

Mucoepidermoid Carcinoma of the Hard Palate: a Rare Cause of Hypervascular Tumor

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A highly vascular tumor of the head and neck, with the exception a true vascular lesion, has rarely been observed. We report a rare case of a large, highly vascular, mucoepidermoid carcinoma (MEC) of the hard palate in a 28-year-old woman. The highly vascular channels were identified by ultrasonography and angiography. This case is noteworthy in that a large, highly vascular tumor of the minor salivary gland simulated a vascular lesion. When preoperative imaging demonstrates large vascular channels, preoperative angiography will benefit surgical management and embolization should be considered if possible.

Key Words: Mucoepidermoid carcinoma, hard palate, hypervascular tumor, angiography, embolization

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is one of the most common salivary gland malignancies. As its name implies, it is composed of a mixture of cells, including mucus-producing, epidermoid or squamous, and intermediate types.¹ In addition to these essential cell types, clear cells are represented in varying proportions in MECs, and some tumors have been reported to be almost predominantly composed of clear cell elements.^{2,3}

An MEC occurs most commonly in the parotid gland, with the minor glands being the second most common site, particularly the palate. Minor salivary gland tumors appear as asymptomatic

swellings, which are sometimes fluctuant with a blue or red color, which can be mistaken clinically for a mucocele or a vascular lesion.^{1,4}

Vascular mass lesions in the head and neck are usually benign vascular tumors. They include hemangioma, lymphangioma, and arteriovenous malformations, which are mostly congenital. However, the presence of marked peritumoral vascularity may indicate a hypervascular tumor.⁵ In these situations, the clinical differentiation between true vascular lesions and highly vascular tumors may be difficult.

This article reports a rare case of a large, highly vascular, clear cell variant of MEC, which was initially diagnosed as a hemangioma arising from the hard palate.

CASE REPORT

A 28-year-old Korean woman visited the Department of Oral & Maxillofacial Surgery, Chuncheon Sacred Heart Hospital, Hallym University in October 1997 for evaluation of left palatal swelling. The mass had been first noticed approximately 1 year prior, and had grown slowly in size. Her medical and family history was not remarkable. Oral examination disclosed a dome-shaped, soft and pulsatile mass, measuring 3.0 × 3.0 cm, on the palatal side of the left second premolar to the soft palate. The mass was covered with an intact mucosa of partly blue to red color (Fig. 1). The left maxillary third molar had been buccally displaced with severe mobility, and was sensitive to percussion. There was bleeding from the palatal gingival sulcus. When the mass was

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Fig. 1. Photograph at initial examination showing a dome-shaped, soft bluish mass of the palatal side of the left second premolar to the soft palate, which was covered with an intact mucosa.

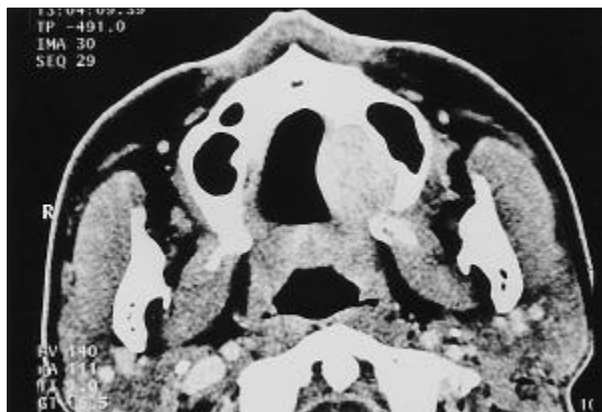


Fig. 2. CT scan demonstrating a well-demarcated and low attenuated, solid mass with a homogenous enhancement in the left palatal region without bony destruction.

aspirated, a bloody fluid was collected. As a result, an initial diagnosis of a vascular mass such as hemangioma was made.

