Peripheral ameloblastoma- A rare case report

Sayali Jadhav[1], Zaneta D'souza[2*], Tabita Joy[3], Manisha Sardar[4], Arush Thakur[5]

[1,5]Dental Surgeon, [2]Post Graduate Student, [3]Professor and Head, [4]Associate Professor, Dept. of Oral Pathology and Microbiology, [1-4]Government Dental College and Hospital, St. George Medical Hospital Campus, Mumbai, Maharashtra, [5]Shree Balaji Hospital, Kangra, Himachal Pradesh, India

***Corresponding Author: Zaneta D'souza**
Email: zanetadsouza@gmail.com

Abstract
Peripheral ameloblastoma, also called as extraosseous ameloblastoma is a rare variant of ameloblastoma. It comprises of 2-10% of all ameloblastomas. This lesion can be mistaken for any other peripheral lesion both clinically and radiographically. Hence a correlation between the clinical, radiographical and histopathological findings is inevitable. Herewith we present a case of peripheral ameloblastoma in a 47-year-old male patient affecting the lingual alveolar mucosa of the mandibular region with complete clinical, radiological and histopathological details.

Keywords: Gingiva, Ameloblastoma, Odontogenic Tumors.

Introduction
A myriad of exophytic pathologic lesions which are localized to the gingiva. They could be infectious, inflammatory, neoplastic, developmental, traumatic or miscellaneous in origin and they are designated by the misnomer 'Epulis'. These growths may not always be benign like pyogenic granuloma, peripheral giant cell granuloma, but maybe aggressive like peripheral ameloblastoma, or malignant like oral squamous cell carcinoma, fibrosarcoma, and melanoma. Hence it is important that a localized gingival overgrowth should be investigated and assessed first through an incisional biopsy before treatment planning. Herewith we present such an unusual case of a localized gingival overgrowth in a 47 year old male patient.

Case Report
A 47 -year- old male patient reported, with a chief complaint of a painful growth in lower right posterior tooth region since, 6 to 8 months. There was no history of trauma, however, patient had a habit of frequent use of a tooth-pick. Medical history was non-contributory. Intraoral examination revealed a well-demarcated, firm, non-tender, sessile exophytic growth with a smooth surface and which was pale pink to red in color (Fig. 1). It extended from distal of 43 to distal of 45 region lingually and measured approximately 3x2 cm in diameter. Involved teeth were vital. For the radiographic diagnosis, the patient was advised an Orthopantomography (OPG) in which, there was horizontal bone loss between 43 to 45. Additionally, he was also advised a cone beam computed tomography (CBCT) which showed peripheral cuffing of the bone in the lesional area. Based on our findings, a clinical diagnosis was made as peripheral giant cell granuloma with a differential diagnosis of pyogenic granuloma and a fibrous epulis. Incisional biopsy was taken and histopathological examination was done.

The incisional biopsy showed surface epithelium, and fibrocellular connective tissue stroma with numerous odontogenic epithelium arranged in islands and strands under scanner view (Fig. 2A, Fig. 2B). Under the low power view most of the follicles and strands were composed of central stellate reticulum like cells and hyperchromatic basaloid cells (Fig. 3). The cells at the periphery showed reverse polarity that resembled ameloblasts. Few islands also showed acanthamatous changes within the central stellate reticulum like cells. Following the incisional biopsy, excisional biopsy was also done which showed similar findings. Correlating the clinical, radiological and pathological findings a final confirmatory diagnosis of peripheral ameloblastoma was made.

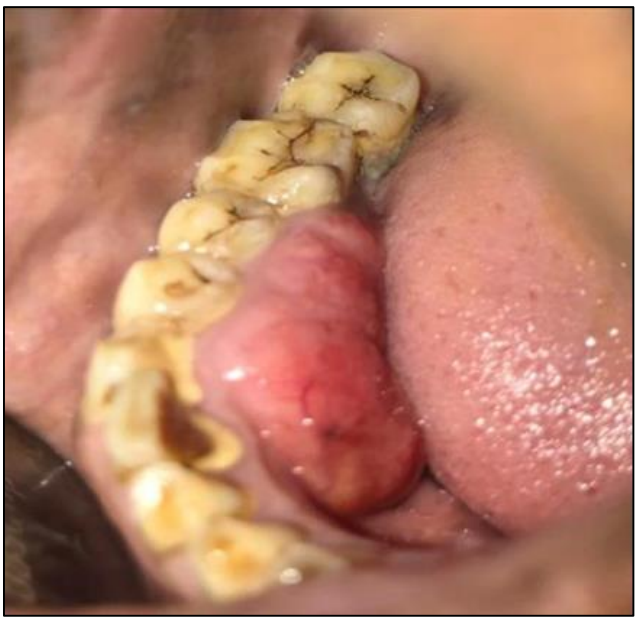

Fig. 1: A well-defined, sessile, smooth surfaced, pale pink to reddish, tender growth observed on lingual gingiva extending antero-posteriorly from 43 to distal of 45 and supero-inferiorly from lingual cusp of involved teeth to lingual vestibule

