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Gastric metastasis from salivary duct carcinoma mimicking primary gastric cancer



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ABSTRACT

INTRODUCTION: We present a very rare case of gastric metastasis mimicking primary gastric cancer in a patient who had undergone surgery for salivary duct carcinoma.

PRESENTATION OF CASE: A 67-year-old man had been diagnosed as having right parotid cancer and had undergone a right parotidectomy and lymph node dissection. The histological diagnosis was salivary duct carcinoma. One year after the surgery, a positron emission tomography-computed tomography scan using fluorodeoxyglucose (FDG) revealed an abnormal uptake of FDG in the left cervical, mediastinal, paraaortic, and cardiac lymph nodes; stomach; and pancreas. On gastroduodenoscopy, there was a huge, easily bleeding ulcer mimicking primary gastric cancer at the upper body of the stomach. Biopsy revealed poorly differentiated adenocarcinoma. Therefore, we were unable to differentiate between the primary gastric cancer and the metastatic tumor using gastroduodenoscopy and biopsy. Because of the uncontrollable bleeding from the gastric cancer, we performed an emergency palliative total gastrectomy. On histological examination, the gastric lesion was found to be metastatic carcinoma originating from the salivary duct carcinoma.

DISCUSSION: In the presented case, we could not diagnose the gastric metastasis originating from the salivary duct carcinoma even by endoscopic biopsy. This is because the histological appearance of salivary duct carcinoma is similar to that of high-grade adenocarcinoma, thus, resembling primary gastric cancer.

CONCLUSION: When we perform endoscopic examination of patients with malignant neoplasias, a possibility of metastatic gastric cancer should be taken into consideration.

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1. Introduction

There are many histological types of salivary gland neoplasms. They include benign and malignant tumors of epithelial, mesenchymal, and lymphoid origin. The 2005 World Health Organization Classification of salivary gland tumors is complex and includes 10 benign and 23 malignant entities of epithelial origin [1]. Salivary gland malignancies have an estimated incidence of 0.5–2.5 per 100,000 individuals [2]. Among the malignancies, salivary duct carcinoma is characterized by an aggressive behavior, early metastases, local recurrence, and a high mortality rate [3] and con-

stitutes <1.8% of all major salivary gland tumors [4]. This cancer predominantly arises in the parotid gland (72%), followed by the submandibular gland (15%) [4]. However, gastric metastases from head and neck carcinomas are rare, and their incidence among all gastric metastases has been reported to range from 1% to 6.2% [5–7]. We report a very rare case of a 67-year-old man with salivary duct carcinoma metastasized to the stomach.

2. Presentation of the case

A 67-year-old man underwent a right parotidectomy for parotid cancer at our hospital. The histological report showed salivary duct carcinoma with capsular invasion. The patient received adjuvant chemoradiotherapy after the surgery. A neck ultrasonography at 1 year after the surgery showed that the left supraclavicular lymph node was swollen. Histological examination by biopsy revealed metastatic carcinoma. A computed tomography scan showed gas-

Abbreviations: FDG, fluorodeoxyglucose; CT, computed tomography.

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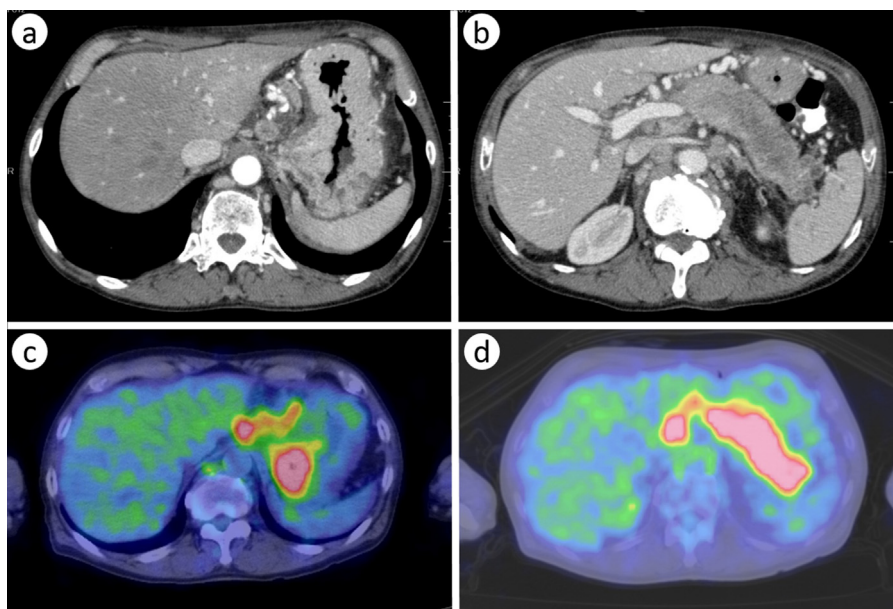


Fig. 1. Computed tomography (CT) scan and positron emission tomography–computed tomography (PET–CT) scan using fluorodeoxyglucose (FDG). (a), An abdominal CT scan demonstrated gastric wall thickening with an enlarged perigastric lymph node; (b), An abdominal CT scan demonstrated pancreatic enlargement; (c), A PET–CT scan using FDG demonstrated FDG-avid spots in the stomach and cardiac lymph nodes; (d), PET–CT demonstrated FDG-avid spots in the pancreas.

