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Case Report

Secondary peripheral ameloblastic carcinoma of the palate: A case report and literature review

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ABSTRACT

Secondary peripheral ameloblastic carcinoma is an odontogenic malignant tumor that arises from benign peripheral ameloblastoma and exhibits cellular atypia. We describe the case of an 82-year-old male referred to our department with a hemorrhagic mass on the right palate that developed 6–7 years earlier. Clinical examination revealed a mass 40 × 30 × 10 mm in size affecting the hard palate that exhibited relatively clear boundaries, granular superficial appearance, and hemorrhaging. Computed tomography and magnetic resonance imaging revealed compressive resorption of the bone surface in contact with the tumor. A biopsy was performed; the histopathological diagnosis of the biopsy was peripheral ameloblastoma without any findings of malignancy. The patient subsequently underwent tumor resection. The resected specimen of peripheral ameloblastoma exhibited malignancy. The histopathological diagnosis was secondary peripheral ameloblastic carcinoma. The recurrence was not confirmed. However, CT imaging performed revealed nodular pulmonary shadows 14 months after surgery. We consulted with the Department of Respiratory Medicine, and a biopsy was planned. However, the biopsy was not performed because the patient was admitted for the treatment of pulmonary tuberculosis. A malignant lesion appeared in the liver 21 months after surgery. The patient developed multiorgan failure during that month and died. A review of seven reported cases demonstrated that our case is typical with regard to clinical course, symptoms, and clinical, imaging, and histopathological findings.

1. Introduction

According to the 2005 World Health Organization (WHO) classification of odontogenic tumors, secondary peripheral ameloblastic carcinoma is an odontogenic carcinoma that arises from benign peripheral ameloblastoma and exhibits cellular atypia on histopathological examination [1]. There are only six reported cases of this disease, and it seems extremely rare. Here we report the case of a patient with secondary peripheral ameloblastic carcinoma and perform a review of the literature.

2. Case report

2.1. Clinical course

An 82-year-old man was referred to our department in January 2013 with a hemorrhagic mass on the right palate that developed 6–7 years earlier. Although the mass had gradually enlarged, it was asymptomatic and remained untreated. The patient had a medical history of hypertension, hypertrophic cardiomyopathy, hyperuricemia, renal cancer, and anemia. Additionally, the patient had poliomyelitis as a child, causing equinus deformity of the right foot and gait disturbance.

Initial intraoral examinations revealed a solid mass lesion with clear boundaries on the patient's hard palate measuring 40 × 30 × 10 mm. The tumor was broad based, with a granular superficial appearance. It was soft on palpation and hemorrhagic (Fig. 1). There was no palpable cervical lymph node.

Contrast-enhanced computed tomography (CT) revealed a neoplastic lesion that was uniformly internally contrast enhanced, and there was compressive resorption of the bone surface in contact

☆ Asian AOMS: 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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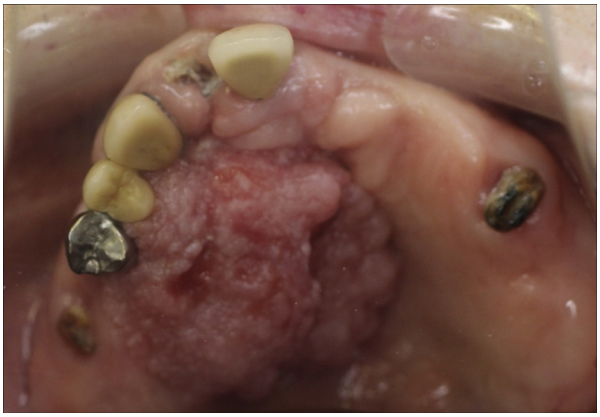

Fig. 1. Intraoral findings. We observed a 40 × 30 × 10-mm mass with clear boundaries affecting the hard palate.

with the tumor. The tumor showed the same signal intensity as the muscle on T2-weighted magnetic resonance imaging (MRI). No lymphadenopathy was evident in the submandibular and cervical areas (Fig. 2).

A cytology sample was taken by scraping the hemorrhagic area in the center of the lesion. The smear showed atypical epithelial cells, suggestive of epithelial dysplasia or carcinoma *in situ* and corresponded to Papanicolaou Classification IV; therefore, the tumor was considered malignant. In January 2013, a tissue biopsy of the anterior tumor was obtained under local anesthesia. The histopathological diagnosis was peripheral ameloblastoma (Fig. 3).

