

CASE REPORT

Rare case of rectal carcinoid with synchronous primary carcinoid tumors of the lung misdiagnosed as lung metastases

Fuminori Teraishi,^{*,†} Rhohei Shoji^{*} and Toshiyoshi Fujiwara^{*,†}^{*}Department of Gastroenterological Surgery, Okayama University Graduate School and [†]Department of Minimally Invasive Therapy Center, Okayama University Hospital, Okayama, Japan**Key words**

lung metastases, neuroendocrine tumor, rectal carcinoid.

Accepted for publication 3 July 2024.

Correspondence

Dr Fuminori Teraishi, Department of Gastroenterological Surgery, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, Kita-ku, Okayama 700-8558, Japan. Email: pkc1940h@okayama-u.ac.jp

Declaration of conflict of interest: The authors disclose no conflicts.

Abstract

A 68-year-old woman was referred to our hospital for rectal surgery after a pathological diagnosis of rectal carcinoid with venous invasion following endoscopic submucosal dissection of a 5 mm-sized submucosal tumor in the lower rectum. Chest CT showed nodules in the left upper lobe and right lower lobe, but positron emission tomography and somatostatin receptor scintigraphy showed no hyperaccumulation in the lung nodules. CT-guided needle biopsy was performed on the nodular lesion in the left upper lobe, which showed focal growth of tumor cells with a high N/C ratio and positive synaptophysin, leading to a diagnosis of pulmonary metastasis of rectal carcinoid. Since the patient was asymptomatic and did not wish to undergo surgery or chemotherapy, she was followed up strictly with sufficient informed consent. Three years have passed since the diagnosis, and there is no tendency for the lung metastasis to increase, and no other new lesions have been observed. The disease had not progressed and remained stable. Therefore, immunohistological analysis of the lung biopsy specimen was performed again, which was positive for TTF-1 and negative for CDX2. Consequently, the diagnosis was changed to primary lung carcinoid tumors, and the patient remains under follow-up with no disease progression.

A 68-year-old woman was diagnosed with a 5 mm submucosal tumor in the lower rectum (Fig. 1a) and underwent endoscopic submucosal dissection (ESD) for diagnostic treatment. ESD specimen showed the tumor was approximately 5 × 5 mm in diameter, well demarcated, and negative for margins. Pathology results confirmed rectal carcinoid (neuroendocrine tumor; NET G1) with venous invasion (Fig. 1a), and the patient was referred to our hospital for rectal resection. Although chest CT revealed small nodules in the left upper lobe and right lower lobe (Fig. 1b), positron emission tomography and somatostatin receptor scintigraphy showed no hyperaccumulation in the pulmonary nodules. A CT-guided needle biopsy was performed on the nodular lesion in the left upper lobe (Fig. 1c), and pathology revealed focal proliferation of tumor cells with a high N/C ratio and positive synaptophysin (Fig. 1d), consistent with a diagnosis of NET G1. This led to a clinicopathological diagnosis of lung metastasis from rectal carcinoid. Since the patient did not wish to undergo rectal and pulmonary resection or systemic chemotherapy, we decided to closely follow her up. Three years have passed since the diagnosis, and there is no increasing trend in lung tumors (Fig. 1e), nor have new lesions been observed. The disease had not progressed and remained stable. Therefore,

immunohistological analysis of the lung biopsy specimen was performed again, which was positive for TTF-1 and negative for CDX2. Consequently, the diagnosis was changed to primary lung carcinoid tumors (Fig. 1f), and the patient remains under follow-up with no disease progression.

Carcinoid tumors, arising from neuroendocrine cells, have the potential to metastasize to various organs, with the lungs being one of the most common sites of distant spread.¹ Although carcinoids are generally considered slow-growing and indolent tumors, lung metastases can significantly impact prognosis and management. Clinically, lung metastases from carcinoid tumors may be asymptomatic or present with symptoms such as cough, dyspnea, chest pain, or hemoptysis. Radiographic evaluation, including chest X-ray, CT scans, or somatostatin receptor scintigraphy, is essential for detecting and characterizing pulmonary lesions and assessing their impact on lung function.

Treatment strategies for lung carcinoid tumors depend on various factors including the extent of disease, tumor burden, presence of symptoms, and patient preferences.² The choice of treatment modality should be individualized based on tumor biology, extent of metastatic disease, and patient-specific factors. In our case, the clinical diagnosis of pulmonary metastasis of rectal

