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CASE REPORT

# Spontaneous gastrosplenic fistula in primary gastric lymphoma: case report and review of literature

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## Case report

A 66-year-old woman presented to the accident and emergency department with complaints of left upper quadrant pain that radiated to the chest. She had been generally unwell over the previous 2 weeks and gave a history of a similar episode a year earlier. She was a non-insulin-dependent diabetic and had hypertension, asthma, ischaemic heart disease, rheumatoid arthritis, psoriasis, gout and a previous deep vein thrombosis. She was apyrexial, with a normal pulse and blood pressure. On examination there was left upper quadrant tenderness with normal bowel sounds. Investigations at the time of admission were as follows: CRP 246 mg/l, WBC count $13.2\times10^9$, Hb 8.7 mg/l and prothrombin time 20.2 s. Chest and abdominal radiographs showed no abnormality. US examination was inconclusive because of the woman's large body habitus and therefore CT was performed using oral contrast agent owing to difficulty in cannulating any accessible vein. CT revealed a direct communication between the gastric lumen and the spleen through a fistulous tract.

Following CT the woman underwent an upper gastrointestinal endoscopy, which showed a large infiltrating tumour with central ulceration replacing most of the fundus of the stomach; this lesion was biopsied. An associated acute gastric ulcer of 0.25 cm was noted on the lesser curvature of the stomach. Following the endoscopy, CT was repeated using both oral and intravenous contrast medium to stage the lesion, as shown in Fig. 1. This demonstrated the fistula, thickening of the gastric wall around the fundus and gastro-oesophageal junction, and enlarged coeliac axis nodes. There were low attenuation areas in the spleen but no significant enlargement, as shown in Fig. 2. The findings suggested a gastric tumour with invasion and fistulation into the spleen, splenic metastasis and local lymphadenopathy. Histologically the gastric lesion represented a diffuse large-B-cell non-Hodgkin's lymphoma. As the woman had significant comorbidities and was a high risk for general anaesthesia, no surgery was planned and medical management was continued in consultation with the haematologists. The patient died after 2 months of chemotherapy.

## Discussion

The first gastrosplenic fistula was reported from Belgium in 1962. The authors described a case of gastrosplenic fistula with a characteristic radiographic appearance due to the presence of air in the spleen, which they termed "aerosplenomegaly".[12] There have been 12 reported cases of gastrosplenic fistula in the literature, of which 7 were neoplastic,[2-5,9,12,14] 4 resulted from perforation of benign gastric ulcers[13] and 1 from gastric Crohn's disease.[1] Among the neoplastic causes of gastrosplenic fistula, lymphoma was the underlying malignancy in 5 cases, of which 4 primaries were in the spleen and 1 in the stomach.[3] Lymphosarcoma and adenocarcinoma of the stomach were found in the remaining 2 cases.[2-5,9,12] Fistulation followed chemotherapy in 4 cases[2,3,5] and was spontaneous in 3 cases, but in the latter the spleen was the primary site of origin of the tumour.[4,9] Ours is probably the first reported case of spontaneous fistulation from a primary gastric lymphoma.

### Pathogenesis

Splenic penetration by benign gastric ulcers is

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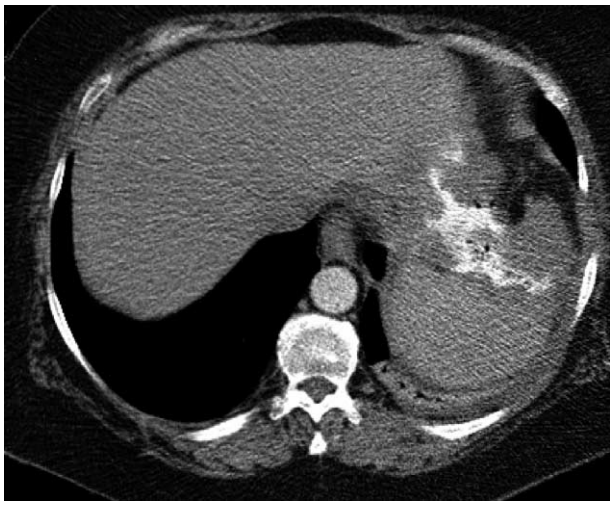

**Figure 1** Axial CT with oral and intravenous contrast showing the oral contrast in the gastrosplenic fistula flowing into the spleen.

divided into early penetration characterized by inflammatory reaction around a confined perforation which invades the spleen, and advanced penetration in which the ulcer crater extends into the splenic parenchyma. This latter type is termed a gastrosplenic fistula.[7] No such categories have been suggested in the literature for penetration by malignant lesions.

