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Hypervascular mucoepidermoid caricinoma of the palate

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SUMMARY

Salivary mucoepidermoid carcinoma is the most common malignant salivary gland tumor. Low or intermediate grade tumors are slow-growing and can be mistaken for benign lesions on both physical exam and imaging studies. We present a case of hypervascular mucoepidermoid carcinoma of the hard palate that was initially thought to represent a benign vascular lesion. To our knowledge, only two prior cases of hypervascular mucoepidermoid carcinoma have been previously reported. This is the first case showing the MRI features of this tumor and its temporal evolution on CT over several years. Our patient ultimately underwent angiographic tumor embolization so that her lesion could be safely biopsied without significant bleeding risk. We present this as a potential diagnostic pitfall and explain how the treatment for hypervascular mucoepidermoid carcinoma varies compared to conventional tumors.

Key words: mucoepidermoid carcinoma, hypervascular, tumor embolization.

INTRODUCTION

Salivary mucoepidermoid carcinoma (MEC) is a rare tumor, accounting for under 0.5% of all malignancies (1). However, it is the most common malignant salivary gland tumor. MEC occurs in major salivary glands about half of the time, most commonly in the parotid gland (2). In the remaining 50% of patients, MEC occurs in minor salivary glands, most commonly involving the palate. One large study of 376 patients with salivary MEC found that the majority of tumors are low or intermediate grade, which portends a better prognosis (1). However, the slow growth of intermediate and low grade MEC can mimic benign lesions and delay diagnosis and treatment.

We report an unusual case of minor salivary gland MEC involving the palate which mimicked a benign vascular lesion. To our knowledge, vascular MEC has only been previously reported twice.

CASE REPORT

A 61-year-old woman presented to our institution after having excessive bleeding while having a maxillary tooth extraction. She was originally found to have abnormal soft tissue swelling around the tooth during a routine physical exam, and her local oral surgeon recommended tooth extraction. During attempted extraction, there was immediate profuse bleeding from her tooth socket. Angiography was performed, which showed a vascular mass in the right hard palate region. She underwent embolization of right internal maxillary and right ascending pharyngeal arteries, as well as selective embolization of left internal maxillary branches supplying the lesion, providing temporary symptomatic relief.

The patient presented to our institution for further work-up per recommendations from her local physicians. Physical exam revealed a large pulsatile mass in the right palate. A sinus CT was obtained, which confirmed a large destructive lesion centered in the right palate (Figure 1, A-B). On MRI, this was T1 hypointense with T2 hypointense, enhancing nodular components (Figure 1, C-F). The mass was felt to be indeterminate but possibly consistent with a vascular lesion given its previously reported angiographic findings. However, malignancy remained in the differential. The patient was asymptomatic and preferred not to undergo surgery or biopsy; therefore, 6 month follow-up CT imaging was recommended, though the patient deferred this as she continued to feel well.

She had no further issues with this lesion until 9 years later, at which time she was referred for additional extractions of teeth three and four for dental

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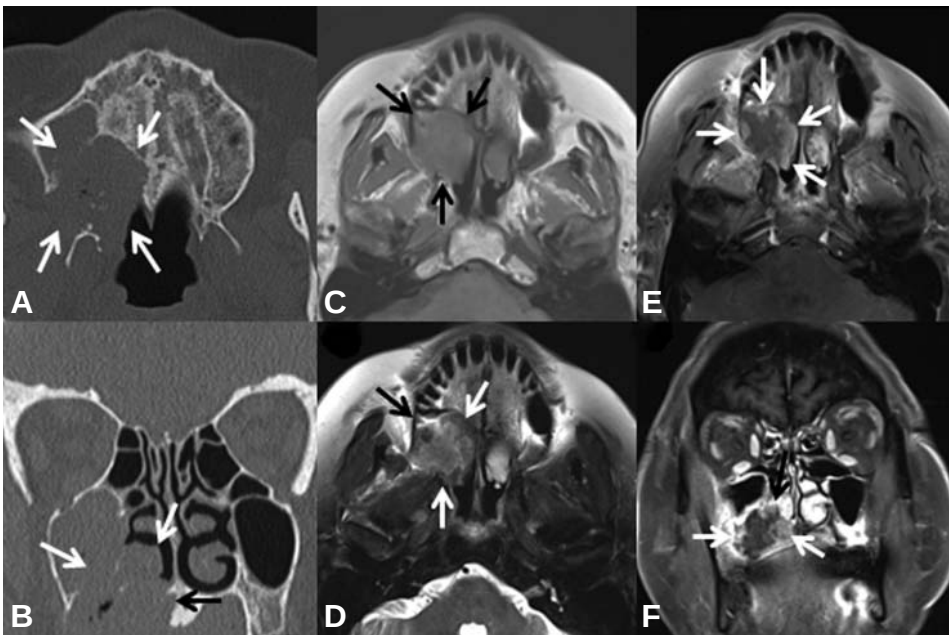

