



Salivary duct carcinoma: a case report with cytological and pathological features

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Abstract

A 55-year-old Japanese man presented with rapidly growing tumor in the left parotid region without any symptoms. Based on the fine-needle aspiration cytology report of parotid epithelial malignant tumor (suggestive of salivary duct carcinoma), the left parotidectomy was performed. Histopathology and immunohistochemistry examinations revealed features of salivary duct carcinoma. Although salivary duct carcinoma comprising a small proportion of salivary gland tumors and is known to be aggressive, he is free from recurrence and metastases 36 months after the surgery and radiation therapy.

Keywords: Salivary duct carcinoma, parotid gland, fine-needle aspiration cytology, histopathology, immunohistochemistry

Introduction

Salivary duct carcinoma is a rare malignant epithelial tumor, which accounts less than 1% of epithelial salivary gland neoplasms [1]. Seventy-five percent of salivary duct carcinomas occur in the parotid gland [1]. The peak incidence is in the seventh decade of life, and its occurrence in patients under age 50 year is uncommon [1]. Because of similar histologic features, salivary duct carcinoma is often compared to ductal carcinoma [2], atypical ductal hyperplasia [3,4] of the breast, but the majority of patients are men. This malignancy is one of the most aggressive salivary gland malignancies [1]. Local invasion, frequent lymphatic and hematogenous metastasis, and poor prognosis characterize the biologic behavior of this malignancy.

Because of its low incidence [1], the cytological and pathological features have not been elucidated sufficiently. This case report illustrates both the cytological and pathological features of salivary duct carcinoma that developed in a Japanese adult male.