Gastrosplenic fistulas can arise from various underlying malignancies, such as adenocarcinoma, lymphoma, lymphosarcoma, leiomyoma and leiomyosarcoma. However, fistulation in general is more common in lymphoma than in adenocarcinoma because of the absence of a desmoplastic reaction.[2,11] In the abdomen, lymphomas are of the histiocytic or large-cell diffuse type and are seen in the stomach more commonly (56%) than in the spleen. In general such lymphomas are more aggressive and require chemoradiation[8-10] which may hasten the process of tumour lysis and contribute to the formation of the fistulous tract because of loss of tumour tissue or acute tumour lysis syndrome following therapy.[2,3]

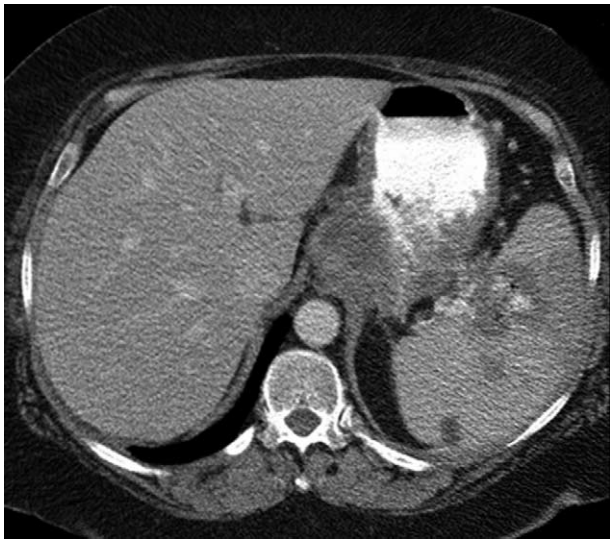

**Figure 2** Axial CT showing low density lesions in the spleen which are possible metastasis.

Of the splenic lymphomas, those associated with splenomegaly and central necrosis have a higher tendency to adhere to and fistulate into other organs. A study of gastrointestinal tract lymphomas by Rosenfelt et al. revealed treatment-related bleeding and perforation as a complication in 25% of cases that failed to enter complete remission.[10] Spontaneous tumour necrosis is not common, and if it does occur it is usually in the terminal stage of the disease or in individuals with disseminated disease who are not evaluated further.[5] An autopsy study of 323 persons with untreated gastrointestinal lymphomas revealed gastric lesions in 128. In only 2 cases was fistulation noted, emphasizing the rarity of the condition even at postmortem.[11]

## Presentation

Left upper quadrant pain is the most commonly described presentation of gastrosplenic fistulas, as in our case.[2,4,5,9] A review of 10 cases of large-cell lymphoma of the spleen by Harris et al. in 1984 showed 9 of the 10 patients to be over 50 years of age and presenting with left upper quadrant pain at the time of diagnosis. In this series 1 case was found to have a gastric fistula.[9] By means of erosion of the splenic artery, these gastrosplenic fistulas can present as upper gastrointestinal bleeding.[14]

## Diagnosis

In recent years CT has been the imaging technique by which most fistulas are diagnosed.[2-7] The classical finding is the presence of oral contrast medium in the spleen, as shown in Fig. 1. On non-contrasted CT, the presence of an air/fluid level within the spleen should raise suspicion of a fistula. The site of the air/fluid level may correspond to the site of primary splenic lesion.[5] Homogeneous hypodense lesions noted in the spleen could represent areas of splenic infarction due to involvement of the splenic artery by the gastric tumour. They can also be due to haematoma, liquefaction of splenic tissue after penetration or metastasis to the spleen.[5] Specific signs of penetration of the spleen are the loss of fascial planes and soft-tissue

densities extending between the two organs. In a report by Glick et al. on preoperative recognition by CT of splenic penetration by benign gastric ulcer,[7] a low-density linear area within these soft-tissue densities was presumed to represent the fistulous tract.

The role of upper gastrointestinal barium studies in these patients is limited, because lesions penetrating into surrounding structures may give a misleading radiographic image due to the fibrous reaction stimulated by penetration.[13]

Endoscopy may reveal the gastric lesion as an ulcerated cavity on the greater curvature, a direct communication or gastric folds converging on the greater curvature with bright red central oozing.[4] In 1 case based on endoscopic findings of a fistula, an endoscope was passed into the splenic pulp and the lining of the fistula was noted to consist of necrotic material.[2] Caution has to be exercised in obtaining a biopsy of a mass in the walls of such lesions, as the mass could represent a splenic haematoma. Therefore CT before the endoscopy and biopsy may be of value to the endoscopist.

### Management

Surgery must be the method of management of gastrosplenic fistula because of the possibility of erosion into the splenic artery leading to catastrophic bleeding. Chemotherapy is the mainstay of management of the underlying lymphoma. Delayed surgical intervention was, however, successful in 1 case where the patient was followed up by outpatient endoscopy and later underwent operation after the closure of the fistula.[3] Patients presenting with bleeding may undergo splenic artery embolization followed by splenectomy and gastric resection.[14]

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