Gastrohepatopleural Fistula Complicating A Gastric Diverticulum: A Rare Event And A Difficult Diagnosis

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Citation
R Orlando, F Pomerri, F Lirussi. Gastrohepatopleural Fistula Complicating A Gastric Diverticulum: A Rare Event And A Difficult Diagnosis. The Internet Journal of Gastroenterology. 2008 Volume 7 Number 2.

Abstract
A 68-year-old woman developed a gastrohepatopleural fistula with pleural effusion as a result of a gastric diverticulum complicated by a hepatic abscess. The diagnosis required multiple imaging studies, two gastroscopies, and a barium study of the upper gastrointestinal tract. The patient underwent laparotomy with surgical removal of the fistula, which represents a definite intervention if performed early.

INTRODUCTION
Gastropleural fistula is a rare condition characterized by a communication between the stomach and the pleural space [1,2,3,4,5]. It is an uncommon complication of a number of conditions such as major pulmonary and oesophageal resections [4] and gastric lymphoma [2,3]. Also, a gastropleural fistula has been peptic ulceration [1,5,6] and perforation of an oesophageal hiatal hernia [6]. Moreover, it may arise from perforation of a gastric diverticulum through the diaphragm [6]. However, this latter condition is rare, because the diaphragm forms a thick barrier between the stomach and the thoracic cavity. Thus, a fistulous communication between the abdominal and the pleural cavity implicates the erosion of an intra-abdominal abscess, following gastric perforation through the diaphragm with a resulting pleural effusion [1,3]. The diagnosis of gastropleural fistula is usually made by upper endoscopy, radiographic contrast examination, or at surgery (n). We describe a case of a solitary diverticulum of the posterior wall of the gastric antrum complicated by the onset of an abscess of the right hepatic lobe and subsequently by a pleural fistula with pleural effusion.

CASE REPORT
A 68-year-old woman was admitted to hospital in February 2005 complaining of right abdominal pain described as a growing or sickening pain, not relieved by food, but only by non-steroidal antinflammatory drugs (NSAIDs). No other associated symptoms such as sweating and tachycardia were present. The pain was deep, constant, of middle intensity and radiated to the back. Also, it became more noticeable with breathing and forced the patient to lie on her left side. She denied alcohol abuse and smoking. Her past medical history included tuberculous annexitis and latero-cervical lymphadenitis and hypertension, but no previous peptic ulcer disease and/or abdominal surgery. Moreover, there was no family history of gastrointestinal diseases. Her bowel habits were referred as normal.

On admission, the patient was afebrile, alert and oriented. Her vital signs were normal. Her abdomen was soft and non-tender. No jaundice, hepatosplenomegaly or stigmata of chronic liver disease were present and Murphy sign was negative. The 10th and 11th right intercostal spaces on the middle and anterior axillary line were aching on digital pressure. The remainder of the physical examination was normal.

Laboratory tests showed WBC= 7.800/mm$^3$ (n.v. 4.200-12400/mm$^3$), Hb= 10 g/L (n.v. 11.5-14.7), Hct 30 (n.v. 34-45), platelets 132.000/mm$^3$ (n.v. 150000-450000). Routine laboratory tests (protein profile, glucose, urea, creatinine, electrolytes, amylase and urinalysis) were within the normal range. Liver function tests (total bilirubin, conjugated bilirubin, alkaline phosphatase, gamma-glutamyl transferase, albumin, prothrombin time) were also normal. Neoplastic markers were negative; ESR was 54 mm/h (n.v. 2-39) and C-reactive protein was 225 mg/L (n.v. < 6).

ECG, chest and abdominal x-ray were normal; in particular, there was no evidence of rib fractures in the right side of the chest. An abdominal ultrasound was performed, which showed no signs of free fluid or evidence of pathological
conditions in the upper part of the abdomen such as liver disease, cholelithiasis, bile duct obstruction, nephrolithiasis, or Caroli’s disease.

Thoracic computerised tomography (CT) revealed dense pulmonary bands consistent with fibrosis in the right and left apex, middle lobe, and inferior right lobe, which was consistent with the previous tubercular illness.

An upper gastrointestinal tract endoscopy showed mild antral gastritis and a patent pylorus with no evidence of fistulous tract formation. Biopsy of the stomach was negative for Helicobacter pylori. Colonoscopy showed no significant abnormalities.

The vertebral column X-ray revealed dorsal and lumbosacral spondylosis. NSAIDs and proton pump inhibitors (PPIs) were prescribed for a presumptive diagnosis of costochondritis. In March 2005 she underwent a NMR of the chest and upper abdomen showing a negligible fluid collection over the right hemidiaphragm and another small fluid collection between the diaphragm and the liver surface (fig. 1).

**Figure 1**
Figure 1: Sagittal magnetic resonance view of the inferior chest and upper abdomen showing two fluid collections (close arrows).

In November 2005, a second NMR of the chest and upper abdomen revealed an expanded fluid collection above the right hemidiaphragm and the presence of a hyperdense 5-cm large area in the contiguous hepatic parenchima, which was compatible with a peripheral abscess in the right lobe of the liver (fig. 2 and 3).

**Figure 2**
Figure 2: Magnetic resonance of the abdomen showing perihepatic and pleural thin fluid collection (open arrows).

