# Gastrosplenic Fistula Due to Splenic Lymphoma: Two Case Reports and Literature Review

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# Abstract

### Purpose

Gastrosplenic fistula (GSF) is a rare and potentially fatal complication of various diseases, of which lymphoma is the most common cause. We aim through our work to relate two cases of GSF and to review literature.

### Methods

We reviewed two cases treated in our department of GSF and made a research in Pubmed using the keywords “Gastrosplenic fistula” and “Splenic lymphoma”.

### Results

GSF is a rare condition that can occur spontaneously or after initiation of chemotherapy. It arises from the rapid growth of tumour and invasion of surrounding organs. Diagnosis may be difficult to make and confused with splenic abscess. Treatment modalities include surgical resection, chemotherapy or a combination of both. Here we report two cases of GSF due to diffuse large B cell lymphoma patients. The first case is of a 54-year-old woman with a spontaneous fistula in the stomach. The second one is of a 48 year-old- male patient presenting a fistula after chemotherapy. Both patients died after surgery.

### Conclusion

GSF is a rare but dangerous condition in which surgery is currently the preferred treatment.


# Introduction

Gastrosplenic fistula (GSF) is a rare manifestation of stomach or spleen lesions [1, 2]. In most cases, this complication is due to diffuse large B cell lymphoma (DLBC) [3] of the spleen or the stomach. This condition occurs because of the aggressive nature of this tumor. The tumor invades the gastric or spleen wall leading to the formation of a fistula. Hodgkin’s lymphoma represents the second most frequent GSF etiology followed by histiocytic lymphoma. It can happen spontaneously or after chemotherapy. The management of these lesions is still challenging to this day. Here, we report two cases of GSF complicating a splenic DLBC lymphoma. The first case is of a spontaneous fistula. The second, is of a GSF occurring after the start of chemotherapy.

# Case Report

On physical examination, her general condition was preserved, a painful 13 cm splenomegaly was noticed without hepatomegaly. There were no palpable lymph nodes and no clinical sign of haemorrhage. Laboratory studies revealed a hypochromic microcytic anaemia (haemoglobin: 8.3 g/dl), leucocytes and platelets were normal. A thoracoabdominal computed tomography (CT) revealed a splenic mass

measuring 13.5 cm. This mass was adherent to the stomach. There was an intra-abdominal and retroperitoneal lymphadenopathy. Trans-parietal biopsy was denied because of the haemorrhagic risk and a surgical biopsy was preferred instead. Intraoperative findings revealed a locally advanced large splenic mass, invading the stomach and adhering to the left lobe of the liver. This mass also invaded the left diaphragm and compressed the pancreas.

Histopathology concluded to a diffuse large B cell lymphoma positive for CD20, Bcl6, Bcl2, Ki67% (90%). Further investigations included a PET- scan that concluded to an active lymphatic spleen lesion with lymphadenopathy above and under the diaphragm. The tumour was classified stage III according to Ann-Arbor classification system and the patient was to start chemotherapy within few days. Two days later, she presented at emergency room for a severe abdominal pain. A CT scan revealed a rapid progression with a 21cm necrotic mass of the higher spleen pole, “aerosplenoemgaly “due to multiple gastrosplenic fistula with the gastric corpus **(Fig. 1).** The largest fistula measured 1cm and connected the mass with the left wall of the stomach. A gastrectomy with splenectomy wad planned. The patient died from a septic shock.

Biopsies of the axillary lymphadenopathy and of the pulmonary mass were performed and concluded to a DLBC lymphoma. Both of them stained positive for CD20, bcl6, bcl2 and Cmyc and Ki67 was 80%. The tumour was classified stage IV of the Ann-Arbor classification. Chemotherapy with Rituximab, Adriablastine, cyclophosphamide, vincristine and prednisone (RCHOP) was initiated. After the second cycle, he presented at our emergency for worsening of his dyspnoea with an abdominal pain. CT scan revealed a large 16.8 x 15.9 cm necrotic mass in the spleen. It was communicating with the stomach via a gastrosplenic fistula (GSF) with a presence of a posterior pleuro-splenic fistula (Fig. 2). This mass infiltrated the greater curvature of the stomach, the body and the tail of the pancreas. The CT scan also showed multiple suspicious kidney nodules. The patient underwent surgery but died a week after.

