

Gastrosplenic fistula due to splenic large B-cell lymphoma

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A gastrosplenic fistula (GSF) is an unusual complication arising from a variety of primary gastric or splenic malignant lesions and less commonly from benign diseases. Splenic large cell lymphoma may be a main cause of this distinctive complication. We report a case of 62-year-old male with spontaneous GSF due to pathologically proven splenic large cell lymphoma who was diagnosed by computed tomography and treated successfully by surgical management.

Key words: Computed tomography, gastrosplenic fistula, splenic lymphoma

INTRODUCTION

A fistulous tract between the stomach and the spleen is a very rare manifestation. The first gastrosplenic fistula (GSF) was reported in Belgium in 1962. The authors described a case of GSF with a characteristic radiographic appearance due to the presence of air in the spleen, which they termed "aerosplenomegaly".^[1] We searched using Medline engine with the term "gastrosplenic fistula" and found only 24 case reports up to 2012. Splenic malignant lymphoma, especially the diffuse, large cell type, may be one main cause of this distinctive complication because of its tendency to gastric wall invasion and extensive tumor necrosis. GSFs not only occur after chemotherapy for splenic lymphoma,^[2] but also occur spontaneously.^[3] We report a case of spontaneous GSF in a 62-year-old patient with splenic, diffuse, large cell lymphoma. On abdominal computed tomography (CT), a fistulous tract between the gastric lumen was visualized which verified by endoscopical biopsy, the patient was successfully treated by surgical management.

CASE REPORT

A 62-year-old man was admitted to our hospital (the Second Hospital of Shandong University, Jinan, China) on May 2009 with complaints of left upper quadrant

pain that radiated to the back and a history of 8 kg weight loss in the last 2 months in conjunction with fatigue, dyspepsia, and episodic fever in the last 2 weeks. The pain was accompanied by nausea, which worsened to the point where he was unable to lie on his left side. His medical history was negative except appendectomy for acute appendicitis 15 years ago. On the physical examination, there was left upper quadrant tenderness with normal bowel sounds, no peripheral lymphadenopathy or neurological deficit was found, but splenomegaly with a palpable tip was established.

Routine admission laboratory tests were as follows: CRP 284 mg/L (normal range, 0–10 mg/L), WBC count $14.6 \times 10^9/L$ (normal range, $4.0\text{--}10.0 \times 10^9/L$), Hb 92 g/L (normal range, 120–160g/L), and albumin level of 29 g/L (normal range, 30–35g/L). tumor markers were: carcinoembryonic antigen, 2.03 ng/mL (normal range, 0–3.4 ng/mL); a-fetoprotein, 3.48 IU/mL (normal range, 0–11.3 IU/mL); and CA19-9, 13.44 U/mL (normal range, 0–39 U/mL). The chest and abdominal radiographs showed no abnormality. Abdominal CT scan showed the presence of air in the spleen, and a 1.6cm fistulous tract between the stomach and the hilum of an enlarged spleen, with a large splenic abscess [Figure 1]. Esophagogastroduodenoscopy showed next to the cardia in the greater curvature of the stomach an ulcerative lesions with regular margins (about 3 cm wide, 7 cm deep) and the rest of the gastric mucosa without alterations. Cold forceps biopsy specimens of the ulcer and splenic tissue were obtained, which on immunohistochemistry showed large tumor cells with vesicular nuclei and prominent nucleoli; the cells stained positive for CD20, suggesting diffuse large B-cell lymphoma. Bone-marrow biopsy or flow

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Figure 1: A fistulous tract between the stomach and the hilum of spleen was shown, with the presence of air in the spleen and a large splenic abscess

cytometry did not reveal any other evidence of disease.

Laparotomy was performed after the patient's albumin level improved to 38 g/L and correction of his anemia. Under general anesthesia, the abdomen was opened *via* a 18 cm-long midline incision. On exploration, the spleen was found to be densely adherent to the diaphragm, lateral abdominal wall, and greater curvature of the stomach by an abscess. The adhesions were separated with digital palpation. Following the dissection of the spleen, an inflamed and necrotic 7 cm of splenic segment and approximately 2 cm of fully perforated fundus were found leading to the GSF. The patient underwent splenectomy, gastric wedge resection of the greater curvature, and distal pancreatectomy. The GSF was confirmed in the gross specimen. The pathological report of spleen surgically removed was large B-cell lymphoma, consistent with endoscopic biopsy. Postoperatively, the patient received intensive care for 3 days and was able to resume a full diet on day 4. Recovery was uneventful. Adjuvant chemoradiotherapy was started on the 16th postoperative, day and the patient remained well at follow-up.

