral and maxillofacial surgeons should be aware of peripheral odontogenic neoplasms, as they are likely to encounter in their clinical practice, because of their location in gingival tissues.
- Histopathological examination can provide a true insight to the incidence and prevalence and as well as prognosis of peripheral odontogenic neoplasms.
- Careful histopathological evaluation may be helpful to differentiate peripheral DA from SOT, a close contender.

References

1. Bologna-MR, Mosqueda-TA, de Almeida-OP, Toral-RV, Martínez-MG. Peripheral desmoplastic ameloblastoma: Histopathological and immunohistochemical profile of a case. *Med Oral Patol Oral Cir Bucal*. 2010 Nov 1; 15 (6):e846-9.

2. Curran AE, Byerly PD. Peripheral desmoplastic ameloblastoma: report of a rare case. *J Oral Maxillofac Surg*. 2008 Apr; 66(4):820-5. doi: 10.1016/j.joms.2007.04.033.

3. Eversole LR, Leider AS, Hansen LS. Ameloblastomas with pronounced desmoplasia. *J Oral Maxillofac Surg* 1984; 42(11):735-40.

4. Curran AE. Peripheral odontogenic tumours. *Oral Maxillofacial Surg Clin N Am* 16 (2004) 399–408.

5. Smullin SE, Faquin W, Susarla SM, and Kaban LB. Peripheral desmoplastic ameloblastoma: report of a case and literature review. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008; 105: 37-40.

6. Oteri G, Lentini M, Pisano M, Cicciù M. Peripheral desmoplastic ameloblastoma in adolescent age: clinico-pathological and immunohistochemical analysis of a case. *Open Dent J*. 2014 Sep 29; 8:159-63. doi: 10.2174/1874210601408010159. ECollection 2014.

7. Barnes L. Surgical pathology of the head and neck. 3rd edition. Informa healthcare. Odontogenic tumours. p. 1218- 21.

8. Barnes L. Surgical pathology of the head and neck. 3rd edition. Informa healthcare. Odontogenic tumours. p.1228 - 30

9. H.P. Philipsen, P.A. Reichart , H. Nikai , T. Takata , Y. Kudo. Peripheral ameloblastoma: biological profile based on 160 cases from the literature. *Oral Oncology* 2001; 37: 17-27.