**Figure 3**
Figure 3: Magnetic resonance of the abdomen featuring a round irregularly shaped peripheral abscess in the right liver lobe (close arrow).

The outcome of the radiological procedures did not allow any definite diagnosis while the patient started complaining of a persistent pain in the right upper abdomen and in the right lower thorax. Thus, she was admitted to hospital again and underwent an abdominal CT scan. A peripheral liver abscess between the 8th and the 5th hepatic segment and gas collection extending from the diaphragm adjacent to the hepatic surface to the gastric antrum were detected (fig. 4).
A barium study of the upper gastrointestinal tract was therefore performed, demonstrating a 4-mm large fistula extending from the posterior wall of the gastric antrum to the right hepatic lobe. It was 3 cm in length and penetrated in the liver region forming a saccular structure of 3.5 x 1.5 cm (fig. 5).

A second upper gastrointestinal endoscopy showed the presence of a small diverticulum with a circular opening located in the posterior wall of the gastric antrum. The pyloric orifice was narrowed, and did not allow the passage of the endoscope (Olympus GIF 2T10). The mucosa surrounding the orifice appeared normal. The patient, therefore, underwent laparotomy, which showed extensive and dense adhesions between the bowel loops, the gastric wall, and the abdominal wall. Following adhesion lysis, a fistula between the gastric antrum, the liver, the right subphrenic region and the right pleural cavity was found and was removed. Therefore, the final diagnosis was gastrohepatopleural fistula with liver abscess. After surgical repair, the patient experienced an uncomplicated post-operative course and was discharged on day eight after the operation. Over the two years of follow-up, she remained virtually asymptomatic and continued her activities of daily living.

DISCUSSION
The case presented here appears of clinical relevance for at least two reasons: first, the gastrohepatopleural fistula associated to the hepatic abscess, and second, the relatively prolonged interval between the onset of symptoms and the final diagnosis. The gastropheuple fistula is an uncommon
condition and is facilitated by the corrosive action of the gastric juices \((\gamma, \delta)\). The diagnosis of gastropleural fistula is usually made with contrast radiology, upper GI endoscopy, or at operation. The prognosis of a fistula extending from the upper GI tract to the pleura seems to depend upon the delay from diagnosis to surgical intervention due to the high risk of diverticulitis. Thus, it is important to take into account this condition early in the patient's management \((\gamma, \delta)\).

Gastropleural fistula should be considered in the differential diagnosis of thoracic empyema, especially when there is a longstanding history of peptic ulceration \((\gamma)\). Bini et al \((\gamma)\) described a case of spontaneous biliopeumothorax following gastropleural fistula due to gastric perforation by means of a nasogastric tube in a patient undergoing Billroth II gastric resection for adenocarcinoma. Small diameter fistulas may be managed with PPIs therapy \((\iota)\). Surgery is warranted in cases presenting with long-lasting symptoms or complications or in the smaller-diameter tracts not resolving with medical therapy and also in large-calibre fistulas, which are less likely to respond to PPIs therapy \((\iota)\).

Gastric fistulas may also be managed by laparoscopy, as this procedure is associated with less morbidity and, should be considered in alternative to laparotomy especially in pediatric patients \((\iota)\). The exact aetiology of the gastrohepatopleural fistula in our case remains unknown. One can speculate that the fistula complicated a gastric diverticulum which, in turn, was secondary to a peptic ulcer formation. In such instance, the gastric wall might have adhered to the liver surface, resulting in gastrohepatic fistula, liver peripheral abscess, and abscess rupture into the pleural space. This sequence of events, however, does not seem to be the case in our patient, since she never complained of the characteristic symptoms of peptic ulcer disease \((\iota)\).

Alternatively, gastric diverticulum formation may have been the consequence of an increased intragastric pressure, possibly in association with local muscular weakness in the antral wall \((\iota)\). Finally, one could hypothesize that the presence of TB-related intra-abdominal adhesions might have led to the formation of a traction diverticulum.

The prolonged interval between the onset of symptoms and the final diagnosis is due to the observation that gastric diverticula are mostly asymptomatic throughout life. If anything, large diverticula are responsible for non-specific upper gastrointestinal symptoms such as vague upper abdominal or epigastric pain. They usually require surgical treatment \((\iota, \theta)\) because of the risk of complications such as haemorrhage \((\iota, \theta)\), carcinoma, perforation with a resulting subphrenic abscess and/or gastropleural fistula \((\iota, \theta)\), severe gastroesophageal reflux and \((\iota)\), pyloric obstruction \((\iota, \theta)\). In fact, large diverticula are associated to stasis of food residues with bacterial overgrowth causing acute diverticulitis \((\iota)\).

In conclusion, the case reported here emphasizes the difficulties of establishing the diagnosis \((\iota)\), which often requires repeated endoscopic and, above all, radiological procedures. With such a rare condition, it is also difficult to find out an aetiology and the prognosis in such patients relates to early diagnosis and treatment. Thus, the clinicians should consider the presence of a gastropleural fistula in the differential diagnosis of right upper quadrant abdominal pain, especially when symptoms are aspecific and exploration of the gastrointestinal tract by endoscopy is inconclusive.

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