

CASE REPORT

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Massive pleural effusion post-emergency cesarean in undiagnosed congenital diaphragmatic hernia

Nurul Liana Roslan, Lim Nikkie, Mahathar Abd. Wahab

ABSTRACT

Introduction: Bochdalek's hernia is a congenital diaphragmatic hernia (CDH) resulting from inadequate closure of the membrane over the left side of the hemidiaphragm. Patients will start to develop symptoms upon displacement of abdominal contents into the thoracic cavity. This case report highlights the radiological modality findings that can be used in diagnosing a late-presentation CDH in adults.

Case Report: A 35-year-old postpartum woman developed shortness of breath, left-sided chest pain, episodes of vomiting and epigastric tenderness within 24 hours post-emergency cesarean section. An initial diagnosis of left tension hydrothorax was made based on the clinical and radiological findings. Left diagnostic and therapeutic thoracocentesis using landmark technique was performed and brownish fluid was obtained upon aspiration. Post-procedural chest X-ray (CXR) reported cystic-like features over the left hemithorax. Therefore, computed tomography (CT) thorax was proceeded and confirmed the left diaphragmatic hernia with a mediastinal shift to the right. However, the patient's condition worsened and required emergency open thoracotomy with left diaphragmatic hernia repair. Intraoperative findings showed left Bochdalek's hernia

complicated by multiple visceral organ perforations secondary to previous thoracocentesis. The patient was discharged well after a few weeks of hospital stays.

Conclusion: The diagnosis of late-presentation Bochdalek's diaphragmatic hernia remained challenging and can be easily missed. Therefore, a comprehensive evaluation is required prior to performing an invasive diagnostic procedure. Chest X-ray and ultrasound are useful diagnostic aid in the absence of CT thorax and magnetic resonance imaging (MRI) modalities.

Keywords: Bochdalek's hernia, CDH, Diaphragmatic hernia, Pleural effusion

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Nurul Liana Roslan¹, Lim Nikkie², Mahathar Abd. Wahab³

Affiliations: ¹Emergency Physician, Emergency and Trauma Department, Hospital Kuala Lumpur, Kuala Lumpur, Malaysia; ²Medical Officer, Emergency and Trauma Department, Hospital Kuala Lumpur, Kuala Lumpur, Malaysia; ³Consultant Emergency Physician, Emergency and Trauma Department, Hospital Kuala Lumpur, Kuala Lumpur, Malaysia.

Corresponding Author: Nurul Liana Roslan, Emergency Physician, Emergency and Trauma Department, Hospital Kuala Lumpur, Kuala Lumpur, Malaysia. Email: drnurullianaroslan@gmail.com

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INTRODUCTION

It is undeniably rare and difficult to diagnose congenital diaphragmatic hernia in adults [1]. The reported incidence in adult population was estimated to be approximately 0.17% [2]. The patient frequently presents with subtle symptoms upon displacement of abdominal contents into the thoracic cavity, which requires thoughtful analysis and makes the diagnosis more challenging [1, 2]. This case report emphasizes the findings from radiological modalities that can be used to diagnose a late-presentation CDH in adults.

CASE REPORT

A 35-year-old woman underwent emergency cesarean operation following fetal distress after attempts of a normal delivery failed at her 38 weeks of pregnancy. Intraoperatively, fundal pressure was performed to facilitate passage of the fetus, and panniculus retraction had to be done as the patient was pre-morbidly obese; nevertheless, the patient remained stable throughout the operation.

Few hours after the operation, the patient developed episodes bouts of vomiting with epigastric tenderness, shortness of breath, and left-sided chest pain. A right-sided tracheal deviation and diminished breath sounds over the left side of the chest were detected during a physical examination. Vital signs included a body temperature of 37.3°C, a blood pressure of 100/60 mmHg, a heart rate of 140 beat per minute, an oxygen saturation of 85% on oxygen flow of 15 liters per minute, and a respiratory rate of 25 per minute. Despite the initial resuscitation, her respiratory distress worsened. Her full blood count showed leukocytosis with a left shift, while arterial blood gas was suggestive of hypoxic respiratory failure characterized by a lower than 60 mmHg arterial oxygen tension (PaO₂) without hypercapnia. The urgent CXR showed a "complete white out" left lung field suggestive of massive pleural effusion with a mediastinal shift to the right (Figure 1). As the patient was hemodynamically unstable with an acute respiratory distress syndrome, diagnosis of tension left hydrothorax was made. A left thoracocentesis was then performed as part of emergency decompression measures using the landmark technique instead of real-time ultrasound guidance due to a lack of equipment availability. Approximately 1.6 liters of brownish fluid were aspirated. However, there was no clinical improvement.

