Extraosseous Ameloblastoma of the Buccal Mucosa: A Case Report of a Rare Entity

Abstract

Extraosseous ameloblastoma is the peripheral counterpart of the usual well known central ameloblastoma. The most commonly involved sites include the lingual gingiva of the mandible, lips, and palate, with only a few reports of peripheral ameloblastoma in the buccal mucosa cited in the literature. This article aims to describe a case of peripheral ameloblastoma of buccal mucosa, its presentation, the dilemma in diagnosis, and management. We emphasize the need to include this rare entity in the differential diagnosis of buccal mucosal swellings.

Keywords: *Buccal mucosa, peripheral ameloblastoma, per-oral resection*

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Background

Extraosseous ameloblastoma is the extraosseous counterpart of the usual well known central ameloblastoma that occurs within the jaws. This entity was initially mentioned in literature by Kuru in 1911 when he described a case of intraosseous ameloblastoma that had eroded alveolar bone and clinically presented as a "peripheral ameloblastoma." However, the first true peripheral ameloblastoma was reported by Stanley and Krogh in 1959.

PA has a reported incidence of only 1%–5% of all ameloblastomas.[1] The sites usually involved are the gingiva, lips, and palate with the most common site being the lingual gingiva of the mandible.

Literature search reveals only a few reports of peripheral ameloblastoma in the buccal mucosa.

We report a case of extragingival peripheral ameloblastoma of the buccal mucosa.

Case Report

A 55-year-old female reported to us in the department of the head-and-neck oncology with complaints of swelling in the right cheek for the past 1 year which was gradually increasing in size. She gave no history of pain, bleeding associated with the swelling. She was a known hypertensive and on medication. She also gave a habit history of tobacco chewing for the past 20 years. On local examination, approximately 4 cm × 4 cm swelling is noted over the right lower cheek overlying the masseter muscle region. Intraorally, submucosal swelling was noted in the right buccal mucosa with minimal induration and no ulceration. There was no cervical lymphadenopathy. A computed tomography (PNS) showed a peripheral enhancing lesion in the right cheek, measuring 3.2 cm × 2.8 cm with no sclerotic or lytic lesion within it [Figure 1].

A guided biopsy from the lesion was done, which reported scant foci of atypical cells arranged in sheets and nested patterns, and excision biopsy was suggested for further characterization.

The clinical findings, imaging, histology were discussed in a multidisciplinary tumor board with broad differentials, and the surgery was planned. The tumor was excised with adequate margins intraorally, under general anesthesia, and all aseptic precautions. Intraoperatively, the mass was encapsulated adherent to masseter muscle, with a well-maintained plane with the mandible, maxilla, and zygoma [Figure 2]. The excised specimen was sent for histopathological evaluation.

Macroscopically, a single gray-brown nodular lesion measuring 3 cm × 3 cm × 2.8 cm, firm inconsistency was noted. The cut surface was grayish-white solid. Focally, the lesion abutted the inked surgical margin [Figure 3a].

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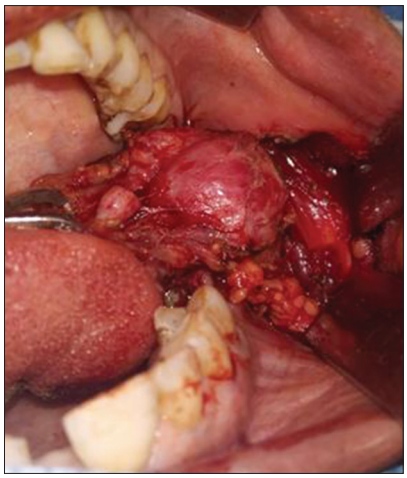