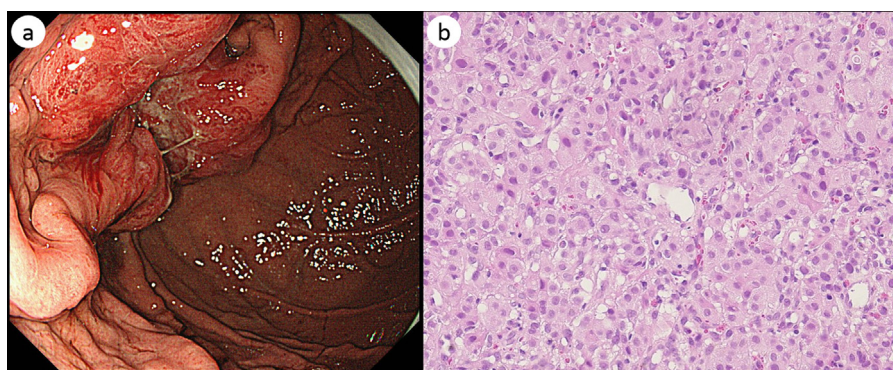


Fig. 2. (a) Gastroduodenoscopy examination. The endoscopic findings demonstrated a large ulcerative lesion at the upper body of the stomach; (b) A biopsy specimen revealed poorly differentiated adenocarcinoma [hematoxylin and eosin (HE), ×400].

tric wall thickening with an enlarged perigastric lymph node and pancreatic enlargement (Fig. 1a and b). A positron emission tomography–computed tomography scan using fluorodeoxyglucose (FDG) showed that there were intensive FDG-avid spots in the left cervical, mediastinal, paraaortic, and cardiac lymph nodes; stomach; and pancreas (Fig. 1c and d). A complete blood cell count revealed a hemoglobin level of 10.5 g/dL, hematocrit of 32.4%, and platelets of 206,000/ μ L. A tumor marker study revealed that the alpha-fetoprotein level was 2.5 ng/mL, carcinoembryonic antigen level was 11.1 ng/dL, and the carbohydrate antigen 19-9 level was 11.0 U/mL. On gastroduodenoscopy, there was a huge, easily bleeding ulcer at the upper body of the stomach (Fig. 2a). Biopsy revealed poorly differentiated adenocarcinoma (Fig. 2b). Therefore, we could not differentiate between primary gastric cancer and a metastatic tumor by gastroduodenoscopy and biopsy. Bleeding from the gastric cancer, which was difficult to control by proton pump inhibitor therapy and endoscopic hemostasis, continued until the hemoglobin level decreased to 5.0 g/dL, and the patient needed blood transfusion. Therefore, we performed an emergency palliative total gastrectomy to control the bleeding. Macroscopically, a clearly demarcated large ulcer (70 mm in size) was seen on the posterior wall of the subcardia (Fig. 3a and b). Histologically,

mixed large and small solid nests were observed, which invaded the serosa layer with lymphatic permeation (Fig. 3c). The cancer cells were diffusely positive for the HER2 oncogene (Fig. 3d). These findings were similar to those obtained for the previous surgically resected specimen of salivary duct carcinoma. Consequently, the cancer was consistent with a metastatic carcinoma originating from the salivary duct carcinoma. The patient made an uneventful postoperative recovery and received postoperative chemotherapy. However, he died from tumor regrowth 8 months after the gastrectomy.

3. Discussion

Salivary gland neoplasms are very rare and appear with an incidence of 0.5–2.5 per 100,000 individuals [2]. Salivary duct carcinoma is a particularly uncommon adenocarcinoma arising from the ductal epithelium of major salivary glands. These cancers predominantly arise in the parotid gland (72%), followed by the submandibular gland (15%) [4]. In salivary duct carcinoma, distant metastases are frequent. The most frequent sites involved (in the order of occurrence) are the lung, bone, and brain [8], whereas the stomach is an unusual site for metastases. It has been reported that

