Case Report

Solitary Splenic Metastasis of Lung Cancer Presenting as Benign Cystic Disease

Katsunari Matsuoka, MD, Tetsu Yamada, MD, Takahisa Matsuoka, MD, Shinjiro Nagai, MD, Mitsuhiro Ueda, MD, and Yoshihiro Miyamoto, MD

Solitary splenic metastasis from primary lung cancer is extremely rare. Here, we demonstrated a solitary splenic metastasis of primary lung cancer that was difficult to distinguish from benign cystic disease. A 69-year-old-female was diagnosed as middle lobe lung cancer. Although preoperative abdominal computed tomography (CT) demonstrated a low-density splenic nodule, fluorodeoxyglucose-positron emission tomography (FDG-PET) revealed no fluorodeoxyglucose uptake in the splenic nodule. Therefore, the nodule was diagnosed as benign cystic disease and middle lobe lobectomy was performed. Postoperative pathologic examination demonstrated papillary-predominant adenocarcinoma with mucin, and the tumor was diagnosed as primary lung cancer. However, the splenic nodule continued to increase postoperatively. Splenectomy was undergone 30 months after the pulmonary resection and the splenic tumor was diagnosed as the splenic metastasis of lung cancer. In the 24 months since the splenectomy, no recurrence has been observed in the absence of treatment. Splenectomy was an effective treatment for solitary splenic metastasis of lung cancer in this case. FDG uptake in the splenic tumor was not evident due to marked mucus production.

Keywords: solitary, splenic metastasis, lung cancer

Introduction

Solitary splenic metastasis from primary lung cancer is extremely rare.^{1,2)} Here, we report a case of solitary splenic metastasis from primary lung cancer presenting as benign cystic disease in view of negative fluorode-oxyglucose-positron emission tomography (FDG-PET) findings and a cyst-like appearance on computed tomography (CT) and magnetic resonance imaging (MRI).

Department of Thoracic Surgery, National Hospital Organization Himeji Medical Center, Himeji, Hyogo, Japan

Received: April 22, 2019; Accepted: July 28, 2019 Corresponding author: Katsunari Matsuoka, MD. Department of Thoracic Surgery, National Hospital Organization Himeji Medical Center, 68 Honmachi, Himeji, Hyogo 670-8520, Japan Email: katsmatsuoka@gmail.com



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Case

A 69-year-old-woman was referred to our hospital because of an abnormal shadow on a chest roentgenogram. Roentgenography and CT of the chest demonstrated a nodular shadow with an irregular margin in the right middle lobe (Fig. 1a). CT demonstrated a low-density area within the nodule. The preoperative level of carcinoembryonic antigen (CEA) was high at 19.6 ng/mL. The nodule was diagnosed as adenocarcinoma by transbronchial biopsy with a bronchofiberscope. Abdominal CT showed a low-density nodule in the spleen (**Fig. 1c**). FDG-PET revealed uptake of FDG in the lung nodule (Fig. 1b), but FDG uptake was not observed in the splenic nodule (Fig. 1d). Because of the low-density area revealed by CT and the lack of FDG uptake in the splenic nodule, a benign cystic lesion such as lymphangioma or hemangioma was suspected. Therefore, we performed right middle lobectomy with lymph node dissection. Postoperative pathologic examination demonstrated papillary-predominant adenocarcinoma with mucin,

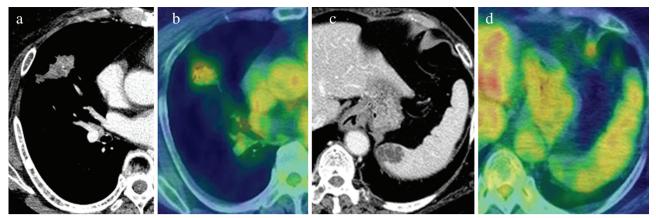


Fig. 1 Chest CT demonstrated a nodular shadow with an irregular margin in the right middle lobe (a). There was a low-density area within the nodule. FGD-PET demonstrated uptake of FDG in the lung tumor (b). Abdominal CT demonstrated a low-density mass in spleen (c). No FDG uptake was evident (d). CT: computed tomography; FGD: fluorodeoxyglucose; PET: positron emission tomography

