Primary splenic malignant lymphoma mimicking metastasis of rectosigmoid cancer: A case report

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INTRODUCTION: Primary splenic malignant lymphoma is quite a rare disease, and its preoperative diagnosis is difficult.

CASE PRESENTATION: An 80-year-old man was diagnosed with advanced rectosigmoid cancer with liver and splenic metastases, for which he underwent single-incision laparoscopic high anterior resection for the primary rectosigmoid cancer. After chemotherapy, he underwent laparoscopy-assisted splenectomy and open partial hepatectomy of segment 3 and segment 5/6 of the liver. The resected specimen of the spleen showed primary splenic malignant lymphoma.

DISCUSSION: A diagnosis of primary splenic malignant lymphoma seems to be made only occasionally at splenectomy. Patients with primary splenic malignant lymphoma treated by curative resection at an early clinical stage have a more favorable prognosis. Laparoscopy-assisted splenectomy is useful for reducing surgical invasiveness.

CONCLUSION: Primary splenic malignant lymphoma should be included among the differential diagnoses of splenic tumors in patients with colorectal cancer with multiple metastases. Curative resection might be a therapeutic option for the treatment of primary splenic malignant lymphoma. Laparoscopy-assisted splenectomy is a useful strategy for reducing surgical invasiveness.

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1. Introduction

Preoperative diagnosis of primary splenic malignant lymphoma (PSML), which is a relatively rare disease, is difficult. A review of previous literature showed that the initial diagnosis of PSML is made only occasionally at splenectomy [1]. We report herein a rare case of PSML mimicking splenic metastasis of rectosigmoid cancer. This work has been reported in line with the SCARE criteria [2].

2. Case presentation

A previously healthy 80-year-old man visited our hospital for evaluation of elevated biliary enzyme levels in November 2016. He had no symptoms, such as fever, weight loss and night sweats. Laboratory tests demonstrated alkaline phosphatase (ALP) of 471 U/dl (normal 105–340 U/dl) and lactate dehydrogenase (LDH) of 570 U/L (normal 110–230 U/L). The serum levels of carcinoembryonic antigen (CEA) and carbohydrate antigen 19-9 (CA19-9) were 62.5 ng/ml (normal 0–5 ng/ml) and 452.4 U/ml (normal 0–37 U/ml), respectively. Computed tomography (CT) scan showed rectosigmoidal wall thickening in the pelvis, a liver tumor in segment 5/6, splenic tumors (Fig. 1a), and a left lung tumor. The left lung tumor, located in segment 10, presented pure ground-glass opacification with pleural indentation. Colonoscopy showed a type 2 rectosigmoid tumor, 45 mm in size, located 15 cm from the anal verge. Examination of biopsy specimens of the rectosigmoid tumor revealed moderately differentiated adenocarcinoma. Positron emission tomography with 2[18 F]-fluoro-2-deoxy-d-glucose (FDG-PET) revealed a rectosigmoid tumor with a maximum standardized uptake value (SUVmax) of 11.6, a left lung tumor with a SUVmax of 3.1, a liver tumor and splenic tumors with excessive uptake values, consistent with the CT scan findings. There were no excessive uptake values in the thoracic and abdominal lymph nodes. These findings led to a preoperative diagnosis of advanced rectosigmoid cancer with liver and splenic metastases and primary left lung cancer. Therefore, the patient first underwent single-incision laparoscopic high anterior resection in December 2016. Examination of the surgical specimen revealed moderately differentiated adenocarci-
nomal penetrating the serosa, with regional lymph node metastases. Six courses of XELOX (capecitabine and oxaliplatin) plus panitumumab chemotherapy were administered after rectosigmoid cancer surgery. CT scan performed after chemotherapy demonstrated significant reduction in size of the hepatic and splenic tumors (Fig. 1b), although the tumor in segment 10 of the left lung had enlarged. On dynamic contrast-enhanced magnetic resonance imaging (MRI), T1-weighted imaging showed a signal-hypointense mass in segment 3 of the liver, in addition to the already-known tumor in segment 5/6 (Fig. 2). The serum levels of CEA and CA19-9 decreased to 8.3 ng/mL and 7.7 U/mL, respectively. In May 2017, the patient underwent laparoscopy-assisted splenectomy and open partial hepatectomy of segment 3 and segment 5/6 of the liver. First, the spleen was mobilized from the retroperitoneum using the three-port laparoscopic technique (Fig. 3). After mobilization of the spleen, a J-shaped subcostal incision was made and splenectomy and partial hepatectomy of segment 3 and segment 5/6 of the liver were performed. Intraoperative evaluation showed no lymph node swelling in the abdominal cavity. Pathological findings of the liver tumors were compatible with metastases of rectosigmoid cancer. Macroscopically, the splenic tumors were whiter and firmer than liver tumors (Fig. 4). Microscopically, the splenic tumors demonstrated a diffuse proliferation of large lymphoid cells that were composed of diffusely proliferating atypical cells with irregular medium, to large round or oval nuclei and a high nucleus/cytoplasm (N/C) ratio (Fig. 5). Immunohistochemically, the tumor cells were positive for the B-cell marker CD20, but not for CD3, CD5, and CD10.
The medium primary lymphoma, recurrence period, tumor issues. Ki-67 was diagnosed as a diffuse B-cell lymphoma.

