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Peripheral Ameloblastoma – An Incidental Finding in A Patient with Intraosseous Ameloblastoma: A Case Report and Review of Literature

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Abstract

Peripheral Ameloblastoma (PA) is a benign odontogenic tumor, arising from the cell rest of Serres, reduced enamel epithelium and basal cells of the surface epithelium. It is a rare odontogenic neoplasm occurring commonly in the mandibular gingiva that clinically resembles other peripherally occurring lesions like pyogenic granuloma, peripheral ossifying fibroma, peripheral giant cell granuloma, and papilloma. We report a case of peripheral ameloblastoma on the left side of lower jaw which was identified on subsequent reviews in a 32-year-old male patient who underwent enucleation for intra-osseous ameloblastoma on right side of mandible previously in our institution.

Keywords: Peripheral ameloblastoma, ameloblastoma, odontogenic tumor, molecular markers.

Introduction

Ameloblastoma is a benign odontogenic tumor with local destruction and recurrence potential arising from the cell rest of Serres, reduced enamel epithelium and basal cells of the surface epithelium(1). The three types are solid/multicystic ameloblastoma, unicystic ameloblastoma and the peripheral ameloblastoma(2). In general, peripheral (extraosseous or soft tissue) odontogenic tumors are rare lesions that occur in the soft tissue overlying the tooth-bearing areas of the mandible and the maxilla. Therefore, peripheral ameloblastoma is a rare entity and accounts for 1% of all the types of

ameloblastomas(3). It is a lesion of the soft tissues solely but presents with the histologic features of intra-osseous ameloblastoma. Another lesion that appears in the gingiva with histological resemblance to peripheral ameloblastoma is the basal cell carcinoma(4). However, some consider it to represent the same lesion with no change in surgical treatment.

Here we report a case of peripheral ameloblastoma on the left side of lower jaw which was identified on subsequent reviews in a patient who underwent enucleation for intra-osseous ameloblastoma on right side of mandible previously in our institution under General Anaesthesia.

Case report

A 32-year-old male patient presented with complaints of a swelling over the gingiva for a period of one month with no progressive increase in size or pain. On examination, he had a diffuse swelling over the buccal gingiva of left cuspid mandibular region. The mucosa over the lesion was normal with a smooth surface and firm consistency. On further history taking, patient revealed about his previous surgery where he was operated on right cuspid region of mandible for unicystic ameloblastoma by enucleation and chemical cauterization with Carnoy's solution under General anaesthesia an year back. Patient did not have regular follow up due to personal reasons. Six months later, he presented with this lesion as he suspected a recurrence owing to the slight increase in size. There was no relevant past medical history or allergy to known drugs. No cervical lymphadenopathy was present on examination.

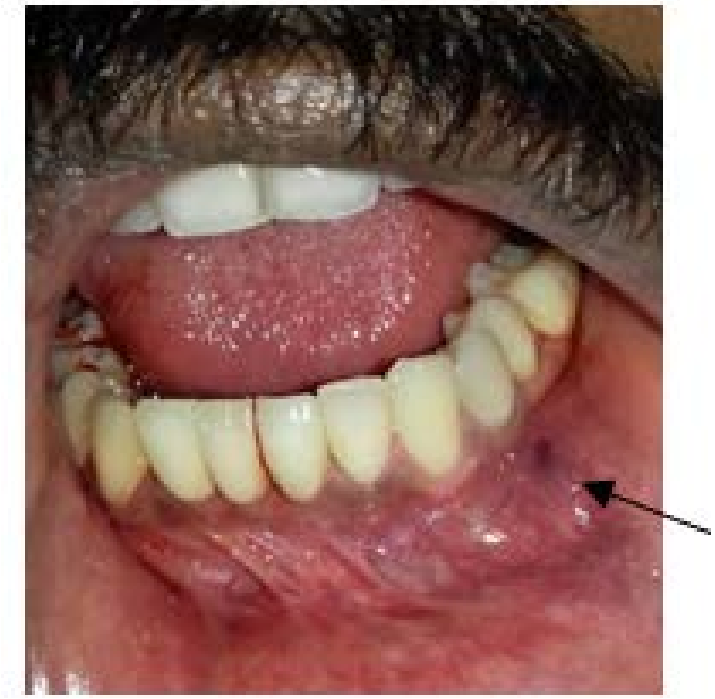

Fig. 1: Clinical picture showing a diffuse expansile lesion with obliteration of buccal vestibule over the left mandibular cuspid region.

OPG showed a diffuse radiolucent area in the operated site. On comparing the post-enucleation and pre-enucleation radiographs, it was understood that the lesion was healing with satisfactory bone formation. No evidence of abnormality suggesting the suspicious lesion on left side was detected. Mandibular occlusal radiograph also did not reveal any obvious lesion.

