

Peripheral Ameloblastoma –A Unique Presentation

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Abstract: Peripheral ameloblastoma (PA) is a benign extraosseous odontogenic soft tissue tumour that is found confined to the gingiva or alveolar mucosa. Although PA is less aggressive than other classical subtypes it has the same histological characteristics of intraosseous ameloblastoma. We report a clinical case of PA of alveolar mucosa covering the left posterior maxillary tuberosity having the clinical presentation of an ulcer thereby highlighting the importance of histological examination in diagnosis.

Keywords: Benign, Peripheral ameloblastoma, Odontogenic tumour, Palatal lesions

I. Introduction

Peripheral ameloblastoma (PA) is a rare, benign, extra osseous odontogenic tumour that is believed to arise from remnants of the reduced enamel epithelium, cell rests of the dental lamina, or from basal cells of the surface epithelium⁽¹⁾. It was first reported in literature by Stanley and Krogh in 1959⁽²⁾. The PA mostly presents clinically, as a slow-growing, firm, painless mass which is either sessile or pedunculated with a smooth surface and the overlying mucosa appears normal. It is usually confined to the gingiva or alveolar mucosa and can cause a depression of the underlying bone or exhibit a “cupping” effect due to pressure resorption⁽²⁾. PA shows a male predilection (2:1) and lesions have a higher incidence in the mandible than in the maxilla (2.4:1). They occur most commonly in the mandibular lingual premolar region followed by the mandibular anterior region. The most common site of involvement in the maxilla is the soft palatal tissue adjacent to the maxillary tuberosity⁽²⁾. The purpose of this paper is to present a case of PA that occurred in the left posterior alveolar mucosa covering the maxillary tuberosity.

II. Case Report

An 87-year-old male reported to Department, presented with a complaint of a painless ulcer on the left side of upper jaw (Figure 1). The lesion had initially been noticed as a small painless nodular mass which exhibited gradual and slow growth over the last decade. About six months ago the patient inadvertently bit the swelling causing an ulceration which has not since resolved. He reported no accompanying motor, sensory or neural disturbances or any other symptoms, except for a mild discomfort on consumption of hot and spicy foods. The patient's medical history revealed that he was a diabetic for the last twenty six years and is on oral hypoglycaemic drugs. He had undergone total extraction due to periodontitis seventeen years ago and had never undergone prosthetic rehabilitation. His family and personal history were non-contributory. On general examination the patient appeared to be of moderate build and nourishment and the vital signs were all within normal limits. No gross facial asymmetry or abnormalities were noted on extraoral examination (Figure 1) and the lymph nodes were non tender and not palpable.

On intraoral examination a solitary ulcer was noticed on the left posterior part of the hard palate (Figure 2) over the maxillary tuberosity area. On inspection the ulcer was roughly ovoid in shape found to be approximately 1.5cm x 1cm, with well-defined margins and surrounded by normal mucosa. The floor of the ulcer appeared to be covered by a thick fibrinous exudates and yellowish slough. The findings during inspection were confirmed on palpation and the ulcer was found to be tender, the edges sloping and the base was firm in consistency. Apart from the presence of the exudate no discharge or bleeding was elicited during palpation. Hard tissue examination revealed that completely edentulous upper and lower arches.

Maxillary left lateral occlusal radiograph (Figure 3), OPG (Figure 4) and posteroanterior skull (PA skull) (Figure 5) radiographs doesn't reveal any bony involvement in relation to the lesional area. We did an incisional biopsy of the lesion and the histopathologic sections showed islands of odontogenic epithelial cells showing ameloblast-like peripheral cells with polarized nuclei and central reticulum network (Figure 6). The histopathological examination revealed orthokeratinized stratified squamous epithelium in association with a fibrovascular connective tissue along with underlying connective tissue containing follicles and strands of odontogenic epithelium. The follicle exhibited peripheral ameloblast like columnar cells with reversal of polarity. There was also evidence of squamous metaplasia and keratin pearl formation. The neoplastic odontogenic epithelial islands were seen very close to the lower border. Based on these characteristics the lesion was histopathologically diagnosed as peripheral ameloblastoma.

As the patient is not willing for an excisional biopsy the patient is under close follow up and as of now lesion does not show any change in size.

III. Discussion

Odontogenic tumors refers to a group of lesions that originate from the teeth-forming tissues, i.e. the epithelial or the ectomesenchymal component or from both. These unique pathologies of the jaws arise mostly within jaw and rarely within the soft tissues of jaw as seen in our case. Peripheral ameloblastoma accounts for 28.9% cases of peripheral odontogenic tumor and is the second most common peripheral odontogenic tumor, preceded by odontogenic fibroma⁽⁷⁾. Their clinical department can vary from innocuous non-neoplastic hamartomas to aggressive tumours, such as central ameloblastoma. Peripheral ameloblastoma is a rare extraosseous variant of central ameloblastoma that mimics the histologic characteristics of its intraosseous counterpart but occurs in the soft tissues. It usually occur without bone involvement, except for superficial erosion or depression of the bone crest called as saucerization" or "cupping" effect^(3,5).

Peripheral ameloblastomas usually originate from remnants of the dental lamina or from basal cells of the oral epithelium^(6,7). Cytogenetic study of a case of peripheral ameloblastoma found some genetic alteration in the patient and reported trisomy 7, differently from the intraosseous counterpart of ameloblastoma in which loss of chromosome 22 has been described. This unveils the fact that tumorigenesis of these lesions is different^(8,9).

