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Spontaneous Gastrosplenic Fistula Resulting From Primary Gastric Lymphoma: Case Report and Review of the Literature

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ABSTRACT

A fistulous tract between the stomach and the spleen is a rare manifestation. Spontaneous gastrosplenic fistula formation resulting from primary gastric lymphoma is extremely rare and should be managed as an emergency. To date, four gastrosplenic fistulas originating from gastric lymphoma have been reported, of which three were spontaneous and one occurred following chemotherapy. We report a case of spontaneous gastrosplenic fistula in a 35 years-old-man with gastric malignant B-cell non-Hodgkin's lymphoma who was diagnosed by computed tomography and endoscopically, followed by successful treatment with total gastrectomy and splenectomy.

Key Words: Gastrosplenic fistula, Gastric lymphoma, Surgical Treatment

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Introduction

Direct communication between two abdominal visceras usually results from a congenital, traumatic, inflammatory, neoplastic or an iatrogenic process affecting one or both organs. Gastrosplenic fistula formation resulting from primary gastric lymphoma is extremely rare and should be managed as an emergency (1, 2). Eighteen previously reported cases of gastrosplenic fistulas secondary to malignancy are cited in the literature; four of these developed from a gastric lymphoma (1-16). We report a case of spontaneous gastrosplenic fistula in a patient with gastric malignant B-cell non-Hodgkin's lymphoma successfully treated by total gastrectomy and splenectomy. This article is the fourth case of gastric lymphoma leading to spontaneous gastrosplenic fistula reported in the literature.

Case Report

A 35 year-old-man presented to our hospital with complaints of progressively increasing left upper quadrant pain radiating to the back, fatigue for 1 year which increased in the last 8 months and a weight loss of 4 kg in the previous 15 days. His pain was postprandial and increased with flatus and was relieved by using proton pump inhibitors. He did not complain of other symptoms (e.g., fever and sweating). He was a heavy smoker (2 packages/day) for 10 years, but his history was otherwise unremarkable.

Physical examination revealed neither peripheral lymphadenopathy nor palpable mass lesion in the abdomen. Leukocyte count was 9.200/mm^3, liver function tests, renal function tests, hemostasis parameters and tumor markers (carcinoembryonic antigen, α-fetoprotein, CA-19-9) were totally normal. Abdominal ultrasonography revealed a 54x53 mm, lobulated, air-containing hypoechoic cystic lesion between the gastric fundus and splenic hilum, indicative of splenic abscess. Abdominal computed tomography (CT) showed irregular thickness of the gastric corpus wall and extravasation of the contrast agent through the splenic hilus (Figure 1). Upper gastrointestinal endoscopy revealed a 3 cm dimensioned necrotic deep ulcer at the gastric fundus and a fistula was

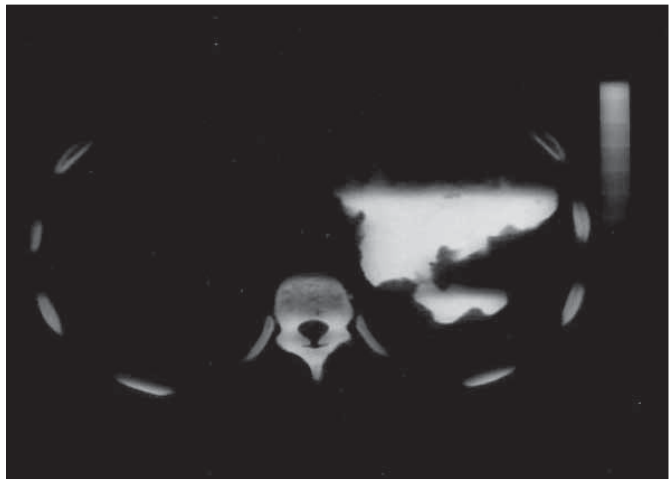

Figure 1. Abdominal axial contrast-enhanced computed tomography shows extravasation of the contrast agent through the splenic hilus