The post-procedural CXR is necessary to evaluate the improvement of pleural effusion by assessing the reduction of the air fluid level and the lung expansion. However, the first post-procedural CXR was incorrectly performed in the supine position (Figure 2), therefore, the semi-erected and lateral decubitus CXRs were done. The repeated CXRs (Figures 3 and 4) showed an improvement of air fluid or hydro-aerial level in the left hemithorax with persistent deviation of mediastinal organs to the right side. However, upon reviewing of the initial CXR post-procedure, a cystic-like structure was found over the left hemithorax (Figure 2) which called for immediate further investigation to rule out the likelihood of a visceral organ herniation in the diaphragm. An urgent CT thorax was ordered and diagnosis of the left diaphragmatic hernia was established. The CT revealed that the normal outline of left hemidiaphragm was not clearly visible with collapsed left lung including multiple other organs (stomach, duodenum, small bowel loops, pancreas, spleen, left liver lobe) within the left hemithorax. The right main bronchus appeared to have mild narrowing due to the compression of patient's distended stomach

and the great vessels were displaced to right hemithorax as well (Figure 5).

The patient was then intubated for impending respiratory failure, given volume resuscitation, and started on inotropic support for refractory shock. Empirical antibiotics were given. Emergency open thoracotomy and left diaphragmatic hernia repair proceeded in the operation theater. Intraoperative findings noted a Bochdalek's hernia with a size of 9×7 cm. The left thorax was filled with a spleen, the whole stomach, small intestine and a part of the pancreas. There were also approximately 1 liter of gastric content. Repair of stomach perforation was done. She was admitted to intensive care unit post-operatively and was discharged well after 54 days of hospitalization.

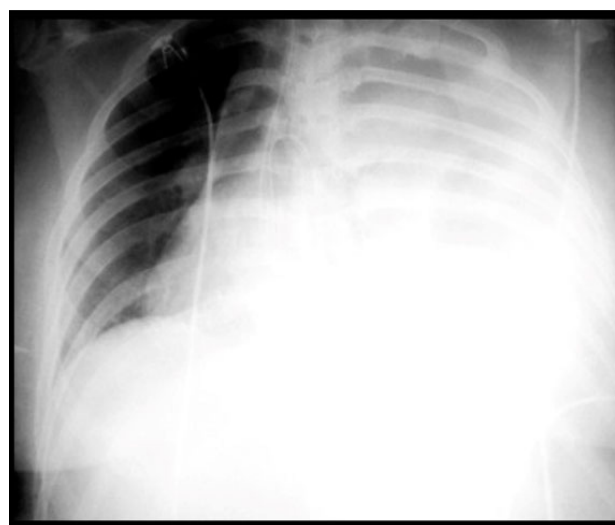


Figure 1: The first chest X-ray showed "complete white out" left lung field. Notice the vague bowel shadows within the left thoracic cavity.

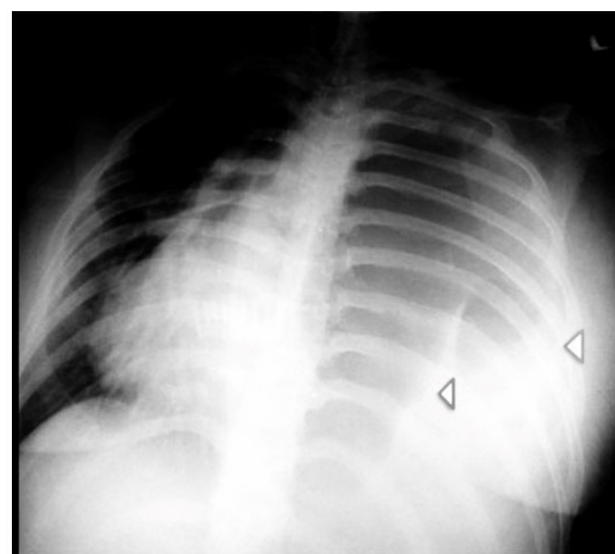


Figure 2: The chest X-ray after left thoracocentesis that was initially done in the supine position. A suspicious cystic-like lesion (arrow heads) at the left hemithorax suggestive of bowel loops, with persistent shift of the mediastinum to the right.

