Gastro-pleural fistula – a rare complication of chronic traumatic diaphragmatic hernia

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Abstract
This report describes the delayed presentation of a gastro-pleural fistula due to a strangulated diaphragmatic hernia after penetrating thoraco-abdominal trauma. It highlights the difficulties faced in the diagnosis of acute diaphragmatic injuries and the potential significant consequences of such missed injuries. The need for a high index of clinical suspicion and appropriate evaluation of thoraco-abdominal penetrating wounds is paramount. The ideal investigatory tool continues to be evaluated, with direct visualization of the diaphragm garnering most support.

INTRODUCTION
Traumatic diaphragmatic herniation is a well-recognized complication of penetrating and blunt thoracoabdominal injury. When the initial diaphragmatic injury goes undetected, there is a delayed clinical presentation with chronic visceral herniation and significant concomitant morbidity.

The key to management is early detection and repair of the diaphragmatic injury. Various strategies, including the use of newer imaging techniques, have been proposed to improve the detection of diaphragmatic injuries in the acute setting. Despite these techniques, missed diagnoses still occur. The following unique case is indicative of the continued problem.

CASE REPORT
A 28-year-old man who had sustained a stab wound to the left lower anterior chest one year previously (managed solely by tube thoracostomy at that time) presented with a short history of severe jaundice and acute respiratory distress. The significant examination findings included a scar at the sixth left intercostal space in the anterior axillary line, a left pleural effusion and mild abdominal tenderness localized to the epigastrium. A left basal thoracostomy tube was sited and drained three liters of dark, bilious fluid containing particulate matter.

A clinical diagnosis of a traumatic diaphragmatic hernia with strangulated bowel and entero-pleural fistula was made. Blood investigations revealed anaemia (haemoglobin 9.3g/dl), leucocytosis (WBC 16.3 x litre), prerenal azotemia (urea 16.1μmol/l; creatinine 118μmol/l) and hypoalbuminemia (albumin 29g/dl). Liver function tests showed a direct hyperbilirubinemia with deranged intrinsic liver enzymes. An arterial blood gas check confirmed marked hypoxia with pO₂ of 60mmHg. Cultures were taken and he was commenced on empirical broad-spectrum antibiotic therapy. Chest radiography prior to thoracostomy tube insertion revealed a left hydropneumothorax (Fig. 1).

After initial drainage, the repeat chest radiograph suggested the tip of the thoracostomy tube to be inferior to the diaphragm (Fig. 2).

Figure 1
Figure 1: Chest radiograph demonstrating a left hydropneumothorax
The patient was taken to the operating theatre where, through a thoraco-abdominal approach, herniation of 90% of the stomach was noted through a 3cm defect in the left hemidiaphragm (Fig. 3). The herniated proximal stomach was gangrenous and perforated with copious purulent exudate present in the left hemithorax. The nasogastric tube was noted to be protruding through the perforation (Fig. 4).

The proximal necrotic segment of stomach was excised and the esophageal and gastric resection margins were closed. After peritoneal lavage, a Stamm’s gastrostomy and Witzel’s feeding jejunostomy were fashioned. The diaphragmatic defect was then repaired primarily with interrupted horizontal mattress polypropylene sutures. The surgical incisions were closed after copious lavage of the left hemithorax and peritoneal cavity. Finally, a cervical esophagostomy was sited for diversion.

Post-operatively, culture-directed antibiotics were commenced and enteral nutrition was established. The patient’s postoperative course was complicated by a subphrenic abscess and left thoracic empyema, both of
which required operative drainage. An esophago-pleural fistula which was subsequently identified radiologically resolved spontaneously. His initial hospital stay totalled 45 days. He was readmitted four months later and a stapled esophago-gastric anastomosis was performed. The cervical esophagostomy was closed later. His subsequent recovery was uneventful and he remains well after two years follow-up.

**DISCUSSION**

Traumatic diaphragmatic hernia was first described in 1541 by Sennererus in a patient with delayed stomach herniation through a left-sided diaphragmatic injury. Despite the knowledge of its occurrence, the true incidence of diaphragmatic herniation remains unknown. This is likely due to subtle clinical presentations.