The peripheral ameloblastoma has increased predilection of occurrence in the mandible than in the maxilla. In mandible lingual gingiva area, followed by the anterior area and in the maxilla, soft palatal tissue of the tuberosity area is most affected^(1,2,10). In our case palatal tissues over the maxillary tuberosity was affected. There are reports of misdiagnosed extralingival peripheral ameloblastomas - buccal mucosa and floor of the mouth - but most of the lesions later checked them into other categories of peripheral tumors such as basal cell adenoma or an ameloblastoid variant of a misdiagnosed squamous cell carcinoma^(1,11,12). Peripheral ameloblastoma which frequently occur between the fifth and the seventh decades of life has increased males predilection than females (1.9:1)⁽²⁾. In our case a male patient in the eighth decade of life presenting as an ulcer in the left maxillary tuberosity area.

Any peripheral hyperplastic swelling superficial to the alveolar ridge, such as peripheral hyperplastic fibroma, pyogenic or peripheral giant-cell granuloma and fibroma should be included in the differential diagnosis of peripheral ameloblastomas^(1,6). Performing a detailed imaging survey of patients with peripheral ameloblastoma is extremely important to exclude bone involvement by the tumor mass, which could otherwise culminate in misdiagnosis, recurrences and modification of the patient prognosis. In the imaging studies, this patient doesn't exhibit any involvement of the bone tissue. The involvement of cortical bone by the peripheral tumor, can lead to doubt regarding the biological behaviour, whether it is a hamartoma or persistent hyperplasia rather than a neoplasia^(1,13).

The histological differential diagnosis of the lesion are peripheral squamous odontogenic tumor, peripheral odontogenic fibroma, odontogenic gingival epithelial hamartoma and intraoral basal cell carcinoma^(2,14). The most common histologic types of PA are follicular and acanthomatous patterns^(1,10). Other histologic features rarely encountered are the calcifications, dentinoid, bone-like or cementum-like material⁽¹⁾. The acanthomatous type is well known for presence of calcifications^(13,15-17), although it is is not characteristic of peripheral ameloblastomas⁽¹⁾. The lesions which shows dystrophic calcification might be a sign of a potentially malignant feature^(13,17) as the earlier reported cases showed features of ameloblastic carcinoma. Occasionally, calcification may be a consequence of an intense apoptotic activity, and the terminal

differentiation of keratinocytes is considered to be a variant of the apoptotic process^(20,21). As the present case did not show any signs of malignancy, the latter supposition is more likely to be the cause of the calcifications. Several studies have demonstrated that peripheral ameloblastomas may present indolent behaviour compared with the central variant^(10,22,23). Thus, the treatment of choice is a conservative supraperiosteal excision with adequate disease-free margins^(1,4). Tumor recurrence is uncommon and, although very rare, some peripheral ameloblastomas were reported to undergo malignant transformation into ameloblastic carcinoma^(24,25). In the present case, the treatment consisted of surgical resection of the tumor.

IV. Conclusion:

Although the current case had clinical appearance of reactive ulcerative lesions, only the microscopic examination revealed the diagnosis of PA. Therefore, it is important to include peripheral ameloblastoma as a differential diagnosis of ulcerative lesions palatal mucosa. In short appearances can be deceiving and so it is imperative that we do histopathologic examination in all cases as far as possible.

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Legends



Figure 1:extraoral view of patient

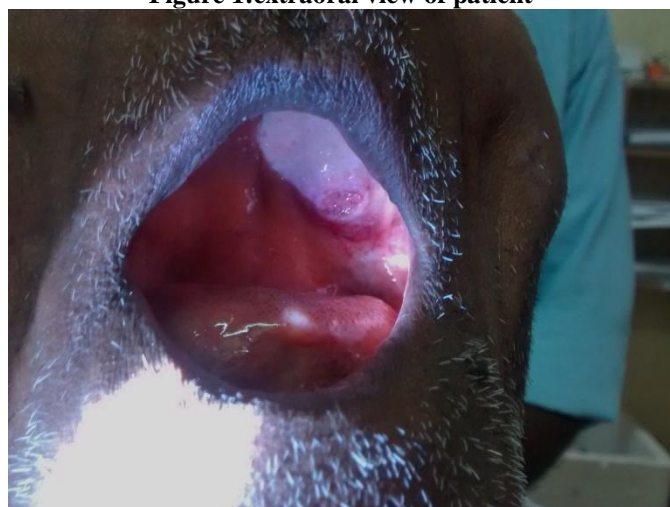


FIGURE 2: intra oral view of the lesion



Figure 3: maxillary occlusal radiograph

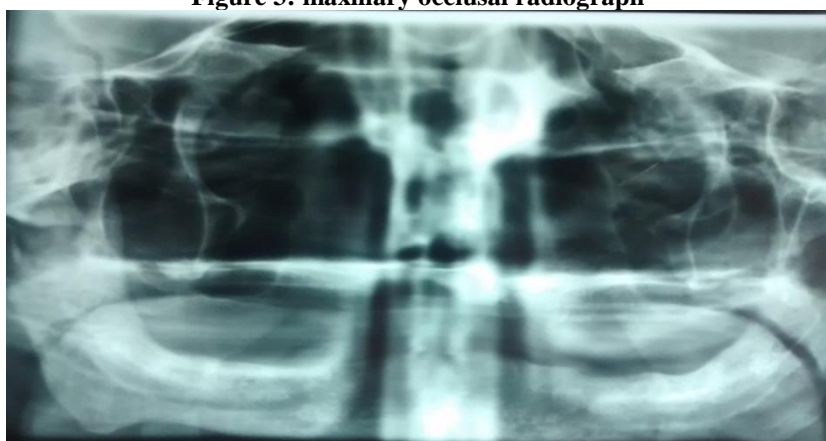


FIGURE 3:orthopantamogram of the patient



FIGURE 4: skull view