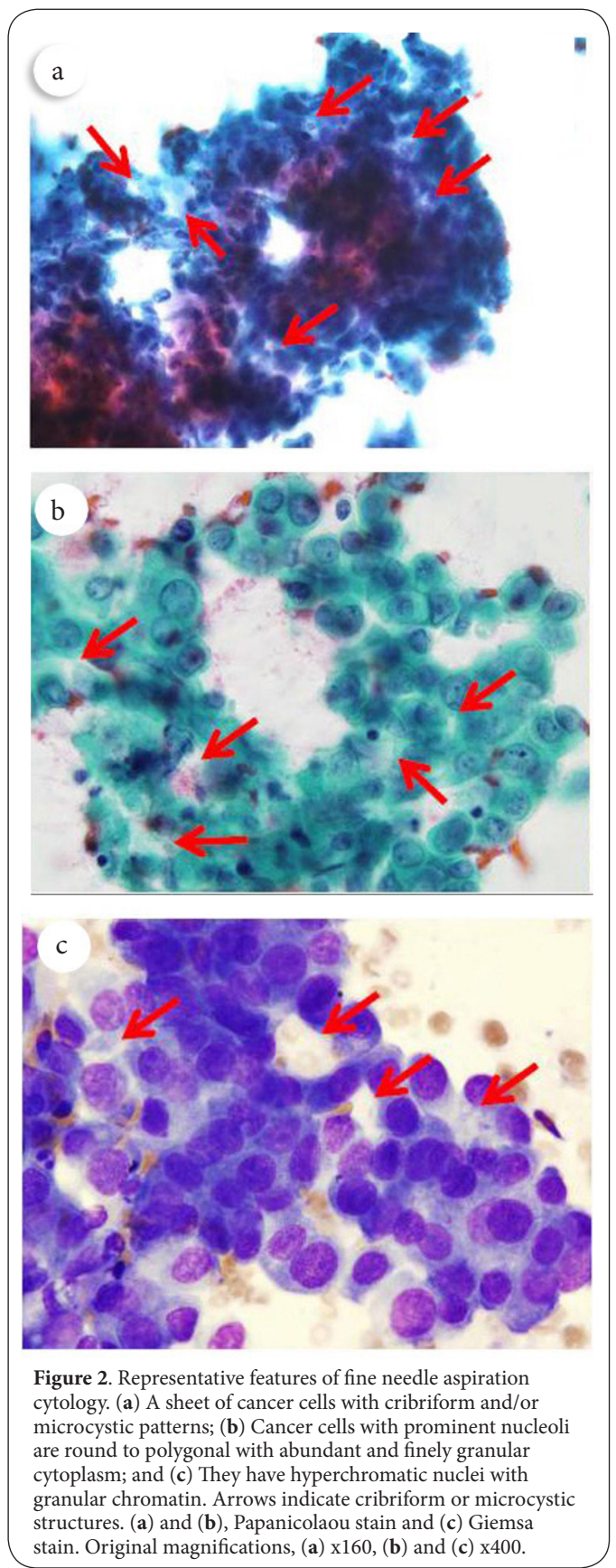
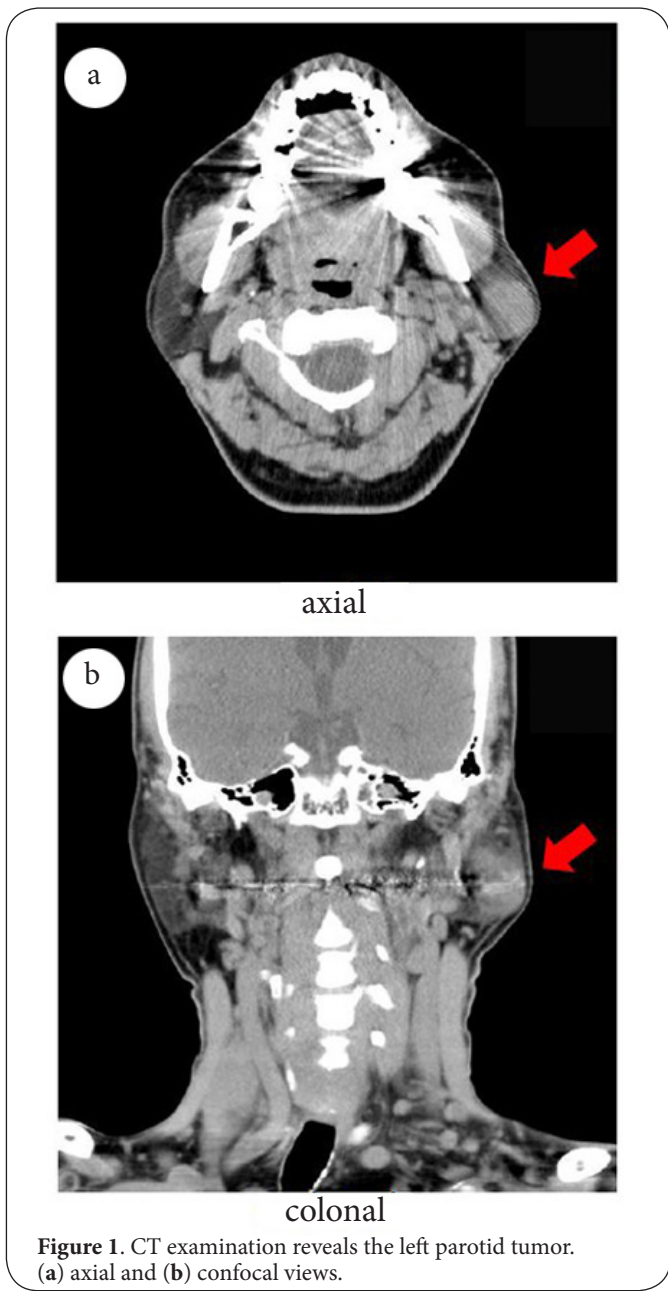
Case presentation

On June 9, 2011, a 55-year-old man presented our hospital with a progressively growing tumor mass on the left side of his

