

## PERIPHERAL AMELOBLASTOMA- A CASE REPORT WITH EMPHASIS ON ETIOPATHOGENESIS

<sup>1</sup> Raghavendra Kini<sup>2</sup> Vathsala Naik<sup>3</sup> Anjali Shetty<sup>4</sup> Smit Singla<sup>1</sup> Professor<sup>2</sup> Professor and Head<sup>3</sup> Assistant professor<sup>4</sup> Post graduate student<sup>1-4</sup> Dept of Oral Medicine and Radiology, A.J. Institute of Dental Sciences, Mangalore, India

### ABSTRACT:

Peripheral ameloblastoma, a rare and unusual variant of odontogenic tumour, comprises about 1% of all ameloblastomas. The extrasosseous location is the peculiar feature of this type of tumour, which is otherwise similar to the classical ameloblastoma. It appears in the gingiva and oral mucosa and it usually does not show any bone involvement on radiographs, except for saucer shaped erosion of underlying alveolar bone. Recurrence is considered uncommon. We report a case of peripheral ameloblastoma of maxillary gingiva.

**KEYWORDS:** peripheral ameloblastoma, odontogenic tumours, alveolar mucosa, maxilla

### INTRODUCTION

Peripheral ameloblastoma (PA) is a rare, benign epithelial odontogenic tumor, occurring in the soft tissues overlying the tooth bearing areas of maxilla and mandible. Clinically it presents as a firm, slow growing, and dome shaped swelling with a smooth surface covered with normal mucosa. Ideally they do not show any bony changes on radiographs.<sup>1</sup> Conservative surgical excision with a small margin of healthy tissue is the recommended treatment. We present and analyse a case of peripheral ameloblastoma in the anterior region of maxilla, with special emphasis on etiopathogenesis.

### Case report

A 40 years old male patient presented to our department with a swelling in the gingiva of right maxillary region. He had noticed the swelling a month back. The swelling was gradually increasing in size and was totally asymptomatic. Past dental and medical histories were non-contributory. On intraoral examination a solitary, well defined dome shaped swelling was present in the interdental area of 13 and 14 measuring about 1.5 cm in size. Swelling was non tender and firm in consistency with normal overlying mucosa (**Fig.1**). The teeth in the vicinity were non tender, undisplaced and vital. Based on history and clinical examination a provisional diagnosis of peripheral ossifying fibroma was made. Peripheral odontogenic tumors and gingival cyst were considered in differential diagnosis.

Periapical and maxillary standard occlusal radiograph did not reveal any bony abnormalities. An excisional biopsy was performed under local anaesthesia, during

which time; bony erosion was noticed beneath the lesion (**Fig.2**). The specimen was fixed in 10% formalin and sent for histopathological examination. Histologically it comprised of deeper tissue islands, strands of odontogenic epithelium with peripheral tall columnar cells with centrally stellate reticulum like cells suggestive of peripheral ameloblastoma (**Fig.3**). The healing was uneventful and the patient has been followed up at regular intervals for past nine months and he is disease free (**Fig.4**).

### Discussion

Ameloblastoma is a benign epithelial odontogenic tumor with three main clinical variants- solid/multicystic (86%), single cyst (13%) and peripheral ameloblastoma (1%). So peripheral ameloblastoma (extrasosseous ameloblastoma) is a rare tumor occurring in the soft tissues of the tooth bearing areas of maxilla and mandible.<sup>2</sup> Several authors refer to Kuru<sup>3</sup> as having reported on the peripheral ameloblastoma for the first time. But what Kuru described was not a PA, infact it was intra-osseous ameloblastoma, which penetrated through the alveolar bone and got fused with oral epithelium giving an impression of a peripheral lesion. The first completely documented case of a PA is attributed to Stanley and Krogh.<sup>4</sup>