Fig. 1. Axial and coronal CT images in bone windows (A-B) show a soft tissue mass centered in the hard palate with invasion the inferior right nasal cavity and maxillary sinus (A and B, arrows). Axial T1W (C), axial T2W (D), and axial and coronal T1W postgadolinium images (E-F) show that the hard palate mass is T1 isointense to muscle (C, arrows). It has T2 hypointense peripheral components (D, arrows) that correspond to enhancing areas on the postgadolinium images (E-F, arrows).

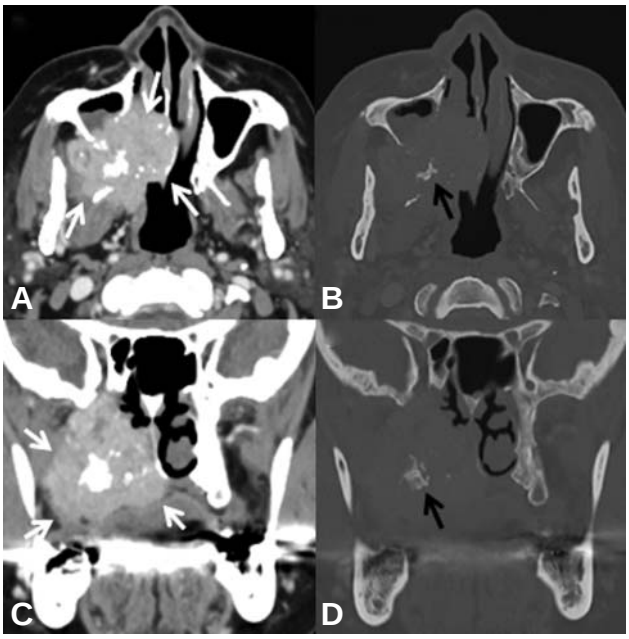

Fig. 2. Axial and coronal contrast enhanced CT images windowed for soft tissue (A, C) and bone (B, D) obtained 9 years after the initial images demonstrate interval enlargement of the hard palate mass (A and C, white arrows) with new osseous destructive changes (B and D, black arrows).

issues. Given her prior bleeding with attempted tooth extraction, a follow-up sinus CT was obtained to reassess her known palate lesion. This showed interval enlargement of the mass with more prominent osseous destructive changes, suggesting possible malignancy rather than a vascular lesion (Figure 2, A-D). MRI was attempted but was limited due to anxiety. She was referred to angiography for repeat embolization of the lesion, the goal being to facilitate her tooth extractions and allow for biopsy of the mass without risk of significant bleeding. Angiography demonstrated a hypervascular right maxillary mass predominantly supplied by a maxillary branch of the right middle meningeal artery as well as the distal right internal maxillary artery (Figure 3, A-D). These branches were embolized using 250 to 350 um polyvinyl alcohol (PVA) particles. Post-embolization angiogram showed the mass to be 95% devascularized (Figure 3, E-F).

The patient underwent uneventful third and fourth tooth extractions. Biopsy of the mass revealed intermediate grade mucoepidermoid carcinoma. The patient declined surgical management and is undergoing interval imaging follow-up.

DISCUSSION

Salivary MEC can be challenging to diagnose, particularly when the tumor is low or intermediate grade. Typical imaging features of low/intermediate grade MEC on CT are a circumscribed mass with cystic spaces and enhancing solid components. Ultrasound features are nonspecific but can include heterogenous echotexture, indistinct margins, irregular shape, and absence of posterior acoustic enhancement (3). On MRI, these tumors typically have low to intermediate T1 and T2 signal with variable enhancement. Marked vascularity is not generally associated with MEC, however, making our patient's lesion atypical. Vascularity is best assessed by digital subtraction angiography (DSA). While MRI features such as avid enhancement of prominent flow voids can suggest vascularity, these features are not always present; our patient's MRI did not show flow voids, and only portions of the mass enhanced.

To our knowledge, hypervascular MEC has only been previously reported twice (4, 5). Interestingly, both of these prior patients also presented with smoothly marginated hard palate masses

initially thought to represent vascular lesions. This is the first report to concomitantly demonstrate the imaging findings of hypervascular MEC on CT, MRI, and DSA. Additionally, this is the first case showing the temporal evolution of this tumor on CT over several years.

