Case Report

Spontaneous Gastrosplenic Fistula in Primary Gastric Lymphoma: Surgical Management

Mustafa Kerem, Omer Sakrak, T. Utku Yilmaz, Fatma Ayca Gultekin, Ayse Dursun and Abdulkadir Bedirli, Departments of General Surgery and *Pathology, Faculty of Medicine, Gazi University, Ankara, Turkey.

Gastrosplenic fistula formation resulting from primary gastric malignancy is rare and should be managed as a matter of emergency. We report a patient who was diagnosed with primary non-Hodgkin’s lymphoma and who underwent surgical treatment for gastrosplenic fistula. [Asian J Surg 2006;29(4):287–90]

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Introduction

Gastrosplenic fistula formation resulting from primary gastric malignancy is rare and should be managed as a matter of emergency. Here, a patient who was diagnosed with primary non-Hodgkin’s lymphoma and who underwent surgical treatment for gastrosplenic fistula is reported.

Case report

A 57-year-old man, a farmer, presented to a local hospital on May 2004 with abdominal and back pain and a history of a 7 kg weight loss in the last 3 months in conjunction with episodic epigastric pain and dyspepsia. He had neither night fever nor jaundice. On the first physical examination, no peripheral lymphadenopathy or neurological deficit was found, but splenomegaly and cardiac failure were established. Ultrasonographic examination showed that there were cystic and nodular formations with suspicion of malignancy in the spleen. Some millimetric lymph nodes were seen on carinal section of the mediastinum in thoracic computed tomography (CT). Further investigation with abdominal magnetic resonance imaging showed thickening of the corpus of the stomach, hepatomegaly with 17 cm cranial–caudal length and an air-containing mass in the spleen. Nodular areas in the spleen were interpreted as necrotic areas due to lymphoma. There were no signs of intra-abdominal lymphadenopathy or bone-marrow pathology, which can affect the prognosis of the disease. Tumour markers were: carcinoembryonic antigen, 1.65 ng/mL (normal range, 0–3.4 ng/mL); α-fetoprotein, 0.964 IU/mL (normal range, 0–11.3 IU/mL); CA19-9, 15.22 U/mL (normal range, 0–39 U/mL). Bone-marrow biopsy or flow cytometry did not reveal any other evidence of disease. Due to cardiological comorbidities such as dilated cardiomegaly, first to second degree mitral insufficiency, minimal aortic insufficiency, second degree tricuspid insufficiency, left atrial dilatation, pulmonary hypertension and 27% ejection fraction ratios, the patient was referred to our centre for further treatment. On the first examination in our clinic, the patient had abdominal pain radiating to the back and tenderness over the epigastrium. Ultrasonographic examination revealed no other pathology than the nodular lesion, measuring 35 × 40 mm, which enlarged the inferior border of the spleen. On CT, there was a collection measuring 7 cm in diameter in the upper pole of the heterogeneous spleen. Thick-walled collection containing air densities, indicative of splenic abscess or gastrosplenic fistula, had pushed the greater curvature of the stomach towards the anterior...
side. Also, there was a smooth bordered, hypodense, heterogeneous area measuring 3 cm in diameter next to the lower border of the spleen (Figure 1). In the level of the portal hilus, there was a low attenuation area that deviated the para-aortic region to the left and could not be differentiated at the borders of the stomach, body of the pancreas and hilus of the spleen. Further evaluation with positron emission tomography (PET/CT) showed prominent thickening in the fundus and lesser curvature of the stomach with high 18F-FDG attenuation. There was also a mass lesion that occupied a huge part of the spleen, with central hypovascularity accompanying high 18F-FDG attenuation around it like a ring. With regard to the results of both CT and PET/CT, it was interpreted as invasion and infiltration of the gastric tumour to the spleen.

While planning gastroscopy, the patient underwent emergency operation because of the acute abdomen. Under general anaesthesia, the abdomen was opened via a midline incision. On exploration, the spleen was densely adherent to the anterior wall of the abdomen and diaphragm, and the splenic hilus was adherent to the splenic flexura. There was thickening along the posterior wall of the stomach from the fundus to cardia. The posterior surface of the fundus was densely adherent to the spleen. Also present were perisplenic oedema, inflammation, wall thickening on the transverse mesocolon, lymphadenopathy (reaching 2 cm in diameter) along the para-aortic, splenic hilus and coeliac truncus. The spleen was detached from the anterior abdominal wall and diaphragm. Following the dissection of the upper pole of the spleen, an inflamed and necrotic 7 cm of splenic segment and approximately 6 cm of fully perforated fundus were found leading to the gastrosplenic fistula.