Computerized tomogram (CT) demonstrated a well-demarcated and low attenuated, solid mass with a homogenous enhancement in the left palatal region, measuring 3 cm in length. It also revealed a thinning or displacement of the maxilla without evidence of bony erosion (Fig. 2). However, the CT findings were inconsistent with the physical findings. Therefore, ultrasonography was subsequently performed, and revealed a hyperechogenic mass with surrounding hypervascularity. Digital subtraction angiography was done to detect the vascular branch and prevent severe bleeding during surgery. The results revealed a hypervascular mass with feeding vessels supplied from the distal branch of the left internal maxillary artery (Fig. 3A). After superselective embolization of the distal internal maxillary branches with particles, good devascularization was achieved (Fig. 3B). Two days later, surgical excision of the mass and extraction of the left maxillary third molar were performed under general anesthesia. The mass was relatively well defined, and there was neither copious bleeding and nor evidence of bony invasion. The tooth was not involved with the mass. The postoperative course was uneventful, and there was no sign of recurrence during 5 years of follow-up.

Grossly, the cut surface of the tumor was relatively well defined, and showed a homogenous

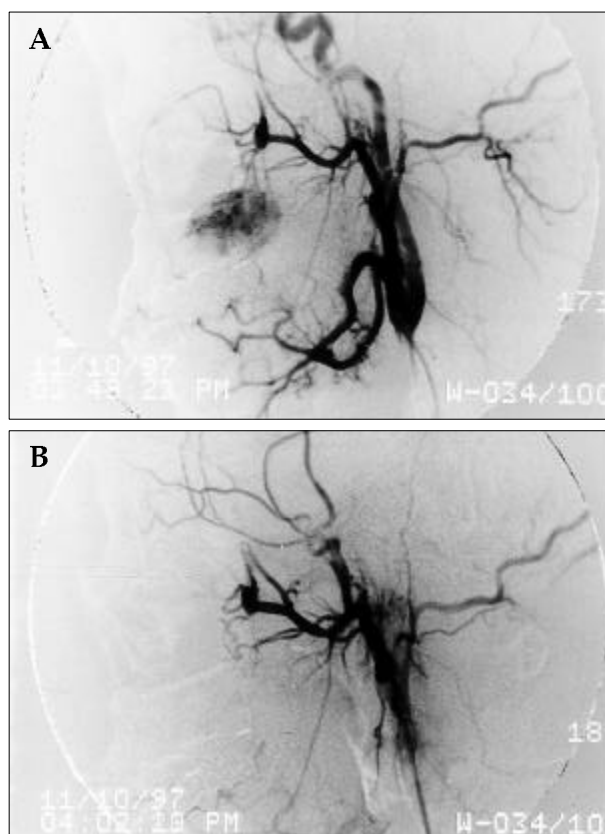


Fig. 3. A. Digital subtraction angiography showing a hypervascular mass with feeding vessels supplied from the distal branch of the left internal maxillary artery. B. Near-total devascularization of the tumor after superselective embolization of the distal internal maxillary branches with particles.

pink to gray-yellowish, soft mass with a hemorrhagic area (Fig. 4). Microscopic examination



Fig. 4. Cut surface of the tumor showing a relatively well-defined, homogenous, pink to gray-yellowish, soft mass with a hemorrhagic area.

revealed a tumor chiefly composed of solid sheets or cords of clear cells. Some areas of squamous differentiation were found, but keratinization was not observed. These epidermoid cells appeared to have a gradual transition to clear cells (Fig. 5). Some of the clear cells contained glycogen, as evinced by the periodic acid-Schiff (PAS) staining with or without diastase digestion. Although vascular proliferation was inconspicuous within the tumor parenchyma, there was prominent thick walled vascular ectasia in the peritumoral connective tissue. Emboli were present within the vessels, and some areas of the tumor revealed multifocal coagulative necrosis, indicating the effects of pre-operative embolization.

Immunohistochemical staining demonstrated that most of the tumor cells reacted positive to Cytokeratin (DAKO, Denmark), but negative to S-100 (BioGenex, SanRamon, CA, USA), Vimentin, and CEA (both DAKO, Denmark). It is believed that these findings are compatible with a clear cell variant of MEC.

