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Case Report

Epithelial-myoepithelial carcinoma arising in the oral floor: Report of a case and review of the literature

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ABSTRACT

Epithelial-myoepithelial carcinoma (EMC) is an extremely rare low-grade malignant salivary gland tumor that most commonly occurs in the parotid gland, but can also arise in minor salivary glands. We report here a case of EMC localized in oral floor in a 66-year-old female who presented with left side of oral floor swelling of four years duration. CT scan revealed a nonhomogeneously enhancing mass in the left side of the oral floor extending to the midline of the oral cavity. After establishing a clinical diagnosis of a benign tumor arising in the left side of oral floor, a biopsy of the lesion was performed under local anesthesia. The histological diagnosis was suspected pleomorphic adenoma, however, it could not deny the possibility of malignancy, because of that there were any evidence of nuclear atypia or mitotic figures. Then, the patient underwent a wide local excision, where was realized immunohistochemical analysis, staining the duct-like structures, where the inner layer of epithelial cells positively for AE1/AE3 and CK 14, whereas outer layer of myoepithelial cells stained positively for calponin and vimentin antibodies. Based on these findings, the final histological diagnosis was EMC. The patient was discharged from the hospital, and she is being followed up about 5 years without evidence for neither recurrences nor metastases.

1. Introduction

Epithelial-myoepithelial carcinoma (EMC) was first reported by Donath et al. in 1972 [1]. EMC is defined by the World Health Organization (WHO) classification in 1991 as a rare low-grade malignant neoplasm and account for than 1% of salivary glands tumors [2,3]. Histopathologically, the tumor is characterized by well-defined tubules with two cell types: an outer layer of myoepithelial cells with clear cytoplasm surrounding an inner lining of eosinophilic cuboidal epithelial cells [4] resembling intercalated ducts. It has been suggested that it derives from the intercalated ducts of the salivary glands, because the tubular growth pattern of this tumor epitomizes this phenotype [5,6]. This tumor arising most frequently (80%) in the parotid gland, but lesions have also been reported in submandibular glands (10%) and minor salivary glands (1%) [1,7]. This paper reports a very rare case of EMC arising from oral floor, as well as discusses the findings immunohistochemical, emphasizing the possibilities involved in the genesis of this neoplasm, the clinical characteristics, diagnosis, and treatment of this case, and that provides a review of the literature.

2. Case report

A 66-year-old women was referred to our division on March 28th, 2010, complaining of swelling with ulceration in the oral floor. She had noticed the lesion for about 4 year and complaining of lesion that was “gradually swollen”. In extra-oral examination was not observed any alterations. In intra-oral examination showed a fixed firm mass of 32 mm × 17 mm with a part of ulcer was detected in the left side of oral floor and of normal color (Fig. 1). No cervical lymphadenopathy was found. A computed tomographic (CT) scan showed a nonhomogeneously enhancing mass in the left side of the oral floor extending to the midline of the oral cavity (Fig. 2). Magnetic resonance imaging (MRI) showed a soft-tissue mass in the left oral floor extending to the midline of the oral cavity. The axial and coronal T1-weighted image of the lesion showed a uniform low signal intensity, and that the axial and coronal T2-weighted image showed well-defined nodular mass with a moderately high,

☆ 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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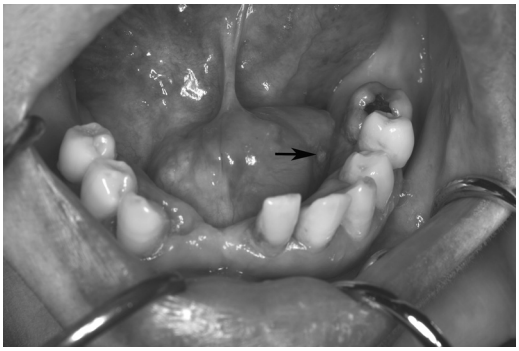

Fig. 1. Clinical view showing a fixed firm mass of 32 mm × 17 mm was detected in the oral floor with regular surface and an ulceration (arrow).

slightly nonhomogeneous signal intensity (Fig. 3A–D). No regional and distant metastases were found by contrast enhanced CT and PET-CT. There was no history of smoking or alcohol consumption. After establishing a clinical diagnosis of a benign tumor arising in the left side of oral floor, a biopsy of the lesion was performed under local anesthesia on April 5th, 2010. The histological diagnosis was suspected pleomorphic adenoma, however, it could not deny the possibility of malignancy, because of that there were atypical cells. Then, a wide local excision including the mandibular anterior gingiva, periosteum and the left sublingual gland was immediately performed with a 10 mm soft tissue margin under general anesthesia on April 20th in order to protect the Wharton's duct (Fig. 4A). With careful dissection, lingual nerve was identified and protected. On surgical exploration, it was found that the mass involved the sublingual gland. It was difficult to delineate the exact origin of the pathology (sublingual or other minor salivary gland); hence it was decided to sacrifice the left sublingual gland and its duct on the affected side (Fig. 4B). After excision, the right and left Wharton's duct was relocated and the raw surface on mylohyoid muscle was covered with a full-thickness skin from right inguinal region. The post-operative course was uneventful. The patient is being followed up 5 years without evidence for neither recurrences nor metastases (Fig. 4C).

