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CASE REPORT

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Bochdalek hernia masquerading as pleural effusion in a young adult

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Abstract

Bochdalek hernia (BH) is a congenital diaphragmatic hernia which rarely presents in adulthood. We report on a 25-year-old man admitted with 3-year recurrent and self-limiting abdominal pain and vomiting. Chest radiograph showed left pleural opacity which shifted position in the decubitus film suggesting pleural effusion. Before attempting drainage, point-of-care (POC) thoracic ultrasound (TUS) demonstrated features of intrathoracic gastrointestinal content above the left hemi-diaphragm. Computed tomography of the thorax confirmed a large left BH. Acute recurrent symptoms 3 months post-discharge was associated with BH enlargement and early bowel ischemia, which was successfully managed by emergency surgery. This case illustrates the importance of pre-procedural imaging with POC-TUS before pleural procedures in all cases of suspected or apparent pleural effusion, and of prompt surgical treatment of symptomatic BH despite clinical stabilization.

KEYWORDS

Bochdalek hernia, CXR, pleural effusion, thoracic ultrasound

INTRODUCTION

Bochdalek hernia (BH), usually presenting neonatally, is a congenital diaphragmatic hernia caused by incomplete fusion of the posterolateral diaphragmatic foramina. Approximately 150 adult cases have been reported in the literature, mostly presenting with non-specific symptoms and radiological manifestations. A high index of suspicion is required to avoid misdiagnosis and potentially disastrous outcome(s).[1] We report a case which masqueraded as pleural effusion on chest radiograph (CXR), and highlight the importance of pre-procedural confirmatory imaging and prompt surgical treatment.

CASE REPORT

A 25-year-old non-smoking and non-drinking Chinese male presented to the emergency room with left-sided colicky abdominal pain and vomiting undigested food for 2 days, with similar self-limiting symptoms recurrently for 3 years, occasionally accompanied by cough, sputum, breathlessness and chest pain. Physical examination was normal apart from percussion dullness and reduced air entry over the left posterior chest wall. Complete blood count, liver and renal function tests, serum amylase, blood gas and electrocardiogram were normal. Erect postero-anterior (PA) CXR revealed a left hemithorax opacity with "meniscus sign" (Figure 1A) and position-shift in the decubitus film (Figure 1B), suggestive of free-flowing pleural effusion. Before the planned pleural drainage, point-of-care thoracic ultrasound (POC-TUS) using a phased-array transducer at the posterior left hemithorax in the sitting position revealed multi-layered area with hyper-echoic content above an ill-defined left hemi-diaphragm (Figure 1C), and a tubular shadow on directing the transducer cranially (Figure 1D), both suspicious of intestinal structures. Computed tomography (CT) of the thorax confirmed a large left BH with occupation of the left pleural cavity by intraperitoneal fat, a large part of the stomach and parts of the transverse and descending colon, and compressive left lower lobe atelectasis with rightward mediastinal displacement (Figure 2A, B). Surgical consultation recommended conservative management as symptoms had spontaneously