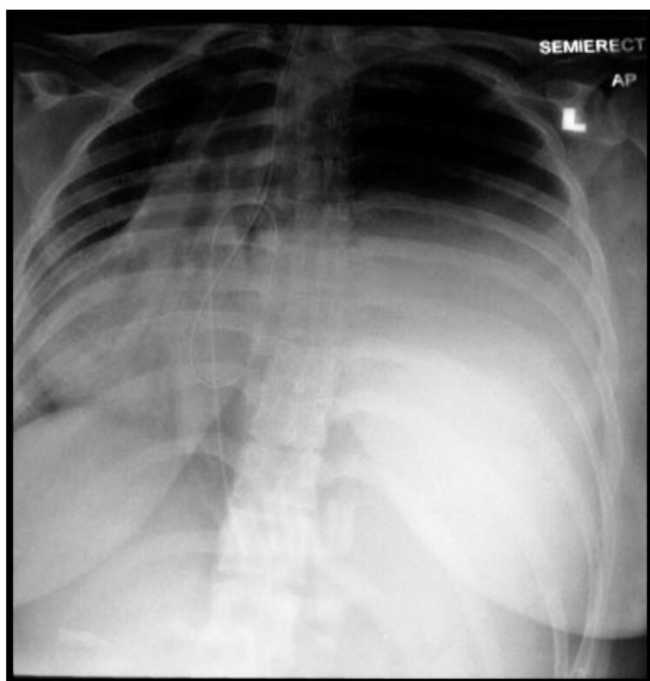


Figure 3: The chest X-ray that was repeated in semi-erected position. Improvement of the air fluid or hydro-aerial level can be seen in left hemithorax with persistent deviation of mediastinal organs to right side.

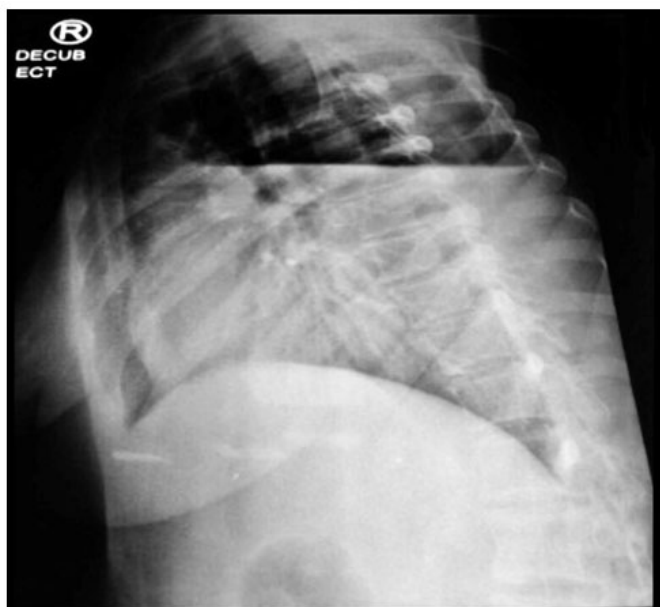


Figure 4: The lateral view of the chest after left thoracocentesis. Air-fluid level or hydro-aerial level can be seen in the hemithorax. There is no focal elevation of the diaphragm seen in this view.

DISCUSSION

Bochdalek's hernia, named after a Czech Anatomist in the year of 1848, is a congenital diaphragmatic abnormality resulting from inadequate closure of the posterolateral



Figure 5: A CT thorax shows the air fluid level and herniation of the visceral organ within the left hemithorax with a discontinuity in the left hemidiaphragm.

pleuroperitoneal membrane in the lumbocostal triangle [1, 2]. The defects frequently presence over the left side of the diaphragms, which the abdominal contents, including stomach, bowel loops, liver, spleen or fat tissues, may be displaced into the thoracic cavity [3]. In comparison to the left side, herniation at the right side is rare—probably attributed to the protection provided by the liver [2, 3].

The late-presentation CDH usually presented with sudden onset of symptoms (epigastric discomfort, nausea, vomiting, shortness of breath, and chest pain) due to mechanical or pressure changes in the thoracoabdominal cavities [2, 4, 5]. A sudden rise in intra-abdominal pressure pushes visceral organs toward the weak hiatus and contributes to the development of a diaphragmatic hernia. Therefore, history of heavy physical exertion, direct trauma over the torso or any abdominal manipulations before symptoms onset should raise suspicion [4–6].

This condition is difficult to diagnose especially in adult population [3]. A large study done by Thomas and Kapurin found that 38% of patients with CDH presented as acute surgical emergencies were initially diagnosed incorrectly [6]. It may be misdiagnosed as pleural effusion, pneumonia, tension pneumothorax, lung cysts, or atelectasis [3]. An incorrect diagnosis will cause undesirable delays in treatment and unwanted complications [7] which happened in our patient.

