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Case Report

Peripheral ameloblastoma of the mandible: A case report

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ABSTRACT

The extra-osseous/peripheral ameloblastoma (PA) is a relatively rare subtype of ameloblastoma, which develops in the soft tissues of the gingiva and oral mucosa. In this article, we describe a PA found in the gingiva of the right mandibular premolar region in a 70-year-old male. On oral examination, a firm, pedunculated mass with a granular surface and partial ulceration was found on the lingual gingiva of the right mandibular first premolar to the first molar. The pathological diagnosis of the incisional biopsy specimen was highly suspected to be PA with difficulty to differentiate from basal cell carcinoma (BCC). As the clinical history and features could not help to rule out the possibility of BCC, en bloc resection with safe margins was performed. Immunohistochemically, the tumor cells showed positivity for CK19 and p63 but Ber-EP4 was negative for tumor cells. The final histopathological diagnosis was peripheral ameloblastoma.

1. Introduction

According to the 2005 World Health Organization classification, benign ameloblastoma is divided into four types: solid/multicystic, extra-osseous/peripheral, desmoplastic, and unicystic [1]. The extra-osseous/peripheral ameloblastoma (PA) develops in the soft tissues of the gingiva and mucosa and shows an innocuous clinical behavior. It comprises about 2–10% of all ameloblastomas. PA exhibits histopathological cell types and patterns similar to those of solid/multicystic type ameloblastoma [1,2]. Most cases of PA arise from the tooth bearing region and differential diagnosis include various inflammatory diseases and benign and malignant tumors. PA and basal cell carcinoma (BCC) have similar histological appearance and growth patterns and it is often hard to differentiate them [3]. In this article, we present a case of PA which was difficult to distinguish from BCC based on clinical features and histological examination of the initial biopsy specimen. Immunohistochemical staining of Ber-EP4 for diagnosis is also discussed.

2. Case report

A 70-year-old male was referred by his dentist to our clinic for evaluation of a painless gingival swelling in the right mandible. He noticed the swelling one year before and the swelling had gradually increased in size. He had a past history of angina pectoris and hypertension and these were well controlled by medication. On oral examination, a firm, pedunculated mass with a granular surface and partial ulceration was found on the lingual gingiva of the right mandibular first premolar to the first molar (Fig. 1). There was no obvious lymphadenopathy or other abnormalities in the head and neck region. Computed tomography (CT) demonstrated a mild depression of the lingual alveolar bone in the right mandibular premolar region (Fig. 2). Magnetic resonance imaging (MRI) showed a heterogeneous high signal intensity mass in T2-weighted images, measuring 15 mm × 10 mm and a low signal intensity mass in T1-weighted images (Fig. 3A and B). Peripheral bone invasion was not found. The pathological diagnosis of the incisional biopsy specimen was highly suspected to be PA with difficulty to differentiate from BCC because of the small size of the biopsy specimen. As the clinical history and features could not help to rule out the possibility of BCC, en bloc resection with safe margins was performed. The post-operative course was uneventful and there was no recurrence during the one-year follow-up period.

Histopathologically, the resected specimen showed that the tumor had continuity with the surface epithelium. The tumor islands consisted of a central mass of loosely connected stellate reticulum-like cells with acanthomatous areas surrounded by a

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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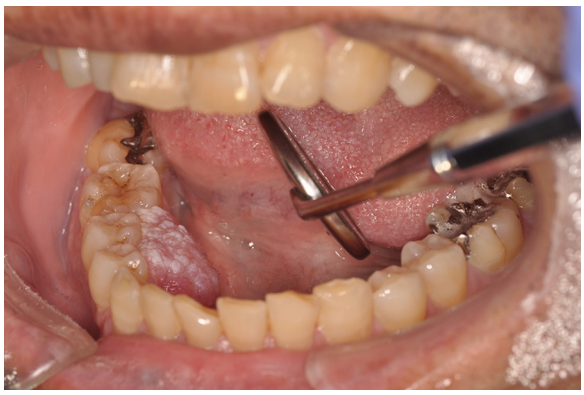

Fig. 1. Intraoral photograph of a painless firm and pedunculated mass with granular surface on the lingual attached gingiva of the right first premolar to the first molar in the mandible.

layer of columnar cells with well-polarized nuclei (Fig. 4A and B). Immunohistochemically, the tumor cells showed moderate positivity for CK19, strong positivity for p63 but Ber-EP4 was negative for tumor cells (Fig. 5A and B). The final histopathological diagnosis was peripheral ameloblastoma.

3. Discussion

Though PA and BCC exhibit similar growth patterns and histological features, they are supposed to be two distinct and separate entities [3,4]. PA is thought to arise from extraosseous rests of dental lamina (rests of Serres) beneath the oral mucosa or from surface epithelium having potential to differentiate into odontogenic epithelium, and BCC is thought to arise from pluripotent basal cells present within the surface epithelium and adnexal structures [1]. BCC is a malignant lesion; therefore, this should be the first consideration in differential diagnosis [3].