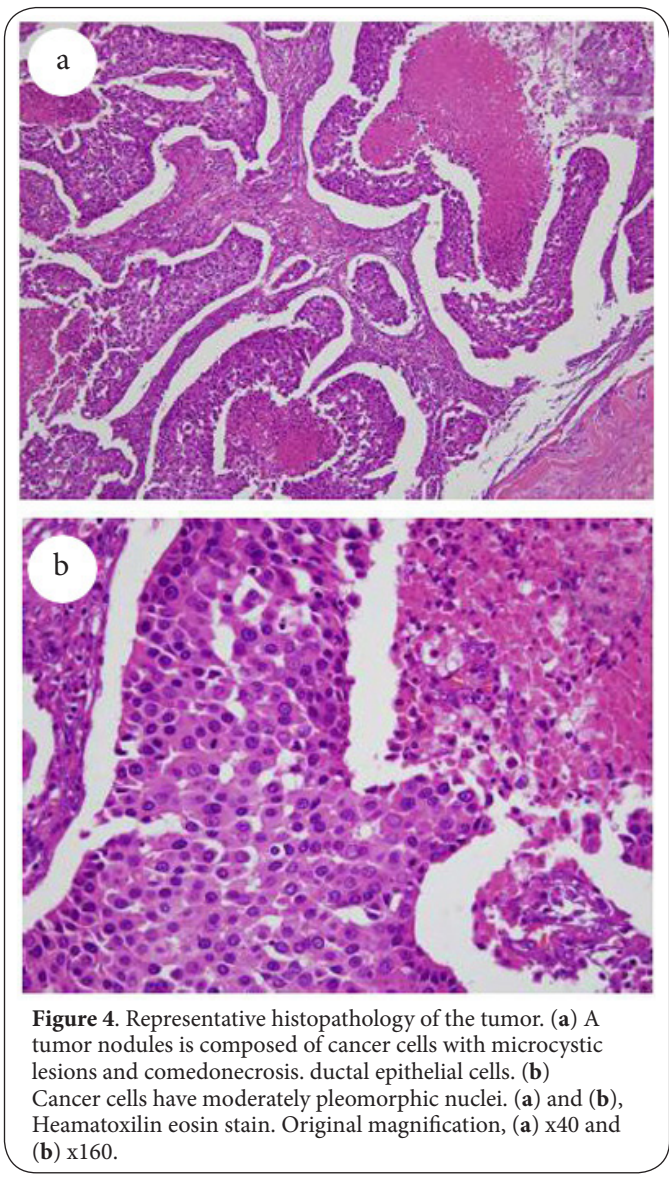
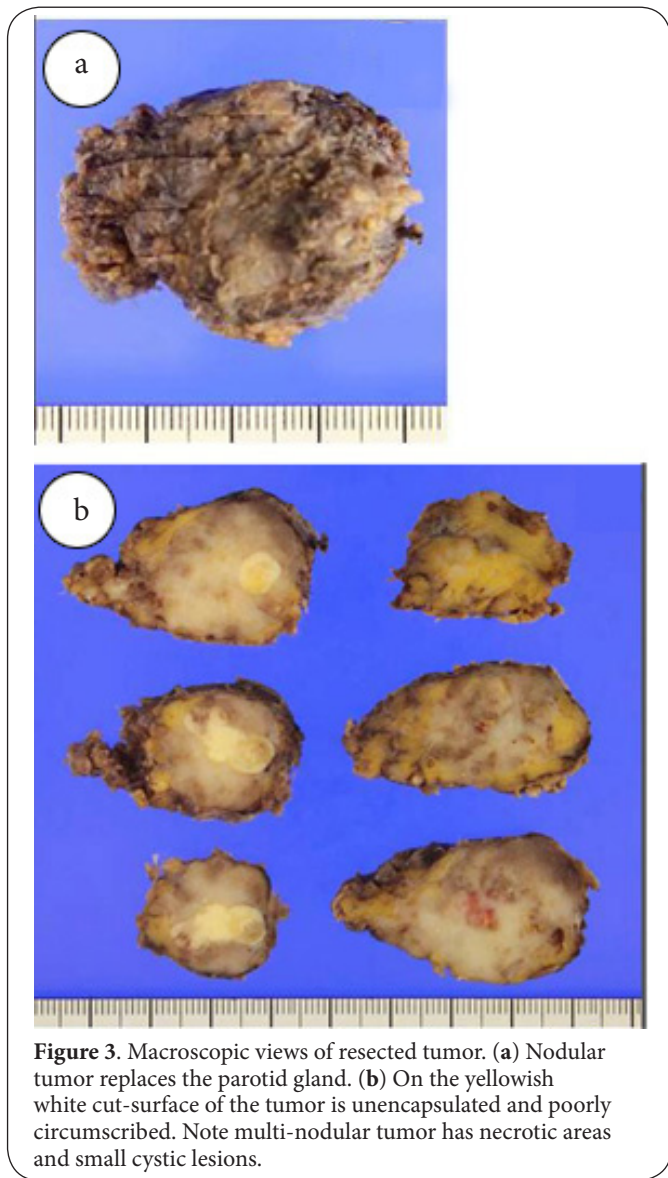
neck. He had no history of diseases. Physical examination revealed a single painless, firm, and mobile 3 cm mass in the parotid area. Facial nerve function was intact. At presentation, the results of routine serum biochemistry tests were normal, with the exception of the level of total bilirubin (1.5 mg/dl), blood sugar (118 mg/dl). Computed tomography (CT) scan showed a tumor mass (2.4 cm in diameter) almost entirely replacing the left parotid gland (Figure 1).