The movement of fluid into the pleural space is a normal physiological response of pregnant women owing to the pressure changes in the systemic circulation (an increase in blood volume and hydrostatic pressure, and a drop in colloid osmotic pressure). In addition, repeated Valsalva maneuvers, increased intrathoracic pressure, and decreased lymphatic drainage in the pleural space, driven by the elevation in systemic venous pressure during labor will further worsen the pleural effusion [8].

Gourgoulianis et al. reported that 23% of postpartum women within 24 hours of normal delivery had pleural effusion [8]. Nonetheless, they did not find any significant correlation with age, weight-gain during pregnancy,

duration of labor, use of intravenous fluid, or oxytocin administration [8]. Interestingly, a prospective pilot study on the disease prevalence found that pleural effusion is not rare during pregnancy, instead, patients mostly do not exert any symptoms [9]. A case study by Caliskan et al. observed the same presentation with our case, in which both of the patients had an uneventful vaginal delivery of their first child [5]. However, upon detecting an unexplained air-fluid levels over the left hemithorax on CXR, they were managed to perform a barium enema orientated CT to diagnose diaphragmatic herniation, in contrast to our case, which warranted an emergency decompression due to suspected tension hydrothorax [5]. Therefore, it is paramountly important that incidental findings of pleural effusion during the postpartum period necessitate further investigations prior to any invasive intervention.

Recognizing the limitation of CXR to identify diaphragmatic injury or rupture is poor, with 12–66% probability of missed diaphragmatic rupture, this radio-imaging modality remains the initial investigation in assisting physicians to suspect diaphragmatic herniation in both asymptomatic and symptomatic patients [10]. Up to 60% of diaphragmatic injuries can be detected and diagnosed with first radiography [10]. Diaphragmatic hernia should be suspected if there is an elevation of the hemidiaphragm with a large opaque shadow within the lung field, obscuring the distinct diaphragmatic outline, or if there is an incomplete shadow over the diaphragm with a contralateral shift of the mediastinum [2, 6, 11]. The CXR's lateral decubitus projection will also be useful in locating the herniation [10]. A pathognomonic symptom of diaphragmatic hernia is the visceral gas distribution (with or without hydro-aerial levels) within the lung field, as well as focal constriction of the viscus at the site of tear (collar sign) [10, 12]. Furthermore, the absence of nasogastric tube tip visualization at the stomach fundus could provide valuable information about any abnormalities of the diaphragm [5, 10, 11]. In addition to the aforementioned CXR features, penetration of the CXR film plays a role in interpretation. In our center, the radiological images will be adjusted using the windowing tools to facilitate the visibility of anatomical structures, before printed into film by radiographers. Due to the limited access to the digital viewing systems, thus it was very challenging to find any abnormalities on an under-penetrated CXR film.

Bedside lung ultrasonography (BLUS) can be used to confirm the diagnosis of diaphragmatic injury as pleural effusion, pulmonary contusion, atelectasis, and phrenic nerve palsy can mimic or mask diaphragmatic injury on CXR [10]. The diaphragm can be identified in longitudinal axis, from the anteromedial to the lateral chest wall, using a curvilinear transducer (5–8 MHz) [2]. Diaphragmatic hernias should be suspected when there is a discontinuity of the diaphragmatic contour, focal elevation of the diaphragm with a wide-angled waist [2], presence of peristaltic bowel movement within the

hemithorax [13], and absence of visceral organ such as spleen and kidney below the diaphragm [14]. However, BLUS has limitations in diagnosing diaphragmatic hernia in the presence of gas-filled intestinal loops, aerated lungs, and acoustic shadowing from the ribs [12].

When there is clinical or radiological suspicion of diaphragmatic herniation, the CT scan has been accepted as the modality of choice. However, it has only been demonstrated that CT thorax has variable sensitivity (61–87%) and specificity (72–100%) to detect diaphragmatic injury, with a better sensitivity in detecting left-sided abnormalities attributed by soft tissue-fat contrast [13, 15]. There are a few features that have been mentioned in the literature, but they are mostly associated with traumatic diaphragmatic injuries [15–18]. For example, “diaphragmatic discontinuity,” which is the most prevalent sign (95.7%) seen with CT imaging and has a sensitivity and specificity of 36–82.7% and 88–95%, respectively, and “dangling diaphragm,” which has a sensitivity of 54% and specificity of 98.3%. Both of these are direct signs of diaphragmatic injuries. A few of the indirect signs that have been described are “thickening of the diaphragm” (69.6%), “organ herniation within the hemithorax” (65.2%), “dependent viscera” (59.5%), and “collar sign,” [15–18]. Magnetic resonance imaging (MRI) can provide better soft tissue resolution, it may be necessary in a stable patient with an equivocal CT result [16]. However, both CT and MRI studies required the patient to be stable.

