Case Report

Extraosseous ameloblastoma of maxillary gingiva: A rare case

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ABSTRACT

Extraosseous ameloblastoma (EA) is a rare odontogenic tumor which comprises about 1% of all ameloblastomas. Extraosseous location is the peculiar feature of this type of tumor, which is otherwise similar to the classical ameloblastoma. The exact etiology of EA is not yet known. The clinical appearance may vary; it presents as a slow-growing, firm, painless mass with a sessile or pedunculated base with a smooth surface. The radiographic findings are nonspecific; diagnosis is based on histopathological features. It poses a diagnostic and therapeutic challenge due to its morphology and biological behavior. This article aims to elucidate rare case of EA of maxillary gingiva in a 40-year-old male patient and also an attempt has been made to discuss its pathogenesis in light of current information from the literature.

Key words: Ameloblastoma, extraosseous ameloblastoma, peripheral ameloblastoma, peripheral odontogenic tumor

INTRODUCTION

Ameloblastoma is the most common benign neoplasm of odontogenic epithelial origin. It usually occurs in the jaw bones and surrounding tissues. It can be classified into three subgroups: Intraosseous multicystic, intraosseous unicystic, and extraosseous ameloblastoma (EA).[1] EA was first reported by Kuru in 1911. Stanley and Krogh defined the clinical and histopathologic characteristics in 1959.[2] The lesion often grows without symptoms and presents as a painless swelling. Although it is usually confined to the gingiva or alveolar mucosa, it may cause a depression of the underlying bone or exhibit "cupping" effect due to the pressure resorption.[3] This case is unique since it involves maxillary gingiva, whose clinical findings were non-specific.

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The purpose of this paper is to present an unusual case of EA and to discuss its pathogenesis and controversies.

CASE REPORT

A 30-year-old male patient reported with a complaint of painless swelling in the right maxillary gingival region since 1 month. Swelling started as a nodule and progressed to size of 1 × 1 cm. On examination, there was a well-defined, sessile, non-tender swelling with respect to right maxillary canine and first premolar. Overlying mucosa was smooth, stretched with shiny appearance [Figure 1].

Based on history and clinical examination differential diagnosis of pyogenic granuloma, peripheral giant cell granuloma and peripheral odontogenic fibroma were considered. Intraoral periapical radiographs and panoramic radiographs revealed no bony changes. Surgical excision was done under local anesthesia and specimen on histopathological

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examination revealed islands of odontogenic epithelium with peripheral tall columnar cells with well-polarized nuclei and loosely arranged stellate reticulum like cells in the center suggestive of EA [Figures 2 and 3]. The course has been uneventful after the surgical removal. The patient was advised to visit regularly for examination. After 2 years of follow-up, no recurrence was seen.

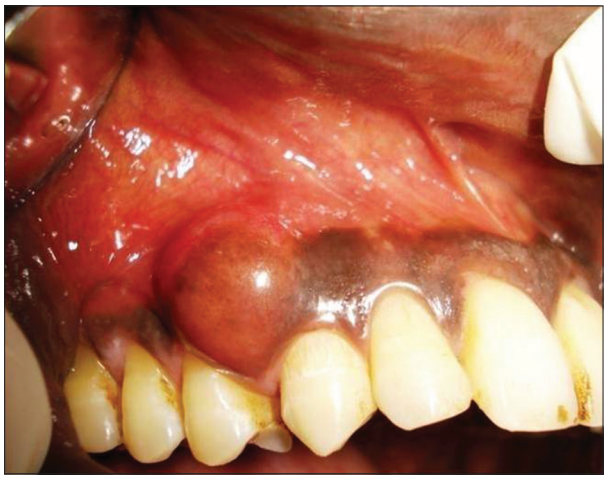

Figure 1: Photograph showing well-defined nodular swelling in maxillary gingiva

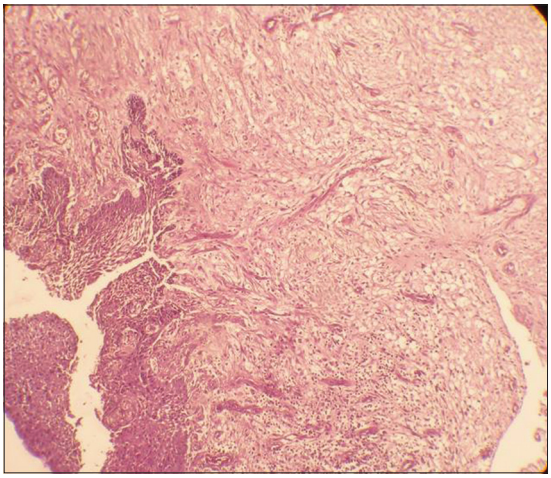

Figure 2: Photomicrograph (×4) shows intact overlying squamous epithelium and odontogenic islands