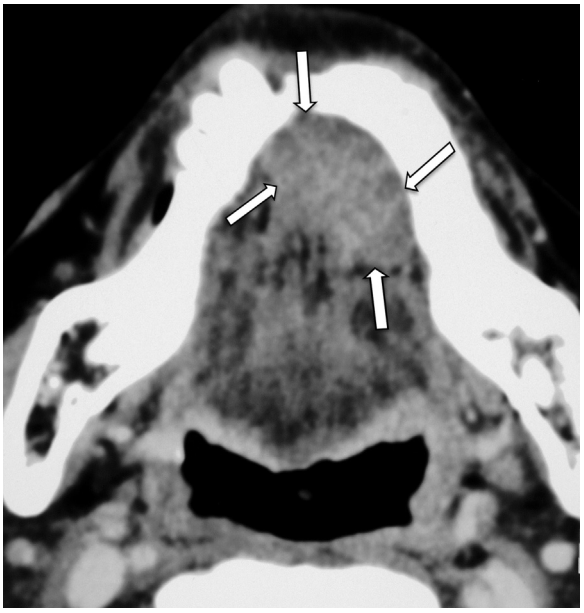

Fig. 2. Axial contrast-enhanced CT scan showed a nonhomogeneously enhancing mass in the left side of the oral floor extending to the midline of the oral cavity (arrows).

3. Pathological findings

Macroscopic findings: The surgical specimen was about 55 mm × 30 mm × 25 mm in size and showed hard elasticity with a white cut surface. Cut surface revealed an encapsulated, well-circumscribed, lobulated mass (Fig. 5A). *Microscopic findings*: Microscopic examination showed a well circumscribed, encapsulated tumor arranged in a lobular pattern (Fig. 5B). Each lobule comprised ductal and myoepithelial-like elements. The ductal components showed eosinophilic cuboidal intercalated duct like cells bordering small lumina, and were surrounded by pale acidophilic myoepithelial-like cells (Fig. 5C). Nuclear atypia was mild, and mitotic figure count was very low. Immunohistochemical findings: The proliferative activity of tumor cells was relatively high, because of that the positive immunoreactivity for Ki-67 was about 10% (Fig. 6A). The ductal components indicated strongly positive for cytokeratin AE1/AE3 and CK 14 (Fig. 6B and C). On the other hand, the neoplastic myoepithelial-like elements revealed strong positive immunoreactivity for vimentin and calponin (Fig. 6D and E). However, the special staining for periodic acid Schiff (PAS) (before and after diastase digestion) and Mucicarmine were both negative. Hence, a final diagnosis of EMC was made. Conclusively, the pathological TNM (pTNM) staging classification was made as pT2N0M0, stage II. Furthermore, it was judged to occur from sublingual gland from what this tumor mass was surrounded by a certain level of hearty salivary gland tissue.

4. Discussion

EMC is a low-grade malignancy, only rarely have high-grade or dedifferentiated EMC cases been reported. This unusual salivary tumor was included in the WHO classification of salivary gland tumors in 1991. EMC arising most frequently (80%) in the parotid gland, but lesions have also been reported in submandibular glands (10%) and minor salivary glands (including the sublingual gland) (1%) [1,7]. Thus, this tumor arises in the salivary glands, predominantly the major salivary gland, especially the parotid gland [6]. However, they have rarely been observed in tissues other than the salivary glands, such as the breast, lacrimal gland, lung and nasal cavity [6,8–10]. Furthermore, this tumor occurs in older persons (sixth decade and beyond), and has a female predominance [11]. The patient here was 66-year-old female, such as in the mean age reported in the literature. Interestingly, the parotids have the lowest level of malignancy (15–30%), with the submandibular gland (40%) and the minor salivary gland having a 50% malignancy rate, while the rate of malignancy in the sublingual gland is 70–90% [12]. Furthermore, it is sometimes very difficult to decide the differential diagnosis of origin of the salivary gland tumor in oral floor. Eventually, we might have no choice but to depend on pathological findings.

After updating the WHO classification of salivary gland tumor in 1991, the 44 reports, we had read extensively on the clinical features and prognoses of EMC including the present case, are summarized in Table 1. According to this search results, the fine needle aspiration (FNA) was performed in 18 cases, however, no case was diagnosed as an EMC. Although it has come across the reports which were performed the fine needle aspiration (FNA) occasionally [13,14], it seems quite difficult to diagnose EMC by FNA, because of that it is necessary to find the biphasic cell pattern, duct-forming cell and myoepithelial cells out. To provide a definitive diagnosis in treating with EMC, it is particularly important to use a total excisional specimen. For comparison, the pathologic differential diagnosis includes clear cell carcinoma, mucoepidermoid carcinoma, acinic cell carcinoma, sebaceous carcinoma, myoepithelioma and clear cell oncocytoma. Pleomorphic adenoma and