

## Gastropleural Fistula: A Rare Complication of Ewing Sarcoma

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Gastropleural fistula (GPF) is a rare condition that can occur as a consequence of prior pulmonary surgery, trauma, or malignancy. Conservative management usually fails, and gastrectomy and even thoracotomy is often required, especially in debilitated patients. We present a patient with GPF who had a history of Ewing's sarcoma. Diagnosis of GPF was confirmed by upper gastrointestinal system endoscopy and radiographic contrast examination, and the patient underwent a laparoscopic wedge resection of the fistula. To our knowledge, this is the first report of a GPF, in the formation of which recurrence of Ewing's sarcoma had played a role and in the treatment of which wedge resection of the fistula was performed. Laparoscopic treatment of GPF may be associated with less morbidity and should be considered as the initial procedure of choice.

Key words: 1. Fistula  
2. Gastrectomy

### CASE REPORT

A 30-year-old man was admitted to our clinic with complaints of abdominal pain and fever. He had a history of Ewing's sarcoma diagnosed 18 months earlier on his chest wall and he had undergone pneumectomy with negative excision margins, followed by chemotherapy. On physical examination, the patient had decreased breath sounds on his left hemithorax and tympanic percussion. His saturation was 91% despite oxygen treatment given at a 10<sup>-1</sup>/min rate through an oxygen mask. After oral administration of methylene blue, leakage of the substance from the thoracic tube was detected. A contrast-enhanced computed tomography and barium study showed a gastropleural fistula between the stomach and the thorax. Gastroscopy confirmed the diagnosis of a fistula, with a massive amount of pus coming from the thorax. Treatment with ampicillin/sulbactam and fasting relieved his abdominal pain and fever. After oral feeding was started, his symptoms

worsened; therefore, the patient was administered parenteral nutrition. Since the conservative treatment failed, the patient underwent laparoscopic surgery. The gastropleural fistula was in the greater curvature of the stomach in the fundic area. Wedge resection of the fistula was performed by three trocars and a 60 GIA stapler (Ethicon Endo-Surgery, Cincinnati, OH, USA), and the diaphragm was repaired with a prolene graft. The thoracic tube was extracted on the second postoperative day, and the patient was discharged on the fifth postoperative day. Pathologic evaluation of the specimen demonstrated the recurrence of Ewing's sarcoma, which may have played a role in the formation of the gastropleural fistula.

### DISCUSSION

Gastropleural fistula (GPF) is a rare condition that can occur as a consequence of prior pulmonary surgery, trauma, or malignancy [1]. Markowitz and Herter [1] first described gas-

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tropleural fistula in 1960. They described the causes of gastropleural fistula as intrathoracic perforation of the stomach in hiatal hernia, traumatic diaphragmatic hernia with perforation of the stomach, and intraperitoneal gastric perforation with erosion of a subphrenic abscess via the diaphragm [2]. The diaphragm is an effective barrier to the spread of infection. Transdiaphragmatic spread of infection occurs by means of spontaneous diaphragmatic perforation. Fistulation between the stomach and the pleural cavity through the diaphragm is an uncommon diagnosis. It has been previously reported as a complication of pulmonary resection, trauma (especially due to traumatic diaphragmatic hernia), peptic ulcer disease, and malignancy [3]. Some case reports have indicated a predisposition to GPF after the oral intake of steroids or anti-inflammatory drugs, when the cause is gastric perforation [4].

Ewing's sarcoma is a rare cause of chest wall tumors that comprises 5% of all thoracic tumors and 0.04% of all primary tumors. Because of its high grade malignancy, a multidisciplinary approach including wide resection of the tumor, chemotherapy, and also radiotherapy after the resection should be considered [5]. According to a recent review, subphrenic pathologies less frequently lead to formation of a GPF, generally due to diaphragmatic erosion. Supradiaphragmatic conditions, such as infections, intrathoracic operations, and fistulas due to procedures (forceful intercostal tube insertion or nasogastric tube placement after gastric adenocarcinoma resection) can also result in GPF [6].

To our knowledge, this is the first reported case of a GPF caused by Ewing's sarcoma invasion, which presented 1.5 years after the operation. The diagnosis of GPF is usually determined by contrast radiology, upper gastrointestinal endoscopy, and pleural fluid testing [7]. The use of methylene blue as a marker, and the testing of the pleural fluid for pH or bile salts have been suggested as easy bedside tests for GPF

[8]. Conservative management usually fails in GPF, and gastrectomy and even thoracotomy are often required, especially in debilitated patients [3].

In conclusion, although GPFs are relatively uncommon, they require prompt surgical intervention. Laparoscopic treatment of a GPF may be associated with less morbidity, especially in debilitated patients, and should probably be considered as the initial procedure of choice.

## CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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