A spontaneous adult right-sided Bochdalek hernia containing perforated colon

Alistair AP Slesser[1] • Hannah Ribbans[2] • Dominic Blunt[3] • Rex Stanbridge[4] • Gordon N Buchanan[5]

[1]Department of General Surgery, Watford General Hospital, Watford, UK
[2]Imperial College London, UK
[3]Department of Radiology, Charing Cross Hospital, London, UK
[4]Department of Cardiothoracic Surgery, Hammersmith Hospital, London, UK
[5]Department of Colorectal Surgery, Charing Cross Hospital, London, UK
Correspondence to: Alistair AP Slesser. Email: alistairslesser@hotmail.com

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Bochdalek hernias containing perforated colon are rare and require colorectal and thoracic surgical expertise to ensure a good outcome.

Case report

A 37-year-old woman was admitted to a district general hospital with a five-day history of shortness of breath, vomiting and right upper quadrant and right shoulder tip pain. She was passing flatus but had not opened her bowels for five days with no relevant previous history, except a previous uneventful pregnancy by vaginal delivery three years previously. On examination she was tachycardic (110 bpm), normotensive and had saturations of 95% on 4 L of oxygen. Abdominal examination was unremarkable. Laboratory tests revealed: Hb 12.0 g/dl, WCC 18600/mm^3, CRP 453 mg/L, Bilirubin 46 μmol/L. An arterial blood gas demonstrated: pH 7.452, p02 10.4, Lactate 1.0 and B.E −3.7. No clear clinical diagnosis was made. Computed tomography (CT) performed two days later showed herniation of the right colon through a posterior defect in the right hemi-diaphragm with free gas in the thorax and abdomen suggesting perforation (Figures 1 and 2).

The patient was transferred the same evening to our teaching hospital surgical unit for specialist care with a temperature of 37.5˚C, tachycardia 125 bpm, respiratory rate of 30/min and saturations of 98% on 35% of oxygen. Respiratory examination revealed absent breath sounds in her right chest. Immediate combined thoracotomy via a right thoracotomy incision and midline laparotomy revealed significant faecal contamination of the chest, a 4-cm Bochdalek hernia with a necrotic perforated right colon in the chest. No sac was found and the colon was reduced. The lung was debrided and decorticated. The necrotic bowel was resected and an ileocolic side to side primary stapled anastomosis was performed. The chest and abdomen were thoroughly lavaged with over 20 L of saline. A primary suture repair of the diaphragmatic defect was performed, both per abdomen and reinforced from the thorax, and chest drains were placed. After two days in intensive care, the patient made an uneventful recovery. At six-month follow-up, the patient was well and with no evidence of hernia recurrence on surveillance CT.

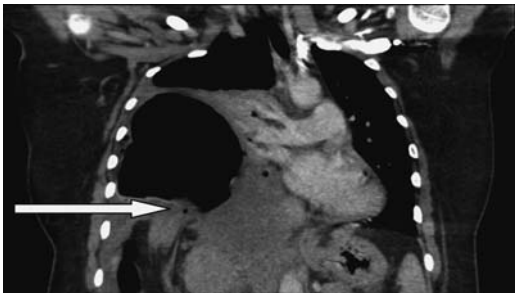
Discussion

The clinical presentation of Bochdalek hernias is rare in adulthood, with the majority of these hernias being left-sided.[1,2] Right-sided Bochdalek hernia presenting in adulthood is extremely rare with fewer than 20 cases reported.[3] A literature search was undertaken using the term 'Bochdalek hernia' in combination with either 'emergency' or 'colon'. A total of 11 articles describing adult right-sided Bochdalek hernia containing colon requiring surgery were found. The 12 cases, including the present case, are discussed in Table 1.[1,4–13]

Diaphragmatic hernias were recognized as far back as the late 17th century.[3] Vincent Alexander

Reviewer
Maryam Alfa-Wali

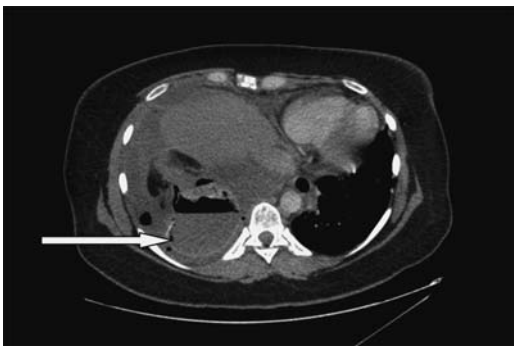
Figure 1
Coronal preoperative chest CT demonstrating herniation of colon into chest with free air (arrow)