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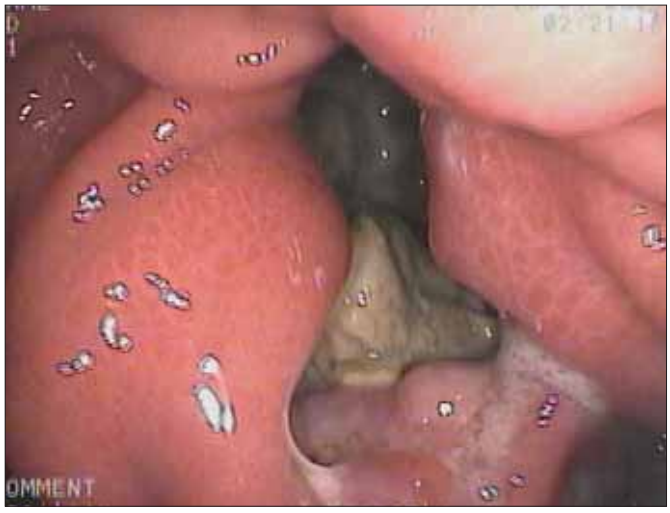

Figure 2. Necrotic deep ulcer in the gastric fundus

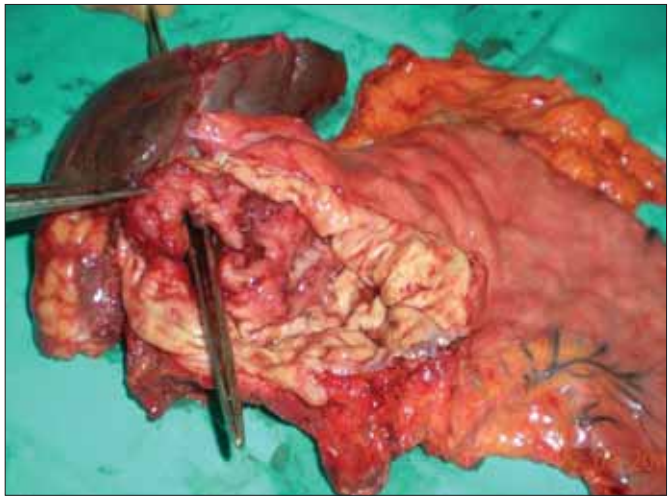

Figure 3. Operation specimen shows fistula orifice on gastric fundus

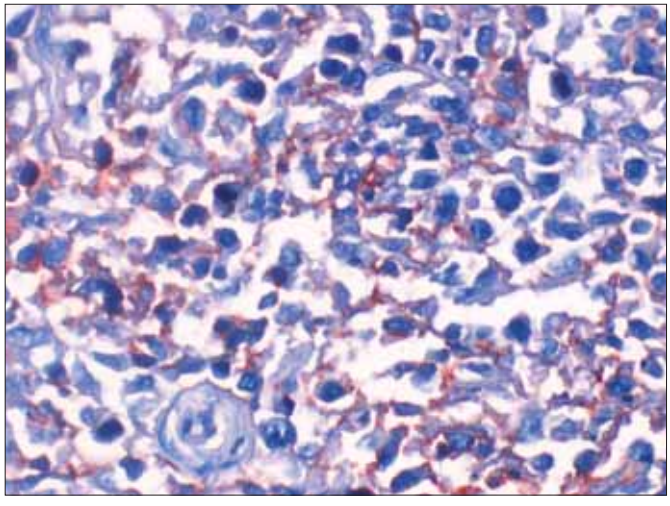

Figure 4. Diffuse atypical lymphatic cell infiltration below the mucosa (staining positively for CD20)

seen spreading towards the splenic hilum (Figure 2). Multiple biopsies were taken from this apparently malignant ulcer. Histological evaluation revealed a non-Hodgkin's lymphoma staining positively with CD20 and leucocyte common antigen and negatively for cytokeratin.

During laparotomy, a gastric tumor with perforation was detected at the gastric fundus. The posterior surface of the fundus was densely adherent to the spleen. Spreading between the spleen, great curvature of stomach and pancreas, an abcess with dimensions of 15x10 cm was found. Following the dissection, a gastrosplenic fistula was found leading to an inflamed and necrotic 8 cm of splenic segment and approximately 5 cm of perforated fundus. After drainage of the abcess, total gastrectomy, splenectomy, and roux-n-Y esophagojejunostomy were performed.

Macroscopically, an ulcerative and infiltrative gastric tumor measuring 6x5x2 cm which infiltrated the spleen in a 9x5.5 cm area was observed (Figure 3). Pathological evaluation revealed infiltration of a malignant B-cell non-Hodgkin's lymphoma, positive for leucocyte common antigen (LCA) and CD20, and negative for cytokeratin (CK), epithelial membrane antigen (EMA) and CD3 (Figure 4). The tumor was seen in the serosa of the stomach, whereas no tumor was seen in the surgical border.

The patient was discharged on the 8th day postoperatively without any complications and received chemotherapy with cyclophosphamide, hydroxydaunomycin, oncovin and prednisone (CHOP regimen).