DISCUSSION

GSF is most commonly associated with some gastric or splenic diseases, especially lymphoma of the gastric or splenic origin.^[1-20] Other etiologies include benign gastric ulcers,^[21-24] colorectal adenocarcinoma,^[25] gastric adenocarcinoma,^[26] splenic abscess,^[27-29] and Crohn's disease.^[30] Of the 20 case reports of GSF associated with lymphoma, 17 were splenic lymphoma and 3 were gastric lymphoma.^[11,13,16] Sixteen involved non-Hodgkin's lymphoma, and diffuse large B-cell lymphoma was the histologic subtype definitively identified in all but four of these case reports.^[4,8,10,18] It has been suggested that the tendency for this type of lymphoma to invade the gastric wall and cause widespread necrosis was required for the formation of fistula.^[9,18] GSF is more likely to occur spontaneously in patients with lymphoma (seen in

13 of the 20 case reports); however, there are seven reports of fistulas that developed following recent treatment with chemotherapy,^[2,5,6,10,12,15,18] suggesting that rapid tumor cell death in the context of gastric wall infiltration can contribute to the evolution of this complication.^[6,7] It is worth noting that an existed thin fistula between the stomach and spleen would become even worse under the conditions of chemotherapy.^[7]

The classic presentation, as first described by Harris *et al.*^[9] in 1984, consisted of left upper quadrant pain, fever, and weight loss. Their article compared 10 patients with large cell lymphoma. Our case was typical of diffuse large cell lymphoma. Of the 20 case reports of GCF associated with lymphoma reviewed by us, the most common presenting signs and symptoms of GSF were left upper quadrant or epigastric pain (12 in 20) and weight loss (10 in 20). Other symptoms were fever, fatigue, dyspepsia, etc.. The most consistent physical exam finding is splenomegaly, present in 85% of the 20 case reports (17 in 20). There are three reports of patients with lymphoma presenting with massive hematemesis as a result of GSF formation with subsequent erosion of gastric contents into the splenic vessels.^[3,10,19] However, the incidence rate of upper gastrointestinal bleeding in patients with GSF resulting from lymphoma was lower than from benign peptic ulcers.^[18]

Abdominal CT is superior to other radiological tests in the diagnosis of GSF.^[13] Air-fluid levels or the presence of air in the spleen should alert us to GSF. The fistulous tract can indicate the proper diagnosis because the presence of air does not exclusively indicate an abscess. CT visualization of the fistulous tract or retrograde filling of the splenic cavity by orally administered gastrointestinal contrast medium can indicate the correct diagnosis. Upper gastrointestinal endoscopy can confirm the GSF by direct visualization of the fistulous opening. CT scans provided the definitive diagnosis of GSF in 13 of the 20 patients with lymphoma,^[15-18,20] while endoscopy and upper GI only provided definitive diagnoses of GSF in three and two cases, respectively.^[5,8,10,17,20] Some experts believed endoscopy was not necessary when correct diagnosis was provided by CT.^[20] In our case, endoscopy was employed to obtain biopsy specimens for pathological diagnosis preoperatively.

Managements of GSF associated with lymphoma is worth considering. Most reported patients with GSF underwent splenectomy with partial gastrectomy, and sometimes with distal pancreatectomy. Despite the fact that many reports urged operation including splenectomy must be done as early as possible once GSF occurred,^[2] other therapies such as preoperative splenic artery embolization,^[3] endoscopic treatment with continued chemotherapy,^[15] laparoscopic partial gastrectomy, and drainage of the splenic fluid collection without splenectomy^[8] were also carried out effectively in

selected patients. However, for patients with refractory splenic malignancy, en-bloc resection of the spleen and the infiltrated portions of stomach contributing to the fistulous tract remains the safest and most effective treatment option.

In conclusion, GSF may occur secondary to splenic large cell lymphoma, not only after chemotherapy but also spontaneously. CT scans and endoscopy contribute to the correct diagnosis of GSF. Operative treatments should be of greater concern in most patients with GSF caused by lymphoma.

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