Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology xxx (xxxx) xxx–xxx

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case report

Carcinoma ex pleomorphic adenoma without malignant findings upon clinical in the palate

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ARTICLE INFO

Keywords:
Carcinoma ex pleomorphic adenoma
Pleomorphic adenoma
Minor salivary gland
Palate

ABSTRACT

Carcinoma ex pleomorphic adenoma (CXPA) generally develops from long-standing and recurrent cases of pleomorphic adenoma (PA). As a salivary gland tumor, the histopathological finding is vary, and the diagnosis of a benign or malignant tumor may not be reflected by imaging data. We here present a case of CXPA in the palate that showed no malignant findings upon clinical, imaging and histopathological examination of a biopsy specimen, but that was confirmed to be malignant upon surgical resection. A 72-year-old man was referred with a painless mass in the right-side palate that exhibited increasing swelling. Histopathological and immunostaining investigations of the biopsy specimen revealed PA, in addition to clinical and imaging evaluation. However, as that was may be malignant considering the prolonged course the onset, we informed to patients for possible postoperative dilation surgery and adjuvant therapy. Histopathological diagnosis of the resected specimen was CXPA with an intra-capsular carcinoma. The carcinoma component was a mucoepidermoid carcinoma. The clinical course of this patient provided two important insights. First, CXPA in the palate can present findings indicative of PA in clinical examination and imaging. Second, surgical management for PA considering the potential for CXPA was beneficial. In cases of PA that onset was suspected to be many years previous, surgical management should be undertaken that considers the potential presence of CXPA, even if the preoperative diagnosis does not suggest malignancy.

1. Introduction

Carcinoma ex pleomorphic adenoma (CXPA) is a salivary gland tumor that arises from a preceding long-standing and recurrent pleomorphic adenoma (PA) [1]. As histopathological findings is various in a salivary gland tumor, characteristic findings of a benign or malignant tumor may not be confirmed by imaging approaches [2]. With regard to minor salivary gland tumors in particular, comprehensive diagnosis based on histopathological findings is important in addition to clinical and imaging results. Here we present a case of CXPA in the palate that showed no evidence of malignancy upon clinical, imaging and histopathological examination of a biopsy specimen, but which was confirmed to be malignant upon surgical resection.

2. Case report

A 72-year-old man was referred to our department for a detailed examination of a long-term painless mass in the right-side palate that was exhibiting increasing swelling. There was no paralysis, trismus, or swelling of the cervical lymph nodes. A 2 × 2 cm mass that was elastic and soft with a clear boundary was identified, but no abnormalities in the surface mucosa were detected in the right-side palate (Fig. 1). Computed tomography imaging scans showed a well-defined tumor with a clear compressived bone resorption image on the right-side side palate and no invasive destruction in the surrounding bone, nor calcification inside the tumor. Furthermore, magnetic resonance imaging showed a well-defined tumor with a smooth band edge. A T1-weighted image revealed a tumor with low density, while a T2-weighted image revealed the tumor had an inhomogeneous center of low and high density. Additionally, a contrast-enhanced T1-weighted image revealed a non-uniform contrast effect (Fig. 2A, B). The clinical diagnosis was a minor salivary gland tumor. We planned to perform resection of the tumor under general anesthesia. Two weeks prior to surgery, to confirm the type of salivary gland tumor and obtain a definitive diagnosis as to

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https://doi.org/10.1016/j.ajoms.2018.02.009
Received 29 September 2017; Received in revised form 13 February 2018; Accepted 22 February 2018

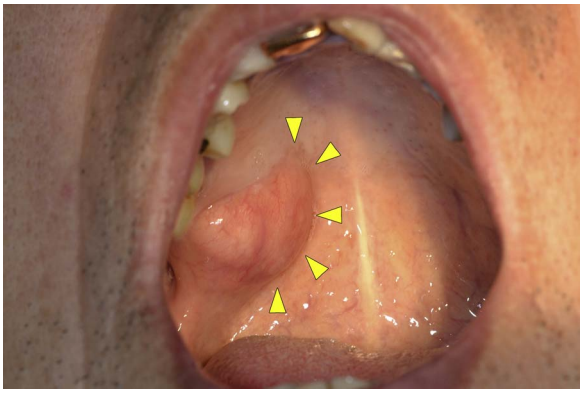

Fig. 1. Clinical view of the mass in the right-side palate (arrow).

whether it was benign or malignant, we took a biopsy from the central part of the tumor under local anesthesia. Histopathological findings of the biopsy specimen was suggested PA. On immunohistochemical staining, tumor cells were positive for epithelial membrane antigen (EMA), CAM5.2, and smooth muscle actin (SMA), calponin, S-100 protein. We diagnosed PA from the histopathological and immunostaining findings of the biopsy specimen. Because of ulceration in the biopsy section, we then performed cytology, which revealed a class 2 tumor without atypical cells. Although PA was diagnosed based on the above-mentioned results, we considered the possibility of CXPA as it was suggested that the course from onset was longer. For surgery, we set the safety margin to 1 cm, with the resection including adjacent normal tissue. Histopathological diagnosis in the resected specimen was CXPA with intra-capsular carcinoma (Fig. 3A–C); tumor cells of carcinoma component were predominantly squamous epithelial cells. However, some tumor cells were positive for mucicarmine. We diagnosed low-differentiated mucoepidermoid carcinoma with predominant squamous epithelial cells because of some mucous cells presented (Fig. 4).