Discussion

A fistulous tract between the stomach and the spleen is a very rare manifestation (13). Gastric cancer, colorectal adenocarcinoma, Crohn's disease, benign gastric ulcer and splenic or gastric lymphoma are commonly known causes (17). There have been 18 reported cases of gastrosplenic fistula related to malignancy (Table 1) (1-16). To date, four gastrosplenic fistulas originating from gastric lymphoma were reported, three were spontaneous (1, 2, 11) and one occurred following chemotherapy (7). This is the fourth case of spontaneous fistulas originating from gastric lymphoma reported in the literature.

Malignant non-Hodgkin's lymphomas arising from the stomach are generally of the histiocytic or diffuse large cell type (56%), which are more aggressive and require chemoradiation This may hasten the process of tumor lysis and result in a track or 'fistula' between organs because of loss of tumor tissue or acute tumor lysis syndrome following therapy (3, 10, 18, 19). The tumor cells are reduced faster than the gastric stroma cells developing for repair purposes. This formation triggers a deficiency within the stomach wall (10). Spontaneous tumor necrosis is not common and, if it occurs, it is usually in the terminal stage of the disease or in individuals with disseminated disease who are not evaluated further (6) In this case the fistulation is due to necrosis of the large tumor without any treatment.

In recent years, CT has been the imaging technique by which most fistulas are diagnosed (4, 6, 9, 10). On noncontrasted CT, the presence of an air/fluid level within the spleen

Table 1. Previously reported cases of gastrosplenic fistulas resulting from malignancies

Author	Publication Year	Mechanism	Pathology
De Scoville	1967	n.a	Splenic Lymphosarcoma (two cases)
Bubenik	1983	After chemotherapy	Histiocytic (Large Cell) Lymphoma
Harris	1984	Spontaneous	Histiocytic (Large Cell) Lymphoma
Krause	1990	After chemotherapy	Gastric Adenocarcinoma
Hiltunen	1992	After chemotherapy	Gastric Large Cell Lymphoma
Delgado	1994	Spontaneous	Primary Splenic Lymphoma
Blanchi	1995	Spontaneous	B-cell Centroblastic Lymphoma (two cases)
Carolin	1997	After chemotherapy	Histiocytic (Large Cell) Lymphoma
Yabuki	2000	Spontaneous	Gastric Lymphoma
Bird	2002	Spontaneous	Primary Splenic Lymphoma
Choi	2002	Spontaneous	Large Cell Lymphoma
Pizzirusso tases	2004	After chemotherapy	Colorectal Adenocarcinoma Splenic Metas-
Kerem	2005	Spontaneous	Gastric Lymphoma
Puppala	2005	Spontaneous	Gastric Lymphoma
Al-Ashgar	2007	Spontaneous	Splenic Hodgkin's Lymphoma
Arıbas	2008	After chemotherapy	Splenic Large Cell Lymphoma

n.a: Data not available

should raise suspicion of a fistula (6). Gastroscopy was used in two cases in the literature; endoscopy may reveal the gastric lesion as an ulcerated cavity on the greater curvature, a direct communication to spleen or gastric folds converging on the greater curvature with a bright red central oozing as seen in our patient (2, 9).

Although chemotherapy is the mainstay of management of the underlying lymphoma, most centres employ a multimodal treatment programme for patients with gastric lymphoma. Among the gastrointestinal lymphomas, postoperative complication rates are very high, especially in emergency cases (5% to 27%) (11, 18, 19), but surgery must be the preferred treatment for the management of gastrosplenic fistulas because of the possibility of erosion into the splenic artery leading to catastrophic bleeding, which is the most threatening complication (4, 7, 12). Surgical treatment has also often been advocated for establishing an accurate pathological staging and reliable histological examination (18, 19). Some fistulas were successfully treated conservatively (9, 10), but the majority were treated by splenectomy and en bloc resection of part of the stomach. In our case, total gastrectomy, splenectomy, and esophagojejunostomy were performed without any postoperative complications. Adjuvant chemoradiotherapy was started on the 20th postoperative day.

In conclusion, despite its rarity, gastrosplenic fistulas may occur spontaneously secondary to a gastric lymphoma. Awareness of this condition might lead the clinician to early recognition by CT scan and upper gastrointestinal endoscopy. Surgery must be the preferred method of managing gastrosplenic fistula in order to avoid further catastrophic bleeding from the spleen.

Conflict of Interest
No conflict of interest was declared by the authors.

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