Bilateral lung metastasectomy in carcinoma of the ampulla of Vater

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Abstract
The efficacy of lung metastasectomy is well established in several cancers, including colorectal cancer. However, little is known about the result of lung metastasectomy in carcinoma of the ampulla of Vater. Only two case reports have reported the efficacy of metastasectomy in ampullary cancer patients with pulmonary metastasis. We report the result of bilateral lung metastasectomy in a patient with ampullary cancer. A 63-year-old woman underwent pylorus-preserving pancreaticoduodenectomy for carcinoma of the ampulla of Vater. About three years after the surgery, two non-calciﬁed lung nodules in the right lower and left upper lobes had developed. Wedge resections of both lung nodules were performed and the pathological examination showed that the lung nodules were pulmonary metastases from the ampullary cancer. Ten years after the lung surgery, the patient is well and there is no evidence of recurrence. Surgical resection could be considered in patients with pulmonary metastasis from ampulla of Vater cancer, even when the metastases are bilateral.

Introduction
Carcinoma of the ampulla of Vater is a rare tumor with an incidence rate estimated at six per million persons per year, but has steadily increased over the past 30 years.1,2 The only curative treatment for ampullary cancer is surgical resection: pancreaticoduodenectomy. Even after curative resection, relapse occurs in some patients. In general, the intra-abdominal area is the site of recurrence, and extra-abdominal metastasis usually develops in advanced stage. Only two cases of lung metastasectomy of ampullary cancer have been reported.3,4 Herein, we report a case of bilateral lung metastasectomy in a patient with ampulla of Vater cancer.

Case report
In June 2003, a 63-year-old woman underwent pylorus-preserving pancreaticoduodenectomy for carcinoma of the ampulla of Vater. Histological examination revealed a well-differentiated adenocarcinoma of the ampulla. The tumor was 1.2 × 1.0 × 0.5 cm³ in size with duodenal wall invasion, the resection margins were clear, and there was no metastasis in nine regional lymph nodes. It was classiﬁed as pT2N0M0, stage IB. Blood chemistry data were unremarkable, and carcinoembryonic antigen (CEA) and carbohydrate antigen (CA)19-9 levels were within normal limits. The patient was a non-smoker and her past medical and family histories were unremarkable.

After the surgery she was followed up with regular abdomen-pelvis computed tomography (CT) imaging. In May 2006, about three years after the surgery, an abdomen-pelvis CT showed a 1 cm sized mass-like lesion in the supraceliac area. Positron emission tomography/CT was performed to differentiate the possibility of metastatic lymph node or non-speciﬁc ﬁbrosis, and showed no hypermetabolic lesion in that area. However, two mild hypermetabolic lung nodules were found. The following chest CT showed two non-calciﬁed lung nodules in the right lower (1.5 cm) and left upper lobes (1 cm) (Fig 1). Percutaneous needle aspiration was performed and cytological examination revealed a few atypical cells suggestive of malignancy.
In July 2006, wedge resections of the right lower and left upper lobes were performed and pathologic examination revealed 1.5 cm sized nodules in both lobes. The resection margins were clear. The two nodules were well demarcated, focally necrotic, and consisted of tall columnar cells forming irregular tubules. The histological features were similar to those of the previous ampullary adenocarcinoma (Fig 2). Furthermore, the immunohistochemical properties of both lung nodules and ampullary tumors were similar. They both showed immunopositivity for cytokeratin 7 and 20, and immunonegative for mucin 2 and thyroid transcription factor-1 (Fig 3). These findings indicated that lung lesions favor metastatic adenocarcinoma from ampullary cancer rather than primary lung cancer. After surgery, postoperative chemotherapy with oral tegafur/uracil plus leucovorin was administered for a year. As of May 2016, 10 years after the lung surgery, the patient remained well with no evidence of recurrence.

**Discussion**

Ampullary cancer is a rare tumor and accounts for 12.7–32.2% of periampullary cancer. According to a recent study based on Surveillance, Epidemiology, and End Results cancer registry data, the curative resection rate of ampullary carcinoma is 40% and the postoperative five-year survival rate is 37.8%. Hsu et al. reported that 42% (57/135) of patients who underwent pancreaticoduodenectomy recurred and showed 31 liver metastases, 26 locoregional recurrences, nine peritoneal carcinomatoses, seven

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**Figure 1** Chest computed tomography showed two non-calcified lung nodules in the (a) left upper (1 cm) and (b) right lower lobes (1.5 cm).

**Figure 2** The histological features of the lung tumor are similar to those of the previous ampullary adenocarcinoma (hematoxylin and eosin staining). (a,b) Ampullary cancer and (c,d) lung tumor.
bone metastases, and six other sites of metastases, including the lung. The clinical course after lung metastasectomy in ampullary carcinoma has rarely been reported. To date, only two case reports have shown the results of lung metastasectomy in patients with lung metastasis of ampullary cancer. In 2005, a case of repeated hepatectomy and bilateral lung resection for metastasis from carcinoma of the papilla of Vater after pancreaticoduodenectomy was reported in Japan. The patient died 12 years after pancreaticoduodenectomy. In 2011, another case of solitary lung metastasectomy of ampullary cancer was reported with a relatively short follow-up period (14 months after lung surgery).

Although lung metastasectomy is a well-established procedure for colorectal cancer, definite indication is not defined in many other cancers, including ampullary cancer. The International Registry of Lung Metastases, the largest registry, has researched 5206 patients from 18 thoracic surgical units. Data shows that five-year and 10-year survival rates are better in complete pulmonary metastasectomy cases and demonstrated a survival benefit of lung metastasectomy in various cancers. When deciding whether to perform lung metastasectomy, many factors need to be considered, such as primary tumor histology, number and size of metastases, or the extent of resection. In a survey of the practice of pulmonary metastasectomy among thoracic surgeons, 17% of surgeons responded that bilateral pulmonary metastases are a relative (16%) or absolute (1%) contraindication for surgical resection. However, our case demonstrates that long-term survival is possible after metastasectomy, even in cases of bilateral lung metastasis in ampullary cancer.

In conclusion, surgical resection could be considered in patients with pulmonary metastasis in ampulla of Vater cancer, even when metastases are bilateral.

Disclosure
No authors report any conflict of interest.

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