Differential considerations for a palatal mass include MEC, other salivary gland tumors, hemangioma, odontogenic cysts, mucoceles, and primary or metastatic osseous tumors (6). Clinical and imaging evidence of hypervascularity or excessive bleeding would generally suggest a hemangioma, but such findings are not definitive as our patient illustrates. Odontogenic cysts can be differentiated from MEC on CT or MRI due to the presence of a soft tissue mass in MEC, but radiographs in MEC can show deceptively benign lucency causing MEC to be mistaken for benign odontogenic lesions (7). Mucoceles are typically circumscribed and have a bluish dome appearance on physical exam, which can be mistaken for MEC. However, unlike MEC, they have uniform low attenuation on CT (8). Rarely, MEC can have intralesional calcification and hyperattenuation that can mimic fibro-osseous lesions on CT (9).

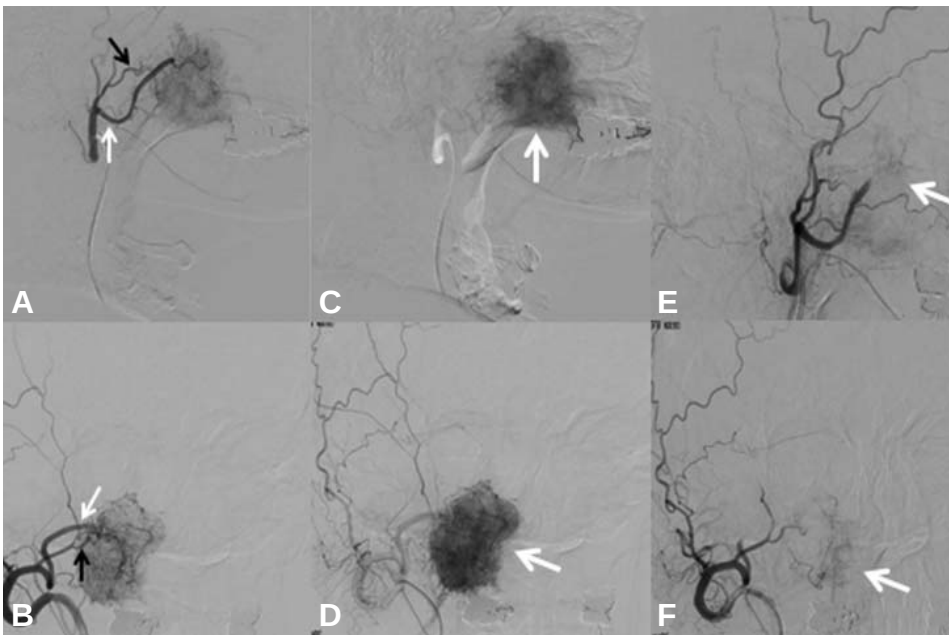

Fig. 3. Lateral and anteroposterior digital subtraction angiographic images during the early arterial phase (A and B) demonstrate a hypervascular mass predominantly supplied by the internal maxillary artery (A and B, white arrows), with minor supply from a maxillary branch of the middle meningeal artery (A and B, black arrows). The extent of hypervascularity of the mass is best seen later in the arterial phase on lateral and anteroposterior views (C and D, arrows). Lateral and anteroposterior images obtained after tumor embolization with polyvinyl alcohol (E and F) show marked decrease in vascularity of the mass.

Treatment for MEC is variable. For low or intermediate grade tumors, wide local excision is often sufficient (10). Higher grade tumors frequently require radiation as well. Additionally, close imaging follow-up is recommended for higher grade tumors, as local recurrence in not uncommon. Although MEC is rarely as markedly hypervascular as seen in our patient, embolization may be warranted in such cases. Embolization may be considered prior to biopsy or resection of the tumor but may also be helpful prior to elective procedures such as tooth extractions.

CONCLUSION

We've presented a rare case of hypervascular mucoepidermoid carcinoma. Our patient's work-up and imaging illustrate several learning points. First, hypervascular palatal masses are not necessarily benign. CT can be invaluable in these patients to suggest malignancy based on aggressive features. Second, hypervascular MEC should be considered in the differential if a slowly enlarging mass is seen on follow-up imaging. Unlike hemangiomas or other benign vascular entities, MEC may show osseous destructive changes. Finally, DSA plays an important role in characterizing and treating hypervascular MEC through prophylactic embolization to lessen bleeding risk.

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