The patient underwent tumor resection under general anesthesia in the month of April of the same year. A safety margin was established 5 mm from the base of the tumor, and we resected it with the periosteum. Because the bone surface was smooth, intraosseous invasion was not suspected. After the outer layer of the bone surface was removed, the raw surface was covered with an artificial dermis composed of atelocollagen. Finally, a splint was installed to apply pressure. The histopathological diagnosis of the resected specimen was secondary peripheral ameloblastic carcinoma.

The patient was disease-free 14 months after surgery. However, CT imaging performed in June 2014 revealed nodular pulmonary shadows. We consulted with the Department of Respiratory Medicine, and a biopsy was planned to confirm whether the shadows reflected primary lung cancer or pulmonary metastases of the ameloblastic carcinoma. However, in the month of August of the same year, the patient was admitted to hospital for the treatment of pulmonary tuberculosis. Because his symptoms did not improve, the biopsy was not performed, and it was not possible to confirm a diagnosis. In January 2015, a malignant lesion appeared in the liver. The patient developed multiorgan failure during that month and died. Consent for autopsy was denied, and we were unable to determine whether the ameloblastic carcinoma was metastatic.

2.2. Pathological findings

A histopathological examination of the resected specimen revealed that the tumor was located extraosseously from the mucosal surface to the periosteum of the maxilla. The tumor mainly consisted of the islands of odontogenic epithelium within thin fibrous stroma accompanied by stellate reticulum-like structures in the center of nests. These histological findings led to a diagnosis of peripheral ameloblastoma; however, an area of high cell density 7 mm in size was evident in the center of the tumor. This area was clearly distinguishable from its surroundings and, within it, scattered mitosis was visible. Anisokaryosis and highly developed tumor cells that had lost polarization and were arranged in dense sheets were also evident (Fig. 4). On the basis of these histopathological findings, the tumor was diagnosed as secondary peripheral ameloblastic carcinoma.

3. Discussion

We reported the case of a patient with secondary peripheral ameloblastic carcinoma. According to the 2005 WHO classification of odontogenic tumors, ameloblastic carcinoma exhibits cellular atypia and can be classified into primary and secondary types [1]. The secondary type arises from benign ameloblastoma and can be further divided into intraosseous and peripheral types (Table 1).

Philipsen et al. re-examined previous reports on peripheral ameloblastoma and reported that malignancy was evident in six cases [2]. However, Lin et al. reported that one of the six cases exhibited no histopathological signs of malignancy; therefore, only five cases matched the current definition of this disease [3]. We searched PubMed for reports on the condition published since 2005

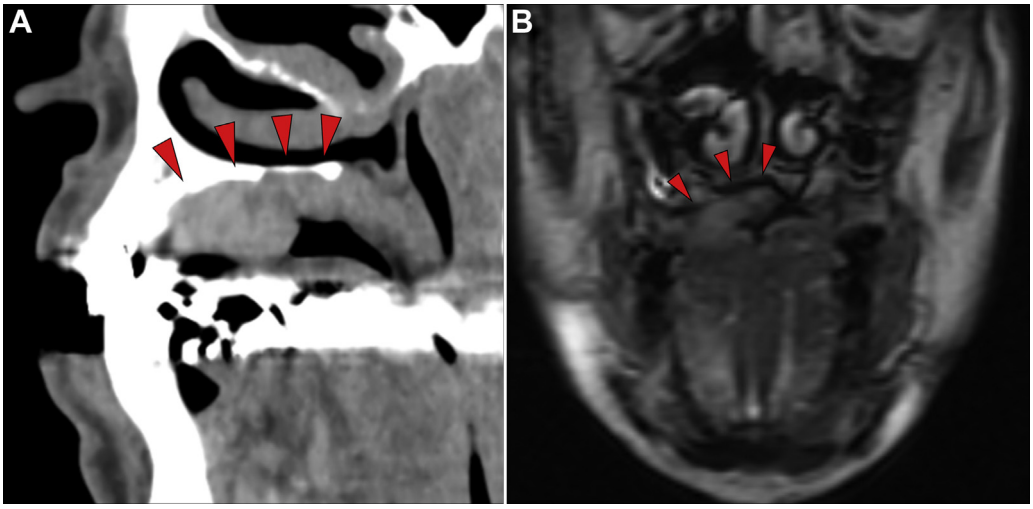

Fig. 2. Imaging findings. (A) Contrast-enhanced computed tomography; (B) magnetic resonance imaging (MRI). Note the compressive resorption of the bone surface in contact with the tumor (red arrowhead; A). The tumor showed the same signal intensity as the muscle on T2-weighted MRI (red arrowhead; B). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)