DISCUSSION

The images and clinical presentation in the current case seemed to indicate a surface lesion. The presence of a blue to red lesion suggested a tumor of vascular or salivary gland origin.⁴ Blood aspiration does preclude a lesion of vascular origin. Moreover, the digital subtraction angiography demonstrated a highly vascular tumor with the

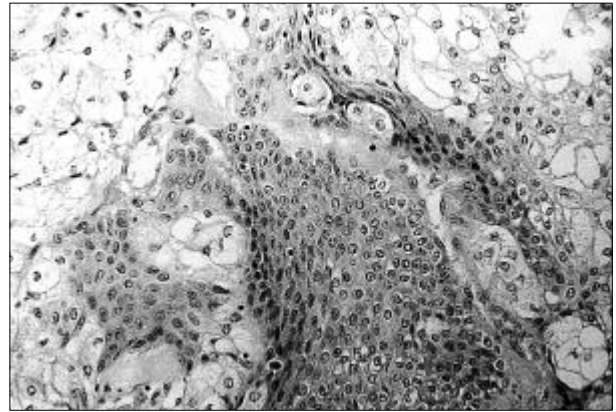


Fig. 5. Photomicrograph showing the tumor chiefly composed of solid sheets of clear cells with areas of squamous differentiation which appear to have a gradual transition to clear cells (Hematoxylin-eosin, $\times 200$).

feeding vessels. As a result, we initially misinterpreted the lesion as a vascular origin. However, the blue color that was observed can be attributed partly to the cystic spaces of the tumor components with hemorrhage or to the tumor-associated vascular ectasia.⁴ On histopathologic examination, the present case was a clear cell variant of MEC with a peritumoral hypervascularity.

A highly vascular tumor of the head and neck, with the exception a true vascular lesion, has rarely been observed.⁵ Previous reports have presented the radiographic appearance of various hypervascular tumors, including metastatic renal cell carcinoma, paraganglioma, ectopic meningioma, olfactory neuroblastoma, merkel cell carcinoma, angiofibroma, and acinic cell carcinoma.⁵⁻⁸ Hypervascular tumors exhibited the following imaging features: dilatation of the feeding vessels, hypertrophy of the peritumoral vessels, intratumoral flow voids, and early and intense enhancement on dynamic, post-contrast, T1-weighted images.^{5,6} In the present case, dilatation of the feeding vessels or hypertrophy of the peritumoral vessels is more likely. However, the cause of this event was not elucidated.

Although it is uncommon, tumor vascularity can interfere with the removal of the tumor by complicating adequate hemostasis. Angiography can be used for identification of feeder vessels prior to surgical intervention, delineation of tumor size and location, and, later, for evaluation of therapy.⁹ In addition, lesions from where a pro-

ductive aspirate is gained, and which have a diameter of 25 mm or more, have better definition with CT and angiography before surgical manipulation.¹⁰ In our case, angiograms demonstrated a hypervascular mass with feeding vessels supplied from the distal branch of the left internal maxillary artery. To our knowledge, there have been no published imaging reports of a highly vascular MEC. Similarly, in a case of acinic cell carcinoma of the maxillary sinus, angiography of the carotid artery showed a hypervascular tumor, which was fed by the maxillary or ophthalmic artery.⁸ Currently, the most expedient technique is that of preoperative embolization, followed by surgical excision of the occluded portion within the subsequent 7 to 10 days.^{10,11} Preoperative embolization was performed to control severe hemorrhage in the present case. The effects on the tissue sections were confirmed by emboli in the peritumoral vessels, and extensive coagulative necrosis of the tumor was evident.

The current case is noteworthy in that the tumor-associated vascular ectasia of the minor salivary gland simulated a vascular mass lesion, which could interfere with the tumor resection. When preoperative imaging demonstrates highly vascular channels, preoperative angiography will benefit surgical management and embolization should be considered if possible.

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