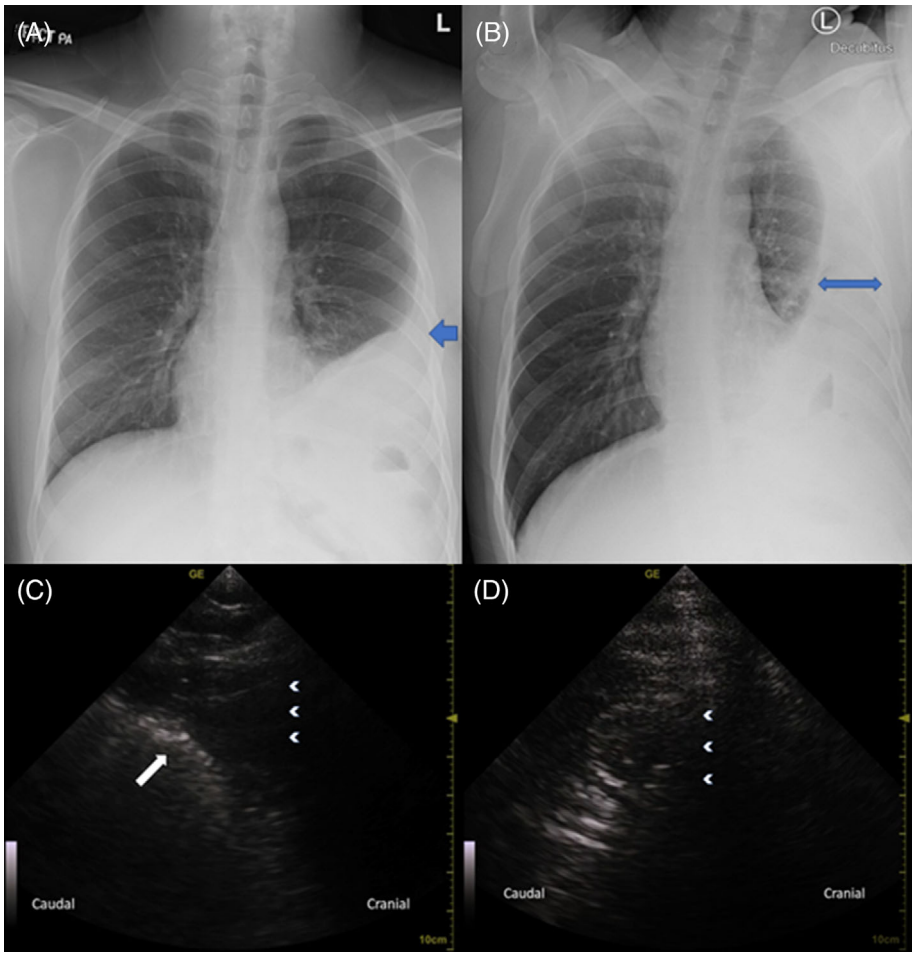

FIGURE 1 CXR erect film showed left hemithorax opacity with "meniscus sign" (arrow) (A) and left decubitus film showed "shifting opacity" (double arrow) (B), suggestive of a free-flowing left pleural effusion. TUS at posterior left hemithorax demonstrated multi-layered area with hyper-echoic content (arrowheads) above an ill-defined left hemi-diaphragm (arrow) (C) and a tubular structure cranially (arrowheads) (D), suspicious of intestinal structures

resolved. The patient was discharged with early cardiothoracic surgery referral.

On admission to another hospital 3 months later with acute recurrent symptoms, the patient was afebrile, had tachycardia despite normal blood pressure, and distended abdomen with left-sided tenderness. White blood cell count was 23.7 $\times 10^9$ cells/L, blood pH was 7.28 and HCO3 19 mmol/L. Urgent contrast CT thorax and abdomen revealed increased intra-abdominal organ herniation in the left pleural cavity, and bowel kinking with minimally diminished enhancement of a bowel wall segment close to the superior mesenteric artery and vein at the hernia defect (Figure 2C, D). Emergency surgery was performed based on suspected bowel strangulation. Intra-operative findings included a 6-cm hernia defect at the postero-lateral aspect of the left hemi-diaphragm, and partial obstruction of small bowel with dusky mesenteric changes. The BH was completely reduced followed by mesh repair of the diaphragmatic defect. Postoperatively, the patient had fully-expanded left lung and uneventful recovery. He remained well on follow-up a year later.

DISCUSSION

Our case illustrates that CXR may be misleading in BH, with CXR findings of "meniscus sign" and "shifting opacity" indicating free-flowing pleural effusion (Figure 1A, B). Pleural drainage without prior confirmatory imaging in a similar case did not result in organ injury because the herniated content was mainly peritoneal fat.[2] BH had also been misdiagnosed as tension pneumothorax by CXR alone, resulting in gastric perforation following chest-tube drainage.[1]

Pre-procedural TUS imaging is essential to differentiate pleural effusions from BH. Only three case reports had described ultrasound findings in adult BH. Two abdominal ultrasounds (AUS) showed disfigured spleen with fragmented echogenic line of hemi-diaphragm,[3] and a sonographic "ring-down" artefact behind herniated fat.[4] Only one report of TUS using a linear transducer described bowel loops with visible peristalsis in the posterior left hemithorax with normal lung sliding immediately cranially, but without visualizing the hemi-diaphragm.[5] In contrast, our POC-TUS could visualize normal gut above an ill-defined