CONCLUSION

Diagnosing late-presentation Bochdalek's diaphragmatic hernia remains challenging as it can be easily missed as massive pleural effusion. Therefore, a comprehensive evaluation is required prior to performing an invasive diagnostic procedure. Imaging such as chest X-ray and bedside ultrasound play an important role during initial evaluation to assess the presence of any associated abnormality in the absence of CT thorax and MRI modalities, and guiding a safe treatment plan.

REFERENCES

1. Brown SR, Horton JD, Trivette E, Hofmann LJ, Johnson JM. Bochdalek hernia in the adult: Demographics, presentation, and surgical management. *Hernia* 2011;15(1):23–30.
2. Karmazyn B, Shold AJ, Delaney LR, et al. Ultrasound evaluation of right diaphragmatic eventration and hernia. *Pediatr Radiol* 2019;49(8):1010–7.
3. Bianchi E, Mancini P, De Vito S, et al. Congenital asymptomatic diaphragmatic hernias in adults: A case series. *J Med Case Rep* 2013;7:125.
4. Nitecki S, Bar-Maor JA. Late presentation of Bochdalek hernia: Our experience and review of the literature. *Isr J Med Sci* 1992;28(10):711–4.

5. Caliskan AC, Bodur S, Aytan H, Gun I, Acu B. An unusual cause of maternal respiratory distress in postoperative-postpartum period: Diaphragmatic hernia. *Eur J Gen Med* 2012;9(2):142–5.
6. Thomas S, Kapur B. Adult Bochdalek hernia—Clinical features, management and results of treatment. *Jpn J Surg* 1991;21(1):114–9.
7. Sridhar AV, Nichani S. Late presenting congenital diaphragmatic hernia. *Emerg Med J* 2004;21(2):261–2.
8. Gourgoulianis KI, Karantanas AH, Diminikou G, Molyvdas PA. Benign postpartum pleural effusion. *Eur Respir J* 1995;8(10):1748–50.
9. Dikensoy E, Balat Ö, Diknsoy Ö. The prevalence of pleural effusion in pregnant women: A pilot study. *Turkish Journal of Medical Sciences* 2006;36(5):291–3.
10. Iochum S, Ludig T, Walter F, Sebbag H, Grosdidier G, Blum AG. Imaging of diaphragmatic injury: A diagnostic challenge? *Radiographics* 2002;22 Spec No:S103–16.
11. Froman A. The value of the routine chest X-ray film in detecting diaphragmatic hernia; A report of 53 cases. *Dis Chest* 1954;26(4):457–63.
12. Eren S, Ciriş F. Diaphragmatic hernia: Diagnostic approaches with review of the literature. *Eur J Radiol* 2005;54(3):448–59.
13. Bothwell J, Della-Giustina D, Laselle B, Harper H. Ultrasound diagnosis of diaphragmatic rupture. *Crit Ultrasound J* 2011;3(3):153–4.
14. Megremis SD, Segkos NI, Gavridakis GP, et al. Sonographic appearance of a late-diagnosed left Bochdalek hernia in a middle-aged woman: Case report and review of the literature. *J Clin Ultrasound* 2005;33(8):412–7.
15. Panda A, Kumar A, Gamanagatti S, Patil A, Kumar S, Gupta A. Traumatic diaphragmatic injury: A review of CT signs and the difference between blunt and penetrating injury. *Diagn Interv Radiol* 2014;20(2):121–8.
16. Wilbur AC, Gorodetsky A, Hibbeln JF. Imaging findings of adult Bochdalek hernias. *Clin Imaging* 1994;18(3):224–9.
17. Kaur R, Prabhakar A, Kochhar S, Dalal U. Blunt traumatic diaphragmatic hernia: Pictorial review of CT signs. *Indian J Radiol Imaging* 2015;25(3):226–32.
18. Desir A, Ghaye B. CT of blunt diaphragmatic rupture. *Radiographics* 2012;32(2):477–98.

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Author Contributions

Nurul Liana Roslan – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Lim Nikkie – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Mahathar Abd. Wahab – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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