



Cystic Salivary Duct Carcinoma Penetrated by Facial Nerve

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Abstract

Salivary duct carcinoma is a rare malignant salivary gland tumor that mainly has solid features. When it occurs in the parotid gland, it can invade the facial nerve and cause facial nerve paralysis. However, in our case, the salivary duct carcinoma exhibited cystic features on computed tomographic imaging, and the facial nerve passed through the cyst. Total parotidectomy with level-I to -III dissections was performed and nerve passing through the tumor was sacrificed. The patient received postoperative radiotherapy and was clinically and radiologically followed-up for every 3 months. Recurrence or distant metastasis was not reported. To the best of our knowledge, this is the first case involving a salivary duct carcinoma with cystic features and facial nerve invasion. Here, we report a first case of cystic salivary duct carcinoma of the parotid gland which uncommonly undergo cystic change and penetrated by facial nerve and successfully resected without causing facial nerve injury.

Keywords

- ▶ salivary duct carcinoma
- ▶ facial nerve
- ▶ parotid gland tumor
- ▶ cystic tumor

Introduction

Salivary duct carcinoma, first described by Kleisner et al. in 1968, is a high-grade salivary gland malignancy.^{1–3} It commonly occurs in the parotid glands of older men. Salivary duct carcinoma is an invasive tumor with local and distant metastases. It is highly malignant, and nearly half of the patients die from the disease within 5 years.⁴ Wide surgical resection and postoperative radiotherapy are necessary to treat salivary duct carcinoma.⁵ Despite these, its prognosis remains poor. Salivary duct carcinoma typically presents as a solid mass, and when it occurs in the parotid gland, it sometimes can invade the facial nerve causing facial nerve paralysis.⁶ To the best of our knowledge, there has been no report of a case in which salivary duct carcinoma undergoes cystic change, and also a case in which this cystic tumor is penetrated by a facial nerve has not been reported. Here, we

report a first case of cystic salivary duct carcinoma of the parotid gland that which uncommonly undergo cystic change and penetrated by facial nerve. To the best of our knowledge, this is the first case of cystic feature salivary duct carcinoma with facial nerve invasion.

Case

A 67-year-old Korean man noticed swelling in the left parotid region. Physical examination showed 3.0 cm × 3.3 cm sized, firm, and painless on palpation, fixed palpable mass located at left parotid gland without any discomfort and facial nerve palsy. The regional lymph nodes were not palpable. The mass was noticed 3 years ago, and it gradually enlarged over the past year. The laboratory data were within the normal limits, and his medical history was unremarkable. Chest radiography showed no evidence of metastasis. Computed tomography

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Fig. 1 Computed tomography image indicating a round cystic mass at left parotid gland. (A) Coronal view of the cystic mass. (B) Axial view of the cystic mass.

imaging (► **Fig. 1**) revealed a cystic mass initially suspected to be a Warthin's tumor. Under intraoperative nerve monitoring, mass and facial nerve which penetrate the cyst and located at inner mucosa of cyst were identified (► **Fig. 2**). There were more tiny masses around cystic mass including yellowish component different from the cyst component. Marginal resection of all the masses was performed with preserving the facial nerve (► **Fig. 3**). The surgical specimen showed encapsulated cyst containing transparent fluid in the superior portion of the parotid gland (► **Fig. 4**). This finding suggested that salivary duct carcinoma uncommonly underwent a cystic change. Histopathological examination revealed a papillary pattern of neoplastic ductal epithelial cells, and the central portion of the cell clusters exhibited comedonecrosis. In addition, androgen receptor was highly expressed on immunohistochemistry (► **Fig. 5**). These ductal epithelial cells were characteristic of salivary duct carcinoma, resembling ductal breast carcinoma. After salivary duct carcinoma was confirmed in the pathological examination, additional surgery was performed. In the second operation, total parotidectomy with level-I to -III neck dissections was performed and the nerve passing through the tumor, which is expected to be the zygomatic branch of the facial nerve, was sacrificed. Then a sural nerve graft was performed for restoring of facial nerve.



Fig. 2 Intraoperative finding. The facial nerve was found at the inner bed of cystic mass.