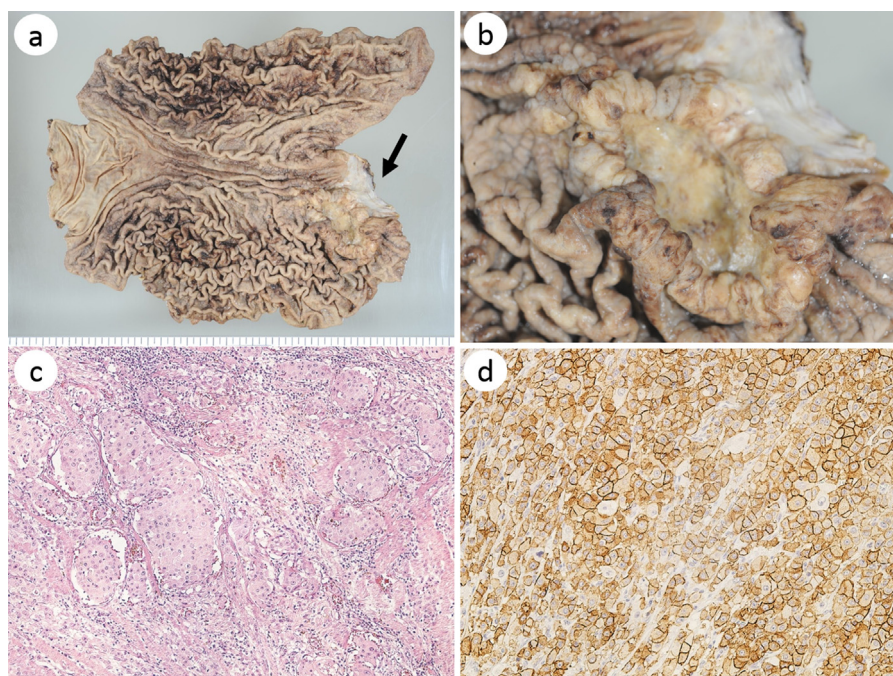


Fig. 3. Macroscopic and histological features of the resected specimen. a, b, Macroscopically, there was a large ulcerative lesion on the upper body (arrow); c, Note the solid nests composed of cancer cells invading the muscularis propria, with marked lymphatic permeation (HE, $\times 100$); d, Immunohistochemically, the specimen was diffusely positive for an anti-HER2 antibody ($\times 200$).

the incidence of metastases to the stomach is 0.8%–5.4% [5,6,9]. Moreover, the lung, breast, and esophagus are the common primary metastatic sites, and metastatic melanoma is also a common malignancy [6,7]. Among metastases to the stomach, gastric metastases from head and neck carcinomas are rare, and their incidence among all gastric metastases has been reported to range from 1% to 6.2% [5–7]. In general, gastric metastases are associated with an advanced stage of disease, and the prognosis is poor. Thus, systemic chemotherapy suitable for the primary cancer is usually chosen as a therapy for gastric metastasis. However, it seems that aggressive surgical treatment is justified in the cases with severe bleeding, obstruction, or perforation because it provides effective palliation [10]. We performed surgical treatment for the gastric metastasis with severe bleeding, which could not be controlled by endoscopic interventions, and succeeded in controlling the bleeding. Gastro-duodenoscopy is useful for the identification of gastric metastases. With regard to the endoscopic appearance of gastric metastases, a submucosal tumor with a central depression, which is called the “bull’s-eye sign,” is well known. However, this typical configuration was found in only 44% of the cases of gastric metastases. The remaining lesions were diagnosed as primary gastric cancers [6]. It has been reported that solitary metastases (similar to primary gastric cancer) were more common than multiple metastases, with the rate of solitary metastases being 62.5%–66.0% [6,7]. For these reasons, endoscopic differential diagnosis of gastric metastases is difficult, such as in our case. It has been reported that endoscopic biopsy is helpful in reaching the final diagnosis because over 90% of metastatic lesions are confirmed histologically [6,7]. However, in our case, we could not diagnose the gastric metastasis originating from the salivary duct carcinoma even by endoscopic biopsy. It is thought that one of the reasons was that the histological appearance of salivary duct carcinoma is similar to that of high-grade adenocarcinoma, thus resembling primary gastric cancer. In conclusion, when we perform endoscopic examination of patients with malignant neoplasias, a close study is required, considering a great risk of metastatic lesions. Furthermore, if endoscopic biopsy is per-

formed, we should provide pathologists with accurate information about the clinical history of the patient.

The histopathological findings on the primary lesion in this case have been reported in a Japanese article [11]. However, this is the first report on surgically resected gastric metastatic cancer originating from salivary duct carcinoma.

Conflict of interest

None.

Funding

No funding was used in this study.

Ethical approval

No ethical approval was required for this case report.

Author’s contribution

Kanefumi Yamashita was a clinician in charge, and wrote the manuscript. Shinsuke Takeno, Yoshikazu Sugiyama, Takayuki Sueta, Kenji Maki, Yoshiyuki Kayashima, Hironari Shiwaku, Daisuke Kato, Tatsuya Hashimoto, Takamitsu Sasaki, and Yuichi Yamashita were also clinicians in charge. Satoshi Nimura was the pathologist in charge.

Consent

A written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

Kanefumi Yamashita.

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