The Ki-67 index was 80–90% (Fig. 6). Therefore, the splenic tumor was diagnosed as a diffuse B-cell lymphoma.

These findings led to a final diagnosis of advanced rectosigmoid cancer with multiple liver metastases of T4aNOM1a stage IVA, and primary splenic high grade B-cell lymphoma stage I, according to Ahmann [1] and Ann Arbor [3] staging. During the 3-months follow-up period, the patient continued to do well without any signs of recurrence or metastasis of rectosigmoid cancer and malignant lymphoma, and hence, curative resection of the lung cancer was considered as the next treatment.

3. Discussion

The clinical course of this patient suggests three important clinical issues. First, PSML should be included among the differential diagnoses of splenic tumors in patients suffering from colorectal cancer with multiple metastases. Second, curative resection might be a therapeutic option for the treatment of PSML. Third, laparoscopy-assisted splenectomy is useful for reducing the length of incisions and the operative invasiveness.

Our experience highlights the fact that PSML should be included among the differential diagnoses of splenic tumors in patients with colorectal cancer with multiple metastases. PSML is the most frequent type of primary splenic malignant tumor, reportedly accounting for 22.2–77.8% of splenic malignancies [4,5]. The incidence of PSML was less than 1% of 5100 lymphomas treated over 18 years at the Mayo Clinic [1]. The initial diagnosis of PSML seems to be made only occasionally at splenectomy [1]. Metastasis to the spleen from non-hematological diseases is very uncommon, and the main sites of primary tumors that metastasize to the spleen are the lung, colon and rectum, ovary, malignant melanoma and breast [6,7]. Metastases to the spleen generally occur in the context of terminal stages of multivisceral metastatic cancer. In our case, preoperative diagnosis of PSML was quite difficult because of the coexistence of liver metastases of the rectosigmoid cancer and elevated levels of CEA and CA19-9.

Our case suggests that curative resection might be a therapeutic option for the treatment of PSML. Sumimura et al. [8] reported that curative resection was mainly performed in patients with stage I PSML according to Ahmann staging [1], and that the 3-year survival rate of stage I (76.0%) was significantly better than that of stage III (16.0%) disease, based on an evaluation of the 71 patients with PSML reported in Japan till now. They concluded that PSML patients treated by curative resection at an early clinical stage have a more favorable prognosis. For stage III patients with involvement of the liver or lymph nodes beyond the splenic hilum, the survival rate in the curative resection group was significantly better than that in the non-curative resection or unresected group [8].

When performing splenectomy, laparoscopy-assisted splenectomy is useful for reducing the length of incisions and the operative invasiveness for the patient. A left subcostal incision is employed in most patients undergoing open splenectomy. In order to obtain good exposure for the resection of tumors in the right lobe of
the liver and spleen, major abdominal incisions, such as a Mercedes incision, might be required. In this case, we could omit the left subcostal incision by mobilizing the spleen laparoscopically from the retroperitoneum, and could resect the tumors in the right lobe of the liver and the spleen using an open procedure with a J-shaped subcostal incision. Experienced laparoscopic surgeons might perform the entire partial right heptectomy and splenectomy procedures, from start to finish, laparoscopically within a reasonable operative period [9].

Precise diagnosis and a multidisciplinary surgical approach might lead to a favorable prognosis in patients with PSML. Patients with hepatic and pulmonary metastases from colorectal cancer may also benefit from aggressive surgical therapy [10]. Besides, PSML patients treated by curative resection at an early clinical stage have a more favorable prognosis. We intended a curative resection of colorectal cancer with multiple organ metastases after chemotherapy, which resulted in a diagnosis of PSML and curative resection of the colorectal cancer, hepatic metastases and PSML.

4. Conclusions

PSML should be included among the differential diagnoses of splenic tumors in patients with colorectal cancer with multiple metastasis. Curative resection might be a therapeutic option for the treatment of PSML. Laparoscopy-assisted splenectomy is useful for reducing surgical invasiveness.

Conflicts of interest

The authors declare no potential conflict of interest.

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Ethical approval

Ethical approval for a case report is not required by our institution.

Consent

Written informed consent was obtained from the patients for the information to be included in our manuscript. His information has been de-identified to the best of our ability to protect his privacy.

Author contributions

Each author participated in writing the manuscript and all agreed to accept equal responsibility for the accuracy of the content of the paper.

Guarantor

Masaki Wakasugi.

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