In most cases, PA is an exophytic growth localized to the soft tissues overlying the tooth-bearing area of the jaws. The age of the patients ranges from 16 to 92 years with a mean age of 50.2 years. About 50% of the lesions occurred between the ages of 40 and 60. The average size of the lesion measures between 1 and 2 cm. PAs are commonly seen in mandible, especially in the premolar region. The maxilla/mandible ratio was 1:2.4 [2]. In addition, the majority of mandibular PA cases occur in the lingual aspect of gingiva. In the maxilla, however, the most common location was the soft, palatal tissue of the tuberosity area. There was no difference in location between the left and right sides of the jaws [2]. In oral BCC, Woods et al. [3] reported that the mean age of occurrence was 66 years and male to female ratio was 1:2. The most common oral site was buccal mucosa.

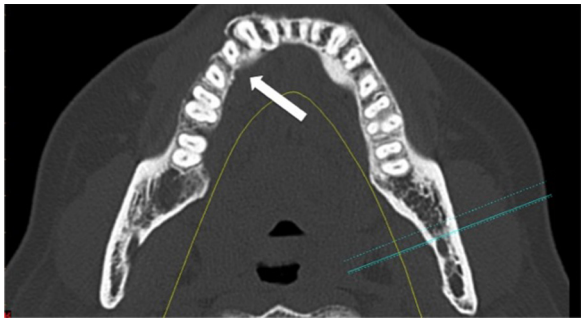

Fig. 2. Computed tomography (CT) demonstrating a mild depression of lingual alveolar bone in the right premolar region of mandible (arrow).

Regarding the clinical features, PA is usually painless and the surface is relatively smooth but, in several cases, it has been described as 'granular', 'pebbly', 'papillary' or 'warty' in appearance [4] and the irregular surface of PA might lead to a misdiagnosis of malignant disease [5]. In oral BCC, the surface commonly showed non-healing ulceration with or without pain [3]. Ulceration was observed in the present case, probably due to a traumatic bite.

In PA, no radiological evidence of bone invasion is usually detected. A superficial erosion of the bone or a superficial bony depression, known as "cupping" or "saucerization" that is thought to be due to pressure resorption may be noticed [4]. BCC may show local bone invasion [6]. Our case also showed a mild depression of lingual alveolar bone in the right premolar region of the mandible.

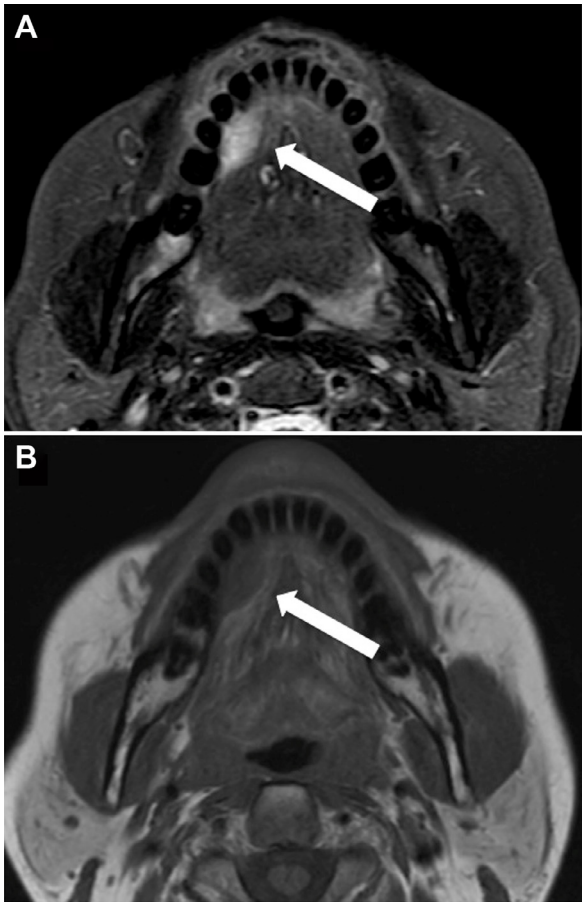

Fig. 3. Magnetic resonance imaging (MRI) findings. (A) A heterogeneous high signal intensity mass in T2-weighted images, measuring 15 mm × 10 mm (arrow). (B) A low signal intensity mass in T1-weighted images (arrow).

The histological feature of PA is identical to that of classic intraosseous ameloblastoma. PA exhibited typical odontogenic differentiation mimicking dental lamina or enamel epithelium connected with a basal layer of gingival epithelium [7]. The basal layer cells of the tumor islands of PA are well polarized in a reverse manner and arranged in a palisading fashion [8]. Most of the differential diagnoses can be ruled out by traditional histological examination. But it is often difficult to distinguish between PA and BCC based on histological examination because PA and BCC share similar histological features [3,4]. According to Woods et al., the main histological differences between BCC and PA are as follows: in BCC, the tumor arises from the surface epithelium, mitotic figures and apoptotic cells are scattered, mucoid ground substance is present and the tumor infiltrates widely throughout the connective tissue often exhibiting a prominent retraction artifact [3].

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