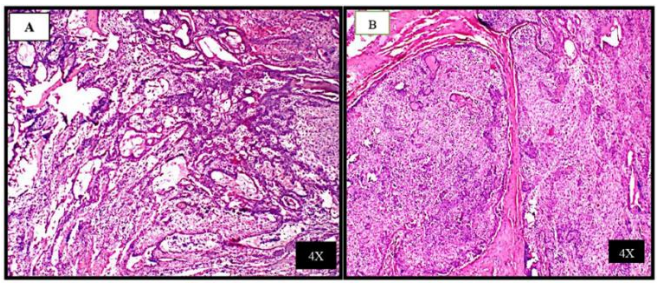

Fig. 2: H&E stained section (4X view) showing lesional tissue composed of hyperchromatic odontogenic epithelial cells in the form of strands (A) and islands. (B)

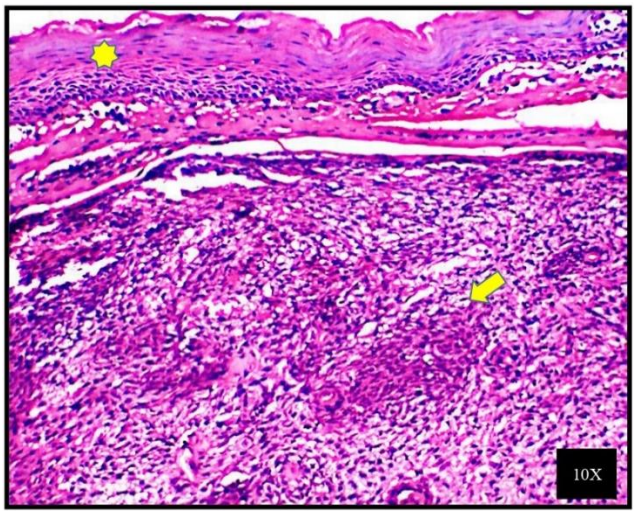

Fig. 3: H&E stained section (10X view) shows parakeratinized surface epithelium (asterisk) separated from underlying lesional tissue. The lesional tissue (arrow) comprises of hyperchromatic odontogenic epithelial cells in the form of islands

Discussion

Amongst all oral lesions, prevalence of odontogenic tumor is 0.8%, of which 30% is ameloblastoma.[1] There are 4 subgroups into which this neoplasm can be categorized namely the conventional type, the unicystic type, peripheral type and the metastasizing ameloblastoma.[2]

The peripheral ameloblastoma (PA) is an exophytic growth that exclusively involves the soft tissues overlying the dentulous areas of the jaw. It is also called as extraosseous ameloblastoma and is an extremely rare tumour.[3] Peripheral ameloblastoma has been considered as a separate entity after Stanley and Krogh in 1911 reported a tumour in the soft tissue occurring in the premolar- molar region of the mandible on the lingual side. [4] WHO (2017) in its current classification has continued to identify this lesion as a separate entity.[2]

Clinically it is a painless, sessile, firm, and exophytic growth, the surface of which is generally smooth. However, "granular" or "pebbly" and "papillary" or "warty" surfaces have also been reported. The color of the lesion varies between normal or pink and red or dark red.[5] In our case the growth was painless, firm and sessile and the surface was smooth.

The tooth bearing areas of the mandible are most commonly involved than that of the maxilla. In the mandible the premolar region is the predominantly involved site.[5,6] Our case showed an involvement of the tooth-bearing area of the mandibular premolar region on the lingual side.

The histopathogenesis of PA is controversial. Both the dental lamina remnants, the basal cells of the surface epithelium and the pluripotent minor salivary gland cells have been reported to be potential cellular sources of origin.[6,7] The diagnostic criteria of peripheral ameloblastoma includes the origin from the overlying epithelium, presence of odontogenic epithelial islands in the lesion and lack of potential to bone infiltration.[6]

Three different zones, the deep tumor islands, the altered surface epithelium overlying the tumor, and the transitional area between "normal" and altered covering epithelium is seen microscopically. The transitional "preameloblastomatous" zone is not always conspicuous and hence, the tumor's origin from the oral mucosa or from remnants of "odontogenic" epithelium cannot be established.[8] The tumour islands show, peripheral palisading of columnar basal cells with hyperchromatic nuclei . Few follicles may show stellate reticulum, while some may depict cystic and/or acanthamaous changes.[8,9]

In comparison with the intraosseous ameloblastoma, PA is noninvasive and has a low recurrence rate. The Dense fibrous tissue of the gingiva and periosteum and the cortical plate of the alveolar process could be an effective barrier to the infiltration of peripheral ameloblastoma [9]

The current treatment of choice is a conservative supra periosteal surgical excision with adequate disease-free margins, and treated cases of PA should be followed up for a longer duration to detect the late local recurrence. [6]

PA should be differentiated from peripheral reactive lesions such as fibrous epulis, pyogenic granuloma, peripheral giant cell granuloma, peripheral odontogenic fibroma, odontogenic gingival epithelial hamartoma, and basal cell carcinoma.[6 10]

Conclusion

Peripheral ameloblastoma has a similar histopathological picture as intraosseous ameloblastoma, however it is exclusive to the soft tissues in the dentulous areas of the jaw. Since it is rare lesion and shows a clinical appearance similar to other reactive, benign and malignant lesions a careful and accurate diagnosis of the lesion will aid in better treatment planning and follow-up. Hence, PA should be considered as a differential diagnosis for all clinically localized gingival overgrowths.

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Conflict of interest

Nil.

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