Fine-needle aspiration cytology from the parotid tumor showed many epithelial tumor cells. They are arranged in cohesive clusters, which have a cribriform architectural pattern (Figure 2a). Individual tumor cells are scattered at the periphery of the cellular clusters. Tumor cells are round to polygonal with abundant and finely granular cytoplasm. They have medium to large, moderately pleomorphic and hyperchromatic nuclei with granular chromatin (Figures 2b and 2c). Based on the cytological findings, we diagnosed this tumor as parotid epithelial malignant tumor (suggestive of salivary duct carcinoma).

The left parotidectomy was performed in July 20, 2011. As shown in Figure 3a, a nodular tumor (2.8 cm in diameter) replaced the parotid gland. Cut-surface of the resected tumor was yellowish white, unencapsulated, and poorly circumscribed



(**Figure 3b**). The tumor was multi-nodular and contained necrosis and small cystic lesions. Pathological examination showed that round and well-circumscribed tumor nodules are composed of moderately pleomorphic ductal epithelial cells (**Figure 4a**). These cells surround a core of comedonecrosis (**Figure 4b**). Solid nests of tumor have scattered necrotic cells with surrounding retraction artifact. These findings suggested salivary duct carcinoma. To confirm the pathological diagnosis, immunohistochemistry using antibodies against gross cystic disease fluid protein-15 (GCDFP15), androgen receptor (AR), human epithelial growth factor receptor 2 (HER2/neu), S-100, and epithelial membrane antigen (EMA) was conducted and showed