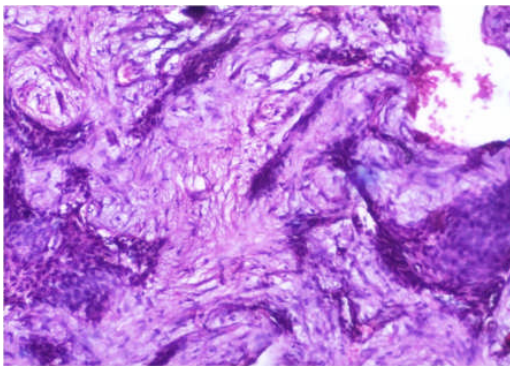
Ameloblastoma is an epithelial odontogenic tumour of the jaw bones which is thought to arise from rests of the dental lamina or from basal cells of the surface epithelium. A recent investigation demonstrated that alterations of the ameloblastin gene form the genetic basis for ameloblastoma<sup>5,6</sup>.



**Fig.1. solitary, well defined dome shaped swelling in the interdental area of 13 and 14.**



**Fig.2 . Bony erosion was noticed beneath the lesion after excisional biopsy.**



**Fig.3. Deeper tissue islands, strands of Odontogenic epithelium with peripheral tall columnar cells with centrally stellate reticulum like cells suggestive of peripheral ameloblastoma.**



**Fig.4. satisfactory healing after one month.**

The origin of PA at cellular level, continues to be a challenge to academicians and researches alike. However two sources of origin should be considered. One, it arises from the remains of the dental lamina located in soft tissues overlying the tooth bearing areas of the jaw bones. In this case, the lesion is totally separated from the surface epithelium by a band of collagenous tissue. Stanley and Krough had mentioned that their case probably arose in this manner from odontogenic epithelial cell rests. Two, it arises directly from the basal cell layer of the overlying epithelium. But however, even the demonstration of fusion between overlying epithelium and tumor tissue will not clearly prove the origin, as the tumor can grow subepithelially and may get fused with the surface epithelium. Even electron microscopic studies have not been able to resolve this challenge emphatically.<sup>1,7</sup>

Peripheral Ameloblastoma is noticed during 4<sup>th</sup> to 6<sup>th</sup> decades of life with an average age of 50 years. It is interesting to note that the average age of occurrence of

PA is much higher than its central counterpart (37.4 years). A slight male predominance has been reported with male/female ratio being 1.9:1. Mandible is most favoured site than maxilla. In mandible it is more often located on lingual aspect on premolar region, while in maxilla its common location being tuberosity area.<sup>8</sup> In contrary in our case swelling was located in maxilla in canine premolar region on facial aspect.

Clinically it presents as a sessile, firm growth of gingiva measuring around 0.5 to 2 cm in size. The surface is usually smooth and pink/red in colour. Clinical appearance was similar in the present case, with swelling measuring about 1 X 1.5 cm, with normal overlying mucosa and smooth surface. Usually they are asymptomatic. In majority of cases there are no radiographic bony changes. In few cases, a superficial erosion of the bone or bony

depression can be noticed, which might be caused due to pressure resorption rather than by the invasion of the tumor.<sup>1,9</sup>

Clinically it needs to be differentiated from other lesions which are common in gingiva<sup>10</sup>.

**Pyogenic granuloma:** it is sessile/peduncled lesion, common in maxillary gingiva. The colour ranges from pink to red to purple. It bleeds profusely with little manipulation. Peripheral giant cell granuloma: common in mandibular gingiva. The colour ranges from red to bluish purple, but usually more bluish than pyogenic granuloma. It can also cause destruction of underlying bone (saucerisation).

**Peripheral ossifying fibroma:** gingiva of maxilla is more often involved than mandible. It is common in younger individuals. The surface can be smooth or lobulated. Early lesions are red in colour but mature lesion appear pale pink. Scattered irregular radiopacities can be seen on the radiograph.

PA can be successfully managed with conservative suprapariosteal surgical excision with adequate distance from margins. Although there were no bony changes on radiograph in our case, but on surgical exploration resorption of cortical plate (saucerisation) was evident beneath the lesion.