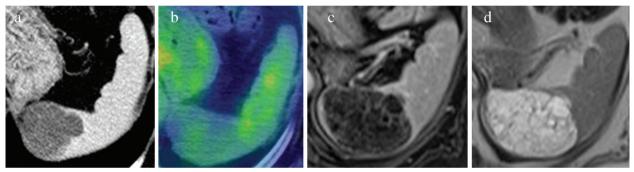


Fig. 2 Abdominal CT demonstrated an increase in size of the splenic nodule (a), although PET examination revealed no FDG uptake in the splenic nodule (b). MRI revealed that the splenic nodule was polycystic, showing low intensity in the T1-weighted image (c) and high intensity in the T2-weighted image (d). CT: computed tomography; FGD: fluorodeoxyglucose; MRI: magnetic resonance imaging; PET: positron emission tomography

and the tumor was diagnosed as primary lung cancer (pT2aN0M0). The CEA value did not decrease after surgery, and chest and abdominal CT demonstrated an increase in size of the splenic nodule (Fig. 2a), although no other abnormal findings were evident. PET examination in the first year after the operation revealed no FDG uptake in the splenic nodule (Fig. 2b). MRI revealed that the splenic nodule was polycystic, showing low intensity in the T1-weighted image and high intensity in the T2-weighted image (Fig. 2c and 2d). The splenic mass showed no solid part, and therefore a benign cystic tumor such as lymphangioma or hemangioma was still suspected. Because the splenic mass continued to increase in size, laparoscopic splenectomy was performed 30 months after the pulmonary resection. Macroscopic and microscopic findings of the excised spleen are demonstrated in Fig. 3. Pathological examination demonstrated adenocarcinoma composed of high columnar epithelium cells with large amount of mucus and the splenic tumor was diagnosed as the splenic metastasis of lung cancer. Thereafter, the CEA level quickly dropped to below the reference value. In the 24 months since the splenectomy, the CEA level has remained below the reference value and no recurrence has been observed in the absence of treatment.

Discussion

Splenic metastasis of primary lung cancer has been reported in 1.2%–5.6% of autopsy cases, but most metastasis involve other organs.^{1,2)} Solitary splenic metastasis is extremely rare and only 31 cases have been reported previously.^{3–5)} The spleen is relatively resistant to metastatic



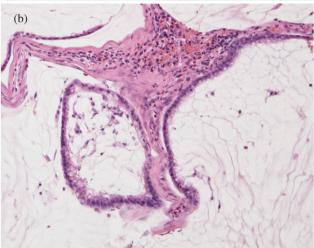


Fig. 3 Macroscopic (a) and microscopic (b) findings of the excised spleen. Pathological examination demonstrated adenocarcinoma composed of high columnar epithelium cells with large amount of mucus.

seeding. Immune cells that play a role in immune surveillance are present in high density in the spleen, help to eliminate the cancer cells before they proliferate, and because of high concentration of antiangiogenic factors, the spleen creates an environment that is not advantageous to the growth of metastatic cancer cells. ^{3,6,7)}

Some reports have described the usefulness of FDG-PET for detection of splenic metastasis.^{8,9)} In the present case, however, FDG uptake was not observed in the splenic tumor. Because a large amount of mucus was produced by the metastatic tumor in the spleen, FDG uptake was

not observed. Previous reports also demonstrated that FDG-PET was limited in the detection of mucinous tumors, particularly in hypocellular lesions with abundant mucin. ^{10,11)} In mucinous tumors, tumor cells are present surrounded by a large amount of mucus, resulting in a relative reduction in the number of viable cancer cells. Because FDG uptake was related to the number of viable cancer cells, the results of FDG-PET tend to be false negative in mucinous tumors. ¹⁰⁾ MRI examination showed low intensity in the T1-weighted images and high intensity in the T2-weighted images, thus mimicking a benign cyst. It should be kept in mind that FDG uptake may not be evident in cancers with marked mucus production.

Splenectomy has been performed in most of the cases reported previously. Although there are no guidelines related to the indication for splenectomy in cases of solitary splenic metastasis, it is reported that long-term remission can be achieved by splenectomy alone in patients with solitary splenic metastasis from lung cancer. 1,4,12) Systemic chemotherapy after splenectomy is a reasonable option, but currently there is no supportive evidence for its efficacy. In the present case, we followed up the patient without additional treatment because the CEA level decreased rapidly to below the reference value after splenectomy, and 24 months later no recurrence was evident. In cases with oligo-metastasis, the clinical course differs from that of normal stage 4 lung cancer, and further studies of additional adjuvant chemotherapy after resection are required.

Conclusion

We have experienced an extremely rare case of solitary splenic metastasis from a primary lung cancer that was difficult to distinguish from benign cystic disease. FDG uptake in the splenic tumor was not evident due to marked mucus production. Splenectomy was an effective treatment for solitary splenic metastasis of lung cancer in this case.

Informed Consent

Informed consent was obtained and the Institutional Review Board approved this retrospective study.

Disclosure Statement

None declared.

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