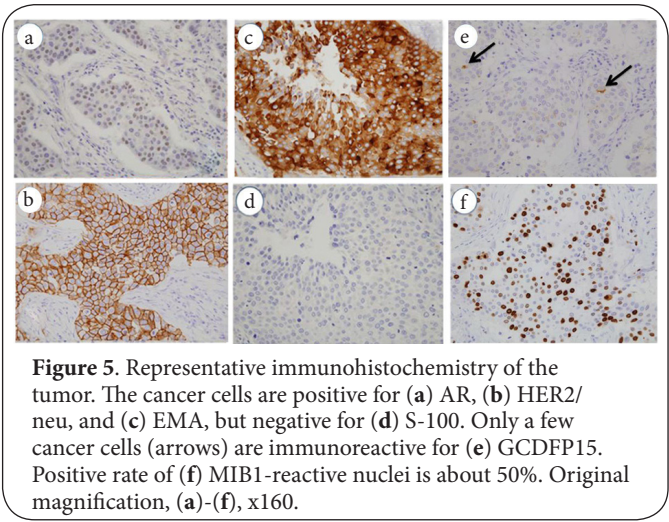


that malignant cells were immunoreactive for AR (Figure 5a), HER2/neu (Figure 5b), and EMA (Figure 5c). However, cancer cells were negative for S-100 (Figure 5d). A few cancer cells were positive for GCDFP15 (Figure 5e). Positive rate of cancer cells against Ki-67 antigen (MIB1) was about 50% (Figure 5f). Other immunohistochemistry showed positive for AE1/AE3 and cytokeratin 7, and negative for α -smooth muscle actin, p53, and p63 [5].

Postoperatively the patient received a total of 50 Gy (2 Gy x 25) of radiation. He is currently under close follow-up every four months and he is well 36 months after the surgery.

Discussion and conclusions

A rare case of salivary duct carcinoma in Japanese man was presented. Cancer cells in this case exhibited pathological and immunohistochemical findings similar to those reported



previously [1,6-12]. Although salivary duct carcinoma is one of the most aggressive salivary gland malignancies, the patient reported here is free of metastasis and recurrence 36 months after the surgery. In addition, this type of cancer strongly expressed HER2/neu is more aggressive than those without HER2/neu expression [1,13]. The reasons of favorable prognosis of this patient are not known. However, some correlation of prognosis to tumor size was reported: less than 3 cm indicated a better prognosis. Therefore, the relatively small size (2.8 cm) of the tumor size in conjunction with low MIB-1-positive index and p53 negative cancer cells [14] may influence the prognosis. Our case suggested a low-grade salivary duct carcinoma [3], but immunohistochemical findings of S-100 and HER2/neu were different from those reported by Brandwein-Gensler et al., [3].

An uncommon and high-grade malignant neoplasm, salivary duct carcinoma, is derived from intralobular and interlobular excretory ducts. Its histopathologic characteristics have a striking resemblance to ductal carcinoma of the breast. In 1968, Kleinsasser et al., [15] first used the term salivary duct carcinoma. Since other salivary gland carcinomas are derived from the duct system and exhibit ductal differentiation, the term salivary duct carcinoma is probably not the best choice to identify a specific category of salivary gland carcinoma. Although Brandwein et al., [16] proposed the term cribriform salivary carcinoma of excretory duct, at present salivary duct carcinoma has become the established terminology.

As to the cytological features of salivary duct carcinoma, viable tumor cells in cytologic specimens are often in a background of necrosis. As found in this case, cancer cells are arranged in cohesive clusters, which sometimes have a cribriform or papillary architectural pattern [1]. Other cytologic features in this case were similar to those reported previously [1]. Therefore, cytological diagnosis of salivary duct carcinoma is possible [7].

The most characteristic histopathological feature is variably sized, rounded, solid or cystic nodules of tumor cells. These findings are similar to those of intraductal carcinoma of the breast [1,2]. Comedonecrosis is characteristic of salivary duct carcinoma and is one of the biomarkers of this malignancy. In this case, we noticed comedonecrosis in the cytologic and histologic specimens. Immunohistochemical observation in this case revealed similar findings reported previously. Cancer cells were strongly positive for AR [17] and HER2/neu [13], but negative for S-100 [1]. However, only a few cancer cells were weakly positive for apocrine marker GCDPF-15, in contrast to previous reports [18,19].

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

Authors' contributions	KH	NK	KN	AS	TS	MM	FE	YY	HS	NW	TN	TY	TT
Research concept and design	✓	✓	--	--	--	✓	✓	--	--	✓	✓	✓	✓
Collection and/or assembly of data	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	--
Data analysis and interpretation	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Writing the article	✓	--	--	--	--	--	--	--	--	--	--	--	✓
Critical revision of the article	--	--	--	--	--	✓	✓	--	--	✓	✓	--	✓
Final approval of article	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

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