Local recurrence rate is much lower in comparison with central ameloblastoma. There are reports of aggressive behaviour of PA invading intracranial structures and malignant transformation. Keeping that in mind, a long term follow up is prudent. Our patient is examined periodically every month, he is disease free since nine months.<sup>11</sup> So PA should be considered in differential diagnosis of swellings located in the gingiva. But, what is more important is the way it needs to be managed; the word "ameloblastoma" sends a strong signal for the surgeons for aggressive treatment plan which is not at all mandated. A simple conservative excision is all that one needs to do in peripheral ameloblastoma.

## References

1. Philipsen H.P, P.A. Reichart, Nikai H, Takata T, Kudo Y. Peripheral ameloblastoma: biological profile based on 160 cases from the literature – Review Oral Oncol 37 (2001) 17-27. [doi:10.1016/S1368-8375\(00\)00064-6](https://doi.org/10.1016/S1368-8375(00)00064-6)
2. Pekiner FN, Ozbayrak S, Sener BC, Olgac V, Inanoglu A S. Peripheral ameloblastoma: a case report. Dmfr 2007; 36: 183-6.
3. Kuru H. Ueber das Adamantinom. Zentralblatt für allgemeine Pathologie und Anatomie 1911; 22: 291-5.
4. Stanley HR, Krogh HW. Peripheral ameloblastoma. Report of a case. Oral Surg Oral Med Oral Pathol 1959; 12: 760-5. [doi:10.1016/0030-4220\(59\)90124-0](https://doi.org/10.1016/0030-4220(59)90124-0)
5. Barnes L, Eveson JW, Reichart P, Sidransky D, editors. The World Health Organization Classification of Tumours. Pathology and genetics. Head and neck tumours. Lyon: IARC Press; 2005. p.285
6. Perdigo PF, Gomez RS, Pimenta FJ, De Marco L. Ameloblastin gene (AMBN) mutations associated with epithelial odontogenic tumors. Oral Oncol 2004; 40: 841-6. PMID:15288841 [doi:10.1016/j.oraloncology.2004.03.004](https://doi.org/10.1016/j.oraloncology.2004.03.004)
7. Carolina Cavalieri, Alessandra Pires Duarte, Marina Goncalves Diniz, Ricardo Santiago Gomez. Current concepts of ameloblastoma pathogenesis. J Oral Pathol Med 2010; 39: 585-91. [doi:10.1111/j.1600-0714.2010.00908.x](https://doi.org/10.1111/j.1600-0714.2010.00908.x)
8. Carolina Cavalieri Gomes, Bruna Goncalves. Garcia, Ricardo Santiago Gomez, Joao Batista de Freitas, Ricardo Alves Mesquita. A clinical case of peripheral ameloblastoma. Braz J Oral Sci 2007; 6(21): 1364-66.
9. Gomes CC, Oliveira CS, Castro WH, Lacerda JC, Gomez RS. Clonal nature of odontogenic tumours. J Oral Pathol Med 2009; 38: 397-400. [doi:10.1111/j.1600-0714.2008.00744.x](https://doi.org/10.1111/j.1600-0714.2008.00744.x)
10. Maryam Seyedmajidi, Mohammadtaghi Hamzehpoor, Soodabegh Bagherimoghaddam. Localized lesions of oral cavity: A clinicopathological study of 107 cases. Research Journal of Medical Sciences 2011;5(2):67-72. [doi:10.3923/rjmsci.2011.67.72](https://doi.org/10.3923/rjmsci.2011.67.72)
11. Nauta JM, Panders AK, Schoots CJF, Verney A, Roodenburg JLN. Peripheral ameloblastoma. A case report and review of the literature. Int J Oral Maxillofac Surg. 1992; 21: 40-4. [doi:10.1016/S0901-5027\(05\)80451-5](https://doi.org/10.1016/S0901-5027(05)80451-5)

## Corresponding Author

**Dr. Raghavendra Kini**

Professor

Dept. of Oral Medicine and Radiology

A.J. Institute of Dental Sciences

N.H.17, Kuntikana, Mangalore

Karnataka. India-575004

raghkini@yahoo.co.in

Telephone – +919845822204