CASE REPORT

Coexistence of salivary duct, myoepithelial and epithelial-myoepithelial carcinomas in the parotid gland: a case report and literature review

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Abstract

Few reports have described two or more histologically-distinct carcinoma types within the same salivary gland. A 62-year-old man presented to our hospital after detecting a mass in the right parotid gland. Computed tomography revealed a tumor (5.1 × 5.0 cm) within the right parotid gland. Tumor resection with lymph node dissection was performed. The proliferation of three morphologically-different tumor cells was demonstrated on histopathologic examination (salivary duct carcinoma [SDC], myoepithelial carcinoma and epithelial-myoepithelial carcinoma [EMC]). The shape of the inner layer of cells in the EMC was similar to the SDC. Specifically, it appeared that the cells were a mixture of the two tumors with reciprocal transfer in the same area. Immunohistochemical staining showed that the SDC cells and the EMC inner cells were positive for AR, HER2 and p53. Thus, we suggest that our case represented a high-grade transformation.

INTRODUCTION

Few reports describe two or more histologically-distinct carcinoma types within the same salivary gland [1]. To date, these cases have been categorized as hybrid carcinomas, collision carcinomas or a high-grade transformation (HGT; [2–5]).

CASE REPORT

A 62-year-old man presented to our hospital after detecting a mass in the right parotid gland. He had no personal or family history of malignancy. On physical examination the mass was 5.0 × 5.0 cm in size with poor mobility. Computed tomography revealed a tumor (5.1 × 5.0 cm) within the right parotid gland that was enhanced by contrast (Fig. 1a). The tumor had an irregular shape with calcifications, a low-density area and the boundary between the tumor and surrounding tissues was indistinct. A malignant tumor of the right parotid gland was suspected, thus tumor resection with lymph node dissection was performed. The cut surface of the tumor was yellow–white with hemorrhage and necrosis (Fig. 1b). The tumor boundaries were ill-defined. The proliferation of three morphologically-different tumor cells was demonstrated on histopathologic examination. First, the cell type was atypical with abundant granular eosinophilic cytoplasm, large nuclei with coarse chromatin and prominent...
entities are not separated but have an identical origin in the category, arising within the same topographical area. Both tumor neoplasms composed of two separate different tumor entities, a hybrid carcinoma or HGT. Hybrid tumors are defined as 'a mixture of cells with EMC inner layer cells exhibiting the same morphology and immunohistochemical staining as a SDC. As a result of acinar metaplasia, the EMC inner layer cells underwent a morphological change, resulting in a SDC-like appearance.

In conclusion, the presence of two or more histologically-distinct tumor entities in one topographical location is rare in salivary glands. When such cases are encountered, we need to consider differential diagnoses based on the potential relationship and pathogenesis of the tumor components, including hybrid tumors, collision tumors, and malignant transformation or HGT [2–5]. First, we excluded collision carcinomas. We considered our case to represent a HGT (from EMC/MC to SDC) for the following reasons: three different carcinomas appeared to be a mixture of cells with EMC inner layer cells exhibiting the similar morphology and immunohistochemical staining as a SDC.

DISCUSSION

The presence of two or more histologically-distinct tumor entities in one topographical location is rare in salivary glands. When such cases are encountered, we need to consider differential diagnoses based on the potential relationship and pathogenesis of the tumor components, including hybrid tumors, collision tumors, and malignant transformation or HGT [2–5]. First, we excluded collision carcinomas. We considered our case to represent a hybrid carcinoma or HGT. Hybrid tumors are defined as a neoplasm composed of two separate different tumor entities, each one of which conforms to an exactly defined tumor category, arising within the same topographical area. Both tumor entities are not separated but have an identical origin in the same topographical area' [2]. We initially diagnosed this as a hybrid carcinoma; however, the precise criteria for a hybrid tumor/carcinoma have not been described in the WHO classification [6]. Moreover, the description, 'hybrid tumors would not show evidence of evolution from one entity to another' according to the diagnostic criteria of hybrid tumors, was deleted in the second edition of Gnepp’s Diagnostic Surgical Pathology of Head and Neck [7]. Hellquist et al. proposed criteria for a hybrid tumor/carcinoma that have not been universally agreed upon, and the number of reported cases is rapidly increasing [3]. In contrast, the concept of HGT in salivary gland neoplasms is most commonly associated with a poorly differentiated adenocarcinoma or undifferentiated carcinoma (hence the older term dedifferentiation). Dedifferentiation is defined as the abrupt transformation of low-grade (LG) well-differentiated carcinomas into a high-grade (HG) morphology, which lacks the original distinct histologic and immunohistochemical features [3, 8]. The HG areas may be clearly demarcated, but a transitional zone is usually present [3]. We consider our case to represent a HGT (from EMC/MC to SDC) for the following reasons: three different carcinomas appeared as a mixture of cells with EMC inner layer cells exhibiting the same morphology and immunohistochemical staining as a SDC. Hamamoto et al. reported that their case had SDC and EMC components and inner ductal cells of double-layered EMC ducts with a similar morphology and immunophenotype to the SDC in the transitional area and suggested that a SDC originated...
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Figure 2: Histopathologic findings of the parotid tumor. (a and b: hematoxylin and eosin staining, ×40 and ×400) The cell type was atypical with abundant granular eosinophilic cytoplasm, large nuclei with coarse chromatin and prominent nucleoli. The cells were proliferative and invasive with duct-like, glandular and solid patterns. (c and d: hematoxylin and eosin staining, ×40 and ×400) The cell type was an atypical spindle-to-epithelioid cell with clear cytoplasm that was proliferative and invasive with trabecular, tubular and solid patterns. (e and f: hematoxylin and eosin staining, ×40 and ×400) The cell type was an atypical duct-like structure comprised of two distinct cell layers. The inner layer consisted of cuboidal cells with eosinophilic cytoplasm and round enlarged nuclei and the outer layer consisted of cells with clear cytoplasm and oval nuclei.

Figure 3: Histopathologic findings of transfer area. (a–c: hematoxylin and eosin staining, ×200) The tumor cells of the salivary duct, myoepithelial and EMC appeared to be a mixture with reciprocal transfer in the same area. From the EMC inner ductal cells [9]. We believe that our case was similar; however, the relationship between MC and EMC/SDC is unclear whether hybrid carcinoma or HGT.

In conclusion, we are of the opinion that our case was a HGT; however, the nomenclature of a hybrid tumor and HGT is ambiguous. Further examination, including next-generation sequencing, is warranted.

Figure 4: Immunohistochemical findings. Most of the cells of the SDC were positive for AR (a, ×200) and HER2 (b, ×200). Most of the cells of the myoepithelial carcinoma were positive for p63 (c, ×200) and S-100 (d, ×200).

Figure 5: Immunohistochemical findings. Most of EMC inner layer cells were positive for cytokeratin AE1/AE3 (a, ×200) and weakly positive for HER2 (b, ×200). Most of the EMC outer layer cells were weakly positive for cytokeratin AE1/AE3 (a, ×200). In the mixed area, most of the EMC inner layer cells were positive for cytokeratin AE1/AE3 and most of the EMC outer layer cells and MC cells were weakly positive for CK AE1/AE3 (c, ×200). Most of the EMC inner layer cells were negative for p63, and most the EMC outer layer cells and MC cells were positive for p63 (d, ×200).

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INFORMED CONSENT

Written informed consent was obtained from the patient’s legal authorized representatives for publication of this case report and accompanying images.

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