Bochdalek in 1848, first described the non-fusion of the posterol-lateral foramina of the diaphragm.[14,15] The Bochdalek hernia is usually congenital, arising due to failed closure of the pleuroperitoneal ducts during the fourth and 12th weeks of gestation with an incidence of 1 in 2200–12,500 births.[16,17] They can, however, be acquired in adulthood due to a 're-opening' of the coronary ligaments.[18] Bochdalek hernias are the commonest form of diaphragmatic hernia and are believed to account for 95% of all cases.[19] They tend to be unilateral but can be bilateral in 14% of cases.[18]

As aforementioned, the vast majority of Bochdalek hernias are diagnosed acutely in neonates and infants with associated respiratory symptoms.[18] They are rarely diagnosed in adults as they tend to remain asymptomatic and are usually incidental findings.[18–20] It is therefore difficult to ascertain the true prevalence of adult Bochdalek hernias. A large retrospective study of over 13,000 abdominal CTs demonstrated a prevalence of 0.17%.[18] However, other imaging studies using multidetector row computed tomography (MDCT) have found higher adult prevalences ranging from 6–12%.[19] Symptomatic adult Bochdalek hernias are usually left-sided possibly due to the caudate lobe of the liver compressing the right pleuroperitoneal canal and thus preventing right-sided herniation.[4,20] This is also probably the case due to the right hemi-diaphragm being fully formed embryologically before the left.[4,16] Interestingly, incidental Bochdalek hernias appear to be mainly right-sided (68%).[18] In addition, Temizöz *et al.* suggest that women have a higher prevalence of asymptomatic right-sided hernia compared to men.[19] Our review seems to confirm this with the majority of patients being women (Table 1). The hernia defect can vary in size from less than 1 cm to a complete lack of a hemi-diaphragm.[16] In approximately 20% there is a hernia sac whereas in the majority of cases, as was seen in our case, there is a direct communication between the chest and the abdomen.[16]

Figure 2
Axial preoperative abdomen/pelvis CT demonstrating postero-lateral right-sided diaphragmatic defect (arrow)



Right-sided symptomatic Bochdalek hernias containing colon are rare; only 12 cases have so far been reported in the literature (Table 1). In nine of the cases, the mechanism was spontaneous, two were postoperative and one was following trauma. To date there are only three reports that describe a left-sided hernia containing perforated colon.[2,20,21] In this review, there was only one other report of a right-sided Bochdalek hernia containing perforated colon found in a patient post hysterectomy.[5] This occurred in the early postoperative phase and it is unclear whether this was related to the patient's surgery. Thus, the present case is unique as it is the only case to present with a spontaneously perforated colon in a right-sided Bochdalek hernia.

It is thought that factors increasing intra-abdominal pressure such as pregnancy, child birth, trauma, coughing, sneezing and large meals can increase the risk of herniation and strangulation of Bochdalek hernia in adults.[4,19] As in the present case, it is recognized that parturition increases the risk of herniation and strangulation.[4] Interestingly, right-sided Bochdalek hernias are rarely symptomatic as the defect is usually larger than left-sided defects.[22] Symptoms are dependent on the defect size and