Splenectomy, proximal gastrectomy, oesophagostomy and pyloroplasty were successfully performed. Several biopsies were taken from the liver, para-aortic and coeliac lymph nodes. Pathological evaluation revealed infiltration of diffuse B cell non-Hodgkin’s lymphoma, staining positively for CD20 and leucocyte common antigen, and negatively for CD3, CD5, CD10, pancreatin and sinaptophysin in an area measuring 10 × 7 × 2 cm in the stomach and 8 × 5 × 4 cm in the spleen (Figure 2). Normal splenic parenchyma was loosely seen between the necrotic areas. Tumour was seen in the serosa of the stomach, whereas no tumour was seen in the surgical border. The patient was discharged on the 5th day after operation without any complaint, and received chemotherapy with cyclophosphamide, hydroxydaunomycin, oncovin, prednisone.

Discussion

Most centres employ a multimodal treatment programme for patients with gastric lymphoma. The role of resection in gastric lymphoma remains controversial, and many patients are now being treated with chemotherapy plus radiation therapy.2,3 Emergency situations, particularly in small bowel involvement, are often a reason for primary surgical intervention. Anecdotal cases of complications (e.g. spontaneous perforation and bleeding) with primary
Chemotherapy or radiation therapy have been considered a reason for primary operation.\textsuperscript{4–6} Surgical treatment has also often been advocated for establishing an accurate pathological staging and reliable histological examination.\textsuperscript{2,3} Here, we report a patient with gastric lymphoma that led to gastrosplenic fistula.

Direct communication between two abdominal viscera is usually the result of congenital, traumatic, inflammatory, neoplastic or iatrogenic processes affecting one or both organs. The development of internal gastrointestinal fistula is a rare process in malignancies and has been reported in less than 1% of gastric cancers.\textsuperscript{7} A fistulous tract between the stomach and the spleen is a very rare manifestation. Our search of the literature led to the finding of a report by de Scoville et al\textsuperscript{8} in which lymphosarcoma of the spleen spontaneously perforated into the stomach. The term “aereosplenomegaly” was used to describe this radiological finding. There have been 13 reported cases of gastrosplenic fistula to date, of which eight have been related to malignancy.\textsuperscript{8–18} Among causes of malignancy leading to gastrosplenic fistula, there were six lymphomas, one adenocarcinoma and one lymphosarcoma.\textsuperscript{9–16} Among the fistula-causing lymphomas, four were in the spleen and two were of stomach origin. Fistula formation followed chemoradiotherapy in five of six lymphoma cases, and developed spontaneously in one case.\textsuperscript{9,11–14,16} Primary gastric lymphoma patients with spontaneous gastrosplenic fistula are not able to undergo any operation due to high anaesthesia risk.\textsuperscript{9} In the present case, surgical intervention was indispensable because of the acute abdomen signs. This is the first case to have undergone surgical treatment due to spontaneous gastrosplenic fistulation.

Among the gastrointestinal lymphomas, postoperative complication rates are very high, especially in emergency cases (5–27%).\textsuperscript{2,3,19–22} In our case, proximal gastrectomy and oesophagogastrrectomy including the tumour were performed without any postoperative complications. Adjuvant chemoradiotherapy was started on the 15\textsuperscript{th} postoperative day.

Perforations to the intra-abdominal space and adjacent organs are more frequently seen in diffuse large cell type of gastrointestinal malignant lymphomas than in other types.\textsuperscript{9,11–14,16} Wide serosal infiltration and necrosis are markedly seen in this type of lymphoma. Also, histopathological diagnosis of splenic lymphomas leading to gastrosplenic fistula and primary gastric lymphoma, which was recently reported as the cause of spontaneous gastrosplenic fistula by Puppala et al,\textsuperscript{9} was diffuse large B cell non-Hodgkin’s lymphoma. The histopathological result of our case was the same.

The rate of perforation due to acute tumour lysis syndrome following adjuvant chemotherapy in GIS lymphoma patients was 5%,\textsuperscript{2,3,20–22} Malignant non-Hodgkin’s lymphoma’s lymphomas arising from the stomach are generally of the diffuse large cell type (56%), which are more aggressive.\textsuperscript{2,3} With the application of aggressive adjuvant chemotherapy in these large cell lymphomas, more prominent tumour lysis syndrome signs can be achieved. This second case of spontaneous perforation reported in the literature was probably due to the necrosis caused by the largeness of the tumour.

Abdominal CT is superior to other radiological tests in the diagnosis of gastrosplenic fistula.\textsuperscript{9} Air–fluid levels in the spleen should alert us to gastrosplenic fistulae. In our case, PET/CT showed lesions in the proximal stomach and the upper part of the spleen as malignancy but was unable to explain the situation. Gastroscopy was used in two cases in the literature.\textsuperscript{9,13} In our case, gastroscopic investigation was planned but not carried out because of development of acute abdomen.

In conclusion, it is better to keep in mind that spontaneous gastrosplenic fistulas can be seen in gastric lymphomas, especially in diffuse large cell type, as a complication of perforations to the intra-abdominal space that occur following adjuvant therapy.

References