CASE REPORT

Transthoracic Littre's hernia presenting with faecopneumothorax following perforation of the Meckel's diverticulum: a late complication of oesophagectomy

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SUMMARY

A Littre's hernia is an unusual phenomenon where a Meckel's diverticulum protrudes through a potential abdominal opening. We wish to present a unique case of a 79-year-old man with respiratory distress following a fall from standing, initially managed as a haemothorax. After a chest drain was placed, bowel contents were drained from the pleural cavity and he was taken to theatre. He had a history of minimally invasive oesophagectomy for cancer and had subsequently developed a diaphragmatic hernia. A blind ending diverticulum with a perforation at its tip was found in the left oblique lung fissure that was subsequently confirmed histologically as a perforated Meckel's diverticulum. The patient had a prolonged stay on the intensive care unit with a left-sided empyema that was managed radiologically prior to discharge. Unfortunately 4 months postoperatively, he passed away from hospital-acquired pneumonia on a rehabilitation ward.

BACKGROUND

A Littre's hernia is the presence of a Meckel's diverticulum (MD) in a hernial sac and they are in themselves very rare. Postoperative diaphragmatic hernias following oesophagectomy are a recognised complication. MD is present in 2%–4% of the population and although it is unusual they can present with spontaneous perforation. This is the first reported case of these combined pathologies occurring in a single patient. Due to the patient's presentation in a critically unwell state, this made the initial diagnosis problematic and the overall outcome poor. The presentation and management of this patient are described to aid those encountering similar cases.

CASE PRESENTATION

A 79-year-old previously independent Caucasian man presented to a major trauma centre 2 days after a mechanical fall in his bathroom with worsening shortness of breath and left-sided pleuritic chest pain; he was asymptomatic before he fell. His medical history included atrial fibrillation (AF), bronchiectasis and a minimally invasive Ivor-Lewis oesophagectomy for curative management of oesophageal cancer 6 years previously. His

regular medications were warfarin, bisoprolol and lansoprazole.

On assessment by the trauma team, he was found to be in respiratory distress and on auscultation had reduced air entry at the left lung base. He was in AF with a fast ventricular response that resulted in decompensation of his systolic blood pressure requiring immediate direct current cardioversion.

INVESTIGATIONS

A mobile chest radiograph was reported showing a large left pleural effusion with surgical sutures over the mediastinum due to previous surgery (see [figure 1](#)). Initial bloods showed a mixed respiratory and metabolic acidosis, raised white cell count ($16.9 \times 10^9/L$), haemoglobin level of 16.8 g/dL, international normalised ratio of 2.9 and a significant acute kidney injury (creatinine 469 $\mu\text{mol/L}$, 1 year before it was 92 $\mu\text{mol/L}$).

The initial diagnosis was a haemothorax and he was managed for multiorgan failure, chest sepsis and haemorrhage with intravenous antibiotics, blood transfusion and immediate reversal of his warfarin. Once stable, he underwent a non-contrast CT scan of the chest, abdomen and pelvis in light of his poor renal function (see [figure 2](#)). The CT showed an effusion in the left hemithorax with associated lung collapse, the previous oesophagectomy gastric pull up was intact and no overlying rib fractures were seen. Small bowel obstruction was seen in the abdomen with a transition point at the left hemidiaphragm. There was no free intraperitoneal fluid or gas or evidence of an MD on the CT.

Following the report, a surgical chest drain was placed into the left side which immediately drained faecal matter raising concerns that a diaphragmatic hernia had perforated, possibly secondary to the insertion of the drain. He was critically unstable at this stage requiring haemofiltration, inotropic and ventilatory support.

DIFFERENTIAL DIAGNOSIS

In view of the mechanical fall and initial chest radiograph with warfarin for anticoagulation, a provisional diagnosis of post-traumatic haemothorax was a potential differential. Due to the raised white cell count and the possibility of a pleural effusion or empyema, chest infection was considered.

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Figure 1 Portable chest radiograph taken on admission showing left-sided pleural effusion.

Following feculent material being drained after the chest tube was inserted, an iatrogenic bowel injury was contemplated. At operation and subsequently following histopathological examination of the resected specimen, the diagnosis was confirmed to be a transthoracic Littre's hernia with Meckel's perforation.