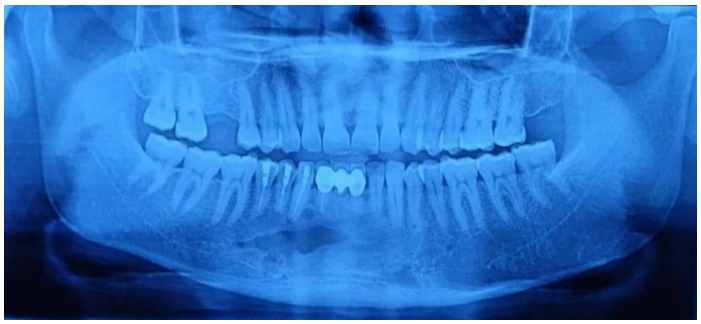

Fig. 2: OPG showing a healing lesion on the right cuspid region with no alteration of bone architecture in left cuspid region where the lesion was suspected.

On surgical exploration, bone was normal whereas the corresponding soft tissue upon dissection yielded a nodular lesion which was firm of size 1 cm. The specimen was sent for biopsy.

Fig. 3: Lesion on surgical exploration

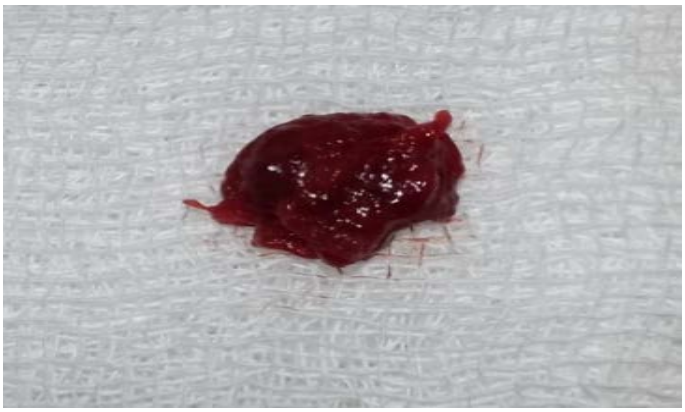

Fig. 4: Lesion post-excision

Microscopic examination of the lesion showed moderate to densely collagenous connective tissue stroma with rests of hyperchromatic cells resembling odontogenic epithelium and few follicles exhibiting peripheral columnar cells with nuclear hyperchromatism, subnucleolar vacoulisation and reverse polarity. A few other follicles exhibited squamous metaplasia within. Flecks of basophilic calcifications were also seen. Vascularity was moderate and stroma showed some muscle fibers. The final histopathological diagnosis was peripheral ameloblastoma.

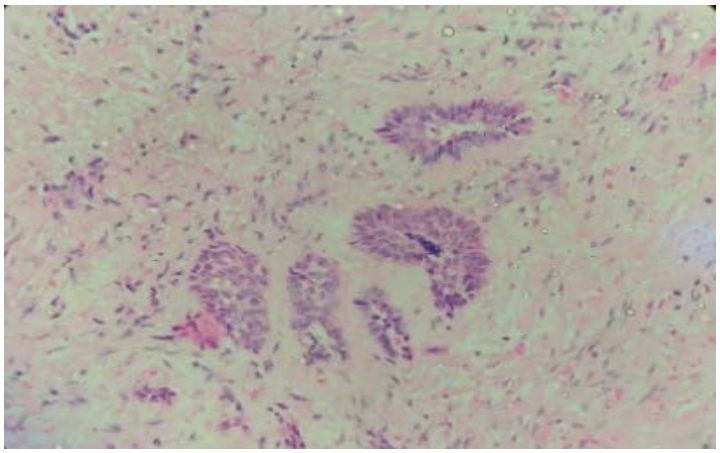

Fig. 5: Ameloblastomatous follicles seen in 10x view.

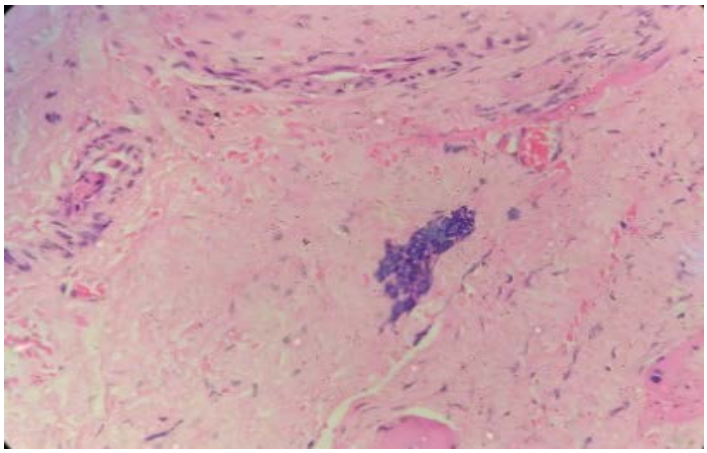

Fig. 6: Odontogenic rests seen in the lesion.