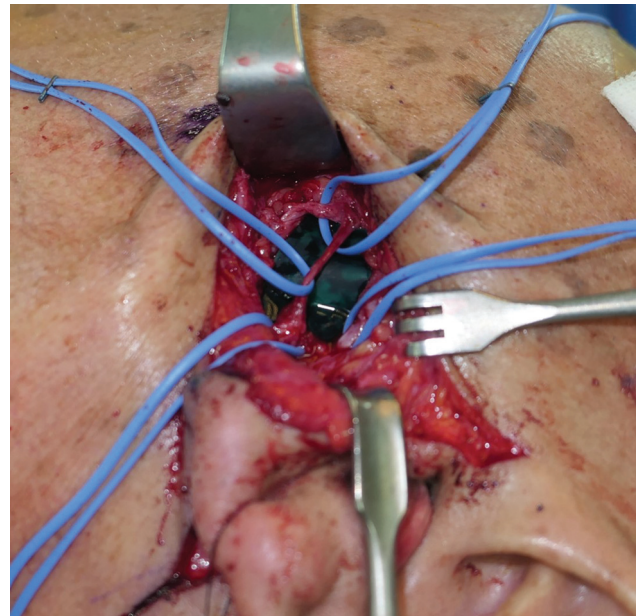


Fig. 3 After total mass excision, the facial nerve was preserved without damage.

No residual tumor was identified in the second surgical pathology examination, and also there was no metastasis in the neck lymph node. In the postoperative nerve conduction study, slight weakness of the zygomatic branch was confirmed but there was no complete palsy. The patient received radiotherapy after the second operation. Follow-up is scheduled for every 3 months to check for recurrence or distant metastasis. Until the latest 10-month follow-up, there is no evidence of recurrence or distant metastasis (► **Fig. 6**).

Discussion

Salivary duct carcinoma is a rare malignant salivary gland tumor that is histologically indistinguishable from mammary duct carcinoma. Salivary duct carcinoma occurs predominantly in males aged >50 years. The most frequently affected site is the parotid gland, followed by the submandibular gland.³ The first-line management of salivary duct carcinoma is wide surgical resection and postoperative radiation or chemotherapy, when indicated by the postoperative pathologic findings, including stage, grade, and perineural invasion. Hormone therapy has been applied as an adjuvant treatment in some cancers, but its efficacy in salivary duct carcinoma has not yet been demonstrated.⁴

Salivary duct carcinoma typically presents clinically as a painless, rapidly growing mass, which develops aggressively with the possibility of early distant metastasis and local recurrence. It typically has solid features and when it occurs in the parotid gland, it sometimes invades the facial nerve and induce facial nerve paralysis.^{4,7} However, in our case, the salivary duct carcinoma underwent a cystic change, and the facial nerve passed through the cyst. However, the patient had no symptoms of facial nerve palsy. Because salivary duct carcinoma typically has a solid feature, the index of suspicion for this disease is lower than that of other benign tumors