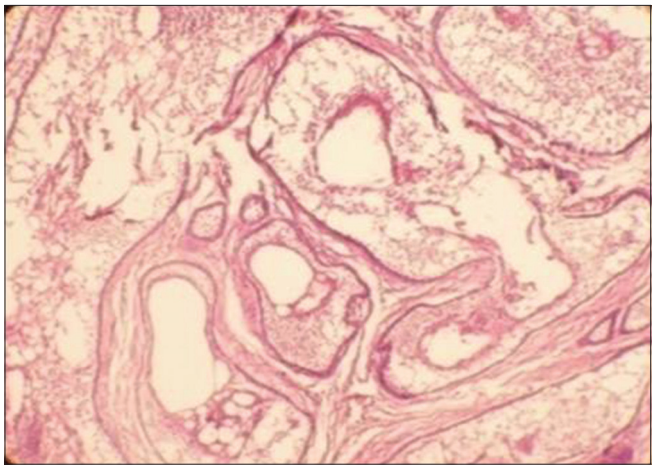

Figure 3: Photomicrograph (×40) shows odontogenic islands lined by tall columnar cells and stellate reticulum-like cells in the center

DISCUSSION

EA is also known as the peripheral ameloblastoma, soft tissue ameloblastoma, ameloblastoma of mucosal origin, or ameloblastoma of the gingiva.[4] Kuru reported the first case of EA in 1911.[5] However, what Kuru described was not a peripheral but rather an intraosseous ameloblastoma having penetrated through the alveolar bone, fused with the oral epithelium and eventually presenting itself clinically as a "peripheral lesion." The first completely documented case of an EA was described by Stanley and Krogh, who defined the clinical and histopathologic characteristics in 1959.[1]

The incidence of EA was reported between the fifth and sixth decades of life. The average age of presentation is 50 years, with a male predominance.[1,6] The present case was reported in a 30-year-old, and this was not in accordance with the literature. EA is typically seen in the retromolar aspect of the mandible or mandibular premolar region followed by the anterior mandible.[3] Other less common region includes the tuberosity of the maxilla. The present case was reported in maxillary canine gingival region which is very rare.

EA originates from two major sources. Those lesions that are located entirely within the connective tissue of the gingiva, showing no continuity with the surface epithelium are very likely to arise from remnants of the dental lamina located in the soft tissues overlying the tooth-bearing areas of the jaw bones. Others may show continuity between the tumor and the superficial epithelium, suggesting a basal cell origin of the tumor.[5]

EA typically presents as a painless exophytic growth of the gingiva with a relatively smooth surface, similar to the present case. However, in several cases, it has been described as "granular," "pebbly," "papillary," or "warty" in appearance. Color of the lesion can range from pink to dark red. During mastication, the EA may become traumatized, and the lesion may thus show an ulcerated surface or may appear keratotic. Usually, the tumor appears as a single lesion, but Gharat *et al*. have described a case which occurred in two different areas of the oral mucosa.[1] A superficial bony depression known as "cupping" or "saucerization" may be noticed radiographically, or at surgery, which is thought to be due to pressure resorption, in contrast to resorption caused by neoplastic invasion.[1,7] Histologically, the tissue reveals an ameloblastic growth under an intact layer of overlying squamous epithelium. The most common

histomorphological cell pattern types are plexiform or follicular. The follicular type is composed of islands and strands of odontogenic epithelium exhibiting columnar ameloblastic cells with well-polarized nuclei and a central mass of loosely connected stellate reticulum-like cells. EA may also exhibit various histological patterns similar to those seen in intraosseous ameloblastoma. The epithelial islands commonly exhibit the squamous metaplasia, with central areas of cyst formation[6] or keratin formation[7] some of the squamous cells in these acanthomatous nests may show ghost cell formation and foreign body reaction to this material within the connective tissue, which is referred to as "ghosting," features associated generally with the calcifying odontogenic cyst.[3] However, now granular cell[8] and desmoplastic[9] and clear cell variants of peripheral ameloblastoma have also been reported; in our case it was the follicular type with an intact layer of squamous epithelium. Peripheral odontogenic fibroma, peripheral squamous odontogenic tumor, and odontogenic gingival epithelial hamartoma are considered in histopathological differential diagnosis. Immunohistochemicaly EA is positive for AE1/AE3, CK8, CK13, CK18, CK14, and CK19.[1,9]

Although EA is considered to be a benign tumor, seven cases of a malignant variety have also been reported. Metastasis has also been reported in one case. Malignant EA has also been reported to arise *de novo* or as dedifferentiated carcinomas from a preexisting benign EA. According to Nagai's classification, EA can be classified into two subtypes: Malignant (metastasizing) EA and peripheral ameloblastic carcinoma.[1,3]

The biological behavior of EA continues to be controversial. The innocuous behavior of the EA seems to be similar to that of a hamartoma rather than a neoplasia. However, some tumors display significant invasive capacity and recurrence potential. Cases of very rare malignant varieties of EA have been published, which showed the typical hallmarks of malignancy.[3] EAs are treated by local soft tissue excision to achieve 2–3 mm margin of normal tissue. The inferior margin should include the periosteum. The recurrence rate of EA is 16–19%.[1,3,10]

CONCLUSION

EA is the uncommon epithelial odontogenic neoplasm. It may arise from dental lamina, developing enamel organ, or basal cells of the oral mucosa. It is important for the dental practitioner to be aware of the clinical features which can lead to an early diagnosis and treatment of this lesion. It poses a diagnostic and therapeutic challenge due to its morphology and biological behavior. EA deserves high academic interest because of its pathogenesis and variable clinical presentation.

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Conflicts of interest

There are no conflicts of interest.

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