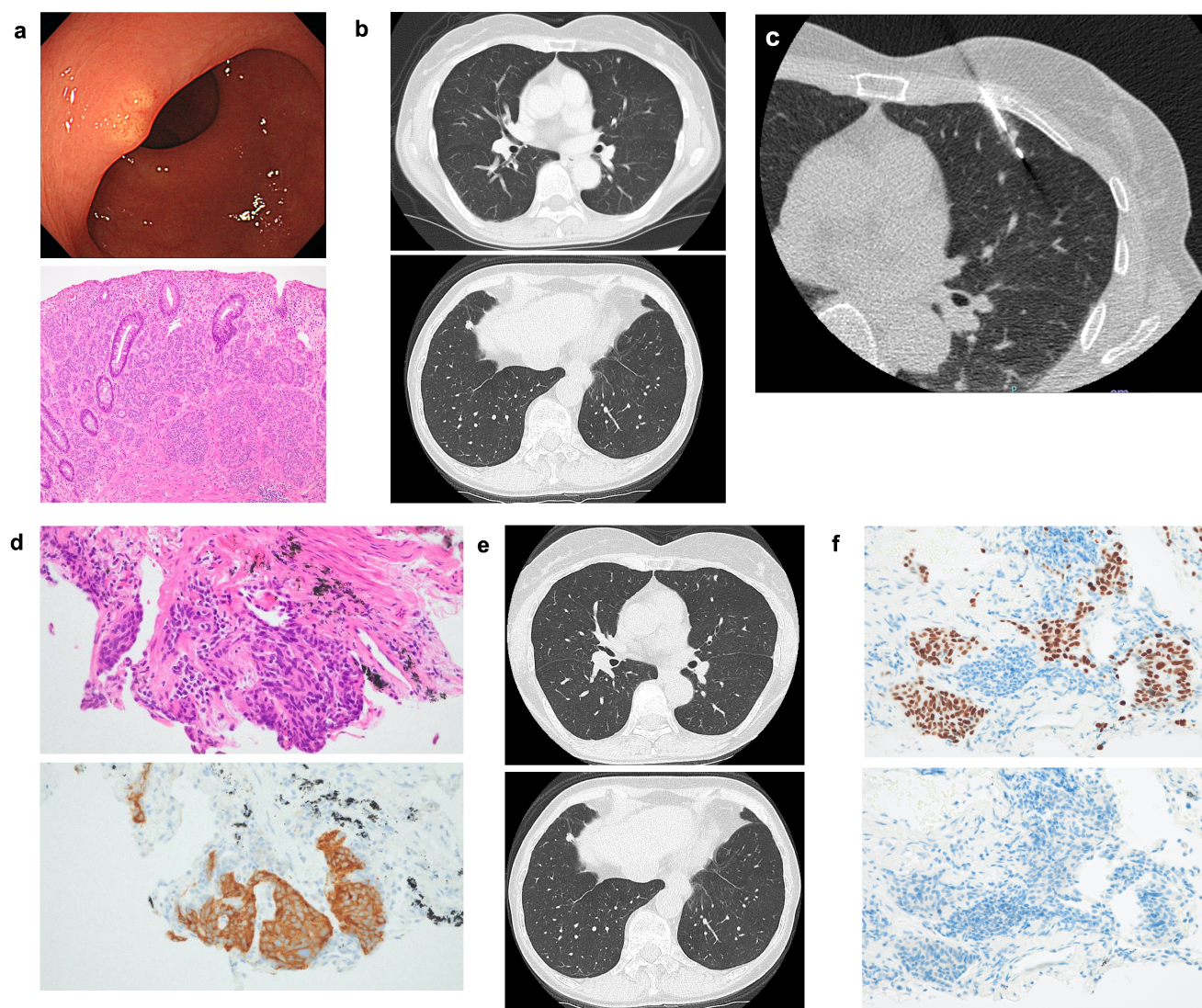


Figure 1 (a) Rectal endoscopy revealed a 5-mm submucosal tumor with a smooth white surface in the lower rectum (upper panel). Histology of the rectal ESD specimen showing focal proliferation of tumor cells with a high N/C ratio (lower panel, H&E stain), which was consistent with a diagnosis of rectal carcinoid. (b) Chest CT scan revealed small nodules in the left upper lobe (upper panel) and right lower lobe (lower panel). (c) A CT-guided needle biopsy was performed on the nodular lesion in the left upper lobe. (d) Histology of the left lung nodule biopsy specimens showing focal proliferation of tumor cells with a high N/C ratio (upper panel, H&E stain, original magnifications $\times 200$) with positive synaptophysin (lower panel, synaptophysin stain, original magnifications $\times 200$), which was consistent with a diagnosis of lung carcinoid. (e) Three years later, chest CT scan showed no change in the left upper lobe lung and right lower lobe lesions. (f) Immunohistochemical staining of the left lung nodule biopsy specimens showing positive TTF-1 stain (upper panel, TTF-1 stain, original magnifications $\times 200$) and negative CDX2 stain (lower panel, CDX2 stain, original magnifications $\times 200$), which was consistent with a diagnosis of primary lung carcinoid.

carcinoid was made based on the clinical course of the disease and the morphology of the lung lesions. Although the patient did not wish to undergo aggressive treatment and could be followed up without disease progression, an immunohistological search should have been performed at the time of initial treatment to differentiate this case from primary pulmonary carcinoid, since the lung is the second most common site of carcinoid after the gastrointestinal tract.³

Informed consent. Written, informed consent was obtained from the patient for publication of this case report and any accompanying images.

Data availability statement. The data that support the findings of this study are available from the corresponding author upon reasonable request.

References

- 1 Riihimaki M, Hemminki A, Sundquist K, Sundquist J, Hemminki K. The epidemiology of metastases in neuroendocrine tumors. *Int. J. Cancer*. 2016; **139**: 2679–86.
- 2 Carvao J, Dinis-Ribeiro M, Pimentel-Nunes P, Libanio D. Neuroendocrine tumors of the gastrointestinal tract: a focused review and practical approach for gastroenterologists. *GE Port. J. Gastroenterol*. 2021; **28**: 336–48.
- 3 Yao JC, Hassan M, Phan A *et al.* One hundred years after “carcinoid”: epidemiology of and prognostic factors for neuroendocrine tumors in 35,825 cases in the United States. *J. Clin. Oncol*. 2008; **26**: 3063–72.