Traumatic diaphragmatic hernias may result from either penetrating or blunt injuries, the former occurring more frequently (24% vs. 0.16-5%). Unfortunately, diaphragmatic injuries are missed in up to 40% of cases in the acute setting. These patients then present after a variable period of delay with significantly greater morbidity and mortality. A wide range of latency periods has been reported, with the longest reported interval being 50 years post injury. These delayed presentations are often complicated, with significant morbidity arising from respiratory compromise due to tension diaphragmatic herniation, fecopneumothorax from strangulated herniated colon, and gastro-pleural fistula from strangulated stomach. Acutely detected diaphragmatic injuries tend to be less dramatic in their presentation, although acute tension diaphragmatic herniation with fatal cardiac arrest has been reported. Detection of diaphragmatic injuries in the acute setting requires a high index of clinical suspicion.

Direct and thorough visualization of the diaphragm is the only reliable investigative method to detect or exclude diaphragmatic injury and thus prevent the sequelae of diaphragmatic herniation. Clinical and radiologic methods, including chest radiography, computed tomography and magnetic resonance imaging are non-invasive but low in sensitivity.

Routine laparotomy for patients with suspected diaphragmatic injury has resulted in high negative exploration rates ranging from 31% to 66%. As such, the development of minimally invasive procedures, including laparoscopy and thoracoscopy, has provided an accurate and safe means of visualizing and repairing isolated diaphragmatic injuries. Laparoscopy, in the acute setting, also allows for the examination of abdominal viscera in patients who do not have a clear indication for laparotomy. The sensitivity, specificity and negative predictive values have been reported as 100%, 87.5% and 96.8%, respectively, in prospective series. Video-assisted thoracoscopy has also been shown to be an accurate, minimally invasive method of identifying these injuries.

Once identified, prompt surgical intervention for diaphragmatic injuries is warranted. Small defects may be repaired primarily with non-absorbable interrupted, figure-of-eight or horizontal mattress sutures. For larger defects, the use of prosthetic materials such as polypropylene or marlex mesh may be necessary. Laparoscopic repair has been evaluated and found to be comparable to open repair with regards to complications and recurrence. Long-term results, however, are lacking.

Having a high index of suspicion and establishing early diagnosis impacts directly on morbidity and mortality. A few case series have quoted mortality rates of 3% for early presentations and 25% for delayed cases. Morbidity is often related to the development of septic complications (septicemia, wound infection, pneumonia) and respiratory complications (atelectasis, respiratory failure).

This patient presented with a traumatic gastro-pleural fistula. This is an uncommon presentation, with only a few cases reported in the literature. Markowitz and Herter were the first to describe the mechanisms of gastro-pleural fistula formation. With regards to traumatic etiologies, the authors noted that direct injury to stomach and diaphragm would lead to acute presentations and that delayed presentations would occur due to strangulation of stomach in a traumatic diaphragmatic hernia.

Most traumatic gastro-pleural fistulae present acutely after direct visceral injury. This patient had a delayed presentation due to a strangulating diaphragmatic hernia. This is a rare presentation, with only two reports of similar presentations described by Johnston and Twente (1952) and Lindskog and Lawrence (1947) where perforations occurred four days after and “long after” trauma in two cases, respectively. The mechanism of perforation postulated involves the local accumulation of acid in the incarcerated, ischemic portion of stomach with resultant gastric auto-digestion.
Non-traumatic gastro-pleural fistulae have been described involving several other mechanisms: gastric perforation due to strangulated intra-thoracic portion of hiatal herniae; diaphragmatic erosion by intra-abdominal abscesses; after gastric perforation from peptic ulcers, nasogastric tubes, gastric lymphomas and gastric bypass operations; and trans-diaphragmatic gastric penetration after an empyema thoracis.

CONCLUSION
The morbidity and mortality associated with delayed recognition of traumatic diaphragmatic hernias is considerable. This is often related to the development of bowel herniation, incarceration and strangulation. The avoidance of these sequelae hinges on the early identification of the diaphragmatic injury. Heightened suspicion of such injuries should be maintained in patients with penetrating thoracoabdominal wounds. Suspicious clinical and radiographic findings should be further evaluated, preferably with direct visualization. In so doing, early intervention is facilitated.

References
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