According to the clinical, radiological and histopathological exams the final diagnosis of peripheral ameloblastoma was made. The patient was kept under follow up and the surgical site healed satisfactorily. Follow up x-rays showed no suspicion after 6 months.

Discussion

The prevalence of peripheral ameloblastoma is 1% of all ameloblastomas(3). The other terms for peripheral ameloblastoma are extraosseous ameloblastoma, soft tissue ameloblastoma and ameloblastoma of mucosal origin. The first case of peripheral ameloblastoma was reported in literature by Kuru in 1911(1). In 1959, Stanley and Krogh defined the clinical and histopathologic characteristics. It is typically a slow, benign, single, sessile, asymptomatic lesion. The most probable source of this lesion is the remnants of dental lamina which are

called as the 'glands of Serres'. Of the three types of ameloblastoma, PA is striking for its location. It can be seen as an exophytic nodular lesion over the gingiva usually in the cuspid region with firm consistency and painless most of the times. The histopathological features are the same as central/intraosseous ameloblastoma. Although both tumors have histological and morphological similarity, they entirely differ in their behavior and character as the Peripheral Ameloblastoma is less invasive locally and less recurrent. In contrast to the intraosseous form that destroys the hard tissue, Peripheral Ameloblastoma does not behave aggressively and the dense fibrous tissues of the gingiva and the periosteum may act as an effective barrier preventing its further infiltration(2). Usually there is no radiological sign associated with the lesion except for some cupping effect/ saucerization of the underlying bone.

The molecular markers that have been studied in ameloblastoma are given in Table 1(5). Moreover, it has also been studied about BRAF and SMO gene mutations in ameloblastoma which suggest multiple gene mutations in solid ameloblastoma and single somatic mutation in case of peripheral ameloblastoma(6). Immunohistochemically, Peripheral Ameloblastoma shows positive reactivity for AE1/AE3, KL1,34, E12, and MNF116 cytokeratin and negative staining for CK8, CK10, CK13, CK17, and CK18(7). Ameloblastoma whether central or peripheral, are essentially devoid of CK7, CK8, CK10, CK18, CK20 and epithelial membrane antigen(8–10).

The differential diagnosis for Peripheral Ameloblastoma includes pyogenic granuloma, peripheral giant cell granuloma, peripheral odontogenic fibroma, peripheral ossifying fibroma, papilloma, and epulis(11). Only few cases of malignant PA have been reported in literature to date. First case of Peripheral Ameloblastoma with malignant transformation was reported in literature by Edmondson et al(12). Local excision including a 1–2 mm margin of healthy tissue remains the treatment of choice and local recurrence is very less.

Table 1: Molecular Markers In Ameloblastoma

Marker	Expression	Indicates
CD 133, Bmi-1, ABCG2	Increased	Oncogenesis, cell differentiation and malignant potential
PCTH 1	Increased	Proliferation of odontogenic epithelium
GLi 1	Increased	Cell proliferation
β Catenin	Increased	Cell to cell adhesion and signal transduction in odontogenic epithelium
BMP – 2,4,7	Increased	Cell proliferation, differentiation, chemotaxis, extracellular matrix production and apoptosis
TGF – β	Activated	Aggressivenss
Syndecan – 1	Decreased	Aggressiveness, invasiveness
Cadherins	Decreased	Aggressiveness
Integrins	Increased	Invasiveness
Claudin 7	Decreased	Invasiveness
Podoplanin	Increased	Invasiveness, tumor metastasis
MT	Increased	Invasiveness, anti-apoptotic and high recurrence
Ki – 67	Increased in peripheral cells	Aggressiveness, cell proliferation

Cyclin D1	Increased	Invasiveness, aggressiveness, poor prognosis and lymph node metastasis
Telomerase	Increased	Oncogenesis, cell proliferation
PCNA	Expressed during late G_1	Aggressiveness, recurrence and malignant potential
FGF -7, 10	Increased	Growth of tumor
Bcl-2, bcl-X	Peripheral cells	Anti-apoptotic, cytodifferentiation
Caspase 3	Central area of tumor islands	Cell death
PTEN	Increased	Aggressiveness
MMP - 1,2,9	Increased	Tumor invasion
PTHrP	Increased	Infiltrative growth and destructive behavior
RANKL	Increased	Osteoclastogenesis tumor expansion
OPN, CD44v6	Increased	Invasiveness, cell migration
CD10	Increased	Growth of neoplastic cells, recurrence
p53	Increased	Increased cellular proliferation and malignant potential

Conclusion

Peripheral ameloblastoma is an uncommon odontogenic tumor. Proper clinical examination with inclusion of peripheral ameloblastoma in the differential diagnosis of any peripheral neoplasm is required. Further confirmatory test is histopathological support of Peripheral Ameloblastoma and immunohistochemistry is additionally helpful in the accurate diagnosis. Although the recurrence rate is less, thorough microscopic evaluation is needed to evaluate for the excision margins along with follow up to avoid future recurrence.

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