TREATMENT

He was taken to emergency theatres 20 hours following his admission for a diagnostic laparoscopy. At operation, the small bowel was seen herniating through the diaphragm and could not be reduced due to significant adhesions. This necessitated an upper midline laparotomy and left-sided thoracotomy through the eighth intercostal space. A splenectomy was necessary following an ischaemic injury from adhesiolysis. The left thorax was grossly contaminated with bowel content. A necrotic and

perforated segment of small bowel was visualised with a blind-ending diverticulum lying within the oblique fissure of the left lung. The herniated bowel was reduced into the abdomen and a 40 cm segment of non-viable mid-ileum was resected followed by an extensive washout. The oesophageal hiatus was repaired and the distal and proximal ends of the small bowel were brought out as a double-barrelled defunctioning ileostomy. Two chest drains were inserted, one anterior to the left oblique lung fissure and one posteriorly.

OUTCOME AND FOLLOW-UP

He stayed on the intensive care unit for a total of 44 days chiefly for support of his ongoing severe sepsis, renal failure and parenteral nutrition. He developed a left lower lobe empyema that required ultrasound-guided drainage before resolution. Once stable, he was discharged to an acute surgical ward where he was fed via nasogastric tube. He was subsequently transferred to a rehabilitation unit, but unfortunately died as a result of hospital-acquired pneumonia on a background of reduced physiological reserve on postoperative day 111.

Histopathological analysis of the resected bowel showed diffuse dilatation with a well-demarcated strictured area 175 mm from the nearest longitudinal margin. Adjacent to this area was a blind-ending diverticulum 45 mm×45 mm×20 mm and at its tip a 7 mm perforation. Microscopically, the diverticulum was confirmed to be an MD with ischaemic changes consistent with strangulation. The removed spleen demonstrated an ischaemic injury. No evidence of malignancy was seen in either specimen.

DISCUSSION

Littre's hernias are an uncommon occurrence with no citable data on their overall incidence. From reported cases, estimated usual sites are 50% inguinal, 20% femoral, 20% umbilical and other locations in 10%.¹ The more rarely reported Littre's hernias include obturator,² Spigelian³ and through previous ventral incisions.⁴ Littre's hernias may also occur internally; however, it must be noted that this is a separate pathology to an adhesional band from an MD causing internal herniation of bowel.^{5–8} MD has herniated through the diaphragm in previous cases; however, these are of congenital aetiology.^{9–10} There are no reports of Littre's hernia through the diaphragm following oesophagectomy.

MD is present in 2%–4% of the population. Of these individuals, 4%–16% will have complications, with perforation being rarely encountered.¹¹ Only five reported cases of traumatic perforation have been described, of which just one was from a low-velocity injury¹² and all were intra-abdominal. In the case presented here, histological features of strangulation in the MD imply that the patient's fall resulted in displacement of their diaphragmatic hernia causing strangulation and perforation over the 48 hours before presentation.

Postoperative diaphragmatic hernias following oesophagectomy in general have a reported incidence of 0.4%–15%, and they may arise as either an early complication or late complication.¹³ Minimally invasive techniques are more likely to result in diaphragmatic hernia than traditional open methods, and their incidence may be under-reported overall.¹⁴ Although again rare, three cases in the literature demonstrate that this complication can subsequently result in a faecoperneumothorax resulting in critically unwell patient presentations.^{15–17}

Given the rarity of the case presented, the diagnosis was not immediately obvious and had the patient been taken to theatres sooner the complications in the chest may have been less severe.

Figure 2 CT scan at T10. (A) Right lobe of liver, (B) right lung, (C) heart, (D) stomach pull-through, (E) herniated small bowel, (F) T10 vertebral body, (G) aorta and (H) left-sided pleural effusion.

Chest radiographs alone are unreliable at detecting diaphragmatic hernias in the acute setting with a quoted sensitivity of just 33%.¹⁸ CT has proven able to detect faecopneumothorax resulting in prompt treatment and patient survival.¹⁷ In this case, due to instability and renal failure, the non-contrast CT was delayed and did not give a conclusive diagnosis. The insertion of the chest drain identified the urgent need for the patient to undergo surgery. Pleural aspiration is a recognised gold standard in the investigation of pleural effusions; it is less invasive than a chest drain, performed easily at the bedside and relatively safer especially under ultrasound guidance.¹⁹ This could have been a useful diagnostic adjunct that may have resulted in an expedited diagnosis. The complication of a faecopneumothorax following diaphragmatic herniation is catastrophic. Clinicians must, therefore, have a low threshold for investigation and treatment of patients with suspected diaphragmatic hernias following oesophagectomy electively to minimise the chances of future complications.