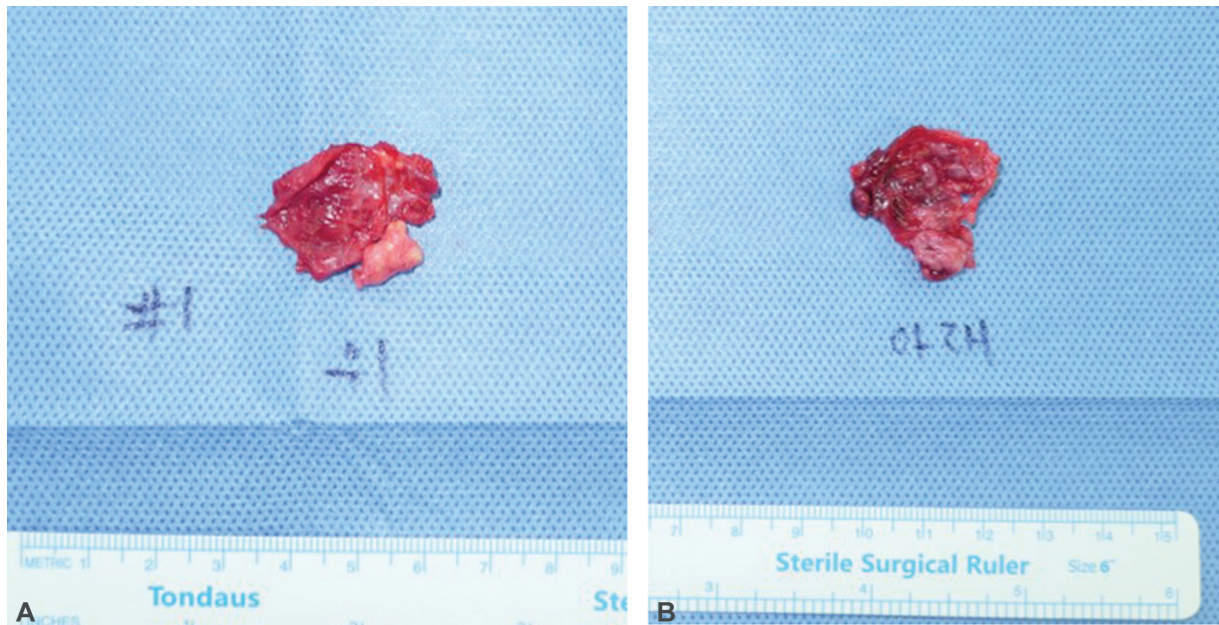


Fig. 4 Divided and excised cystic mass sent for histopathologic examination. (A) Cranial part of the divided cystic mass. (B) Caudal part of the divided cystic mass.

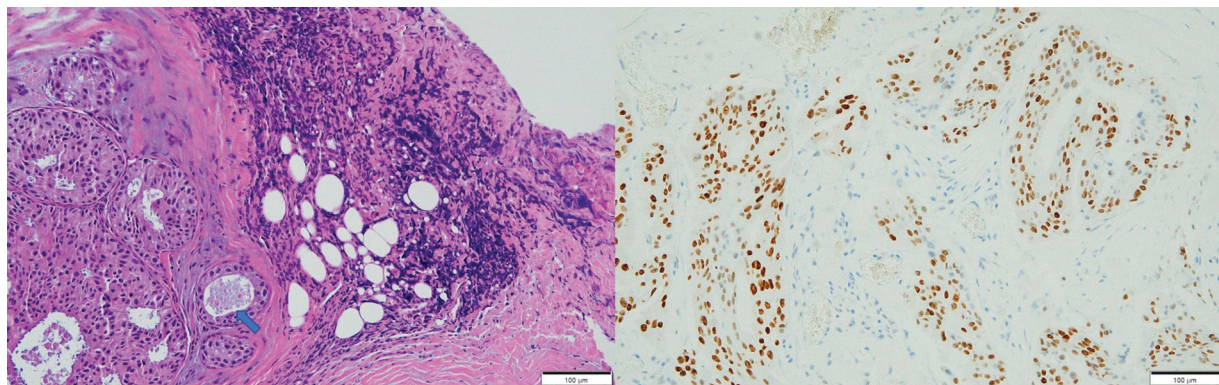


Fig. 5 Histopathological examination revealed intraductal carcinoma pattern with central comedonecrosis (blue arrow) and androgen receptor was highly expressed on immunohistochemistry.



Fig. 6 Preoperative photograph (A) and 10-month follow-up photographs (B).

when evaluating a cystic tumor in the parotid gland. If a malignant tumor is missed in diagnosing a cystic tumor, the prognosis becomes very poor. Therefore, it is crucially important to make an accurate diagnosis.

Although perineural invasion is frequently reported in solid salivary duct carcinoma, it has not been clinically or radiologically reported in cystic feature of salivary duct carcinoma. Enlarged cystic tumors can compress the nerve, but perineural invasion cannot occur. However, in our case, the cystic tumor was penetrated by the facial nerve, and the facial nerve was located in the inner bed of the cystic mass. Therefore, this case is very rare.

Tumor resection with a wide margin is crucial to treat salivary duct carcinoma. As seen in this case, tumors invaded by the facial nerve are challenging to completely excise without damaging the nerve. However, considering that salivary duct carcinoma has possibility of local recurrence and metastasis, complete tumor resection is absolutely necessary even in this case. To prevent a recurrence, radiation therapy is usually recommended after complete surgical resection. After that, follow-up is needed to check for recurrence, both clinically and radiologically.

Author Contributions

Conceptualization: J.U.P. Data curation: Y.K. Formal analysis: Y.K. and J.U.P. Writing-original draft: Y.K. Writing-review & editing: Y.K. and J.U.P. All authors read and approved the final manuscript.

Ethical Approval

The study was approved by the Institutional Review Board of SMG-SNU Boramae Medical Center (Institutional Review Board no. 10-2021-53) and performed in accordance with the principles of the Declaration of Helsinki. Written informed consent was obtained.

Patient Consent

The patients provided written informed consent for the publication and the use of his images.

Conflict of Interest

J.U.P. is an editorial board member of the journal but was not involved in the peer reviewer selection, evaluation, or decision process of this article. No other potential conflicts of interest relevant to this article were reported.

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