After diagnosis was confirmed, we examined the regional lymph nodes for the presence or absence of distant metastasis, which were found to be negative. In the histopathological findings, extracapsular invasion and stump appearance were also negative, thus additional treatment or adjuvant therapy was not administered and the patient was placed under strict observation. Two years after the surgery, local recurrence and metastatic findings have not been identified.

3. Discussion

The clinical course of this patient provided two important insights. First, CXPA in the palate can present with characteristics indicative of PA. Second, surgical management for PA considering the potential for CXPA is beneficial.

CXPA is derived from the malignant transformation of PA, and accounts for approximately 12% of malignant salivary gland tumors originating from approximately 6.2% of PAs [1]. The degree of malignancy differs between intra-capsular and extra-capsular subtypes. In intra-capsular cases, prognosis is good by complete resection. Malignant salivary gland tumors in the palate usually show clinical findings such as ulcers, erosion, or abnormalities in the surface mucosa, and imaging findings such as marginal irregularities, and destruction and invasion of the surrounding bone. However, in cases of early CXPA, these findings may be poor or inadequate. This case emphasizes the difficulty in diagnosing either CXPA or PA, even when PA is suspected from the clinical and imaging findings.

Of the minor salivary gland tumor types, those in the palate have a malignancy rate of approximately 50% [3]. Therefore, considering the incidence of malignant tumors, even in cases that the clinical and imaging findings suggest a benign tumor, histopathological analysis is necessary for reliable preoperative diagnoses that include the extent of any malignancy [4]. This case considered the possibility of CXPA in view of long-term existence for PA. Therefore, fearing of the malignant transformation of these benign tumors that is known to occur after biopsy, we shortened the waiting period from the biopsy to the surgery. Also, we based the resection range on that required for a malignant tumor and informed the patient of the potential need for additional, expansion and adjuvant therapy after the surgery depending upon the histopathological diagnosis of the resected specimens.

This case showed clinical and imaging findings indicative of PA. The clinical course of CXPA demonstrates progressive malignant development whereby malignant cells that have transformed within a PA infiltrate the surrounding tissue in multiple steps [1,5]. Depending on the extent of its progression, it can be classified as intra-capsular carcinoma or extra-capsular-infiltrating carcinoma; in cases of intra-capsular carcinoma, the malignant component does not extend beyond the capsule of the existing PA. Therefore, even if carcinoma is diagnosed, it is difficult to confirm the presence of a malignant tumor from the clinical and imaging findings because the range of the carcinoma is confined within a benign PA. Furthermore, it may also be diagnosed as a PA by biopsy depending on the extent of malignant component. Consequently,

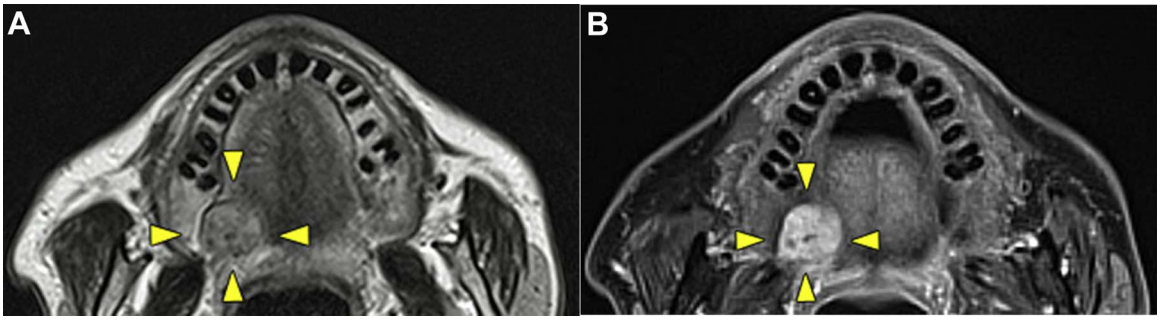

Fig. 2. Horizontal magnetic resonance imaging scans. **A**: T2-weighted image showing a tumor with an inhomogeneous center (arrow). **B**: Contrast-enhanced T1-weighted image showing tumor with non-uniform contrast effect (arrow).