# Discussion

Gastrosplenic fistula is a very rare and potentially fatal condition mainly caused by diffuse large B cell lymphomas (DLBCL), in particular, and lymphomas, in general. Its development is secondary to the aggressive nature of this tumour leading to adjacent organs invasion [4]. GSF occurs spontaneously or after chemotherapy as a result of a rapid regression of the infiltrating tumour [5]. A systemic review of lymphoma related gastric fistulas reviewed 27 cases of spontaneous and chemotherapy induced GSF. Men were more often affected than women (22 cases) [6]. Seventeen cases have been reported to be spontaneous. Most of them presented with abdominal pain, splenomegaly, vomiting and fatigue. Some others presented with hematemesis or melena and epigastric pain [7]. These clinical characteristics were present in our cases. Both of our patients presented initially with common abdominal symptoms. But both of our patients progressed rapidly despite benign early symptoms, which emphasises the importance of early diagnosis and treatment.

In fact, evoking the diagnosis can be difficult as the symptoms are general and nonspecific, CT scan is the key in identifying gastrosplenic fistulas [8]. Indeed, if GSF is not detected early it can progress and involve the diaphragm. In our review of the literature we found only a few cases reported with extensive lymphoma masses in the left upper quadrant, including diaphragm involvement similar to our case [9].

GSF diagnosis can be confused with a splenic abscess due to the presence of air into the mass. The CT identification of the fistulous tract is the key to a right diagnosis. Retrograde filling of the splenic cavity by orally administered gastrointestinal contrast medium with CT imaging, endoscopy of the upper gastrointestinal tract or by PET SCAN could also be helpful in doubtful cases [10]. In a PET scan, the GSF can be visualized as the most hypermetabolic region in an increased activity spleen.

The primary lesion, either splenic or gastric, may be difficult to determine at first sight but changes in CT scan over time may clear up the situation. Imagery typically shows a necrotic mass filled with air, seen in the CT scan as air bubbles. A fistulous tract from the spleen to the gastric lumen can also be visible.

A commonly accepted condition required for GSF to happen is the rapid necrosis of infiltrated lymphoma cells in the spleen capsule. In 1984, a clinical and pathological characteristics study of ten cases of spleen lymphomas [11], identified the distinctive DLBCL characteristic of presenting with large, destructive mass with extensive central necrosis and easy capsular penetration. Invasion of the splenic capsule, of the gastric wall and infiltration of the adjacent tissue is also required. Most of described GSF cases are of small fistulas ranging from 3.5 to 8 cm. If a large part of the gastric wall is infiltrated then a large fistula will arise like in our cases. In our review of literature, we could find only a couple of cases with 15 and 18 cm [12].

The management of this lesion is challenging. Historically only surgery was available as a treatment for gastrosplenic fistulas [13]. In our review of literature, twenty four of the cases reported received surgical treatment. Only six of these, underwent chemotherapy as well. The most common surgical treatment is partial gastrectomy with or without splenectomy. But the resection method can be adjusted to the tumour size and its invasion to adjacent organs. For large tumours, a near total gastrectomy and splenectomy may be needed. Resection of a part of the pancreas was performed in ten of the cases we reviewed. And for some patients, complications such as diaphragm perforation had to be repaired per operation.

However, in some cases a resolution of the GSF has been described after chemotherapy. Ineligible patients for surgery have a poor outcome. In fact, only two cases were reported of patients receiving only chemotherapy as treatment with no surgical intervention. One of these cases died two months after initiation of chemotherapy and one achieved remission after two cycles of chemotherapy but no further information about his outcome were included. Five of the 27 patients we reviewed died. One patient died after receiving chemotherapy. One patient received a Gastric wedge resection, fistulectomy and splenectomy and died a month after because of septic choc. Another patient underwent partial gastrectomy and fistulectomy, he died after 5 months after. Another patient underwent splenectomy, partial gastrectomy, diaphragmatic primary repair and died after because of multi organic failure. Another patient underwent Gastric wedge resection and splenectomy and died later due to post op perforation.

Another received Gastric wedge resection, fistulectomy and splenectomy and died after two months due to progression of his lymphomas [14].

One of our patients received two cycles of chemotherapy before undergoing operation. The second case did not receive any chemotherapy regimen

In conclusion, surgery is the most valid option for a gastrosplenic fistula but it may be very difficult to carry out. Only a few cases of GSF are reported to be treated with chemotherapy or chemotherapy with radiotherapy. Further investigations and studies should be done to better understand how to treat GSF without surgery.