Learning points

- ▶ In patients presenting with features of haemopneumothorax, it is imperative to consider relevant surgical background and early specialist referral.
- ▶ With a history of oesophagectomy, the possibility of diaphragmatic hernia must be considered to reduce the possibility of faecopneumothorax occurring; clinicians therefore need to have a low threshold for investigating this while patients are well.
- ▶ The investigation of complex pleural effusions with non-invasive chest X-ray and CT is not always definitive and can lead to delay in definitive management.
- ▶ With critically unwell patients where there is diagnostic doubt about a pleural effusion, then an ultrasound-guided pleural aspirate is a useful adjunct.

Contributors CW was responsible for assisting in primary surgery and caring for patient postoperatively; consented patient for case report, amended case presentation, wrote discussion, prepared final figures and undertook recommended revisions of the paper. KE was involved in the caring of patient postoperatively; gathered and summarised information from patient notes and wrote first draft of case presentation. KH performed primary surgery and was responsible for care postoperatively and reviewed final drafts of case report and recommended revisions.

Competing interests None declared.

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REFERENCES

- 1 Skandalakis PN, Zoras O, Skandalakis JE, et al. Littre hernia: surgical anatomy, embryology, and technique of repair. *Am Surg* 2006;72:238–43.
- 2 Hakeem AA, Shaheen F, Shafi H, et al. CT findings in obturator hernia with Meckel's diverticulum – a case report. *J Gastrointest Surg* 2009;13:576–7.
- 3 Dixon E, Heine JA. Incarcerated Meckel's diverticulum in a Spigelian hernia. *Am J Surg* 2000;180:126.
- 4 Citgez B, Yetkin G, Uludag M, et al. Littre's hernia, an incarcerated ventral incisional hernia containing a strangulated meckel diverticulum: report of a case. *Surg Today* 2011;41:576–8.
- 5 Williams JA. Meckel through Winslow? Littre. *Br Med J* 1968;1:811–2.
- 6 Aggarwal BK, Rajan S, Aggarwal A, et al. CT diagnosis of Meckel diverticulum in a paracolic internal hernia. *Abdom Imaging* 2005;30:56–9.
- 7 Wu SY, Ho MH, Hsu SD. Meckel's diverticulum incarcerated in a transmesocolic internal hernia. *World J Gastroenterol* 2014;20:13615–9.
- 8 Gerdes C, Akkermann O, Krüger V, et al. Incarceration of Meckel's diverticulum in a left paraduodenal Treitz' hernia. *World J Clin Cases* 2015;3:732–5.
- 9 Basani L, Aepala R, Reddy BM, et al. Meckel's diverticulum and malrotation in a 3-month-old infant. *Afr J Paediatr Surg* 2016;13:47–9.
- 10 Kang IS, Ahn SM, Han A, et al. Giant Meckel's diverticulum associated with a congenital diaphragmatic hernia. *Yonsei Med J* 2004;45:177–9.
- 11 Sagar J, Kumar V, Shah DK. Meckel's diverticulum: a systematic review. *J R Soc Med* 2006;99:501–5.
- 12 Tummers WS, van der Vorst JR, Swank DJ. Traumatic rupture of a Meckel's diverticulum due to blunt abdominal trauma in a soccer game: a case report. *Int J Surg Case Rep* 2016;19:8–10.
- 13 Argenti F, Luhmann A, Dolan R, et al. Diaphragmatic hernia following oesophagectomy for oesophageal cancer – are we too radical? *Ann Med Surg* 2016;6:30–5.
- 14 Benjamin G, Ashfaq A, Chang YH, et al. Diaphragmatic hernia post-minimally invasive esophagectomy: a discussion and review of literature. *Hernia* 2015;19:635–43.
- 15 Markogiannakis H, Theodorou D, Tzertzemelis D, et al. Fecopneumothorax: a rare complication of esophagectomy. *Ann Thorac Surg* 2007;84:651–2.
- 16 Sran H, Shaikh I, Coxon J, et al. Tension faecopneumothorax: a rare presentation of colonic diverticular perforation. *J Surg Case Rep* 2011;2011:5.
- 17 Kim KW, Lee JJ, Kim JS, et al. Fecopneumothorax: a rare case of delayed colon diaphragmatic herniation following esophagectomy. *Indian J Surg* 2015;77:117–9.
- 18 Hanna WC, Ferri LE, Fata P, et al. The current status of traumatic diaphragmatic injury: lessons learned from 105 patients over 13 years. *Ann Thorac Surg* 2008;85:1044–8.
- 19 Schildhouse R, Lai A, Barsuk JH, et al. Safe and effective bedside thoracentesis: a review of the evidence for practicing clinicians. *J Hosp Med* 2017;12:266–76.

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