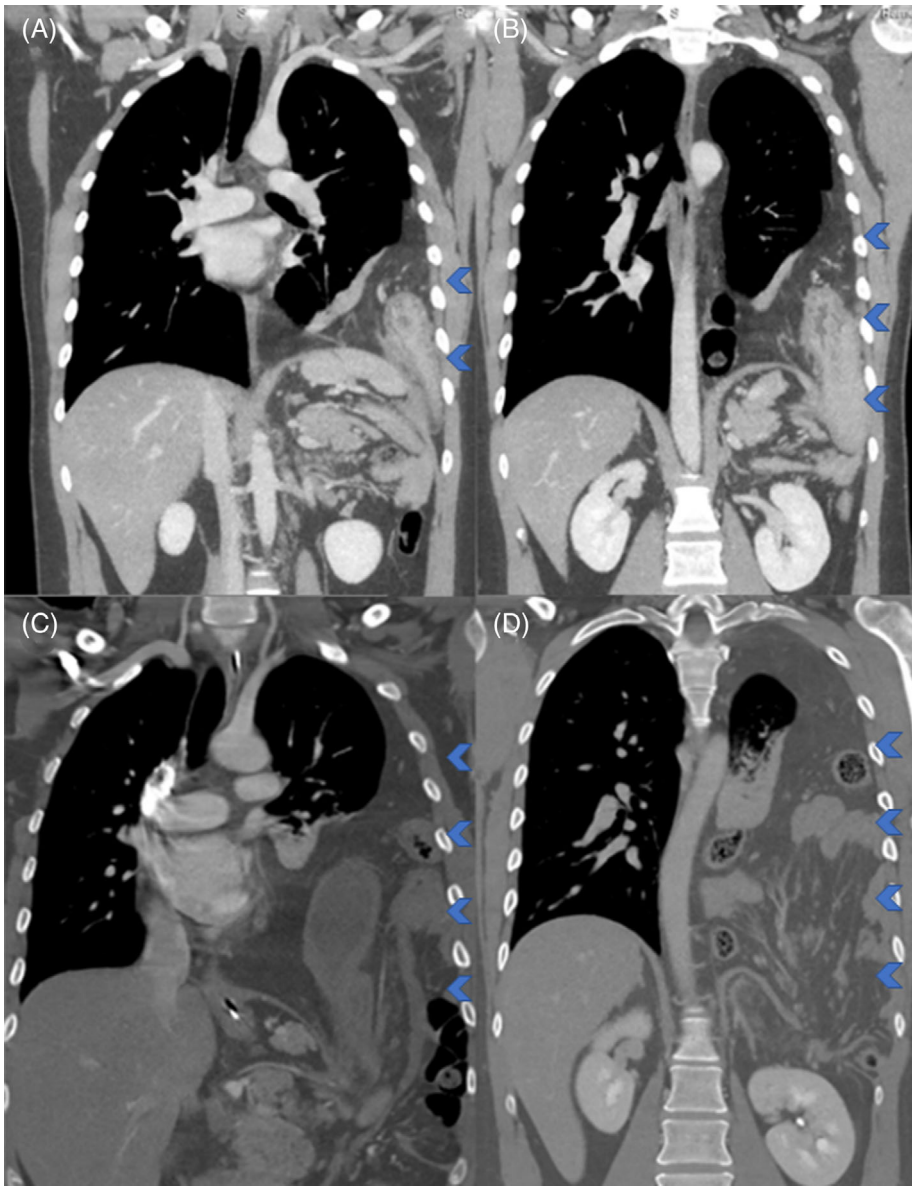

FIGURE 2 CT thorax showed a large left Bochdalek hernia (arrowheads) (A, B), which enlarged on re-admission 3 months later (arrowheads) (C, D)

left hemi-diaphragm (Figure 1C, D). To differentiate pleural effusion from BH, POC-TUS in the sitting position is preferred over AUS, and a low-frequency curvilinear or phased-array probe could provide the penetration required to identify the hemi-diaphragm which may be discontinuous or ill-defined, followed by cranial orientation to evaluate intra-pleural contents. Features suggesting intestinal contents and/or parenchymatous abdominal organs above the hemi-diaphragm would raise the suspicion of diaphragmatic hernia. CT thorax should be performed to confirm the diagnosis. In contrast, pleural effusion is signified by homogeneous hypoechoic fluid collection beneath the chest wall with/without hyperechoic lung atelectasis deep to the collection. Loculations, septations with heterogeneous fluid collections and interspersed hyperechoic tissue strands may be seen in complex effusions.[6]

The following management options may be considered in BH according to the mode of presentation: Asymptomatic (incidental finding): Regular monitoring, shared decision-making on elective surgical repair for large hernias and if symptoms develop. Symptomatic but uncomplicated: Early elective minimal access surgery via thoracoscopic or laparoscopic route. Complicated: Emergency minimal access or open surgery for organ salvage and definitive BH treatment.

This case highlights two critically important points in the clinical management of apparent/suspected pleural effusion. As recommended by the British Thoracic Society (BTS) guidelines,[7] routine confirmatory imaging, in particular POC-TUS, is essential before attempts at pleural drainage. Secondly, surgery should be considered early in symptomatic BH to prevent complications.

In conclusion, we reported a case of large left Bochdalek hernia masquerading as pleural effusion radiologically. A high index of suspicion and confirmatory imaging contributed to the correct diagnosis and obviated potentially disastrous outcomes, and surgical treatment should not be delayed in symptomatic cases of BH.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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