# Declarations

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Authors' contributions:
Feryel KSONTINI: Conceptualization, Writing-original draft, Writing-review & editing
Issad Nefzi: Writing-original draft
Salim Khrouf: Writing-review & editing
Sonia Ouali: Data curation, Supervision, Validation
Asma Zidi: Writing-review & editing
Houcine Magherbi: Writing-review & editing
Mouna Ayadi: Writing-review & editing

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# Figures

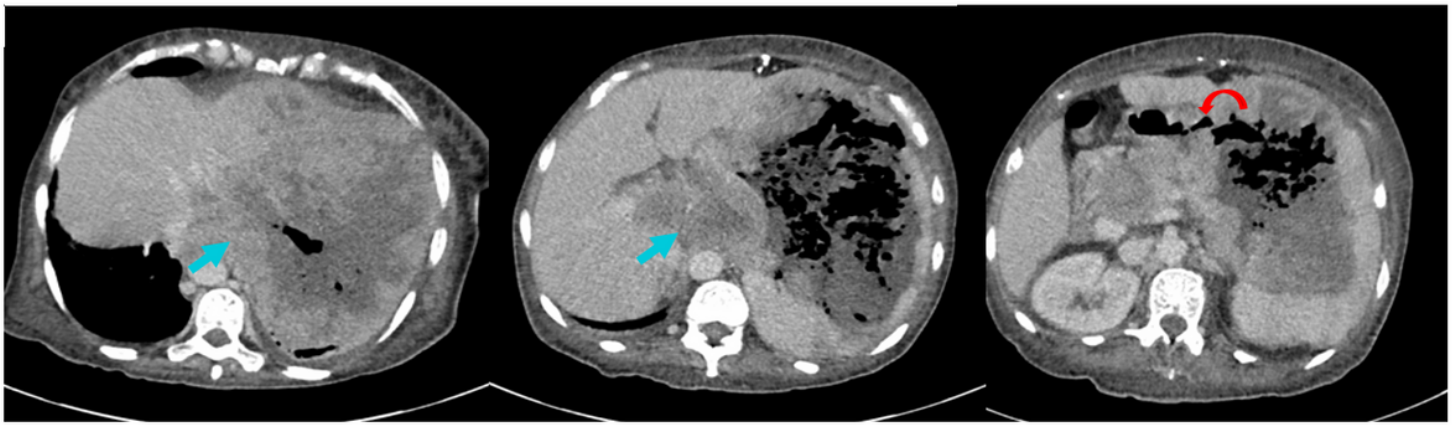

**Figure 1**

Please see the Manuscript file for the complete figure caption.

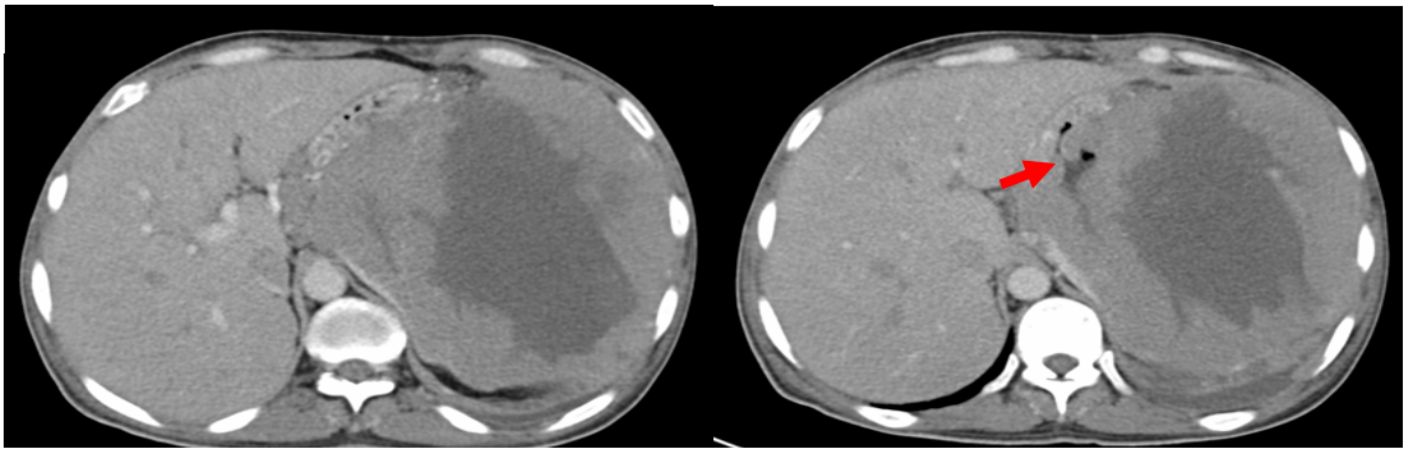

**Figure 2**

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