Figure 1: Intraoperative image of the lesion exposed perorally

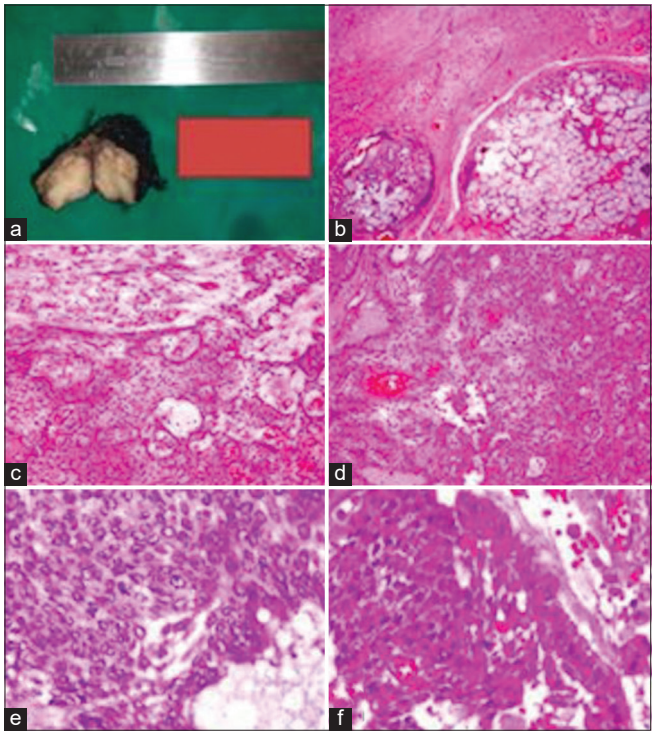

Figure 3: Gross and microscopic images. (a) Grey brown nodular tissue measuring 3 cm × 3 cm × 2.8 cm. Cut surface is greywhite homogenous; (b-f) Benign odontogenic tumor comprising of odontogenic epithelial island and sheets with peripheral palisading. Cells at basal layer show hyperchromasia with subnuclear vacuolation. Areas of unremarkable salivary gland tissue noted (b) (c-f: ×200; e and f: ×400)

Microscopically, the tumor was noted submucosally and showed benign odontogenic tumors comprising odontogenic epithelial islands and sheets with a peripheral palisading arrangement. Cells at the basal layer show hyperchromasia with subnuclear vacuolation. The central core shows stellate reticulum like cells with loosely arranged angular cells that show cystic change. No nuclear pleomorphism increased nuclear-cytoplasmic ratio, or mitotic figures were seen. All the features were suggestive of benign odontogenic tumor-ameloblastoma, extraosseous type [Figure 3b-f].

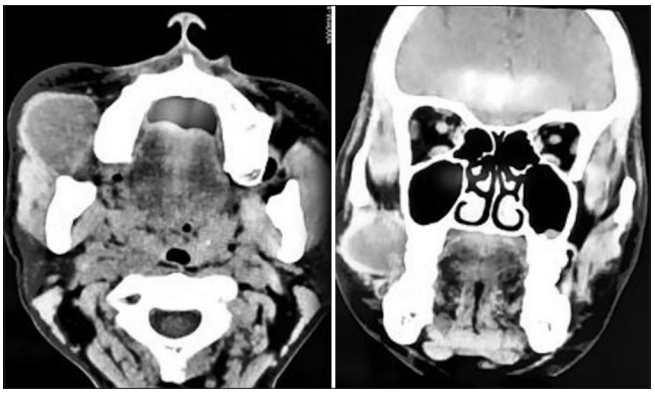

Figure 2: Computed tomography scan showing peripherally enhancing lesion in the right cheek

Discussion

Peripheral ameloblastoma, extraosseous type is a rare entity of the oral cavity categorized as a benign, odontogenic soft-tissue tumor.

The cells of origin in the nontooth bearing region have been postulated as the pluripotent cells of the basal layer of mucosal epithelium from minor salivary glands.

Clinically, these are slow growing and noninvasive in comparison to the central ameloblastoma that is locally invasive causing destruction of bone. The periosteum probably acts as the barrier for tumor invasion into the bone in peripheral ameloblastoma.

This lesion afflicts a higher age group, i.e., an average age of 52.1 years according to Philipsen *et al*. who have done a literature survey of 160 cases.[2] This is a painless, firm neoplasm with exophytic growth. It may also appear as an ulcerated lesion due to trauma from cheek biting. The size ranges from 0.3 to 4.5 cm in diameter with a mean of 1.3 cm.[3] Bone invasion is not a feature of peripheral ameloblastoma although sometimes, saucerization of bone may be noted.

Due to its clinical picture, it may be misdiagnosed clinically as a fibrous epulis, pyogenic granuloma, and peripheral giant-cell granuloma.

The WHO classification of tumors does not contain a histological definition of this rare entity.[4] Histological features of the peripheral ameloblastoma are overlapping with that of basal cell carcinoma.[5] A literature review of 53 cases of peripheral ameloblastoma concluded that there were no distinctive histomorphological features to differentiate between peripheral ameloblastoma and basal cell carcinoma.[6]

The various patterns of histologic appearance commonly noted in peripheral ameloblastoma are similar to that of central ameloblastoma with the acanthomatous pattern reported most commonly. However, granular cell and desmoplastic variants have also been described.[7]

The mainstay of treatment is surgical excision with wide margins and regular follow-up is advised as the recurrence rate reported in the literature is 16%–20%.[2]

Whether this recurrence could be attributed to an incomplete removal of tumor or the biological behavior of tumor *per se* is unclear.[3]

The aim of this case report is to highlight on the clinical, radiological presentation of this rare entity that mimics other lesions in the oral cavity, leading to a misdiagnosis. Histopathological evaluation is the gold standard to be able to reach the diagnosis. It has also been suggested in the literature that, microarray analysis could further help in differentiating this tumor from basal cell carcinoma with which it resembles histologically.[4] The treatment of choice is complete excision with negative margins.

The extraosseous variant of ameloblastoma is a rare entity of buccal mucosal pathologies and it is imperative that the clinicians consider it in the differentials when evaluating buccal mucosal swellings.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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