Table 1
Cases of right-sided Bochdalek hernias containing colon

Referene	*Age*	*Sex*	*Intraoperative findings*	*Operative approach*	*Mechanism*	*Colon*
Present study	37	F	Perforated right colon with faecothorax	Thoraco-abdominal	Spontaneous	Resection, primary anastomosis
Fraser *et al.*[6]	75	F	Colon, small bowel, and right kidney	Laparoscopic/ thorascopic	Spontaneous	Reduced
Laaksonen *et al.*[1]	38	F	Liver and colon	Laparoscopic/ thoracotomy	Spontaneous	Reduced
Kavanagh *et al.*[7]	76	M	Strangulated transverse colon	Abdominal	Spontaneous	Resection, primary anastomosis
Terzi *et al.*[8]	70	F	Colon	Laparoscopic/ thorascopic	Spontaneous	Reduced
Rout *et al.*[9]	35	F	Colon and appendicitis	Abdominal	Spontaneous	Reduced
Court *et al.*[5]	40	F	Perforated colon	Abdominal	Postoperative	Resection, primary anastomosis
Rimpilainen *et al.* [10]	22	F	Liver, gallbladder, right colon, ileum, and right ovary	Thoraco-abdominal	Trauma	Reduced
Kanazawa *et al.*[4]	63	F	Strangulated transverse colon, kidney	Thoraco-abdominal	Spontaneous	Reduced
Zenda *et al.* [11]	69	M	Ileum, colon, gallbladder	Abdominal	Spontaneous	Reduced
Sinha *et al.*[12]	70	F	Stangulated colon with colopleural fistula	Abdominal	Spontaneous/ iatrogenic	Colostomy
Gimovsky *et al.*[13]	20	F	Incarcerated colon	Abdominal	Postoperative	Colostomy

hernia contents and can be pulmonary, gastrointestinal or a combination.[16] Symptoms in adults are predominantly chronic, vague and intermittent in nature but can present acutely with constant and specific symptoms.[4,16] Recurrent abdominal pain, back pain, post pandrial fullness, abdominal distension, a change in bowel habit, nausea and vomiting are gastrointestinal symptoms that can be encountered.[4,16] Respiratory symptoms albeit less common in adults can include shortness of breath, dyspnoea, chest pain and referred shoulder pain.[4,16] Most cases become surgical emergencies when an abdominal organ becomes strangulated.[2]

Auscultation of bowel sounds on chest examination should raise suspicion of a diaphragmatic hernia.[2] Less specific findings include abdominal tenderness, reduced or absent breath sounds and tracheal deviation.[2] Different imaging modalities such as plain chest/abdominal radiographs, ultrasound, magnetic resonance imaging and CT can be used for diagnosis.[19] Chest radiographs have a sensitivity of greater than 70% usually demonstrating air fluid levels, it must be noted that a normal chest X-ray does not exclude a Bochdalek hernia.[2,3,16] The gold standard imaging modality in an emergency or elective case is a double-contrast axial CT.[2,15] MDCTs are used in preference to helical CTs as they have an increased sensitivity.[19] This present case highlights the importance of early CT scanning in reaching an early preoperative diagnosis, particularly since a delay in diagnosis can result in significant morbidity and mortality.[3]

As demonstrated in the reported cases, an open abdominal approach is preferred in an emergency setting for right-sided Bochdalek hernias as the hernia can be better visualized by mobilization of the right lobe of the liver (Table 1).[4,16] In difficult cases such as the present case, a combined thoracic and abdominal approach may have to be undertaken.[16] Minimally invasive techniques may result in reduced morbidity and might also improve the ease of hernia reduction, haemostasis and adhesiolysis.[2,16] Usually, a minimally invasive approach is used in an elective setting, though rarely used for right-sided Bochdalek hernias.[6,16,23] There have, however, been cases whereby minimally invasive techniques have been used for emergency left-sided Bochdalek hernias.[2] Small defects are easier to repair whereas larger defects may involve reduction of intra-abdominal contents.[15] The defect is usually repaired along a 'transverse axis' and may require the use of a polytetrafluoroethylene mesh in the absence of contamination.[2,15,20] Our review demonstrates that although simple hernia reduction is common, colonic resection may have to be undertaken and can result in the formation of a colostomy (Table 1). If a large hernia has been reduced, it is crucial to monitor postoperatively for an abdominal compartment syndrome.[16] Incidental large Bochdalek hernias should be repaired due to the potential associated risks and risk of fatality.[4] The recurrence rate following surgical repair is generally considered low.[24]

Conclusion

Adulthood right-sided Bochdalek hernias are extremely rare. However, this diagnosis should be suspected in patients who have been exposed to factors that have increased their intra-abdominal pressure. Signs and symptoms can be non-specific and the present case highlights the importance of acquiring a CT scan at the earliest opportunity particularly when dealing with an unexplained acute abdomen. This is crucial in the management of these patients, as a delay in diagnosis can increase the risk of mortality. The present case had extensive faecal contamination but did not require a stoma and has not suffered from any chest sequelae or a hernia recurrence. We feel that this good outcome, clearly demonstrates the importance of such patients being operated on in institutions where there is access to both specialist colorectal and thoracic surgeons.

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